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Running Free: The Impact of Frame Running on the Psychosocial Wellbeing and Quality of Life of Novice and Experienced Athletes with Cerebral Palsy

Hamish Johnson

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Abstract

Cerebral palsy (CP) is the most common physical disability in childhood and occurs in approximately 1 in 500 live births. CP is characterised by disorders of the development of movement and posture leading to abnormal movement patterns and affecting limb movement and muscle tone causing activity limitations. The severity of impairments varies between individuals thus their activity limitations will also vary. Secondary impairments of CP include cognitive, behavioural, and emotional deficits. The difficulties with motor ability combined with the presence of any associated impairments can cause a cycle of exclusion and deterioration contributing to an inactive lifestyle with associated health risks.

Physical benefits of physical activity are well researched for children with disabilities. There is less research available on the psychosocial benefits of physical activity, for children with disabilities, including those with CP. The importance of being physically active is well documented, however barriers to physical activity such as a lack of suitable activities, fear of not enjoying it, fear of failure, or fear of social exclusion are further inhibiting physical activity participation of children with CP. Frame running is an adaptive sport that offers the opportunity for children with CP to be physically active, especially those with more severe CP who are further restricted in their physical activity opportunities.

Given the complex and individual nature of CP it can be difficult to identify which aspects of wellbeing (physical, psychological, or social) are most important to measure as the focus of an intervention, rehabilitation, and therapeutic programme. A two-stage Delphi survey involving parents of children with CP, health care professionals and frame running coaches identified the factors that are deemed most important when evaluating the effectiveness of a physical activity intervention for children with CP. Participants of the Delphi survey identified social inclusion, enjoyment and psychological wellbeing as the most important factors, and these results shaped the selection of psychosocial constructs for the following study.

Frame running (formally known as racerunning) is a growing sport, however there is very limited research exploring frame running, and currently no research examining the effects of the frame running participation on the psychosocial wellbeing of children with CP. Five participants took part in a single-case design which examined the effects of a 12-week frame running training programme on the QoL and psychosocial wellbeing of five novice athletes around Scotland. Participants completed outcome measures in their home environment on four separate occasions, baseline, pre-, mid-, and post-training. They were also asked to rate their enjoyment level of each session to identify if frame running is viewed as an enjoyable sport, which is key for promoting engagement and adherence to the sport.

The results did not show any consistent patterns regarding the impact of frame running on the participants' psychosocial wellbeing. However, visual analysis of the data showed some trends towards improvement in some aspects of psychosocial wellbeing, specifically self-esteem and psychological wellbeing, however it is not possible to attribute these results solely to frame running participation. Furthermore, the mean rating of enjoyment (out of 5) across the 12 sessions was high at 4.7, highlighting that frame running is perceived as an enjoyable opportunity for physical activity for children with all severities of CP. The quantitative results provide an insight into the positive impact frame running participation can have on psychosocial wellbeing of children with CP, but further research is needed to confirm these findings in a larger sample to a control population may provide a clearer picture.

With no consistent pattern of the effect of frame running participation on QoL and psychosocial wellbeing established for novice athletes, a subsequent qualitative study explored the perceived impact of frame running on competitive athletes' QoL and psychosocial wellbeing. Furthermore, this study focused on the same psychosocial constructs as the previous study. Ten frame running athletes of local to international level completed the online survey, and through thematic analysis, the identified themes and subthemes suggested that participation has had, and does have, a positive influence on their perceived QoL and psychosocial wellbeing. The athletes reported new and enhanced life experiences through frame running, and highlighted the importance of frame running socially, with athletes describing how frame running gave them the opportunity to build and develop friendships worldwide, giving them a sense of inclusion, belonging, and enabling their social confidence, thus enhancing social development. Furthermore, athletes perceived frame running participation to have enhanced their psychological development with improved self-esteem, development of self-concept and self-confidence, and psychological wellbeing, which are reported to positively impact QoL and health outcomes.

The results from the current thesis provide new insight into the impact of frame running participation on the QoL and psychosocial wellbeing of both novice and competitive athletes with CP. The combined results suggest results suggests that frame running may have a positive influence on psychological development, through self-esteem and self-concept, as well as impacting on psychological wellbeing and in turn enhancing QoL. The current research shows frame running as an enjoyable and accessible opportunity for individuals with CP to be physically active, which has the potential to contribute to an enhanced QoL and psychosocial wellbeing.

Lay Summary

Cerebral palsy (CP) is the most common physical disability in childhood which affects the movement and posture of the individual. The impairments associated with CP can lead to restrictions in the individual's ability to participate in physical activity and/or exercise. The physical benefits of participation for individuals with disabilities, including CP, are well documented, highlighting the importance of being physically active, however the psychological and social effects of physical activity participation are less well researched. Individuals with disabilities can face various challenges in an attempt to become physically active, whether it is a lack of suitable activities, lack of belief in their own ability to participate, or fear of social exclusion. Frame running (formally known as racerunning) is an adaptive sport that offers the chance to children with CP and related conditions to be physically active. It is one of few opportunities for those with more severe CP to be physically active.

The complex and individual nature of CP can make it difficult for therapeutic, rehabilitation programmes and interventions to have a focus as each individual might have different abilities and/or at different stages of development. The first study in the thesis gathered the views and opinions of parents of children with CP, health care professionals, and frame running coaches, on which factors they viewed as most important to measure as a focus of physical activity interventions for children with CP. Over a 2-stage Delphi survey, showed participants reported social inclusion and enjoyment as the top two most important factors, highlight the importance of these constructs in physical activity. The results from this study shaped the constructs measured in the following study, and with limited research involving frame running, it was important for the current thesis to study the effects of frame running participation on the quality of life and psychosocial wellbeing of individuals with CP.

Five individuals with CP took part in a frame running training programme over a 12-week period, and the effects of participation on their quality of life and psychosocial wellbeing were examined. Additionally, participants were asked to rate their level of enjoyment after each training session to evaluate if frame running is viewed as an enjoyable physical activity, which is a key factor in promoting engagement and adherence in physical activity and sport.

The results of the training programme did not show any clear patterns of how frame running participation may have impacted the psychosocial wellbeing and quality of life of the participants. The main takeaway from the study was the high level of enjoyment reported by each of the participants across the 12 training sessions, highlighting the participants perceived frame running as an enjoyable opportunity for children and young people with CP, of all severities, to be physically active.

With no clear patterns regarding the impact of frame running on quality of life or psychosocial wellbeing, a follow-up web-based survey involving current, competitive athletes explored how they perceived frame running to have impacted their quality of life and psychosocial wellbeing. The use of the web-based survey allowed for the 10 athletes to express their opinions, views, and experiences fully, providing valuable insight to how they felt frame running had impacted them. The analysis of the data suggests the athletes feel frame running has had, and does have, a positive influence on their quality of life and psychosocial wellbeing. The athletes highlighted how important frame running is for them socially, as it provided them with the opportunity to build and develop friendships around the world, giving them a sense of inclusion and belonging. Furthermore, most of the athletes felt frame running had a positive influence on their psychological development, improving their self-esteem and self-confidence, and positively impacting their quality of life.

The results from the current thesis provide new insight into the impact of frame running participation on psychosocial wellbeing and quality of life for novice and competitive athletes. Importantly, frame running was recognised as an enjoyable physical activity for individuals with CP and related conditions to participate in, thus providing a potentially valuable opportunity to enhance quality of life and psychosocial wellbeing.

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List of Abbreviations:

CFCS – Communication Function Classification System

CP – Cerebral Palsy

CPISRA - Cerebral Palsy International Sports and Recreational Association

EDACS – Eating and Drinking Ability Classification System

GMFCS – Gross Motor Function Classification System

ICF – International Classification of Functioning, Disability and Health

ICF-CY – International Classification of Functioning, Disability and Health for children and youth

MACS – Manual Ability Classification System

QoL – Quality of Life

SCT – Social Cognitive Theory

SDT – Self-determination Theory

TTM – Transtheoretical Model

VFCS – Visual Function Classification System

WHO – World Health Organisation

Chapter 1 – Introduction

This chapter will introduce the two main components of the thesis, cerebral palsy (CP) and frame running (formally known as racerunning) and present the rationale behind the research. It is important to have suitable opportunities for children with CP to be physically active, particularly for children with more severe CP who are limited by mobility impairments. Frame running is an emerging form of adaptive physical activity that can provide the opportunity for individuals with CP to be physically active from a recreational activity through to International-level competition. The existing evidence base on the effects of frame running is limited, and research evaluating the effect of participation on psychosocial factors has not previously been conducted. The current research has implications for frame running at local, national, and international level, and therefore contributes to the case for its inclusion as a Paralympic event, whilst also providing a baseline for the psychosocial effects of frame running at a novice level.

1.1 Cerebral Palsy – An Introduction

The definition of cerebral palsy is not straightforward as it has been used as an umbrella term relating to a group of disorders of movement and posture (Jystad, et al., 2017). The term has been applied to various groups of individuals with motor impairments and related conditions (Panteliadias & Vassilyadi, 2018). Several attempts have been made to ‘define’ cerebral palsy, with Rosenbaum, Paneth, Leviton, Goldstein, and Bax (2007) putting forward the current accepted definition, which has been adopted by International consensus:

“Cerebral palsy (CP) describes a group of permanent disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing fetal brain. The motor disorders of cerebral palsy are often accompanied by disturbances of sensation, perception, cognition, communication, and behaviour, by epilepsy, and by secondary musculoskeletal problems.” (p.9)

Cerebral palsy is one of the most common motor disabilities and cause of neurological disability during childhood (Compagnone, et a., 2014), with an estimated global prevalence of 1.5 to 2.7 per 1000 live births (Eunson, 2016; Gulati & Sondhi, 2018). However, the prevalence and incidence of CP have varied over time and across geographical locations. CP is seen as a heterogeneous condition in relation to its aetiology, as well as the severity of impairments and types of CP (i.e., there is a large range of CP

conditions and it is non-uniform in terms of the symptoms; Rosenbaum, et al. 2007). The specific causes of CP are unclear as it is not possible to identify a single disturbance or series of disturbances in the fetal brain. A disturbance can be an event or process that damages, interrupts, or influences the typical patterns of brain development, maturation, and formation (Rosenbaum et al., 2007). The brain damage can be caused by a variety of pre-, peri- or post-natal events, including infection, hypoglycemia, and hypoxia, and the disturbances to the brain can appear in the cerebellum, cerebrum, and the brainstem (Rosenbaum et al., 2007; Balik & Givon, 2009; Runciman et al., 2016). Ultimately, the consequences of the disturbances to the brain depend on the severity of the disturbances, and the degree of neurological immaturity. Approximately 80% of children with cerebral palsy are born prematurely (Carroll et al., 2006); and the early brain damage creates a substantial risk of developmental delays and deficits in a wide range of physical and mental functions (Johnson & Almi, 1978).

1.2 Cerebral Palsy – A Complex Condition

Above shows that CP is a complex condition. As a chronic condition, and regardless of the functional profile, individuals with CP require management by a multidisciplinary team to consider their physical, psychological, and social goals and help them develop throughout their childhood, adolescence and into adulthood. As each case of CP is different, managing the condition via various interventions allows focus on different areas, for example, physical and/or surgical therapy, and social support. This is dependent on the functional limitations of the individual, which can make it more difficult for certain goals to be achieved. The limiting mobility impairments for children and adolescents with CP can affect not only their physical activity participation, but also their participation and performance of daily living activities (Fowler et al., 2006). Furthermore, the sensorimotor and developmental issues associated with CP (e.g., vision impairment, cognitive deficits, or muscle spasticity; Rosenbaum et al., 2007) can cause restrictions on their mobility, manipulation of objects and/or communication which can further impact their daily living experiences (Hidecker et al., 2011). The early brain damage restricts experience-dependent learning, and the child is at risk of developing a cycle of deprivation and deficit. There is clear evidence for this in the motor domain. Children with more severe motor impairments (GMFCS levels III-V) typically achieve peak motor function between 6 and 8 years of age, before showing a clinically significant loss of function into adolescence and adulthood (Hanna et al., 2009). However, there is considerable variation in the loss of function between individuals and researchers believe that participation in physical activity is crucial in limiting this deterioration (Damiano, 2006; Hanna et al., 2009). It is, therefore, critically important to facilitate the brain injured child's

participation and hence remaining development in mental, physical, and social activity (Imms, 2007).

Participation in any form of activity, whether it is play, sport, or recreation has been found to have a profound impact on the growth and development of children and adolescence with and without disabilities (Rimmer, & Rowland, 2008). Therefore, physical activity for children with CP is vital for development, and a lack of suitable physical activity may lead to the development of secondary health risks associated with CP, including loss of fitness and/or functional abilities (Zwier et al., 2010). However, facilitating a child with CP to participate in physical activity or sport is no easy task, particularly when the child has mobility and/or cognitive deficits, and there are environmental and/or social barriers that can impede their participation. A low level of mobility or understanding of the activity, or simply the limited availability of a suitable activity or sport, can contribute to low levels of participation. Therefore, it is vitally important to aid participation, and in turn help their development as a child, suitable options are found. Frame running is an emerging and adaptive physical activity that can offer this opportunity to children and young people with more severe levels of CP who do not have access to most other forms of physical activity.

1.3 Frame Running – An Introduction

An individual with CP may be restricted from participating in physical activity or sport because of the lack of suitable or available adapted/mobility equipment. Some of the more common activity options available for individuals with CP are swimming (e.g., Declerck et al., 2016), horse riding (therapeutic horseback riding or hippotherapy; Willgens, & Erdman, 2019), general fitness training (e.g., Verschuren et al., 2007, including strength training (e.g., Dodd et al., 2004), and for more severely impaired individuals, boccia (e.g., Siavoshy & Balurian, 2016), and CP football (International Federation of Cerebral Palsy Football, 2018), including powerchair football (e.g., Jefress & Brown, 2017). Even with these opportunities available, individuals with CP can struggle to find an appropriate and/or enjoyable types of exercise (O'Donnell et al., 2010). Activities accessible to less impaired individuals, such as CP football and swimming, typically provide more opportunity for higher-intensity aerobic demands compared to activities for more impaired individuals (e.g., boccia, powerchair football). For those who are not able to participate in the former activities, the opportunities are extremely limited, and subsequently, low levels of aerobic fitness can lead to future health risks (Janssen & Cramp, 2007).

Figure 1.1 A Frame running Tricycle



Frame running (formally known as racerunning) allows individuals with limited or no independent walking ability to participate in a physical activity/sport as they are able to propel themselves forward using an adapted tricycle (See Figure 1.1) or 'Frame Runner' (Van der Linden et al., 2018). Frame running is predominantly taken up by people with CP but is also suitable for those with similar disabilities affecting mobility and balance, for example, muscular dystrophy or Parkinson's disease. The frame runner, an adapted tricycle, has a three-wheeled frame with a saddle and chest plate which supports the athlete. Athlete's steer using handlebars and, as there are no pedals on the racerunner, the athletes propel themselves forward using their legs despite considerable physical restrictions. Some individuals with CP find it easier to use one leg or use their legs in a simultaneous fashion as compared to the alternating coordination pattern used for running or pedalling a standard tricycle or bicycle, thus widening the level of impaired individuals that can be involved in frame running. The frame runner can be used by children as young as 3 or 4 years through to adulthood. Furthermore, the trikes can be used in social environments and for active travel as well as in exercise and sporting environments, and the versatility of the racerunner is of great benefit to overcome some of the barriers of physical activity participation.

Frame running originated in Denmark in 1991 after an athlete with CP, Mansoor Siddiqi, was not satisfied with participating in backward wheelchair racing, which involved pushing a traditional wheelchair backwards with his feet while looking over his shoulder. The popularity of frame running spread across Denmark as it allowed people with impaired to no walking ability to participate in aerobic exercise. The sport continues to grow and develop, from the first international appearance at the athletics World Cup for the disabled in Berlin 1994, to the annual 'camp and cup' which started in 1997 and opened up to international participants in 2002. The 2019 'camp and cup' was the biggest yet with 112 athletes representing 17

different countries. Frame running was integrated into the World Para Athletics (WPA) programme in 2018, and the sport made its first appearance at the WPA European championships in Berlin that year. Within WPA it is now known as frame running in order to align with the naming of other events. The integration of frame running into the World Para Athletics programme shows the growth and development of the sport over the last 27 years and shows that frame running may be able to provide a platform for people with severely impaired to no walking ability to be included and compete in the International sporting arena.

In Denmark, Sweden, and the Netherlands, frame running is a very popular disability sport, and it is growing in other countries around Europe and worldwide. The popularity of frame running is growing in the UK; there are currently 13 clubs throughout Scotland, England, and Wales. The Cerebral Palsy International Sports and Recreation Association (CPISRA) developed frame running and are still involved in promoting the sport at recreational level and are hoping that the development of the sport will lead to inclusion in future Paralympic games. Currently in frame running athletes with neurological conditions resulting in ataxia, hypertonia, and/or athetosis are classified in one of three classes, RR1, RR2, and RR3, with athletes classified in RR1 having the highest level of mobility limitation, to RR3 athletes classified with lower levels of mobility limitation (van der Linden et al., 2018). Paralympic events require an evidence-based classification system that encourages participation by diminishing the impact the impairment of each athlete has on the outcome of competition events (Tweedy & Vanlandewijck, 2011). As frame running is a relatively new sport, especially at the elite levels of World Para Athletics, the existing evidence base for frame running is limited (van der Linden et al., 2020). The same applies to the evidence base for the effects and impact of frame running on athletes' health and wellbeing only now starting to emerge (Hjalmarsson et al., 2020) with research mostly at planning stages (e.g., Ryan et al., 2020). Published studies on Frame running have so far only focused on physical health, while it is clear that CP is a complex condition that affects many aspects of individual's lives. Therefore, this doctoral research aims to add to this growing evidence by investigating the effect of Frame running using a framework that considers the complexity of CP and its effects on many aspects of individuals' lives in interaction with environmental and personal factors.

1.4 Rationale, Importance & Potential Research Impact

Individuals with CP can have complex issues from birth, through childhood, and into adulthood, so having ways to try and counteract or cope with these issues is very important for improving their QoL. Being physically active is important for everyone with and without disabilities, but more of an emphasis should be made in trying to encourage participation in

physical activity and exercise in children and adolescents with disabilities because of the difficulties they may face in engaging. There are many barriers to participating in physical activity and exercise, so it is important to try and find options or alternatives to overcome barriers and increase levels of participation. The children and adolescents with more severe CP may find it particularly difficult to engage in physical activity and exercise because of physical impairments. Frame running offers these more impaired children and adolescents the opportunity to be physically active and participate in disability sport and provides an opportunity to reduce the physical and mental health inequality between those who are physically active and inactive.

Anecdotally frame running is known to have a positive effect on individuals' lives for children and youth with CP, but for this activity to continue to grow at local, national, and international levels there needs to be more objective and scientific data. Growing the evidence base for frame running is important to allow for a better understanding of the effects that frame running can have on different aspects of a participant's life.

The current research focuses on the effect participation in frame running can have on psychosocial factors and QoL at a novice level, and for more experienced frame running athletes. These results are important for parents, coaches, sporting administrators and health care professionals as they highlight the effects that participating in frame running can have on the participant/athlete. The results may also contribute to the case for inclusion of frame running as a Paralympic event. As a novel study in this area, this research aims to inform future research on the psychosocial benefits of frame running and other disability sports for individuals with CP.

1.5 Scope of Thesis & Delimitations

When it comes to physical activity and exercise interventions for children and adolescents with CP, there is a large number of published studies (e.g., Verschuren et al., 2006; Novak et al., 2013). However, these intervention studies predominantly focus on physical outcomes such as mobility (e.g., McBurney et al., 2003) or strength (e.g., Morton et al., 2005), and the evidence base examining the impact of physical activity participation on psychosocial factors is small in comparison. For example, in a systematic review of exercise interventions for children with CP by Verschuren, Ketelaar, Takken, Helders, and Gorter (2006), only two of the identified studies included psychosocial factors as part of the outcome measures.

The importance of psychosocial factors such as self-esteem, self-concept, and self-efficacy for the development children with CP is not well documented (Barlow & Ellard, 2006). This may be because of the potential difficulty in assessing these factors in children with CP, due to cognitive, behavioural, and/or communication impairments associated with

CP. However, given the importance of psychosocial factors throughout childhood development, and the development and maintenance of a healthy and physically active lifestyle (Zwier et al., 2010), examining the effects of a physical activity participation on psychosocial factors in individuals with CP is essential. These findings can provide valuable insight for the community and their carers.

This thesis will focus on frame running, as it may provide one of few suitable opportunities to children and adolescents with more severe levels of CP to be physically active. The thesis will examine which factors parents, frame running coaches and health care professionals identify as most important outcome measures to evaluate the effectiveness of a physical activity intervention for children with CP. Informed by these results, this thesis will subsequently examine the effects of frame running participation on QoL and psychosocial factors, such as self-esteem, self-concept, and self-efficacy, for children with CP longitudinally, and for experienced frame running athletes retrospectively. Furthermore, it will show how frame running can fill a gap in disability sport by offering a suitable opportunity to children and adolescents with CP to be physically active.

There are many outcome measures that can be selected when studying the effect of a physical activity intervention on an individual's health and wellbeing. In the current research the load on the participants was an important factor to consider particularly with the potential cognitive and communication difficulties associated with CP. Furthermore, covering a multitude of factors could diminish from the final results. To aid in the selection of factors, and thus the outcome measures, an online two stage Delphi study preceded the frame running intervention study. Parents of children with CP, health care professionals, and frame running coaches were asked to identify which factors were most important to measure when evaluating the effectiveness of physical activity interventions for children with CP. The results were used to guide the following research around which factors to measure and thus, which outcome measures to administer. A frame running training programme was incorporated to investigate the effects of participation on the psychosocial wellbeing of children and young people with CP. Consequentially, an online survey of experienced frame running athletes explored their perception of how frame running participation has impacted their psychosocial wellbeing.

The purpose of this research is to provide empirical evidence to back up the subjective and anecdotal evidence suggesting that frame running is a worthwhile and feasible option for engaging this population in physical activity and exercise. Furthermore, it aims to show the effects of frame running on psychosocial factors, as the focus of previous research involving cerebral palsy, and physical activity has been on physical outcomes, and therefore, to provide empirical evidence that frame running can be a feasible option for physical activity alongside other sports.

The current research considers the effect of frame running participation on psychosocial factors, as such, the effects on physical factors such as pain, strength, or mobility, was not examined, and was considered to be outside the scope of the thesis. As previously mentioned, there is a more extensive evidence base focusing on physical factors in comparison to psychosocial factors for children and adolescents with CP (Barlow & Ellard, 2006). Therefore, the current research will add to the physical activity literature with a focus on psychosocial factors.

1.6 Aims, Research Questions & Outline of the Thesis

The aim of this research was to investigate the effects of frame running participation on the QoL and psychosocial wellbeing of children and young people with CP and explore the perceptions of experienced frame running athletes. The views of health care professionals, parents and carers were considered regarding what is important to measure to assess the effectiveness of physical activity interventions for children and adolescents with CP. The thesis aims to answer the following research questions (see section 2.10 for research question rationale):

1. Which domains and factors are viewed as most important by parents, health care professionals, and frame running coaches for evaluating physical activity interventions for children and young people with CP?
2. Does participation in a 12-week frame running training programme have an effect on the perceived QoL of children and young people with CP?
3. Does participation in a 12-week frame running training programme have an effect on the psychological factors: self-efficacy, self-esteem, self-concept, and psychological wellbeing of children and young people with CP?
4. Does participation in a 12-week frame running training programme have an effect on the participation and enjoyment of other activities for young people with CP, and is frame running perceived as an enjoyable physical activity for young people with CP?
5. What impact has frame running had on the psychosocial wellbeing and QoL of competitive frame running athletes?

Chapter one has introduced the two main components of the research, cerebral palsy and frame running, and provided the rationale and scope of the research.

Chapter two critically reviews the literature and research relating to physical activity interventions for children and adolescents with CP, including the current frame running

literature. Furthermore, the International Classification of Functioning, Disability, and Health (ICF), and theoretical underpinning of physical activity participation will be introduced and discussed. The barriers and facilitators to physical activity for individuals with CP are discussed, along with, and the impact of physical inactivity. Finally, any gaps in the literature and knowledge will be identified.

Chapter three discusses the ethical and methodological issues encountered when involving children with disabilities in research. Furthermore, the ethical and methodological issues and challenges encountered in the current research will be discussed, as well as considerations for conducting future research with special populations.

Chapter four will present study one, the two-stage Delphi online survey for parents of children with CP, health care professionals and frame running coaches to identify their views of which factors are most important when evaluating the effectiveness of physical activity interventions for children with CP. The results will be used to introduce the psychosocial constructs selected for the following study.

Chapter five discusses the importance of the psychosocial constructs for physical activity participation and development of children with CP, providing justification for their use. Next, chapter five presents a frame running intervention with novice participants, where the results and effects of participation in frame running on the selected psychosocial factors are described and explained.

Chapter six employs qualitative data collection to investigate the impact of frame running on psychosocial factors of competitive frame running athletes across various countries and levels of competition (e.g., regional, national, or International). The results will add a qualitative insight to the quantitative data from the intervention to show the influence of frame running on psychosocial health at a broader level.

Chapter seven is a general discussion looking which looks over the findings of the thesis, potential impact and directions for future research and the conclusions we can take from the current thesis.

Chapter 2 – Literature review

2.1 Introduction

The previous chapter introduced the central features of this thesis, cerebral palsy (CP) and frame running, and highlighted the importance and potential impact of the current research. Furthermore, each of the studies which contribute to the thesis were outlined, as was the overall aim of the thesis and the research questions.

This chapter will first introduce the classification of CP, and outline the instruments used to help with the functional classification of children with CP. This is followed by a review of the literature outlining the importance of physical activity and exercise for young people with CP. This is followed by a look at the physical activity guidelines for young people with CP, and why physical activity participation is vital for development.

In addition to this, the identification and review of theoretical frameworks which are thought to be important in underpinning of physical activity participation and assist in measuring health and disability, explaining the motivation of exercise, changing health behaviours, including exercise, and identifying psychological predictors which can help to promote and increase physical activity.

The barriers and facilitators to physical activity participation for young people with CP are discussed, outlining the potential struggles young people with CP can encounter when trying to participate in physical activity, as well as factors that can encourage physical activity engagement and participation. A summary of research on CP and inactivity follows, highlighting the impact of being inactive for young people with CP.

A critical overview of frame running and the existing research then provides an insight into the sport, and what it can contribute to the world of disability sport. The comparison between frame running and other suitable physical activity, exercise, and sporting opportunities for young people with CP follows. Furthermore, a critical review of the literature provides an insight to what is currently available for this population, and why frame running can be an important addition to disability sport research.

Finally, any gaps in the research and/or knowledge are brought to the forefront, outlining the rationale for the current research, and this is highlighted by the research questions which are addressed throughout the current thesis.

2.1.1 Classification of Cerebral Palsy

There are various types of CP, with the four established types being spastic CP, dyskinetic CP, ataxic CP, and mixed CP (e.g., spastic and dyskinetic CP in the same person), and these can be classified based on the nature and distribution of the motor abnormalities

(Cans, 2000; Sadowska et al., 2020). A diagnosis of CP normally occurs between the age of one and two years (Herskind, 2015), but various symptoms and signs can be evident from early infancy (Gulati & Sondhi, 2018). Historically, classification has been focused mainly on a combination of topographical distributions (i.e., the pattern of the affected limbs), for example, diplegia, hemiplegia, and quadriplegia, and the motor impairment (i.e., predominant movement abnormality), for example, spastic and dyskinetic (Groter, et al., 2004).

The topographical distributions classify CP by identifying the limbs, parts or areas of the body affected by the disturbances in the brain, however, topographical distribution only relates to spastic CP, as dyskinetic and ataxic typically affect the whole body. Spastic CP can be characterised by increased muscle tightness, joint stiffness, and an abnormal movement and/or posture pattern, and can be distributed unilaterally or bilaterally, and is the most common CP, applying to approximately 80% of those with CP (Himmelman et al., 2009). Spastic unilateral CP is diagnosed if limbs on one side are affected (i.e., hemiplegia) while spastic bilateral CP is diagnosed if limbs on both sides are affected (Rosenbaum, 2003). In diplegia the legs are mainly affected, while in quadriplegia all four limbs are affected. Dyskinetic CP is also characterised by abnormal movement and/or posture patterns, but also accompanied with uncontrolled, involuntary, and recurring movements (Himmelman et al., 2009). Ataxic CP is characterised by weakness and problems with coordination and balance. Individuals with ataxic CP typically show an unsteadiness while walking on a wide base (Ingram et al., 1964). Hypotonic is also mentioned in older literature as a type of CP, but it is absent in more recent classification literature, as the majority of individuals classified with “hypotonic CP” during infancy will later develop ataxic, spastic, or dyskinetic CP. That is not to say that individuals do not continue to have hypotonic CP, as this type of CP can continue due to the involvement of cereo-cerebellar circuits (Gulati & Sondhi, 2018). However, classification of CP should now consider additional characteristics for a fuller understanding. For example, Rosenbaum et al. (2007) explained that for classification of CP, four major dimensions should be assessed (see Table 2.1)

Classifying CP by using anatomical and neuroimaging findings, and/or causation and timing alone is not possible as there is not enough information to make informed and clear-cut decisions. Depending on the various subgroups of CP, between 25% to 80% of individuals have accompanying impairments (Odding et al., 2006). The presence or absence of accompanying impairments, for example, attentional, cognitive, or behavioural deficits, hearing and vision impairments, can be used to aid classification of CP (Rosenbaum, et al. 2007). As mentioned, the subtypes of CP have been based, in the past, on the locations of the lesions, degree of impairment, or part of the body involved, but this does not provide

enough information about the individual's functional ability during their daily life (Eliasson et al., 2006).

Table 2.1 Components of Cerebral Palsy Classification from Rosenbaum & Colleagues (2007)

1. Motor Abnormalities	
a. Nature and typology of the motor disorder	The observed tonal abnormalities assessed on examination (e.g., hypertonia, hypotonia) as well as the diagnosed movement disorders present, such as spasticity, ataxia, dystonia, athetosis.
b. Functional motor abilities	The extent to which the individual is limited in his or her motor function, including oromotor and speech function.
2. Accompanying Impairments	
The presence or absence of later developing musculoskeletal problems and/or accompanying non-motor developmental or sensory problems, such as seizures, hearing or vision impairments, or attentional, behavioural, communicative and/or cognitive deficits, and the extent to which impairments interact in individuals with cerebral palsy.	
3. Anatomical and neuro-imaging findings	
a. Anatomic distribution	The parts of the body (limbs, trunk) affected by motor impairments or limitations.
b. Neuro-imaging findings	The neuroanatomic findings on CT or MRI imaging, such as, white matter loss or brain anomaly.
4. Causation and timing	
Whether there is a clearly identified cause, as is usually the case with post-natal CP (e.g., head injury) or when brain malformations are present, and the presumed time frame during which the injury occurred, if known.	

2.1.2 Functional Classification of Cerebral Palsy

Mild, moderate, and severe are the most commonly used terms to classify CP, but these are too general given the wide variety and severity of associated impairments and are a more subjective method of classification. As the previous section indicated, attempting to classify CP and the severity of each case, has been based on type of CP (e.g., ataxic, dyskinetic, or spastic), the effect of motor disorder with regards to varying anatomical areas (e.g., bilateral or unilateral), or the pattern of involvement (e.g., hemiplegia or diplegia; Wood & Rosenbaum, 2000).

Using functional classification scales for individuals with CP provides information on a wider range of skills, abilities, and functionalities for analysis, providing a better and more rounded functional profile of the individuals. The development of scales such as the Gross Motor Functional Classification System (GMFCS; Palisano et al., 1997), Communication

Function Classification System (CFCS; Hidecker et al., 2011), Manual Ability Classification System (MACS; Eliasson et al., 2006), Visual Classification Function System (VCFS; Baranello et al., 2019), and The Eating and Drinking Ability Classification System (EDACS; Sellers et al., 2014) has allowed a better understanding of individuals' functioning by classifying mobility, communication, handling of objects, visual ability, and eating and drinking abilities respectively. Having a more rounded functional profile of an individual can help in guiding families and health care professionals. Table 2.2 provides a summary of the levels of classification for each of the scales.

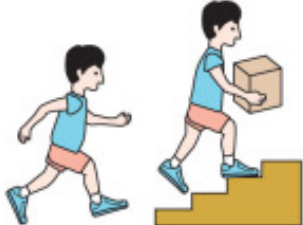
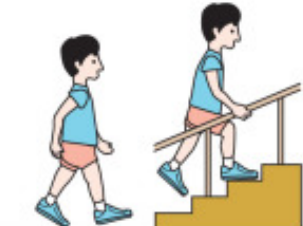

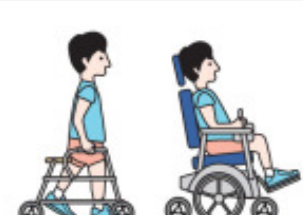

Table 2.2 Summary of the functional classification system levels from Baranello et al., (2019)

Level	GMFCS	MACS	CFCS	EDACS	VFCS
I	Walks without limitations	Handles objects easily and successfully	Sends and receives with familiar and unfamiliar partners effectively and efficiently	Eats and drinks safely and efficiently	Uses visual function easily and successfully in vision-related activities
II	Walks with limitations	Handles most objects but with somewhat reduced quality and/or speed of achievement	Sends and receives with familiar and unfamiliar partners but may need extra time	Eats and drinks safely but with some limitations to efficiency	Uses visual function successfully but needs self-initiated compensatory strategies
III	Walks using a hand-held mobility device	Handles objects with difficulty; needs help to prepare and/or modify activities	Sends and receives with familiar partners effectively, but not with unfamiliar partners	Eats and drinks with some limitations to safety; there may be limitations to efficiency	Uses visual function but needs some adaptations
IV	Self-mobility with limitations; may use powered mobility	Handles a limited selection of easily managed objects in adapted situations	Inconsistently sends and/or receives even with familiar partners	Eats and drinks with significant limitations to safety	Uses visual function in very adapted environments but performs just part of vision-related activities
V	Transported in a manual wheelchair	Does not handle objects and has severely limited ability to perform even simple actions	Seldom effectively sends and receives, even with familiar partners	Unable to eat and drink safely – tube feeding may be considered to provide nutrition	Does not use visual function even in very adapted environments

Note: Gross Motor Functional Classification System (GMFCS); Manual Ability Classification System (MACS); Communication Function Classification System (CFCS); The eating and Drinking Ability Classification System (EDACS); Visual Function Classification System (VFCS).

2.1.2.1 Gross Motor Function

Figure 2.1 Summary of the GMFCS levels from Palisano et al. (2008)

	<p>GMFCS Level I</p> <p>Children walk indoors and outdoors and climb stairs without limitation. Children perform gross motor skills including running and jumping, but speed, balance and coordination are impaired.</p>
	<p>GMFCS Level II</p> <p>Children walk indoors and outdoors and climb stairs holding onto a railing but experience limitations walking on uneven surfaces and inclines and walking in crowds or confined spaces.</p>
	<p>GMFCS Level III</p> <p>Children walk indoors and outdoors on a level surface with an assistive mobility device. Children may climb stairs holding onto a railing. Children may propel a wheelchair manually or are transported when traveling for long distances or outdoors on uneven terrain.</p>
	<p>GMFCS Level IV</p> <p>Children may continue to walk for short distances on a walker or rely more on wheeled mobility at home and school and in the community.</p>
	<p>GMFCS Level V</p> <p>Physical impairment restricts voluntary control of movement and the ability to maintain antigravity head and trunk postures. All areas of motor function are limited. Children have no means of independent mobility and are transported.</p>

The GMFCS allows individuals with CP to be classified objectively with regards to their functional motor abilities. The GMFCS classifies individuals into one of five levels (See Figure 2.1), where the focus is on self-initiated movement, and in particular the ability to walk. The scale classifies motor function into five levels, where individuals classified in level one has independent movement while those classified in level five require complete assistance (Palisano et al. 1997; Palisano et al., 2008). The GMFCS is a well-established

tool to classify gross motor function and is widely used to describe motor function in research populations (Hidecker et al., 2012). However, the GMFCS has some limitations (Eliasson et al., 2006), in that it is not able to predict the functionality of other mobility domains beyond walking. Therefore, the Manual Ability Classification System (MACS) compliments the GMFCS by measuring the functionality of upper extremities.

2.1.2.2 Manual Ability

The brain lesion causing CP, and neurological signs of CP, can be detected through neuroimaging and clinical testing, however, the type and severity of the CP is difficult to determine across the early years, so it is usually confirmed around the age of 4 years. The functionality of a child is very important to a parent, and so it is important to be able to describe the functionality of children showing early signs of CP. The MACS was designed as a tool for children and adolescents with CP, aged 4 to 14, to evaluate their ability to use their hands when handling objects in daily activities (Eliasson et al., 2006). It evaluates their typical manual performance and not their maximal capacity. For the children under the age of 4, showing early signs of CP, Eliasson, Ullanhag, Whaström, and Krumlinde-Sundholm (2017), developed the Mini-MACS to help in classifying the functionality of the younger children. Furthermore, like the GMFCS, the MACS and Mini-MACS have five levels of functionality with increasing levels of impairment. At level one, they have the highest level of functionality and are able to handle objects easily and successfully, and manual ability decreases through to level five, where they are not able to handle objects and have severely limited ability to perform basic tasks (Compagnone et al., 2014).

2.1.2.3 Communication Ability

The prevalence of communication disorders in CP can vary from 31% to 88% (Hidecker et al., 2011) depending on the definition of a communication disorder within CP research. Measuring communication is more challenging compared to object manipulation or mobility because it can be more complex and subjective compared to measuring the ability to perform physical tasks. For example, the ability to complete physical tasks depends on the capacity of the individual, whereas communication is based on the exchange of information between familiar and unfamiliar conversational partners, and the pace of the conversation (Hidecker et al., 2011).

The Communication Function Classification System (CFCS) has been developed to assist the classification of individuals with CP in relation to their communication ability. The CFCS assesses the individual with CP and their ability to communicate with familiar and unfamiliar conversational partners in various environments in order to determine their shared understandings (Compagnone et al., 2014). The CFCS, like the GMFCS and MACS, has

five levels of severity with increasing impairment. At level one, least impaired, individuals are effective in sending and receiving conversation with both familiar and unfamiliar partners, and at level five, most impaired, individuals are rarely able to send and receive conversation, even with familiar partners (See Table 2.2).

The next two functional classification systems, which look at visual ability and eating and drinking abilities, are more recent developments so there is less existing literature; however, they add further domains to the growing functional profile systems for future research.

2.1.2.4 Visual Ability

Visual impairment, of some degree, is one of the associated disturbances of CP, and has been reported to affect up to 50% of children with CP (Baranello et al., 2019). The Visual Classification Function System (VCFS; Baranello et al., 2019) is a functional classification system aimed at classifying how individuals with CP use visual abilities in daily life. Before the VCFS, assessing vision relied on ophthalmological examination, and rarely on the functional ability associated with an individual's vision in daily life. This would normally lead to children with CP beginning rehabilitation without any detailed information about their visual functioning being available to their therapists. The child's visual abilities refer to how a child can use vision purposefully to direct gaze, see, recognize, and interact and explore the environment (Ego et al., 2015). Similar to the previously mentioned classification systems that are already available, the VCFS was developed with five levels of functioning, with level one (I) being highest functioning (able to use visual function easily and successfully in vision-related activities) to level V being lowest functioning (does not use visual function, even in very adapted environments; Baranello et al., 2019). A summary of the levels of functioning can be seen in Table 1.2.

2.1.2.5 Eating & Drinking Ability

The Eating and Drinking Ability Classification System (EDACS; Sellers et al., 2014) is a lesser used functional classification system which classifies the eating and drinking abilities of individuals with CP. Similar to communication difficulties, the prevalence of eating and drinking abilities is unclear and depends on definitions and the assessment tools used, but can range from 27% to 90% (Sellers, et al., 2013). Disturbances of an individual's cognition, communication, perception, and sensation often accompany the motor impairments associated with CP (Rosenbaum et al., 2007). The oral skills that are required for swallowing, eating, and drinking can be negatively impacted by the impairments associated with CP (Sellers et al., 2013), and this can link to risks related to insufficient hydration and nutrition, and the intake of food and liquid into the lungs. The EDACS is used to identify the

most important features of efficiency and safety related to the oral skills that individuals require for eating and drinking. Like the other classification systems, the EDACS describes the abilities across five levels of functionality, where level one is the highest functioning (eats and drinks safely and efficiently; Sellers et al., 2014) and level five is the lowest functioning (unable to eat and drink safely – tube feeding may be considered to provide nutrition; Sellers et al., 2014). The EDACS has not been developed as a diagnostic tool but adds another area of classification for building a more rounded functional profile of individuals with CP in clinical and research settings.

2.1.2.5 The Relationships Between the Functional Classification Systems

The first three classification systems, GMFCS, MACS, and CFCS, were independently created and validated to gain a better understanding of an individual's functional status. Hidecker, et al. (2012) and Compagnone et al. (2014) explored the potential relationships and correlations between the GMFCS, MACS, and CFCS in children with CP to investigate if the scales can be used to predict the functional ability of different scales. Hidecker et al. (2012) examined the potential relationships between the three scales, with parents of children with CP reporting the functionality from each classification system of their child. The results provided complementary information from the three classification systems, and only 16% of the participants had the same level of function in all three scales. Thus, knowing the classification level for one scale rarely leads to being able to predict the classification of the other scales and therefore supports the heterogenous nature of CP.

Compagnone et al. (2014) investigated potential correlations between the three classification scales in children with CP by subtypes (e.g., unilateral, bilateral, hemiplegia, diplegia). The results showed that the three classification systems can be used to build a better functional profile, but the associations are weak and classification from one scale should not be used to assume the functional level on another. For example, children with bilateral CP showed quite extensive motor impairments (GMFCS levels IV and V), but they had the ability to use some method of communication, making themselves understood by both familiar and unfamiliar communication partners which classifies them at levels I and II on the CFCS (Hidecker, et al., 2012).

As mentioned earlier, the similarity between levels of the classification systems may be most consistent at the extremes of each scale, (i.e. level one or level five) and this was indicated by both Hidecker et al., (2012), and Compagnone et al. (2014). Hidecker et al. (2012) described one functional profile as “all V”, and this indicates poor levels of participation and high levels of dependence in daily life. This is in line with a strong statistical association found by Compagnone et al. (2014) across all three of GMFCS, MACS and CFCS which was limited to level V, which is the lowest level of function. As the VFCS and

EDACS are more recent there is less existing research when investigating the potential correlations with the other classification systems. However, a correlation analysis was carried out with the VFCS and other classification systems and Baranello et al. (2019) found that there was just a moderate correlation between them. This further indicates that a child with more severe limitations in motor, communication, or handling functions will not necessarily have severe limitations in visual functioning, and this is in line with the findings from Hidecker et al., (2012), and Campagnone et al. (2014).

The use of classification systems can be useful for researchers given the heterogeneity nature of CP. With wide ranging functional abilities for individuals with CP, the comparison of research can be difficult, as well as conducting systematic literature reviews or a meta-analysis, thus it can be useful for researchers to have clear classification systems. Having definitions and classifications that can be used worldwide means that the data can be combined or compared. With classifications based on function, there is the added point that a researcher can target research on a specific sub-population, for example, non-verbal, non-ambulant, or those with poor manual function.

As well as the comparison or combining of data, the classification systems together can be beneficial in providing a functional profile to researchers and clinicians, as well as informing family members about the roles communication, mobility, and handling objects has for the child and their daily activities. However, it is important to note that the use of these systems should not be used as a test, diagnostic tool, outcome measure, or replace assessment instruments, and should be used to simply describe levels of functionality in their given domains (Rosenbaum et al., 2014). The functional profile of children with CP will influence their ability to be physically active, and may restrict their opportunities, thus leading to decreased physical activity participation and increased bouts sedentary behaviour, heightening the risk of health conditions associated with physical inactivity. The importance of physical activity is discussed in the following section.

2.2 Importance of Physical Activity, Exercise, & Sport

Physical activity is defined as “any bodily movement produced by skeletal muscles that requires energy expenditure. It takes many forms, occurs in many settings, and has many purposes” (UK Chief Medical Officers’ Physical Activity Guidelines, 2019; p. 14). There is a large body of scientific and clinical research that provides evidence of both short- and long-term benefits of participation in physical activity for typically developing children and adolescents (Zweier et al., 2010).

A distinction between physical activity, exercise and sport is important for looking at the contribution and impact on health of individuals, populations, and countries (Kahn et al.,

2012). The terms physical activity and exercise can be viewed as interchangeable as they share some similarities, for example, physical activity and exercise both consist of bodily movement via skeletal muscles; result in energy expenditure; and are positively correlated with physical fitness (Caspersen et al., 1985; Kahn et al., 2012). However, exercise is a component of physical activity with some differences, as exercise needs to be structured, planned and repetitive with the objective of maintaining to improving physical fitness (Kahn et al., 2012; Dasso, 2018). For example, walking 300 minutes per week can be viewed as physical activity, and going to the gym for 90 minutes is exercise (Kahn et al., 2012). There can be grey areas with brisk walking expending more energy than low intensity exercise.

Sport can be viewed as a sub-category of exercise, with sports participation occurring as part of a team or individually, while adhering to a set of rules and having a defined goal (Kahn et al., 2012). Previous research has also suggested that the motivation to participate in exercise and sport can differ (Kilpatrick et al., 2005; Roberts et al., 2015). Participants of exercise identify body-related motives such as weight loss (i.e., intrinsic motives), compared to motives of sport participation identified as enjoyment, competence, and challenge (i.e., extrinsic motives; see section 2.4.1.1 for intrinsic and extrinsic motivation), suggesting the motivation for sports participation to be more desirable than those of exercise (Kilpatrick et al., 2005).

When undertaken frequently, physical activity, exercise, and sport are beneficial for physical and psychological well-being, and overall health (e.g., Hillman et al., 2008, Texeria et al., 2012, Maher et al., 2015). For children and adolescents with CP, the benefit of physical activity and exercise are also well documented (Durstine et al., 2000; Maher et al., 2007), and physical activity can be even more important for this population struggling with impairments that limit their motor ability (Koldoff, et al., 2015). Childhood and adolescence are critical periods for youths with and without disabilities when developing positive health-related behaviours (Rimmer & Rowland, 2008).

2.2.1 Physical Activity for Young People with Cerebral Palsy

Interest and involvement in physical activity can start during childhood, and continue to develop through adolescence, and influence the development of healthy adult lifestyles (Hallal et al., 2006). Thus, promoting participation in physical activity during early life is key for healthy development. On the other hand, psychological, social, and/or physical factors can have a negative impact on an individual's participation of physical activity, contributing to unhealthy and inactive lifestyles. A single factor or a combination of factors can cause unhealthy and inactive lifestyles to persist through childhood and adolescence. However, physical inactivity does not always occur out of choice, and problems caused by inactivity

can arise irrespective of the amount of weekly physical activity individuals are engaged in if they are consistently engaged in long periods of sedentary behaviour (Carlson et al. 2013). Physical inactivity and sedentary levels can both be related to the level of gross motor function in children with CP. Children and adolescents classified with mild gross motor function deficit (GMFCS levels I and II) report higher levels of physical activity and lower levels of sedentary time compared to those classified levels IV and V (i.e., more severely impaired gross motor function; Maher et al., 2007). Although some individuals may not experience difficulties in this area, within the child and adolescent CP population, higher levels of sedentary time and behaviour have been reported in comparison to their typically developing peers, indicating they may be at higher risk of health problems related to more sedentary time and lower levels of physical activity (Nooijen et al., 2014). Furthermore, this may be more relevant now compared to previous years due to the development and easy access to technology, including social media, television streaming services, and gaming.

Participation in physical activity for children with CP can help to improve various aspects of their lives, including functional independence, life satisfaction, and social and emotional well-being (Verschuren, et al., 2012). However, individuals with CP are often at risk of developing secondary health complications alongside the primary neurological disturbance, including increased sedentary lifestyles, low levels of fitness, musculoskeletal fragility, and orthopaedic abnormalities which can limit their ability to participate in physical activity and exercise (Peterson, 2014). There is research providing evidence that improving muscle strength and cardiovascular fitness are important for health (Verschuren et al., 2016), but individuals who have greater mobility or cognitive impairments may not be able to participate in these more moderate to vigorous intensity levels of physical activity. Verschuren et al. (2016) combined findings from studies objectively measuring physical activity levels in children and adults with CP and found 76% to 99% of their waking hours were spent being sedentary, with only 18% participating in light physical activity, and 2% to 7% engaging in moderate to vigorous activities, with the latter only found in the less impaired participants (GMFCS levels I to III).

As previously mentioned, the amount of time many individuals with CP spend sedentary greatly increases the chance of health problems and diseases associated with a sedentary lifestyle developing (e.g., obesity, heart disease, and diabetes). Thus, the importance of physical activity for individuals with CP is paramount to try and counteract the negative impact of physical inactivity and an exaggerated sedentary lifestyle. Whether the aim is to become more physically fit, or improve muscle strength, or simply break up long periods of sedentary behaviour, participation in physical activity or exercise at all levels of intensity is extremely important for children and youths with CP. This can be especially relevant for children and youth with more severe CP (i.e., GMFCS levels IV and V), because

reducing and breaking up long periods of sedentary behaviour may be the only feasible intervention. Changing the behaviour of children and youth with CP is not straightforward and consideration of the contextual factors is going to be necessary. Furthermore, individuals with CP might find it difficult to meet physical activity guidelines because of personal and environmental factors.

2.2.1.2 Physical Activity Guidelines for Young People with Cerebral Palsy

In relation to physical activity guidelines, the World Health Organisation (WHO) published the updated guidelines for all individuals including those living with chronic conditions and disabilities in November 2020. For children and adolescents (age 5-17) including those living with disabilities, the WHO recommends, on average, at least 60 minutes of moderate-to-vigorous intensity physical activity throughout the week, and vigorous-intensity physical activity (including activity that strengthens bone and muscles) at least 3 days a week (Bull et al., 2020).

In comparison to the 2010 physical activity guidelines, the 2020 guidelines show a recognition towards additional health benefits from being physically active, and that health includes not only physical but also social and mental wellbeing (Ding et al., 2020). Furthermore, the inclusion of suggestions for decreasing sedentary time, particularly reducing screen time, highlights the growing interest in the negative effects of excessive sitting time (Ding et al., 2020). Additionally, one of the new messages highlighted in the 2020 guidelines is the idea that any physical activity is better than none and replacing sedentary time with physical activity of any form and intensity can provide some health benefits (Bull et al., 2020). This is important for many individuals with CP as they can spend a large proportion of their time sitting due to mobility impairments associated with CP. However, these guidelines are aimed at more typically developing children and adolescents, and though there is reference to recommendations for individuals living with disabilities, the evidence reviewed covered other health conditions than CP (e.g., spinal cord injury; intellectual disability; stroke; Bull et al., 2020).

Verschuren, Peterson, Balemans, and Hurvitz (2016) combined the information/guidelines provided by the WHO for typically developing individuals and previous research involving individuals with CP and produced physical activity guidelines for the CP population. Their research culminated in guidelines for daily physical activity, cardiorespiratory exercise, and resistance exercise. The guidelines for daily physical activity for individuals with CP was similar to those provided by the WHO, in that the individuals with CP are recommended to participate in moderate-to-vigorous physical activity for 60 minutes a day, for 5 or more days a week. Guidelines for sedentary behaviour were also similar to the WHO guidelines, in that individuals should spend no more than 2 hours a day sedentary,

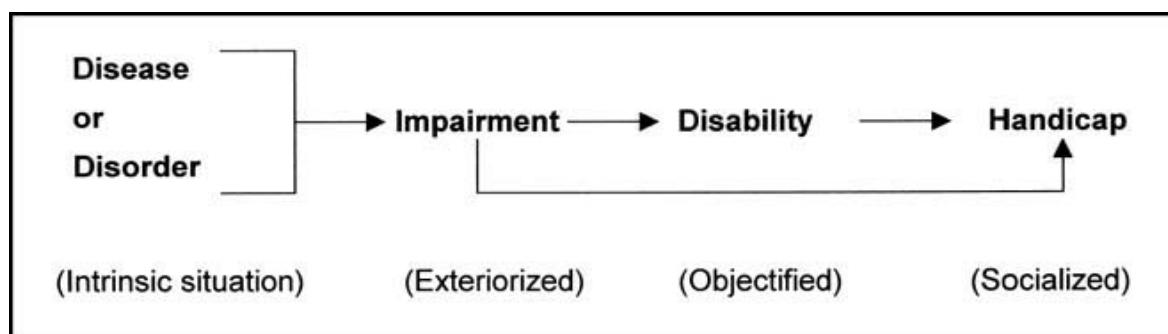
however, this may not be possible for individuals with more severe CP, for example those classified GMFCS levels IV and V. Therefore, a recommendation of a 2-minute break every 30 to 60 minutes from being sedentary is suggested for individuals with more severe mobility impairments.

The guidelines suggested for the CP population by Verschuren et al. (2016) are simply that: guidelines. Due to the wide-ranging mobility levels and associated impairments of individuals with CP, there is no one-size-fits-all solution. Furthermore, the studies used by Verschuren et al. (2016) to build these guidelines recruited more ambulatory compared to non-ambulatory individuals with CP, making it more difficult for the more severely impaired to apply these guidelines and translate the research and become involved in activity programmes for a variety of reasons, including the practical implications of being confined to a wheelchair. For this reason, in an attempt to understand how to manage CP, thus encourage a higher level of physical activity participation by examining the interactive relationship of health conditions and contextual factors, a review and discussion of the International Classification of Functioning, Disability and Health (ICF) framework will follow.

2.3 International Classification of Functioning, Disability, and Health (ICF) Framework

The way in which CP is managed and assessed is dependent on the frameworks which are used to conceptualise the disorders and diseases (Rosenbaum & Stewart, 2004). The World Health Organisation (WHO) published the first edition of the International Classification of Impairments, Disabilities and Handicaps (ICIDH; World Health organization, 1980) in 1980, as part of numerous classifications to understand a wide range of information about different aspects of health. The original ICIDH framework (Figure 2.2) was about classifying the consequences of diseases and disorders and aimed at encouraging people to identify the impact of a disorder on an individual's function and capability to engage fully in their life. The conceptual model represents a cause-and-effect relationship, between the three factors, impairment, disability, and handicap (Fougeyrollas & Beauregard, 2001).

Figure 2.2 Original International Classification of Impairments, Disabilities & Handicaps model (ICIDH; 1980)

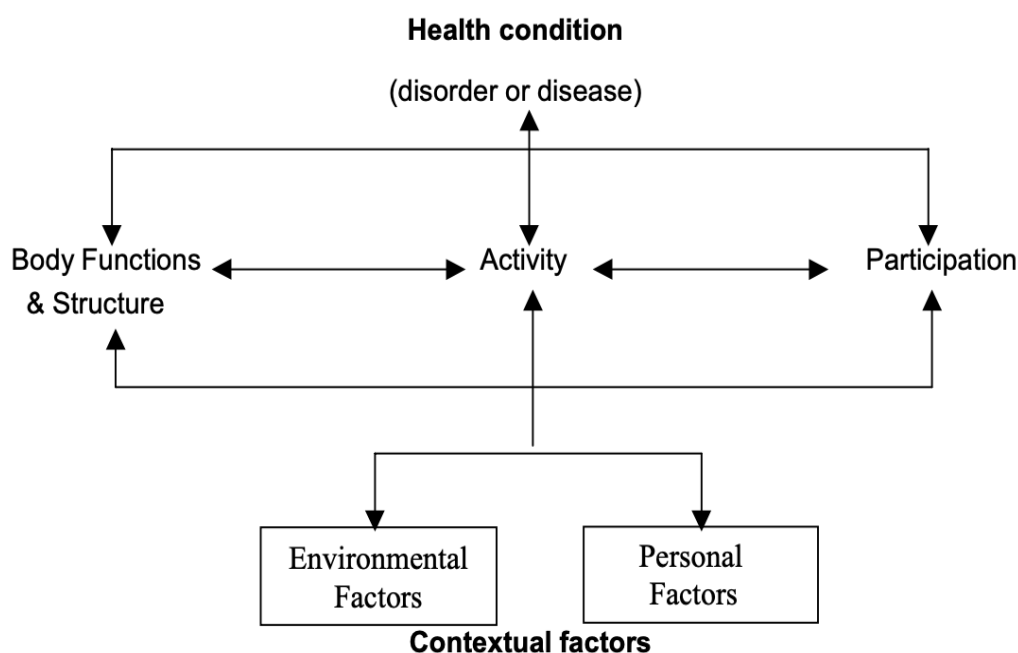


An example of the linear and unidirectional links between the factors has been described as follows:

“...an injury leads to the impairment of an organ’s functions and structures, which then leads to a disability in the person’s behaviour and activities, which generates one or many handicaps or disadvantages concerning social or survival roles” (Fougeyrollas & Beauregard, 2001 p7,).

Despite the intentions of the ICIDH and it being a fresh, new approach for its time in helping to distinguish between the disease itself and the consequences of disease, the uptake was slow. It was not used as a classification framework by many countries as it was viewed as problematic because of the negative depiction of the outcomes of disease or disorder being labelled as “handicap” and “disability” (Rosenbaum & Stewart, 2004).

Figure 2.3 International Classification of Functioning and Disability Framework (ICF; 2001)



The limited use of the ICIDH as a framework led to efforts for it to be refined, revised, and reshaped, and in 2001, the WHO published the International Classification of Functioning, Disability, and Health (ICF; World Health Organisation, 2001) to challenge how we think about health. The ICF is based on the bio-psycho-social model (WHO, 2007), and focuses on “components of health” instead of the consequences of disease (Rosenbaum & Stewart, 2004). The aims of the ICF model (Figure 2.3) are to further develop and advance the ideas of the ICIDH (Tweedy, 2002), and to be a universal classification system, covering all people, unlike the ICIDH which covered just people with disabilities. The new model (ICF framework) of human functioning and disability incorporates all aspects of health and looks to show the interactive relationship between the ideas about health, health conditions, and the contextual factors (Rosenbaum & Gorter, 2011). The structure of the ICF includes description of health conditions which can be categorised in the domains ‘activities and participation’, ‘body functions and structures’, and supplemented by contextual factors which may be either ‘personal’ or ‘environmental’ (Vargus-Adams & Martin 2010). Even though the international classification of functioning, disability, and health in 2001 meant an expansion in age range and coverage of the functional limitations in comparison to the ICIDH, a need was identified to encompass the differences in health conditions, functioning and disability between children and adults (Simeonsson et al., 2006). Hence, in 2007, the World Health Organisation developed The International Classification of Functioning, Disability and Health for Children and Youth (ICF-CY; WHO 2007) to provide a common framework for classifying,

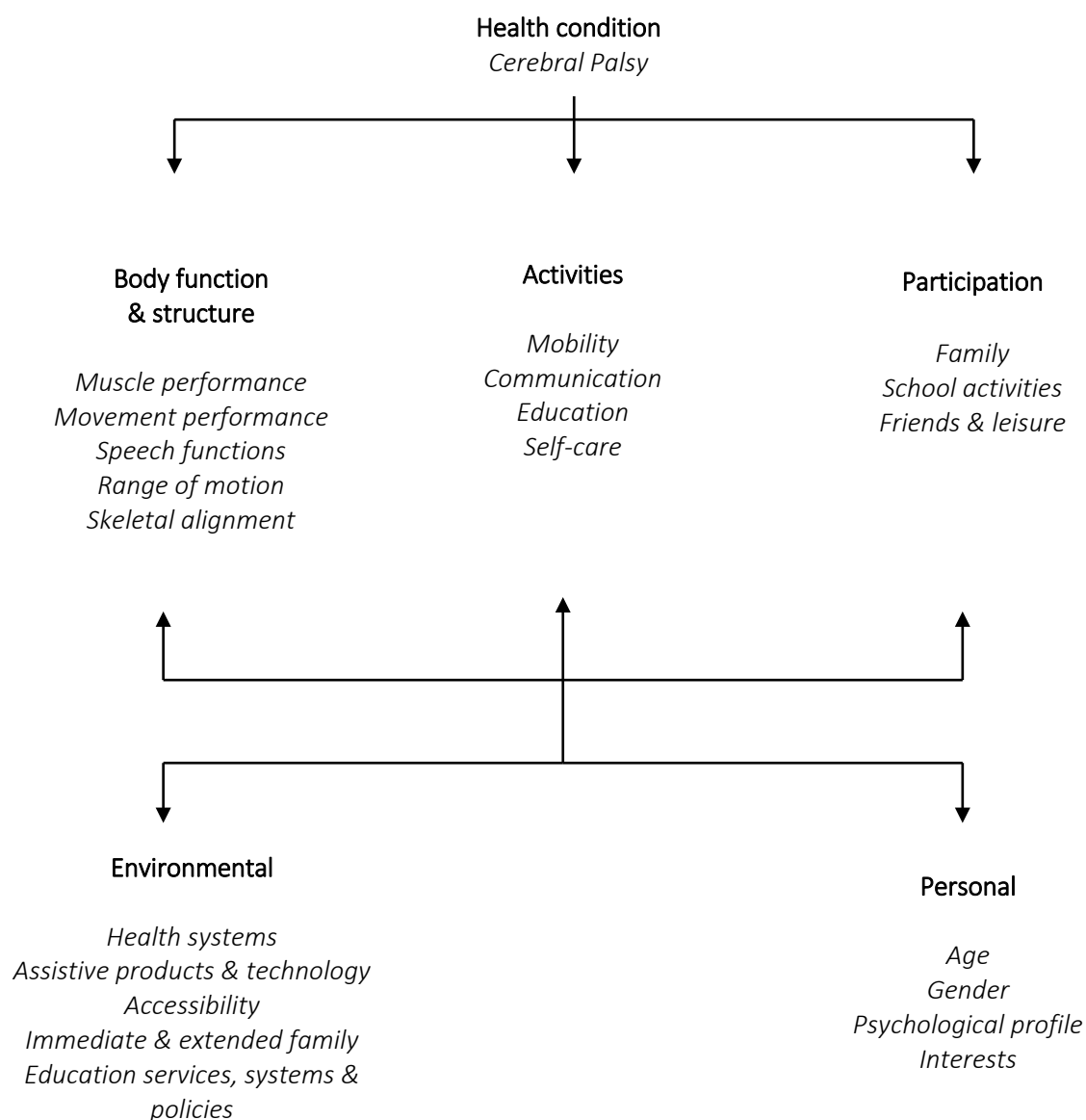
describing, and defining functioning and disability in all children from birth to 18 years of age (Schiariti, et al., 2014).

2.3.1 The ICF-CY framework

The ICF-CY, like the ICF, is a psychosocial model. The conceptual framework and definitions of the ICF-CY are the same as the ICF, however, it contains 231 more paediatric specific codes (e.g., gait pattern, and gestural language; Vargus-Adams & Martin, 2010). Furthermore, it is seen to be useful throughout physical, social, and psychological development and growth as it helps describe and reflect the current levels of function, activity, and participation (Trabaca et al., 2012). The arrows of the ICF-CY highlight the connected nature between the components and contextual factors, which may influence, i.e., have a positive and/or negative impact on the individual's experience of a health condition (see Figure 2.4; McDougall & Wright, 2009). Furthermore, the frameworks can help to plan the most suitable therapeutic, rehabilitation, and/or physical activity/exercise programmes targeting the desired outcomes. This is important for future research due to the heterogeneity of CP, and being able to target relevant outcomes (e.g. improving mobility by increasing physical activity participation or reducing sedentary behaviour) may ultimately help to improve day to day living and QoL.

The ICF-CY, like the ICF, comprises of the components of 'body functions and structures', 'activity and participation', and contextual factors that are 'environmental' or 'personal'. Each of the components aside from 'personal factors' consists of numerous domains/chapters, and within each chapter/domain there are categories that are the most specific units of classification (WHO, 2007). 'Body functions' refer to the physiological and psychological functions of the body; 'body structures' refer to the anatomical parts of the body, (i.e., limbs, organs and their component); 'activity' refers to the execution of a task or action by the individual; 'environmental factors' are defined as external influences on functioning and disability including social, physical and attitudinal environments; 'personal factors' are defined as the internal influences on functioning and disability (WHO, 2007). It is important to note the inclusion of the contextual factors (personal and environmental) because it provides an outlet to record the impact individual characteristics and circumstances have on the child's functioning (Trabaca et al., 2012).

Figure 2.4 International Classification of Functioning, Disability, and Health for Children and Youth with CP (ICF-CY; Trabacca et al., 2012)



The ICF-CY framework is applicable to children with a range of health conditions and has recently been applied to CP research (e.g., Tantilipikorn et al., 2012; Anaby et al., 2017). For example, the ICF-CY was used to identify outcome measures and interventions reported to be used across central Thailand to clarify the management of CP in children in that region (Tantilipikorn et al., 2012). Additionally, applying the ICF-CY framework allows the organisation of motor disorders and accompanying additional disturbances associated with children and adolescence with CP, which can contribute to activity restrictions. From the perspective of the framework, CP can impact on an individual's 'functioning', including 'body structures', (e.g., use of upper and lower limbs), 'body functions' (e.g., cognitive functions), 'activities', (e.g., walking ability), and 'participation' (e.g., playing sports; Novak et al., 2013).

The impact that environmental and personal factors have on activity and participation in children and adolescents with CP are discussed further on in this chapter by looking at the facilitators and barriers to physical activity which are discussed in section 2.5.

The coding, which includes one letter and at least a single digit, and 3-level classification allow for increasing levels of specificity and shows the extent of the functioning and/or disability within that category, of the individual. Components are the first, and broadest level of classification, and each are coded by a letter; 's' body structures; 'f' body functions; 'd' activities and participation, and 'e' environmental factors. Within the components, chapters or domains form the second level of classification, for example, the chapter classification of 'b1' indicates mental functions. Further detail is provided by the categories, the third level of classification within the ICF-CY, with the number of digits expanding with the increasing level of specificity. For example, 'b1' refers to mental functions, 'b126' refers to temperament and personality functions, and 'b1266' refers to confidence (the specificity can expand up to five digits; WHO, 2007).

One of the main advantages of the ICF and the ICF-CY is how it presents a common language that can be used by professionals and across disciplines to describe the health and functioning of individuals with disabilities. The current thesis aims to employ the ICF-CY (study 1) and ICF (study 3) frameworks to assess and categorise areas of interest for evaluating the effectiveness of physical activity interventions, and to analyse the influence of frame running participation on the health and functioning of competitive frame running athletes.

The current thesis aims to employ the ICF and ICF-CY framework to assess and categorise the areas of interest for evaluating the effectiveness of physical activity interventions for children with CP. For children with CP, physical activity participation is one aspect that might positively influence their health and functioning, however there can be barriers to physical participation, and the following section introduces and reviews theoretical frameworks which may underpin participation in physical activity for children with CP.

2.4 Theoretical Underpinning of Participation in Physical Activity for Children with Cerebral Palsy.

Knowing the physical and psychological benefits that can come from regular participation in physical activity and exercise is not enough to motivate individuals to become engaged in physical activity. There are only a small proportion of adults in modern societies that engage in physical activity or exercise to the level that complies with public health guidelines (Sisson & Katzmarzyk, 2008; WHO, 2010). The prevalence of physical activity in youth differs across populations and varies across age ranges, sex, socioeconomic status, and country of

residence (Sisson & Katzmarzyk, 2008). In relation to children and youth with CP, they are reported to spend more time sedentary in comparison to their typically developing peers (Williams et al., 2018), but this can be skewed by mobility impairments associated with CP of all severities. Low levels of moderate to vigorous physical activity in comparison to their typically developing peers can also leave children with CP at greater risk of diseases like obesity and diabetes (Verschuren et al., 2016). Despite it being reported that children with CP may be expending more energy at rest, and performing the same tasks, and participating in the same activities as their typically developing peers (Verschuren et al., 2014), the youth with CP will overall be using less energy in comparison. Breaking the sedentary behaviour and replacing it with light physical activity may be easier than focusing on implementing moderate to vigorous intensity physical activity programmes (Verschuren et al., 2014). However, as previously discussed there are many barriers to regular participation in physical activity for children and youth with CP. Therefore, for a change in exercise behaviour to occur, thus increase levels of physical activity, psychological predictors need to be identified, and the ways in which the predictors can influence exercise behaviour. This will aid the planning and development of interventions aimed at changing an individual's exercise behaviour (Hagger & Chatzisarantis, 2007).

Theories of motivation are key when researching the possibility of change in exercise behaviour, and motivation and enjoyment are two important facilitators in supporting regular physical activity and exercise (Hagger & Chatzisarantis, 2008; Moore et al., 2008; Teixeira, et al., 2012). An individual's lack of motivation to be physically active can be down to a simple lack of interest in physical activity or exercise, where the outcomes are not valued high enough for it to be a made a priority. Another possible explanation for a lack of motivation, is that an individual may believe they are not competent enough to participate, either by not feeling physically fit or skilled enough, or their health condition may limit their ability to participate (e.g., mobility impairment associated with CP; Ryan et al., 2009; Verschuren et al., 2012). Alongside individuals who are unmotivated, there are the people who express personal motivation to engage in exercise on a regular basis, and do so, but only for a short period of time (i.e., a lack of adherence). This may be because they feel like they have to and not because they want to, for example, going to the gym to look better for others. Overall, individuals tend to exhibit low levels of physical activity and exercise because they are either completely unmotivated, not motivated enough to be physically active, or only motivated by external motivational factors which may not lead to sustained physical activity or exercise (Teixeria et al., 2012).

The Self-determination theory (SDT) is an important framework in the current thesis as there is a focus on the acquisition of an individual's motivation to initiate a new health-related behaviour, in this thesis frame running, and how the adherence to the behaviour is

continued (Meyns et al., (2018). Furthermore, previous research has indicated the importance of the SDT in understanding physical activity levels through motivation, which is important to the current research as understanding exercise behaviour and motivation in children with CP could be vital in promoting frame running participation (Texeria et al., 2012), and this is discussed in the following section.

2.4.1 Self-determination Theory

A prominent theory of motivation is the self-determination theory (SDT) and has evolved from research by Deci in the 1970s while investigating the effects of extrinsic rewards on intrinsic motivation (Deci & Ryan, 2011). SDT is a macro-theory, consisting of three sub-theories: Cognitive Evaluation Theory (CET; Deci & Ryan, 1975), Organismic Integration Theory (OIT; Ünlü, 2016), and Basic Psychological Needs Theory (BPNT; Deci & Ryan, 2002). The purpose of SDT is to try and explain motivation and behaviour based on how an individual is motivated, how they are influenced by contextual factors, such as the environment, and aspects of self-perception (Hagger & Chatziasarantis, 2008). Physical activity and sport are one area of research that the SDT has been applied to on many occasions (Deci & Ryan, 2011). A basic principle of the SDT which is relevant to physical activity and exercise, and the CP population, is that it distinguishes between the two types of motivation regulating an individual's behaviour: intrinsic and extrinsic motivation.

2.4.1.1 Intrinsic & Extrinsic Motivation

Intrinsic motivation is being motivated to engage in a behaviour because of the personal rewards and satisfaction, and not because you are looking for an external reward. For example, when someone is intrinsically motivated, they will experience feelings of enjoyment, excitement, and personal accomplishment (Texeria et al., 2012). An important characteristic of intrinsically motivated activities is that individuals experience them as enjoyable and fun, therefore, if the main reason for participating in certain activities is fun, enjoyment, and challenge, then the individuals are intrinsically motivated (Ryan et al., 2008).

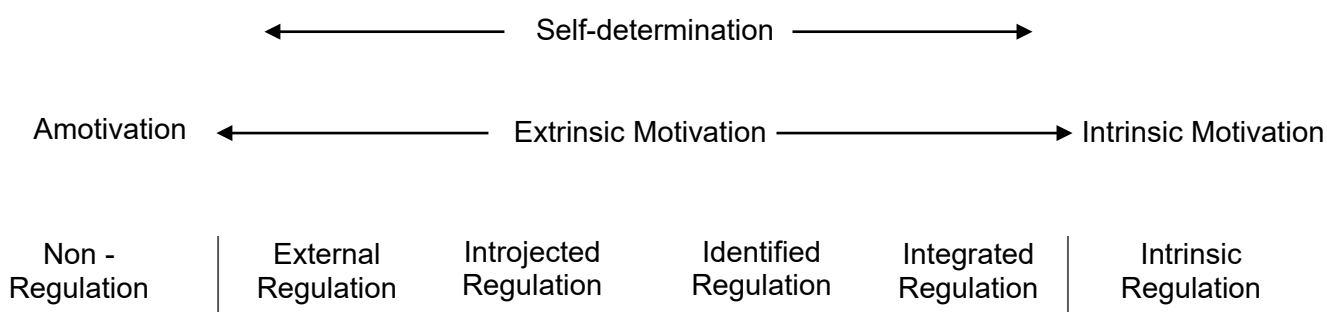
Cognitive Evaluation Theory (CET) was first introduced as a way to account for the effects on intrinsic motivation of external events, for example, feedback and rewards (Deci, 1975; Ryan et al., 2009). It has been developed as a sub-theory of SDT and focuses on the determinants of intrinsic motivation. CET identifies how different social inputs will affect an individual's intrinsic motivation and how environmental and social factors can either help or impede intrinsic motivation. For example, a child with CP may be interested in swimming, but if the teacher/coach was controlling and/or putting too much pressure on them to perform, then the child's level of interest may fall, and they will see swimming as less

enjoyable. This scenario is more prominent in amateur sport, as the main reason these amateur children and adults are participating initially is because of the enjoyment they experience, and if someone else involved becomes too controlling or overly critical then they will dropout because they are no longer experiencing enjoyment or a sense of competence.

In contrast to intrinsic motivation, extrinsic motivation is being motivated to participate in an activity to earn something separate from the activity, and not for enjoyment or satisfaction, for example to earn a reward or to avoid a punishment (Texeria et al., 2012). Exercise is seen as more extrinsically motivated than sport because individuals will more likely exercise to look better or to improve their, and not because they enjoy it, whereas people tend to engage in sport as it is something that they enjoy participating in. However, sport may require extrinsic motivation to participate in regular practice sessions, to develop and refine skills, and to improve overall performance. Most physical activities require a combination of both intrinsic and extrinsic motivation, suggesting the importance of extrinsic motivation in relation to engaging in physical activity (Ryan et al., 2009). Extrinsic motivation, along with intrinsic motivation and amotivation (showing a complete lack of motivation) can be shown to sit on a continuum of self-determination (see Figure 2.6; Biddle et al., 2015).

On the far left of the continuum, is amotivation, another complex category of motivation, when any form of motivation is completely absent for different reasons. For example, someone may not see any value in the activity and so cannot see the link between engaging and the desired outcome, or they simply feel they lack any sort of competence to participate in the activity (Ryan et al., 2009). Intrinsic motivation, sitting on the far right, is the most preferable because individuals will engage and participate because of internal factors suggesting that the likelihood of continued participation is higher. When intrinsically motivated, individuals engage because it is for their own reasons, for example, participating for the fun and enjoyment of the activity (Biddle et al., 2015). Extrinsic motivation, sitting in the middle, is more complex because it is guided by external factors whether environmental or social and can be affected by various factors at one time or a combination. The quality of extrinsic motivation can also differ, with the four main types of extrinsic motivation indicating a move towards either a state of amotivation or intrinsic motivation (see Figure 2.5).

Figure 2.5 Self-determination continuum, with types of motivation, and types of regulation (adapted from Deci and Ryan, 2002)



The four types of extrinsic motivation - external regulation, introjected regulation, identified regulation, and integrated regulation - can be placed along the self-determination continuum and each describe a different quality of extrinsic motivation which moves to the desired state of being intrinsically motivated. External regulation is the first type of extrinsic motivation which shows a step away from a state of no motivation (i.e., amotivation), and describes a behaviour as being controlled by the temptation of rewards or fear of threats (e.g., viewing exercise as a must because it has been prescribed by medical professionals to reduce the individuals risk factors; Biddle et al., 2015). Introjected regulation is next on the continuum, and this indicates that an individual will carry out the behaviour to seek approval, or avoid feeling guilty, which can be a common reason for individuals participating in physical activity (e.g., the feeling of having to go to the gym or you will be overcome by the feeling of guilt; Biddle et al., 2015). Moving towards the self-determination end of the continuum, identified regulation refers to the individual's behaviour being motivated by their understanding of the outcomes and benefits of their actions (e.g., an individual wants to exercise to get fit compared to feeling like they ought to exercise; Biddle et al., 2015). The fourth and most self-determined extrinsic type of regulation, integrated regulation, refers to an individual's behaviour being regulated by their desire to reach and complete important personal goals. It should not be confused with intrinsic motivation because the individual's behaviour is still driven by desire to achieve goals and not carrying out the behaviour for the joy of it (Biddle et al., 2015).

2.4.1.2 SDT & The Basic Psychological Needs

SDT also introduces the concept that to experience optimal motivation there are certain psychological needs that need to be experienced through physical activities (Teixeria et al., 2012), and this can be explained by the basic psychological needs theory (BPNT). The

BPNT, another sub-theory of SDT, offers a framework that tries to explain the roots of self-determined forms of motivation based on three universal, basic and innate psychological needs, which are autonomy, relatedness, and competence (Hagger & Chatzisarantis, 2008; Ryan et al., 2009). Increasing the levels of satisfaction of these three needs will lead to improved feelings of wellbeing (Teixeria, et al., 2012), so this section will briefly introduce each of these basic psychological needs.

Competence is the belief that someone has in their own ability to perform well in the chosen activity. In SDT, competence does not solely relate to the individual's ability and skills, but also to characteristics of the social environment. For example, if a coach or parent is able to provide positive feedback then the individual will feel more competent and this will in turn enhance levels of motivation (Ryan et al., 2009). This can occur in the opposite direction with disengagement and demotivation. The second basic psychological need according to the BPNT is relatedness, which suggests that a sense of a shared experience, sense of belonging and inclusion, and connection with others is going to be vital to somebody's wellbeing, for example, a warm and accepting atmosphere at a sports club. The third basic psychology need is autonomy, which relates to the freedom of choice or self-regulation. When a behaviour is autonomous, it is both intentional and volitional (Ryan et al., 2009). When the sense of autonomy is high, an individual will feel like they are engaged in that activity because they have chosen to and not because they have been told to or pressured into it by somebody else or external factors. Acting autonomously is impacted by social environments, and a low sense of autonomy can leave individuals feeling physically and emotionally depleted by participating in their activity. In physical activity, exercise, and sport, all three of the basic psychological needs require support for motivation, and if they are not supported, then levels of motivation are likely fall and sustaining the activity will be an unattractive prospect.

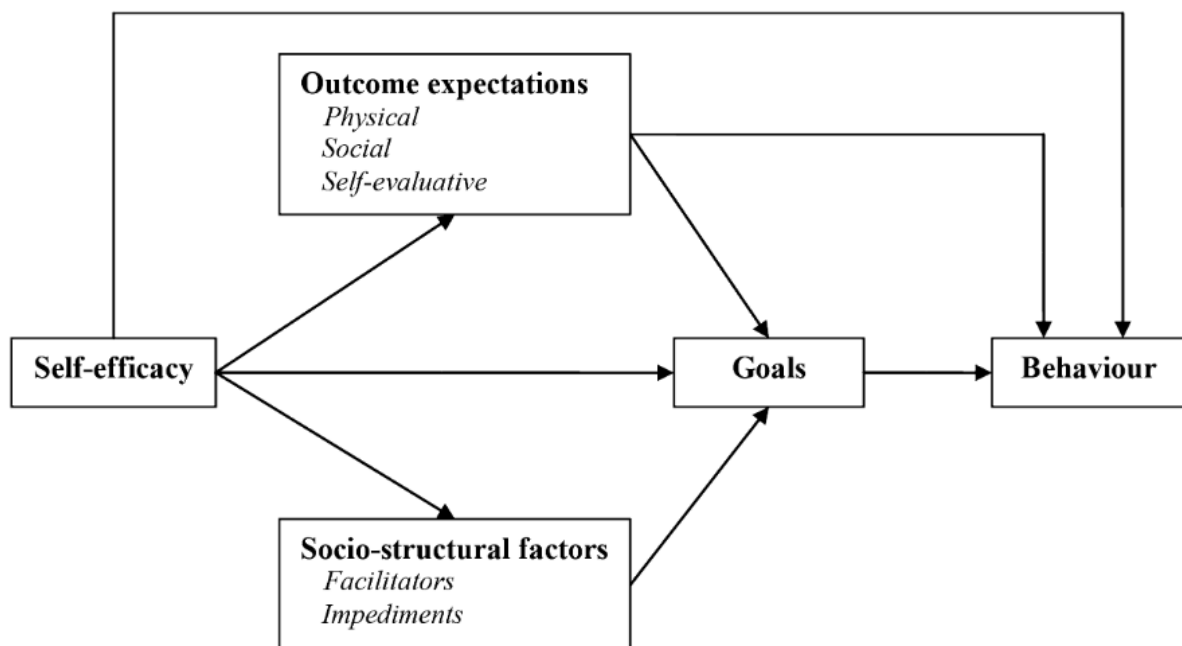
SDT is an important framework in relation to the promotion of health behaviours and physical activity interventions (Ryan et al., 2009). This is because the aim of the SDT is to guide the basic psychological needs of autonomy, competence, and relatedness to develop intrinsically motivated individuals to help with engagement and adherence to physical activity and exercise behaviours. Furthermore, SDT can help researchers understand where the barriers to participation originate, as well as what type of barriers may emerge. For example, a child with CP may not feel they have the motor ability to participate (i.e., lack competence) compared to their able-bodied peers, and so develop a fear of failure and/or exclusion surrounding that activity and are then demotivated to participate in the present and future. Moreover, a child with CP may feel pressurised to take part in an activity by a parent or teacher, which would cause them to feel they have less control. They would see it as less enjoyable as a result, thus demotivating them to participate. Furthermore, application of the

SDT framework can help to develop intrinsic motivation to overcome barriers and encourage promotion of physical activity (e.g., previous research has suggested positive relationships between more autonomous forms of exercise behaviour and motivation; Myens et al., 2018).

2.4.2 Social Cognitive Theory

Physical activity interventions which are informed by previous research and theoretical frameworks are seen to be more effective in comparison to physical activity interventions that are not based on a theory because there is more of a focus on cognitive variables in the former (Young et al., 2014). The focus on cognitive variables highlights their importance when it comes to behaviour, particularly as these are more subject to change compared to other factors. Alongside SDT, Social Cognitive Theory (SCT; Bandura 1986) is another theory that can be examined in relation to physical activity and can be utilised to positively influence physical activity behaviours. Biddle, Mutrie, and Gorely (2015) suggest the key cognitive elements of SCT involves the human ability to think and have the knowledge about the possible consequences from their actions.

Figure 2.6 Social Cognitive Theory Model of Health Behaviour (Bandura, 2004)



There are three main constructs in SCT: self-efficacy, socio-cultural factors, and outcome expectations (Young et al., 2014). Figure 2.6 shows the paths of influence where self-efficacy can affect health-behaviours directly and indirectly, and the level of self-efficacy will influence the outcomes that individuals will visualise, the setting of goals, and the

performing of behaviours (Bandura, 2004). For the purpose of this thesis, it will relate to physical activity, exercise and sport behaviours and goals.

The construct that is seen as pivotal within the SCT is self-efficacy because of the impact it can have on health behaviour and the other constructs. In the health domain, self-efficacy is described as the confidence or belief an individual has to exercise control over their health habits (Bandura, 2004). The level of perceived self-efficacy will impact on an individual's goals and the outcomes that they will expect from their efforts. For example, the higher the perceived self-efficacy, then the higher the goals set, and the more positive the expected outcomes. In comparison, those with low-self-efficacy will set lower goals and believe that the efforts they put in will still result in poor outcomes (Bandura, 2004). Therefore, impacting on the perceived level of self-efficacy is always important when trying to influence health behaviours.

2.4.2.2 Outcome Expectations

Outcome expectations are another important construct of the SCT model, and Bandura (2004) suggest three major groups of outcome expectations: physical (e.g., improving fitness and strength levels from regular participation in physical activity), social (e.g., the idea of more social engagement from improved fitness and strength) and self-evaluative (i.e., the expectation of being able to cope with other challenges from being physically active on a regular basis; Beauchamp et al., 2018). Outcome expectations are representative of an individual's judgement of the possible outcomes that will result from performing, or not performing a certain behaviour. With reference to CP, the outcome expectations of being physically active for children with CP are likely to be different for their typically developing peers because of the impairments associated with CP. It is assumed in SCT that individuals will act in a way believed to lead them to positive outcomes, and thus avoid behaviours that they envisage will produce negative outcomes (Young et al., 2014). For children with CP, the fear of physical activity causing them pain is a barrier to engagement, however research has shown that participation in physical activity can reduce the perception of pain in children with CP (Whitney et al., 2018). The avoidance of being physically active because they expect it to cause pain indicates that the outcome expectations can, whether likely to be realised or not, have an impact on participation and therefore on the physical, psychological and social development of the individual.

2.4.2.3 Socio-cultural factors

Another important construct of SCT is socio-cultural factors. These are the facilitators and barriers to a behaviour and can impact the influence of self-efficacy on the behaviour. As

seen in the SCT model (Figure 2.7), the socio-cultural factors can influence the behaviour indirectly via goals. Barriers and facilitators can be physiological, psychological, social or environmental, and they can impact the goals set which in turn impacts the likelihood of performing or not performing the behaviour. For example, if an individual is attempting to stick to an exercise routine, and their level of efficacy to perform the exercise routine regularly will be impacted by various factors, (e.g., bad weather or tiredness), this may cause them to not carry out the exercise routine because they have a low level of self-efficacy. On the other hand, someone with a higher level of self-efficacy will focus more on positive aspects of the environment (i.e., social engagement at the gym) and this may help them overcome a certain barrier(s) to perform the exercise routine. Barriers and facilitators to physical activity behaviours have been described in greater detail in the following section (see section 2.5).

Bandura (1997) suggests that goals do not always have to be specific, and they can simply act as a guide. Someone with a higher level of self-efficacy is more likely to aim higher when setting goals and be more committed and go further to reach them in comparison to an individual with lower self-efficacy.

SCT is an important model to consider when looking at physical activity interventions because of the importance of self-efficacy when trying to influence health behaviours and there is evidence suggesting it is a useful framework to explain physical activity behaviour (Young et al., 2014). Furthermore, another aspect of the SCT that is important to note in relation to the design of physical activity interventions is continuous change, and influence, that the individual, environment, and behaviour can have simultaneously (Petosa et al., 2003).

A review by Beauchamp and colleagues (2018) highlighted some criticism of the SCT in relation to the physical activity domain. One criticism of the SCT is the role of the sociocultural factors, because it suggests that for an individual to take up a behaviour or make a significant behavioural change, they must overcome different barriers (i.e., personal, or environmental etc.). However, these factors may affect the way in which an individual participates in physical activity in a different way to the process as expressed by the SCT. For example, Bandura (2004) suggests a barrier to healthy living can come from poor health alone, and so in the case of the SCT, barriers to healthy living are to do with the structure of health systems. However, it would make more sense that the perceptions of poor health systems would affect the individual's self-efficacy to healthy living (Beauchamp et al., 2018). In essence, the SCT does not consider the causal link between personal, environmental, and social factors and self-efficacy, and only sees these factors as mediators between self-efficacy beliefs and goals.

A further criticism of the SCT is the issue of direction of causality between self-efficacy and goal pursuits (Beauchamp et al., 2018). Within SCT, it indicates that self-efficacy precedes outcome expectations and goals (Figure 2.6), but the relationship is not bidirectional suggesting that outcome expectations and goals do not influence self-efficacy. It suggests that goals are driven by self-efficacy and so an individual will only set a certain goal because they believe that they have capabilities to reach that goal in the first instance, or vice versa. However, within physical activity outcome expectations and goals can drive self-efficacy beliefs, for example, by providing incentives to be physically active (Beauchamp et al., 2018).

Self-efficacy is a prominent construct in terms of physical activity correlates and determinants (Beauchamp et al., 2018) and despite the two criticisms mentioned above, the SCT has helped to understand self-efficacy and how to influence physical activity behaviours. As self-efficacy is an important construct in the current research, it is important to understand the SCT model. In trying to influence an individual's behaviour, by encouraging them to participate in frame running, the influence of self-efficacy on them engaging in frame running is important, alongside the effect of socio-cultural factors and outcome expectations on the end behaviour of participation in frame running.

2.4.3 Transtheoretical Model

Though physical activity guidelines for children and adolescents with CP do exist, personal and environmental that can affect physical activity participation are not always addressed, therefore limiting, and restricting further their engagement and/or maintenance of physical activity and exercise behaviours. To help in the understanding of physical activity behaviours and what is required to design a suitable intervention to promote health behaviours for children and adolescents with CP, an understanding of the barriers and facilitators can be key. Thus, applying this knowledge to a stage-based approach can be an important intervention design strategy to improve physical activity behaviour. The Transtheoretical Model (TTM) is a popular theoretical framework when applied to physical activity interventions in an attempt to understand and change physical activity behaviours and promote participation. The TTM is a stage-based model, which means that it considers the individual at different stages from even before contemplating to maintaining it long-term. A review by Latimer and colleagues (2012) suggested stage-based approaches produced better results in terms of behaviour change. A knowledge of facilitators and barriers at each stage can be a helpful tool for addressing physical activity behaviour and designing interventions for children and adolescents with CP. Furthermore, having the knowledge of what can inhibit or restrict an active lifestyle at different 'stages' can help researchers alter

environmental and/or personal factors to help progress through the stages (Verschuren et al., 2013).

Originally developed in 1979 by Prochaska, the TTM has been applied to a range of behaviour change scenarios, including sedentary lifestyles which is a common problem for many individuals with CP, and physical activity (e.g., Prochaska & Velicer, 1997; Marshall & Biddle, 2001; Reedman et al., 2017). A systematic review by Hutchison et al. (2009) found that the physical activity behaviour change interventions applying the TTM predominately cite the stages of change as the dominant dimension of the model, but these are only one facet of the multidimensional design of the model.

2.4.3.1. Dimensions of the TTM – Stages of Change

Along with the stages of change, the other facets are self-efficacy, processes of change, and decisional balance (weighing up the pros and cons; Hutchison et al., 2009). The stages of change are the most prominent and well-known dimension of this model, as it recognises that behaviour change happens in a series of stages and not just an “all or nothing” scenario (Marshall & Biddle, 2001). In relation to physical activity behaviour change, the stages of change are labelled as precontemplation, contemplation, preparation, action, and maintenance, and each stage is outlined below (Prochaska & Velicer, 1997; Biddle, Mutrie, & Gorely 2015):

1. *Precontemplation* is the stage where people are not currently physically active and have no intention of becoming physically active in the foreseeable future.
2. *Contemplation* is the stage where people are currently not physically active, but they do intend to become physically active in the foreseeable future (usually measured within the next six months, Prochaska & Velicer, 1997).
3. *Preparation* is the stage where people are currently exercising a little, but not regularly or are intending to become physically active in the near future (usually measured within the next four weeks, Prochaska & Velicer, 1997). Individuals at this stage have normally taken significant action in the previous twelve months, and these are the individuals that physical activity behaviour change interventions/programmes target.
4. *Action* is the stage where people have recently made changes and are newly physically active, but it has been suggested that it is unstable stage and there is a chance of relapse (Biddle et al., 2015).
5. *Maintenance* is the stage which people are currently, and have been, physically active for a significant amount of time, usually measured at a minimum of six months.

The outlined stages suggest that there is a linear progression from one stage to the next, but it is thought that going back and forth between stages (cyclical model) may assist positive behaviour change in the long term as individuals learn from their relapses and mistakes. In relation to physical activity, it has been suggested that it is more of a cyclical progression as more than one attempt may be made to become physically active, to then reach the maintenance stage (Biddle, et al., 2015). For example, a child with CP in the contemplation stage and who is therefore thinking about becoming physically active, may identify environmental barriers (e.g., lack of suitable opportunities) and/or personal barriers (e.g., fear of being at risk of injury). These barriers can hold the individual at the same stage, or even cause the individual to move back to the precontemplation where the idea of becoming active is not even considered. On the other hand, to promote physical activity behaviour and progress to the preparation stage, a knowledge of facilitators can help to change behaviour (e.g., providing a suitable opportunity for the individual to participate, such as frame running).

2.4.3.2 Processes of Change

Another dimension to the TTM is the processes of change, and this is an important dimension as they are the strategies and activities that individuals will adopt and use to progress through the different stages (Prochaska & Velicer, 1997). There are ten basic processes of change and they can be grouped into “cognitive/thinking processes” or “behavioural/doing strategies” (Marshall & Biddle, 2009; Biddle et al., 2015). In relation to physical activity behaviour, cognitive/thinking processes would be most prominent during the action stage as it entails actions like increasing knowledge about physical activity and the benefits of physical activity participation, understanding the consequences of being inactive for the individual, and the potential negative impact to friends and family (Biddle et al., 2015). Behavioural/doing strategies would appear mostly in the maintenance stage because the individual has been physically active for a significant period of time and looking to continue. Therefore, individuals would employ strategies to aid this, including committing to plans of physical activity, and looking for ways to be physically active when barriers are encountered (Biddle et al., 2015). However, the processes of change are thought to appear across different stages of change with cognitive/thinking processes linked to the early stages of change, and the behavioural/doing strategies linked to the later stages of change (Marshall & Biddle, 2001).

2.4.3.3 Self-efficacy

Self-efficacy is another dimension of the TTM and was incorporated into the model from Bandura's self-efficacy theory (Bandura, 1977). Self-efficacy is referred to in the model as the confidence one has in order to deal with and overcome high risk behavioural specific situations without relapsing into unhealthy behaviours (i.e., inactivity; Prochaska & Velicer, 1997). The meta-analysis by Marshall and Biddle (2001) showed self-efficacy to increase across each stage of change, and therefore are more 'ready' to be physically active at each stage of change which is in line with the model.

Self-efficacy as a dimension in the TTM is important to note in relation to the CP population and physical activity participation due to the wide range of potential barriers they often face and have to deal with to be confident in their ability to be physically active in comparison to their typically developing peers (e.g., their belief in their ability to participate without being ridiculed by peers). Self-efficacy was previously mentioned as a key part of the SCT model, indicating that it can be a vital component of behaviour change. Furthermore, it shows that it can be an important factor to investigate in children and adolescents with CP to provide a better understanding of the effect physical activity participation can have on their self-efficacy.

2.4.3.4 Decisional Balance

The final dimension of the TTM is decisional balance, and this is the individual's weighing up of the advantages (pros) and disadvantages (cons) of behaviour change (Prochaska & Velicer, 1997). This can be a successful strategy in helping to change behaviour because it can highlight to the person why it is that they want to change and the negative costs from not changing. For example, perceived pros for becoming physically active would include feeling fitter and stronger from regular exercise, and a disadvantage could be the fear of feeling tired or being in pain. Biddle et al. (2015) suggested that during the initial stages of change, cons will outweigh the pros, hence the lack of intention to change, and the more progress through the stages of change, the more the pros will outweigh the cons. For the CP population, individuals can fear social exclusion, as 'being the odd one out' at a new group activity.

The application of the stage-based approach to understanding change in health behaviours, like smoking or weight loss, gave the initial confidence of applying the TTM to changing physical activity behaviour (Biddle et al., 2015). Using the TTM as a framework to guide and develop changes in physical activity behaviour has been popular, but the effectiveness of the physical activity interventions has been questioned (Hutchison et al., 2009). This is backed up by the critical review by Adams and White (2003) of the

effectiveness of 16 TTM based physical activity interventions. The researchers found that TTM based interventions were more effective for promoting physical activity change over a short term, but there was a lack of evidence supporting the long-term adherence to increased physical activity. Additionally, they reported difficulties in the measurement of the stages of activity change as they differed across the studies indicating the need for a more standardised measure to allow for easier identification and comparison.

2.4.3.5 Criticisms of the TTM

A criticism of TTM based physical activity interventions include the suggestion that physical activity behaviour is more complicated compared to single behaviours (e.g., smoking; Hutchinson et al., 2009), and therefore individuals could be in different stages of change at different times. Furthermore, the TTM is presented as a stage progress model, however, individuals can also move backwards and forwards between stages due to various factors that influence the change in behaviour (e.g., social environment, or self-efficacy; Elezim et al., (2019). Also, Adams and White (2005) suggested that physical activity behaviour is influenced by various factors that are not included in the TTM. They suggest that the TTM ignores social and environmental factors, and the impact that they can have on physical activity behaviour change, and solely focuses on personal motivation (i.e., weighing up the pros and cons). Finally, the difficulty in being able to accurately identify which stage of change somebody currently occupies was seen as another criticism (Hutchinson et al., 2009). This causes difficulty for researchers as they are unable to proceed with the correct information when trying to increase physical activity behaviour impacting the desired outcome. The systematic review by Hutchinson et al. (2009) suggested that for more comprehensive conclusions relating to the efficacy of the TTM based interventions, and to have a complete understanding of the dimensions, future research should be done on all four of the dimensions of the model collectively and individually to better understand the interactions.

2.4.3.6 Summary

Considering a progressive stage model of behaviour change is relevant to the current thesis as the children and adolescents with CP can be viewed at the different stages when they are introduced to frame running. At the different stages, individuals may be facing more complex behaviours to overcome to move from one stage to the next, but the TTM framework may not be entirely relatable to them as for some individuals with more severe CP, who have cognitive impairments, are unable to make decisions for themselves. Therefore, the movement from one stage to the next is out of their control and whether they are willing to

be physically active or not is not up to them but their parents and/or guardians. A parent's influence can be key in promoting physical activity, and this can be done in numerous ways, including providing transport to activity, the organisation of activities, and a showing of positive attitudes towards physical activity (Elezimi et al., 2019). However, the decisional balance for parents with children with severe disabilities who are unable to make their own decisions may be tipped towards less or no participation due to higher levels of risk, time, and financial costs.

Previous research applying the TTM suggests that physical activity programmes should aim to increase self-efficacy and the perceived benefits of physical activity as they are both factors that can help to improve engagement and adherence to participation (Elezimi et al., 2019). It is an aim of the current research to investigate the development of self-efficacy and perceived benefits of physical activity through participation in frame running. However, despite the TTM being important for behaviour change, including physical activity and exercise behaviour, it will only be applied as one of the models to aid interpretation of data rather than as a fundamental model for the design of and application of an exercise intervention, as the identification of stages of activity may not be suitable for the population.

The complexity of behaviour changes and promoting physical activity participation can contribute to an already heightened risk of inactivity for individuals with CP related to the problems with movement and posture experienced (Nooijen et al., 2014). Thus, in an attempt to encourage a higher level of physical activity participation in individuals with CP, an understanding of the barriers and facilitators to participation including social and environmental factors are discussed in the following section.

2.5 Barriers and Facilitators of Physical Activity Participation in Children with CP

Although this may not relate to all individuals with CP, research has indicated that disparities exist in the participation levels in physical activity between children with disabilities and those without (Johnson, 2009). This can partly be explained by barriers to participation in physical activity for children and adolescents with CP, including physical (e.g., limited mobility), psychological (e.g., fear of pain and fatigue) and social (e.g., social exclusion) factors. Alongside the barriers, facilitators of physical activity are also an important area of study, as they can provide information about how to enhance and encourage physical activity in children and adolescents with CP. Numerous reviews of the literature have indicated various barriers and facilitators to physical activity, and can be grouped in various ways but for the purpose of this section they will be identified as personal barriers and

facilitators, and environmental barriers and facilitators with subcategories (Buffart et al., 2009; Verschuren et al., 2012; Shields et al., 2012; Verschuren et al., 2013; Conchar et al., 2016; DeFazio & Porter, 2016). The understanding of barriers and facilitators is necessary for researchers as this can inform the design of effective interventions for promoting participation in children and adolescents with CP. Thus, the following sections will consider each of the categories of barriers and facilitators to provide an insight into physical activity for young people with CP.

2.5.1 Personal Barriers & Facilitators

Personal barriers to, and facilitators of, physical activity participation in children and adolescents with CP can be subcategorised into physiological and psychological factors. Firstly, the physiological factors will be discussed, followed by the psychological factors.

2.5.1.1 Physiological Factors

The main barrier to participation in physical activity/exercise can be the child's disability, and the associated impairments. Given the wide range of mobility impairments that children and adolescents with CP can have, the physical ability of each individual will have a different impact on their participation levels. However, in general, there will be physical limitations, and alongside this, a lack of athletic ability, skills, and coordination, and a lack of energy or feeling of fatigue, which can all be deemed as barriers to participation. As previously mentioned, children with CP undertake low levels of physical activity (Shields et al., 2012), and therefore can be in a poorer health and/or physical condition compared to their typically developing peers. If they are unable to keep up with or need more time to complete a task compared to their peers, then they may be discouraged to take part. Furthermore, the gap in strength and ability between children with and without disabilities is likely to increase as they grow older, increasing the likelihood of low levels of participation (Conchar et al., 2016).

Individuals that are participating in physical activity may find themselves undergoing multiple surgical procedures, which then require a recovery period where little to no participation in physical activity is possible (Conchar et al. 2016). The interruptions due to the surgery and recovery period can cause participants to lose the strength, fitness, and skills that had been gained, and this can discourage and demotivate individuals to participate. This can continue in a negative cycle due to a lack of motivation and belief in their capability to participate in physical activity, and therefore continued poor health and a feeling of fatigue. It is therefore important to identify suitable options that allow an increase in the amount of time individuals spend physically active, and in turn the potential

psychological impact on self-belief and motivation for continued physical activity participation in this population (see theoretical underpinning; section 2.4).

The barriers to physical activity are much more widely researched in comparison to the facilitators, but some facilitators have been shown to be successful in improving participation (Kang et al., 2007). Understanding the possible health benefits of exercise can play an important role in children's decision to become physically active. The knowledge of being able to improve and maintain fitness, muscle strength, and functional independence are very important when it comes to promoting and engaging in physical activity (Buffart et al., 2009). Other reasons include changes that can be seen in the individual's physical appearance including weight loss and muscle building (Buffart et al., 2009). Important facilitators for the cohort with CP are participants using wheelchairs experiencing improved wheelchair skills, and ambulant participants can experience improved walking ability.

It is important to understand that some of the barriers are more permanent and difficult to change than others. For example, a child with more severe CP may not be able to change their physical abilities to a large extent (Verschuren et al., 2013). This highlights the importance of increasing participation via external methods (e.g., more suitable and appropriate opportunities for less ambulant children with CP to be physically active at a higher intensity) or trying to increase participation via increasing levels of motivation to participate by looking at the reasons why someone participates or might participate in physical activity (Teixeira, et al., 2012).

2.5.1.2 Psychological Factors

The positive influence of psychological health from physical activity participation is well documented, however, the negative impact on psychological health of contemplating becoming physically active and participation is not always considered. Prior to taking part there is the possibility of feeling insecure, anxious, ashamed, being seen as an outsider, and not understanding that physical activity can be fun (Verschuren et al., 2012), and all of these factors can demotivate and discourage participation in physical activity. The presence of a cognitive impairment (Verschuren et al., 2012; 2013) is also a barrier to participation as many individuals may find it difficult to understand and learn the necessary skills for the activity. If they have a less visible mobility impairment (e.g., GMFCS level I or II), their ability could be overestimated by a coach or teacher despite the presence of a cognitive impairment. Not understanding or accepting the full extent of their impairment can cause them to feel constrained and failed by their bodies, which can influence choices of activities and furthermore the levels of the activity in which they participate.

Low levels of physical activity may impede individuals from gaining the cognitive benefits from physical activity (Ploughman, 2008). For children with CP, the differing levels

of cognitive impairment can lead to various activity limitations that in turn may affect psychological factors, such as self-concept, psychological well-being, and self-esteem. If these are low, then they may act as further barriers to participation.

Qualitative data from research into facilitators of physical activity showed an individual's attitude towards becoming physically active is important because if someone wants to be active, and believes that they can, then they will (Buffart et al., 2009; Conchar 2016). However, it is unlikely to be that simple because, as mentioned (and will be discussed in coming sections), there are barriers to becoming physically active and then maintaining those levels, that are out with people's control. Having a positive attitude towards becoming and being active is beneficial, but it is only one aspect of the psychology of being physically active. The motivational factors that act as facilitators, including the drive of wanting to win, or achieve a certain goal, being challenged physically, and gaining a sense of enjoyment and fulfilment from participating, which are discussed in relation to theoretical frameworks (see section 2.4).

If the child has the belief that participating in physical activity will provide health benefits and can make them feel more relaxed and fulfilled, then they are more likely to be encouraged to participate. The presence of attitude and motivational factors are not dissimilar to results from typically developing children (van der Horst et al., 2007). The knowledge of these psychological facilitators is important to consider when planning an intervention to encourage and engage children in physical activity participation. Of course, it is not as straightforward as setting out a fun session with everybody achieving what they want, especially with the CP population, as the ability of participants will vary. However, the understanding of these psychological barriers and facilitators can help shape a physical activity intervention or session with the aim of increasing and maintaining levels of participation.

2.5.2 Environmental & Social Barriers and Facilitators

As well as considering the personal barriers and facilitators to participation, the social and environmental factors associated with participation can have an equal, if not bigger influence on the levels of participation for many children and adolescents with CP.

Parental factors can have a big impact on the extent to which a child will participate or whether a child will even try out an activity at all. If a parent is not interested in sport or exercise themselves or does not believe that physical activity is important, then they could struggle to understand the importance and/or potential physical and psychological benefits for their child. Parents of children with CP already face challenges in day-to-day life (Verschuren et al., 2012; Dieleman et al., 2019), and they may not be willing and/or able to

invest more time, money or energy into going to training sessions. The secondary impairments that are associated with CP, for example epilepsy and poor balance, can also put the child at greater risk of injury, or a particular activity may be perceived to be too risky, and then this can discourage parents from allowing them to participate. If their child does participate at some point, the parent may be dissatisfied with the environment (possibly too far away, or the facilities are unclean), or find it difficult to see their child struggling with the sport, and for these reasons they may be discouraged from allowing their child to take part again (Verschuren et al., 2012). Parents can also feel like their child is not going to be accepted in society, and this will carry through to organized sports (Verschuren et al., 2012).

One of the biggest environmental barriers to physical activity for children with CP, particularly those with limited walking ability, is the limited choices of suitable activities, or if they are available, not being able to access them as they are not 'open' to disabilities or there is a waiting list (Verschuren et al., 2012). Sometimes it can be related to a lack of awareness of suitable sport and physical activities, but this can be counteracted by increasing the awareness of the suitable sports and activities and encouraging participation via schools, clubs, or health care professionals (e.g., physiotherapists). If a suitable sport is found, then there are still practical feasibility barriers to overcome. Finding the time to participate can be difficult, along with financial restrictions, and a lack of access to appropriate transport can all be barriers to participation. The members of staff at the sports or community centre may not be aware of the complex needs of children with CP, and therefore may not be trained appropriately to, or open to working with this population. At the sports or community centre, the availability of appropriate facilities for changing or mobility equipment is a further barrier to participation. The barriers involved in having children with CP participate in physical activity and exercise present a considerable challenge to participation (Buffart et al., 2009).

Despite these barriers, parents can also facilitate the participation of their child in physical activity and exercise. Having an understanding of the benefits of physical activity will help parents to encourage their child to take part because they are aware of the potential positive outcomes. The parents can also be key in helping to create the opportunities for the child to be physically active, because the child may feel anxiety or fear of starting something new and the encouragement from parents can ensure that their child is participating in physical activity (Shields et al. 2012).

The social environment can influence the participation of children with CP. On the negative side, both parents and children may feel they are not being accepted by their peers, or other parents, which can result in bullying and taunting, and therefore discourage continued participation (DeFazio & Porter, 2016). On the other hand, the potential development of social and emotional factors for children with CP from participation in

physical activity is very important. Taking part in physical activity allows children to have the opportunity for social interaction and build friendships and relationships, out with the home environment. They can feel like part of a team, have a sense of belonging, and feel like they are accepted by peers, including those without disabilities. Furthermore, having children and adolescents with disabilities be physically active may spread awareness of physical activity to children without disabilities. This may lead to greater facilitation of physical activity as children, adolescents, and parents without disabilities are more aware, and the threat of bullying and taunting may be reduced, thus lowering the risk of mental health disorders for children with CP (Whitney, et al., 2019). The variety of social and environmental factors that influence the ability and desire of a child's participation, whether it be a barrier to, or facilitator of physical activity are all important to consider, especially when designing interventions for children and adolescents with CP.

As discussed in this section, there are a wide range of both barriers and facilitators for young people with CP which can help with or restrict their participation in physical activity. For the individuals that wish to be active, but have more limited mobility, the availability and access to suitable adaptive sports can be one of the main barriers. However, promotion and engagement in physical activity is complex, and not as simple as providing a suitable and accessible opportunity to participate.

2.6 Cerebral Palsy & Inactivity

The amount of sedentary time compared to time spent engaged in physical activity for children with cerebral palsy is a concern (Verschuren et al., 2016). Carlon et al. (2013) carried out a systematic review of the literature relating to the differences between youth with cerebral palsy and their typically developing peers, and their levels of habitual physical activity. The results from the review showed that across all levels of motor function (GMFCS levels I to V) and ages (5 to 18 years), the participation of CP children and adolescents were 13% to 53% less compared to their typically developing peers. The review also indicated that in relation to physical activity guidelines (one hour of moderate to vigorous intensity of physical activity per day for individuals under the age of 18; WHO, 2016), the levels of CP participation were 30% less. The lower levels of physical activity combined with the movement and posture problems experienced by children and adolescents with CP, such as muscle spasticity, increase the risk of sedentary behaviour for children and adolescents with CP, contributing to negative health consequences (e.g., cardiovascular disease; Carlon et al., 2013).

Certain impairments in children with CP, like poor balance and motor function, muscle spasticity and weakness, and cognitive impairments, make it more difficult for them

to participate regularly in physical activity or exercise, especially at a level of higher intensity necessary to maintain and improve physical fitness. The health and social benefits of physical activity for children without disabilities is well recognized (Janssen & LeBlanc, 2010), with a growing understanding of the benefits of physical activity for children and adolescents with disabilities including CP (Andersson & Mattsson, 2000). Impairments from physical inactivity can contribute to a vicious cycle where deconditioning leads to higher levels of physical inactivity and physical and/or mental de-conditioning, and in turn, impairments from the disability can be worsened (Damiano, 2006). Often children and adolescents with CP are already at a disadvantage in comparison to their typically developing peers with respect to reaching optimal levels of physical functioning often due to developmental impairments associated with CP. Physical de-conditioning can include increased levels of fatigue (Berrin et al., 2007), reduced overall fitness and strength levels. The negative physical impact can continue in a cycle of deconditioning because higher levels of fatigue alongside not feeling physically fit can be de-motivating factors for participating in physical activity.

For optimal cognitive, social, physical, and emotional development in children, physical activity is deemed a key component (Verschuren et al., 2016), emphasizing the increasing importance for participation in physical activity for children with CP. Motor ability is key for interaction with the physical world, and the limiting nature of physical impairments in CP can have negative effects on a child's development. With less opportunities to participate in physical activity or exercise situations, children with CP can miss out on gaining appropriate and valuable social and emotional experiences, which can curtail their cognitive and social development. A lack of potentially vital social experiences can lead to children with CP developing low levels of self-efficacy and self-concept, in particular related to their athletic and physical competence (Russo et al. 2008).

Self-concept is a determinant of self-esteem, and it refers to how an individual may view themselves (Shields et al., 2007). With self-esteem having such a significant role in personal development, the way in which somebody views themselves has an impact on their self-esteem and in turn affects their personal development. Individuals with low levels of self-esteem are possibly at higher risk of developing mental illness and suffering with poor physical health in comparison to individuals with high self-esteem (Russo et al. 2008). To combat this, physical activity programs prescribed by therapists and health care professionals have been found to have a positive impact on both perceived physical appearance and athletic competence in children with CP (Russo et al. 2008). The negative impact of not participating in physical activity on one factor (e.g., self-efficacy or self-concept) can have an indirect effect and be detrimental to psychological, physical, and QoL of all children and adolescents, particularly for children and adolescents with CP (Maher et al., 2015).

Given the barriers explored previously (see section 2.5), it is therefore crucial that we consider effective alternatives for children and adolescents with CP to access physical activity and exercise. Furthermore, the level of physical activity in childhood tends to relate to the level of physical activity in adolescents and adulthood (Waltersson & Rodby-Bousquet, 2017), which further indicates the necessity for suitable and effective alternatives for regular participation in moderate to higher intensity physical activity and exercise in young people with CP.

The health risks associated with physical inactivity for children and adolescents with CP highlight the importance of being physically active, however for individuals who wish to be active, but are restricted by limited mobility, the availability and access to suitable adaptive sports can be one of the main barriers. Frame running has the potential to provide an opportunity for individuals with CP, including those who have limited or no walking ability, to improve the physical fitness and reduce the risk of disease by enabling them to participate in moderate to high intensity exercise (Bolster et al., 2017). Thus, the following section will critically review the existing frame running literature and aim to identify any gaps in the knowledge/literature.

2.7 Frame Running & the Existing Research: A critical overview

In previous sections, the importance of participating in physical activity was highlighted, alongside the possible consequences of physical inactivity, particularly in youth with cerebral palsy. The array of barriers and facilitators are important to take into account when trying to promote participation in physical activity. Some of the more prominent barriers for youth with disabilities, apart from the physiological and psychological factors, relate to the practical feasibility of participating, the opportunities for sport and physical activity, and the facility/programme factors (DeFazio & Porter, 2016). Youth with CP, especially those who are more restricted by mobility or confined to a wheelchair, have substantially fewer opportunities to participate in physical activity. The limited choices of suitable activities can mean that the individual has no interest in the available activities or have to travel further to access suitable activities. One suitable alternative, as mentioned in the previous chapter, is frame running, and it can provide individuals who have limited to no walking ability with the opportunity to be physically active at a higher intensity.

With the continuing growth and development of frame running, the inclusion of frame running as a Paralympic event in the future would be likely to have a positive impact on the sport with exposure on the world stage of Paralympic sport. The evidence base for frame running is currently very limited. There are very few studies on the effect of participation of frame running with respect to health and well-being. Studies by O'Donnell, Verellen, Van de

Vliet, and Vanlandewijck (2012), Van der Linden, Jahed, Tennant, and Verheul (2018), and Hjalmarsson, Fernadndez-Gonzalo, Lidbeck, Palmcrantz, Jia, Kvist, Pontén, and von Walden (2020) were some of the first studies to look at performance outcomes for CP athletes using racerunners, and a critical overview of the research will follow.

2.7.1 Exercise Testing of Frame running Athletes

O'Donnell et al. (2010) was the first peer-reviewed publication on Frame running. The nine participants with CP had varying severity and completed a 12-minute Cooper test on a racerunner. A Cooper test is a test of an individual's physical fitness, as participants are to run as far as possible for 12-minutes (Cooper, 1968). The cooper test was adapted for individuals with CP, by measuring the total distance travelled on the frame running tricycles. Heart rate, blood lactate, stride frequency, step length, and pace of the athletes were measured throughout the cooper test. Total distance travelled was also measured for each participant, and the results showed that the performance on the racerunner was dependent on functional ability of each participant. At the beginning of the test, there was an immediate increase of heart rate, and high heart rate was maintained throughout the Cooper test. This highlights that frame running can provide the opportunity for individuals to exercise at high intensity. There were no significant changes in participants' stride frequency, step length, or pace throughout the 12-minute test. These results indicate that the participants did not show significant adverse effects to their muscle tone (lower limbs only) when they are fatigued, again showing the benefits of participation in frame running.

2.7.2 Impairment impact on Frame running performance

Van der Linden and colleagues (2018) conducted the first study assessing the level at which the impairment of body functions affect performance within frame running. Twenty-seven of the participants had CP with a range of CP severity (GMFCS levels II to V). Using a range of impairment measures, the potential impact of each impairment on performance was measured over a 100-metre race. The researchers found that lower limb spasticity, higher passive knee extension, higher isometric leg strength, and higher selective voluntary motor control were all associated with better performance of frame running athletes. This study provides a positive notion regarding frame running, with the inclusion of more severely impaired individuals with CP, GMFCS IV and V, as individuals at this end of the severity scale are more likely to struggle to participate in physical activity and exercise. In comparison to other activities, such as strength or interval training (which are discussed in section 2.8), frame running can provide a feasible and accessible option to those individuals who are motivated to become physically active but are unable to due to different barriers.

The evidence regarding the effects of physical activity and exercise participation within more severely impaired population in comparison to their less impaired and non-disabled peers is also less well known, possibly because of the difficulties of carrying out research with them. However, Van der Linden and colleagues (2018) have shown frame running can provide an opportunity for researchers to look further into the physical, social, and psychological effects of higher intensity exercise in the more severely impaired individuals with CP.

Van Schie et al. (2015) published a conference abstract investigating whether or not frame running bikes would be suitable and enjoyable for exercise and leisure use for children with CP and similar diagnosis. Van Schie and colleagues (2015) carried out 4 projects relating to frame running which included a feasibility study of using the racerunner, a comparison study between using a racerunner and a wheelchair, a reliability test of the 6-minute exercise test using a racerunner, and a pilot study for training with a racerunner.

The feasibility project studied 58 children with CP and similar diagnosis using racerunners and their therapists completed a questionnaire. The qualitative data showed that the majority of children enjoyed frame running but there were some issues highlighted, mainly performance related with problems steering, and trouble with moving the lower limbs. The racerunner versus wheelchair project is important as it provides some evidence, similar to O'Donnell et al. (2010), for using a racerunner as a device for exercise, recreation, and training. Eight participants with CP (GMFCS functional levels II to IV) took part in a 6-minute exercise test, and the distance covered and mean heart rate at the end was compared between using a wheelchair and a racerunner. The mean difference in heart rate at the end of the 6-minute test was significantly higher when using the racerunner (mean HR of 175) than when using a wheelchair (mean HR of 121), and participants recorded heart rates during frame running which are in line with improving physical fitness (minimum HR of 121 to maximum 192 using a racerunner, vs minimum HR 84 to maximum HR 159 using a wheelchair). This research suggests that a racerunner provides higher intensity exercise than a wheelchair, most likely due to the use of larger muscle groups in frame running than wheelchair racing. It should be noted that many individuals with CP who participate in frame running are unable to propel a wheelchair themselves, so for those individuals there is no choice.

Testing the reliability of a 6-minute racerunner test (6MRT) is important as there is a need for a valid and reliable field-based test for children who have limited or no walking ability (Verschuren et al., 2013). Thirty-eight participants with CP, GMFCS levels III to IV, completed a 6MRT once a week over a 3-week period and researchers measured the distance covered on the racerunner and heart rate after six minutes. There was good reliability of the 6MRT, and this has been backed-up by Bolster et al. (2016) who also looked at the reliability and construct validity of the 6MRT in children with CP. The results from both

van Schie et al. (2015) and Bolster et al. (2016) found good reliability in the 6MRT showing it can be used as a method for evaluating the capability of an individual with CP, and the high test-retest reliability found by Bolster et al (2016) indicates it can be used to track initial progress when using a racerunner.

2.7.3 The Effects of Frame running Training & Enjoyment

Similar to Van Schie et al., (2015), Hjalmarsson et al. (2020) used a 6MRT as part of their protocol when investigating the effects of frame running training on cardiorespiratory endurance, heart rate and muscle mass. The study involved 15 participants with CP (GMFCS I, II, III, and IV), aged between 9 and 29 years old, who participated in two frame running training sessions a week over a 12-week period. The participants underwent the 6MRT, physical examination of the lower limbs to investigate participants' range of motion in different areas, and an ultrasound to measure skeletal muscle thickness. The ultrasound and physical examination were carried out before and after the 12-weeks of frame running, and the participants performed the 6MRT on four occasions, before and after, and twice during the frame running sessions at week four and week eight. The main finding from Hjalmarsson et al. (2020) indicated benefits for cardiorespiratory fitness from participation in frame running with all participants improving their performance on the 6MRT. The average improvement was 34%. Furthermore, participants showed a gain in the hypertrophy of the calf muscles post 12-weeks frame running. These results show the positive impact that frame running participation can have on physical health for individuals with CP across a 12-week training period. However, it is important to note the training frequency of twice a week may be difficult for individuals with CP to adhere to, particularly for those who are highly reliant on others for transport to and from training sessions, or there may be financial barriers to participating at that frequency. Verschuren et al. (2016) has recommended that, due to the often-high level of sedentary behaviour and physical inactivity within individuals with CP, initially a low dose of physical activity (i.e., once a week, which does not meet the minimum guidelines) would be sufficient to show improvements in cardiorespiratory fitness.

Similar to van Schie et al. (2015), who reported children to have enjoyed frame running, Bryant and colleagues (2015) examined if the frame running bikes were a suitable and enjoyable form of exercise and recreation for children with CP. The recognition of frame running as an enjoyable activity for children with CP is important for future participation and promoting frame running as a physical activity for this population. Furthermore, as previously discussed (see section 2.4), the construct of enjoyment can play an important role in increasing intrinsic motivation to participate, thus enhancing children's' capacity, confidence and competence (Rosenbaum & Groter, 2011). Both studies exploring enjoyment, van Schie

et al (2015) and Bryant et al. (2015) are published conference abstracts, so results should be interpreted with caution.

As well as enjoyment, Bryant and colleagues (2015) investigated if there were any changes in QoL, gross motor function, and bone health from using the racerunner. Children with moderate to severe CP (GMFCS level IV and V) used frame running bikes three times per week over a 12-week period in a school setting and were measured at baseline and at the end of the intervention. Using the Gross Motor Function Measure (GMFM-88D) to measure ability of the participants, the researchers found a significant improvement in their ability to use the racerunners over the 12-week intervention. The bone density results showed a statistically significant increase in the participants' bone health after the 12-week intervention, showing the potential for benefits after a relatively short period of participation. The quality-of-life data showed no significant changes across any domains of the CP -QoL. However, the qualitative interview data indicated that the children and physiotherapists thought that the racerunners were a feasible and enjoyable mode of physical activity.

It is important to note the inclusion of the participants classified GMFCS level V (n=7), indicating they are normally transported in a manual wheelchair or use a powered wheelchair and have difficulty keeping their head and trunk up against gravity and controlling their arms and legs. The inclusion of these participants in the study shows the accessibility frame running provides to that population in comparison to other modes of physical activity or exercise. It highlights the opportunity provided by frame running to gather evidence on this population and the effects of physical activity and exercise participation. Furthermore, Hjalmarsson et al., (2020) have shown frame running as a favourable and effective training option for physical activity and exercise for individuals with CP to improve their cardiorespiratory fitness and muscle mass in calf muscles. As with the previous frame running research, the sample sizes of the studies are small due to the rarity of the sport in comparison to others and are likely to be convenience samples due to the difficulties in recruiting participants from this population. Therefore, results have to be interpreted with caution and may not be generalised.


Even though the current evidence base behind frame running is limited, there are positive results already evident. The versatility of the racerunner is a major benefit as it can be used in a therapy and school (Bryant et al. 2015), social and sports setting (van Schie et al. 2015), and can be used to promote a healthy lifestyle as well as improve physical fitness (van Schie et al. 2015; Hjalmarsson et al., 2020), particularly for children with limited or no walking ability. To date, the majority of frame running research has looked at physical and performance outcomes of participants, with no high-level evidence published that investigate the potential psychological and social effects to youths with CP when participating in frame running, and this is a gap which this thesis aims to address. All future research involving






frame running will be advantageous to provide a more substantial evidence base for frame running as a disability sport and a potentially viable alternative physical activity and sport option for widening participation.

2.8 Frame running Compared to other Physical Activity and Sports

An important difference between frame running and other disability sports available to individuals with limited or no walking ability is that it provides them with the opportunity to propel themselves forward using their legs, something they may not have been able to do before. Children with CP who have limited or no walking ability are normally classified between GMFCS functional level III and V, with level III being able to walk using a hand-held mobility device or self-propelled wheelchair depending on their upper limb function; level IV relying more on being pushed in a wheelchair or may have the ability to use a powerchair themselves; and level V having the highest level of impairments, with upper and lower limbs severely impaired, and children having no means of independent mobility and are confined to a wheelchair (Palisiano et al. 1997). For these groups of children, the availability of opportunities for physical activity and exercise become fewer with the higher level of impairment. This is particularly evident in the number of activities that provide the opportunity for a high intensity cardiovascular exercise, and this is highlighted in the table below showing the sporting opportunities available for children with CP at each of the five levels of GMFCS (CPISRA, see Table 2.3). The sports listed under GMFCS functional level V show that there are no opportunities for children to participate in physical activity bringing them out of being sedentary, apart from frame running. The plethora of sports available for children with CP, particularly GMFCS functional levels I and II, shows that the opportunity for participation in physical activity is there, but as previously discussed, having the opportunity for activities alone will not solve the problem of promoting and encouraging participation in physical activity for children with CP.

Table 2.3 Cerebral Palsy International Sports & Recreation Association (CPISRA) activity list (<https://cpisra.org/> - reproduced with permission)

 Cerebral Palsy International Sports and Recreation Association

	 GMFCS I	 GMFCS II	 GMFCS III	 GMFCS IV	 GMFCS V
Athletics	Ambulant Athletics – Track and Field	Ambulant Athletics - Track and Field	RaceRunning Seated Athletics - Track and Field	RaceRunning Seated Athletics - Track and Field	RaceRunning Seated Athletics – Track and Field
Swimming	Unaided swimming	Unaided swimming	Unaided swimming		
Football	CP 7-a-side Football	CP 7-a-side Football	Frame Football	Frame Football Powerchair Football	Powerchair Football
Racquet Sports	Standing Badminton Standing Table-Tennis Standing Cricket	Standing Badminton Standing Table-Tennis Standing Cricket Wheelchair Tennis	Wheelchair Badminton Wheelchair Tennis Seated Table Tennis Wheelchair Cricket	Wheelchair Badminton Wheelchair Tennis Seated Table Tennis Table Cricket	Table Cricket
Individual Sports	Taekwondo Para-Cycling (2/3 wheeler) Para Triathlon Equestrian CP Bowls Standing Archery standing Golf Para Shooting Powerlifting Seated Fencing	Taekwondo Para-Cycling (2/3 wheeler) Para Triathlon Equestrian CP Bowls Standing Archery standing Golf Para Shooting Powerlifting Seated Fencing	Para-Cycling (2/3 wheeler) Seated Fencing Para Triathlon Boccia Equestrian CP Bowls Seated Archery seated Wheelchair Slalom Para Shooting Powerlifting	Para-Cycling (2/3 wheeler) Seated Fencing Boccia CP Bowls Seated Archery seated Powerlifting Trap Driving Wheelchair Slalom	Wheelchair Slalom Boccia Para Shooting Trap Driving
Team Sports	Sitting Volleyball Ambulant CP rugby Ambulant Netball	Sitting Volleyball Ambulant CP rugby Ambulant Netball	Sitting Volleyball Wheelchair Basketball Wheelchair Rugby	Sitting Volleyball Wheelchair Basketball Wheelchair Rugby	
Winter Sports	Standing Alpine Skiing Standing Nordic Skiing Snowboarding Para Ice Hockey	Standing Alpine Skiing Standing Nordic Skiing Snowboarding Para Ice Hockey	Sitting Alpine Skiing Standing/Sitting Nordic Skiing Wheelchair Curling Para Ice Hockey	Sitting Nordic Skiing Wheelchair Curling	
Water Sports	Rowing Sailing Canoeing	Rowing Sailing Canoeing	Rowing Sailing Canoeing	Sailing Canoeing	Assisted Sailing

GMFCS Illustrations Version 2 © Bill Reid, Kate Willoughby, Adrienne Harvey and Ken Graham, The Royal Children's Hospital Melbourne ERIC131050

 www.cpisra.org

The evidence base for CP activities has expanded rapidly (Novak et al., 2013) providing a wider range of intervention possibilities. Generally, an intervention aimed at children and adolescents with CP will aim to encourage participation, improve body function and structure, functional skills, and reduce activity limitations (Morgan et al., 2016). Interventions are dependent on the individual or the group of individuals, and therefore can vary across domains, including physiotherapy-based interventions (Antilla et al., 2008), medication and surgical interventions (e.g., orthopaedic surgery; Sakzewski et al., 2009), motor learning interventions (Bar-Haim et al., 2010), and various physical activity interventions including walking, swimming, hippotherapy, strength training, and overall fitness training (Novak et al., 2013; Fleeton et al., 2020). For the purpose of this chapter the focus will be on the different physical activity and exercise interventions to allow for comparison with the current frame running research. It will also indicate the potential of frame running and how it could fill a gap in disability sport, by providing opportunities for children and adolescents with limited or no walking ability to be more functionally independent and/or physically active.

The majority of physical activity and exercise programmes that are designed for young people with CP have focused on improvement of motor function (e.g., mobility, functionality), and this may be because motor impairments are one of the most prominent for this population. On the other hand, there may have been fear to involve children with CP in exercise programmes focusing on other physical outcomes such as cardiovascular fitness, or strength, as it was assumed participation may have adverse effects on the participant (e.g., increased spasticity; Scholtes et al., 2008). However, subsequent research has caused a change in direction for exercise programmes for children and adolescents with CP, as evidence of benefits and/or lack of adverse effects, from participation in a variety of exercise and physical activity programmes is documented. For example, strength training was thought to cause adverse effects on spasticity of young people with CP, and therefore was avoided. However, a systematic review by Dodd, Taylor, and Damiano (2002) shows strength training for children with CP to improve muscle strength without adverse effects, suggesting strength training is a feasible option of activity for young people with CP.

On this basis, the following sections will look at the different physical activity/exercise interventions available for young people with CP. Furthermore, the importance of having these options available and the benefits of participating in these physical activities and exercise programmes for young people with CP will be discussed, thus highlighting the physical activity opportunities that are available, but also what frame running can offer in comparison to the other activities.

2.8.1 Strength Training

Strength training for children and adolescents with CP went through a stage of being discouraged because there was an assumption that it would increase muscle spasticity and have a negative impact on the movement pattern of the participants (Bobath, 1971; Scholtes et al., 2010). However, research has shown that children and adolescents can benefit from strength training in relation to their spasticity, flexibility, and movement (Verschuren et al., 2007). Strength training for children and adolescents with CP could be important because it can target the muscle weakness, whether it is in the upper or lower extremities, which comes from reduced levels of physical activity associated with functional impairments with CP. Strengthening the weaker muscles has the potential to improve movement in individuals with CP, and therefore increase the level of physical activity associated with this population. Alongside the potential physical benefits of strength training, research has shown that there is some evidence that participation in strength training may lead to psychological benefits in children with CP (Darrach et al., 1999; Dodd et al., 2004; Unger et al., 2006).

2.8.1.1 Strength Training & Daily Activity Performance

A single-blind RCT by Bania, Dodd, Baker, Graham, and Taylor (2016) looked at the impact of progressive resistance training (PRT) against usual care on daily levels of physical activity in adolescents with CP over a 12-week intervention period. Participants of the intervention group completed two sessions a week of progressive strength training in a supervised community setting, while the control group continued with their regular care. An activity monitor was given to all participants to measure their daily physical activity levels. The results from Bania indicated no significant increase in recorded daily physical activity levels in young adults with CP in either the intervention or control group. Furthermore, previous research has reported improvements in the perception of strength and ability to perform daily activities in adults with CP following a 10-week PRT intervention (Allen et al., 2004). The difference between the two results may in part be due to the fact that Allen, Dodd, Taylor, McBurney, and Lakin (2004) conducted a qualitative study comprising of semi-structured interviews, compared to the quantitative data from Bania et al. (2016). Improvements in areas such as strength and functional ability may be expected following a PRT intervention, particularly in the CP population due to the mobility impairments and muscle weakness and spasticity associated with CP, and therefore it would translate into improved levels of daily physical activity. However, when objectively measured in Bania et al. (2016), levels of daily physical activity were unaffected in both the intervention and control group despite what participants may have believed post-intervention. A PRT training programme may also be difficult for the CP population to participate in because of mobility impairments, and this is indicated by the inclusion of GMFCS levels II and III participants, suggesting it is not suitable for more impaired individuals.

The quantitative data from Bania et al. (2016) reporting no significant change versus a perceived improvement post PRT intervention suggests that how someone is feeling may not translate directly into significant statistical change. Furthermore, it shows the psychological aspect in relation to being physically active, and the positive impact it can have on their perceived strength and functional ability. Allen et al. (2004) reported the main benefit from the PRT intervention was the enjoyment and social interaction the participants gained, which can aid motivation and adherence, suggesting community-based exercise programmes can be beneficial in promoting social inclusion. This is important to note as the majority of frame running programmes and training sessions are carried out in a community setting and will encourage like-minded individuals to participate in the same activity promoting social interaction. Furthermore, as previously discussed, studies by Bryant et al. (2015) and Van Schie et al. (2015) reported frame running to be an enjoyable and feasible activity.

2.8.1.2 Strength Training & Self-perception

The physical benefits of strength training are well documented, and this is highlighted by a recent systematic review by Fleeton, Sanders, and Fornusek (2020), who looked at strength training and impact on athletes with CP performance. Of the 53 studies included, only one study included a psychological outcome (Unger et al., 2006), suggesting there may be limited evidence on psychological effects of strength training. The importance of looking at a variety of outcome measures could be beneficial in providing a more extensive look at the effects of exercise programmes, not just strength training, and not just physical outcomes or physical activity levels.

Unger and colleagues (2006) carried out a randomized controlled trial to look at the impact of a strength training programme over an eight-week period in adolescents with CP on their gait and perceptions of functional competence and body image. Alongside the gait measurement equipment, the participants completed a self-perception questionnaire to investigate the perceptions of functional competence and body image. To add a qualitative aspect to the research the participants completed two open-ended questions, one related to the perceptions of the body image, "Make a list of all the things you like about yourself and your body", and "Make a list of all the things you do not like about yourself and your body".

The qualitative analysis from the open-ended questions showed that the participants initially made comments about how they did not like their arms and legs, but post-intervention the comments from the experimental group were less directed towards their body parts but more directed towards topics of negative behaviour (e.g., drinking, drug use, fighting, and nail biting). The experimental group showed a slightly greater change in their perception of functional competence in comparison to the control group, but the change was not statistically significant. Despite the questionnaire used by the researchers not being validated, the results from this study have positive outcomes in relation to the participants' perceptions of body image. The qualitative expressions taken from the study can help to back up the significant change in perceptions of body image between the exercise and control group and were reflected further in comments taken from the school therapists indicating that some of the participants had become more confident in social situations, and more likely to participate in activities at school.

This study does indicate effectiveness for the use of a strength training programme for improving perceptions of body image and functional competence. However, more comprehensive research with validated assessment tools could provide a better idea on the effects a strength training programme on areas of self-perception. Also, the participants' GMFCS classification were not listed in this study, but the description table of the participants showed only two out of the 31 participants occasionally used assistive devices (wheelchair and crutches), showing that participants' gross motor function levels were likely

to be either GMFCS levels I or II, and were able to complete the training unassisted. The probability of an individual with more severe CP being able to complete strength training circuits, unassisted, several times a week, are very low, highlighting that strength training interventions may be beneficial in some cases, but the more severely impaired individuals are unlikely to be able to take part.

Another study investigating self-perceptions and how they can be affected by strength training was carried out by McBurney and colleagues (2003). They found children participating in a six-week strength training programme exhibited positive perceptions about their physicality as well as psychological benefits. The 11 participants completed in-depth interviews following their home-based training programme and researchers identified positive and/or negative themes. The participants felt that participating had led to improved physical aspects such as strength, posture, and flexibility. Alongside the physical aspects, there were reported psychological benefits as participants felt they had higher levels of energy and an improved feeling of well-being. One theme that was identified by McBurney et al. (2003) from the qualitative analysis was that the benefits gained were quite individual, and therefore applying the same measures in a quantitative study may not capture a complete picture of the benefits from a training programme. This is similar to other research (e.g., Darrah et al., 1999; Dodd et al., 2004) where different results were found for the same aspect of self-concept after training programmes, suggesting the potential impact of other factors on the outcomes of a training programme. Therefore, to gain a clearer idea of the impact physical activity programmes have on children and adolescents with CP, a case study approach may be more appropriate to provide more multi-dimensional individual results, particularly as CP is such a complex condition which affects each patient differently.

2.8.1.3 Strength Training and Self-concept

The physical benefits gained from strength training for children and adolescents with CP (usually GMFCS I or II) are reasonably well documented (Darrah et al., 1997; Dodd et al., 2003; Unger et al., 2006; Verschuren et al., 2007; Fleeton et al., 2020). However, the psychological benefits are less well documented. Dodd, Taylor, and Kerr (2004) conducted a randomized controlled trial (RCT) evaluating the effects on self-concept of children with CP after a six-week strength training programme.

Self-concept is an important factor when it comes to evaluating the psychological function of an individual because it can relate to life satisfaction (Harter, 1985), with characteristics including scholastic, social, and athletic competence, physical appearance and global self-worth being evaluated (Harter, 1985). Self-concept and its importance as a construct to psychological functioning will be discussed in more depth at a later stage in this thesis (see section 5.1.3.2).

To measure self-concept the RCT by Dodd and colleagues (2004) administered the Self-Perception Profile for Children (SPPC; Harter, 1985) as an assessment tool to look at the children's perceptions of themselves. After baseline measurements, the exercise group (n=10) participated in a home-based strength training programme three times a week, over a six-week period, which targeted both upper and lower limbs. The control group (n=7) continued with their normal routines. Immediately after the six-week training period and at 18 weeks, both groups completed the SPPC questionnaire to assess the effects from the strength training programme on self-concept.

Contrary to the researchers' hypothesis that participation in the training intervention would improve the self-concept of individuals with CP, the exercise group reported a decrease in their self-concept post training programme, while some aspects of self-concept increased in the control group, suggesting that participation in a strength training programme may inhibit self-concept of children with CP (Dodd et al., 2004). The exercise group reported a decrease in scholastic competence, social acceptance, from baseline to six weeks, and baseline to 18 weeks. Furthermore, the control group reported an increase in the scholastic competence and social acceptance domains, indicating statistically significant differences between the two groups for those domains of self-concept from baseline to six-weeks and baseline to 18-weeks. The athletic competence domain for the exercise group remained similar to baseline, however, it appeared to increase for the control group, but the differences were not statistically significant at either 6- or 18-weeks. The negative impact the strength training programme appeared to have on perceived scholastic competence may be related to the time away from doing homework and studying. Similarly, the impact on the perception of social acceptance may have been due to the fact that the strength training programme was a home-based training programme which they may have completed alone or with the assistance of a parent. This would mean that the experimental group had less time to see friends in comparison to the control group. The inhibition on the perception of social acceptance is important to note, because it suggests that participating in exercise or physical activity at home and away from friends may have a negative impact on aspects of the child's self-concept. This shows that environment has a part to play in self-concept of an individual and it cannot be improved solely by participation in a strength training programme.

The research discussed above provide an insight into the potential effect's strength training has on physical and psychological wellbeing, for young people with CP. The physical benefits of strength training for young people with CP (e.g., overall muscle strength, stride length, or isolated muscle strength; Fleeton et al., 2020), which are not detailed in this thesis as they are already well documented and beyond the scope of this thesis, may be complementary with frame running. For example, the strengthening of leg muscles of children with CP (e.g., Scholtes et al., 2010), can improve functionality, and thus help with

frame running performance. Furthermore, van der Linden and colleagues (2018) reported limb spasticity and isometric leg strength to impact on frame running performance over a 100m race suggesting that strength training can complement frame running.

On the other hand, frame running also provides a different opportunity to the children and adolescents with CP to be physically active in comparison to strength training. The studies discussed in this section highlight the limited opportunity for individuals with more severe mobility and/or cognitive impairments to participate in strength training programmes.

Compared to frame running, this is a major difference between the two activities, as frame running has shown to be a suitable option for individuals with all levels of impairment (e.g., Bryant et al., 2015). Another option young people with CP may consider participating in and which is suitable for aerobic exercise is cycling. The following section looks at the various types of cycling that are available to individuals with CP and the effects it can have on their wellbeing.

2.8.2 Cycling

Muscle weakness and reduced mobility in children with CP can limit the participation in typical childhood activities such as running, cycling, jumping, and climbing (Armstrong et al., 2019). The limited ability the children with CP have to take part in the typical childhood activities can lead to decreased levels of habitual physical activity which in turn increases the risks associated with physical inactivity such as a decline in gross motor function (e.g., Hanna, et al., 2009), increased risk in Non-Communicable Diseases (NCDs), decrease in cardiovascular fitness (Mitchell et al., 2015), decrease in muscle strength, volume, and increase in muscle tone (Verschuren et al., 2014). Adapted cycling, whether it is in a static or dynamic form, offers the opportunity as an alternate form of physical activity to children and adolescents with CP who have a lower level of functional mobility (Armstrong et al., 2019).

Children and adolescents classified in GMFCS levels III, IV, and V have been shown to be at risk of losing their motor function as their development continues from child to adolescent to young adult, with the biggest decline in seen in levels IV (Hanna et al., 2009). It is important to note that these are estimations, and there are no specific explanations for the decline of gross motor functions and levels of functionality at different ages. The decline in gross motor function in adolescents and young adults with CP highlights the need for physical activity interventions suitable for the less ambulant population (Verschuren et al., 2008).

2.8.2.1 Non-ambulant Children & Cycling

The muscle weakness associated with CP can contribute to the motor ability limitations and decline for individuals with CP and is more prominent in children who are classified as levels IV and V on the GMFCS (i.e., those who are non-ambulant; Palisano et al., 2008). As discussed in section 2.8.1, children and adolescents with CP can benefit from participation in strength training programmes (Fleeton et al., 2020). However, the majority of the participants of strength training studies are ambulant children and adolescents with CP and participate in exercise interventions such as circuit training and weightlifting, or mixed training combining aerobic and strength training. Children with limited or no walking ability would struggle to participate in these activities because they do not have the physical ability to perform the same exercise routines as ambulant children and adolescents with CP, but they may be able to pedal independently while supported on a static bicycle (Williams & Pountney, 2007).

With a small evidence base involving children with limited to no walking ability, Bryant, Pountney, Williams, and Edleman (2012) examined the effects of an exercise intervention on gross motor function in non-ambulant children with CP. Participants were classified as either level IV or V on the GMFCS, had the ability to pedal independently on a static bicycle that had been adapted, and were able to walk on a treadmill with partial body weight support. The RCT had participants assigned to an adapted bike, treadmill, or control group. The participants in the two exercise groups received exercise training over a six-week period, three times a week, and each session lasted approximately 30 minutes. The control group continued to receive their regular physiotherapy sessions during the six-week period. Using the Gross Motor Function Measure (GMFM)-66 and two dimensions from the GMFM-88 (D – standing and E – jumping, running, and walking), motor function of the participants was assessed at four separate instances: pre-intervention (week zero), immediately post-intervention (week six), and two follow-ups at weeks twelve and eighteen. Two additional measures for the bike and treadmill group, speed and duration of exercise, were measured at week zero and week six.

Results from Bryant et al. (2012) immediately post-intervention reported participants in both the treadmill and static bike group showed significant improvements in their standing ability (i.e., GMFM-88D scores) in comparison to the control group. The small but significant improvements in standing ability for non-ambulant children is notable, as it can have positive implications in terms of future levels of independence which can be a very important factor for an individual's development, particularly for those with more severe impairments. Furthermore, both exercise groups showed a trend towards significant improvement in walking ability post-intervention suggest that being able to have this population participate in an appropriate mode of physical activity and exercise, over a longer period of time than the 6 weeks in the study by Bryant et al. (2012), might lead towards improved mobility.

Importantly, both the static bike and supported treadmill were reported as enjoyable and acceptable forms of exercise for a population with very few suitable opportunities. The results highlight non-ambulant children with CP may have the ability to improve their functional ability if they are provided with an appropriate opportunity.

Williams and Poutney (2007) also examined the effects of a static bike programme on the functional ability of non-ambulant children with CP. The ten participants with CP were classified GMFCS levels IV or V and had the ability to pedal independently on a static bike. The GMFM-66 and GMFM-88 dimensions D (standing) and E (walking, jumping, and running) were used were administered to measure their functional mobility. Participants were measured on four occasions at baseline, pre-exercise period, post six-week exercise period, and six-weeks after the exercise period at follow-up. The static bike used in the study was adapted for the population, providing wrists, trunk, and feet support and participants took part in the exercise three times per week over a six-week period.

The results from the GMFM-88 dimensions D and E showed that eight out of the ten participants improved their standing, walking, running, and jumping scores, and nine out of ten participants improved their overall functional ability with their GMFM-66 scores being statistically significant between pre- and post-exercise periods. After the follow-up period, the improvements in functional gains from the exercise period showed little or no decline which are different to Bryant et al. (2012) who found statistically significant decrease in the bike group and their standing ability (GMFM-88D). However, the Bryant et al., (2012) study was a RCT design, and the decline was in comparison to a control group. This indicates that the difference in the study design can show the difference in results and that they must be interpreted carefully. Williams and Poutney (2007) showed that over a short period of time (six-week training period), functional mobility in non-ambulant children with CP (GMFCS levels IV and V) can be improved using an adapted static bike.

The benefits reported by both Williams and Poutney (2007), and Bryant and colleagues (2012) suggest that non-ambulant children with CP, presented with a safe and suitable opportunity, can positively impact aspects of their gross motor function. With few opportunities to participate/exercise at this capacity, it is important that an option for physical activity is provided for this population to allow them new experiences, such as, training at high intensity and experiencing being out of breath, which previous frame running studies have shown to do (e.g., Hjalmarrson et al., 2020). Thus, frame running can be viewed as a viable option and/or alternative for this population. Furthermore, frame running may be viewed as a complementary option to exercise on a static bike, as an improvement in standing and functional ability from participation on a static bike, could be transferred to help with frame running participation and their walking ability on a racerunner.

Armstrong and colleagues (2019) carried out a systematic review of the efficacy of cycling interventions for the improvement of functionality in children with CP and the results indicate there are a limited number of studies including children classified GMFCS levels IV and V. The two studies discussed are important to the current research because the use of a static bicycle is similar to a racerunner, as it provides support to the participant when participating being an adapted tricycle and allows the participant to experience physical activity at a higher intensity. An additional benefit for frame running, however, is that it does not require the individual to have the ability to pedal, which widens the accessibility of the sport compared to the static bike. Also, frame running provides the opportunity for children with limited or no walking ability to be physically active, similar to the studies by Williams and Poutney (2007) and Bryant and colleagues (2012), and the research for this population is very limited indicating that future research is needed to find more appropriate opportunities for this population.

As discussed in this section, participation on a static bike can provide functional improvements for a population that has limited functional capacity, however frame running can be a viable alternative as it can offer experiences (e.g., higher intensity training) that may not be feasible on a static bike. Importantly, the research has shown there are opportunities, though limited, available for non-ambulant children with CP to be gain benefits from participating in physical active. Another of these activities available to children with CP, including those with more severe impairments, is hippotherapy and therapeutic horseback riding, and the impact from participation will be discussed in the next section.

2.8.3 Hippotherapy & Therapeutic Horse Back Riding

As frequently mentioned throughout the thesis, the opportunity for children and adolescents with more severe CP to experience a sense of freedom is relatively rare due to the support that is necessary in many aspects of their lives. However, horseback riding can provide this opportunity for this population in both therapeutic and or recreational settings. Horseback riding has been a popular therapeutic intervention around Europe after the second World War as it is prescribed to address physical, mental, and emotional issues (Snider et al., 2007).

Horseback riding interventions can be split into two styles, hippotherapy and therapeutic horseback riding (THR; Snider et al., 2007). Hippotherapy is normally provided to the participants who are more severely disabled (i.e., GMFCS levels III to V) by physiotherapists or occupational therapists, and they aim to use the movement from being on the horse to improve a range of factors, including gross and fine motor skills, balance, and posture. In comparison, THR is provided by a riding instructor to the disabled rider who

have minimal CP (i.e., GMFCS levels I or II), and their aim is to teach the basic skills required to control the horse. The Riding for Disabled Association (RDA) is a UK based organisation which offers the opportunity for children and adults with physical and learning disabilities to participate in a variety of riding activities including fitness, therapy, skill development, and the chance to achieve. The type of activities on offer via the RDA highlight the diversity on horseback riding, and how it can be utilised for children and adults with disabilities, including CP, and a number of these are discussed in the coming sections.

It is important to note the difference between therapeutic horseback riding and hippotherapy as hippotherapy is not seen as a recreational opportunity but as the chance to address impairments associated with the disability using the movement of the horse (Casady et al., 2004). An early descriptive paper by Horster and colleagues (1976) suggested that participation in horseback riding in the form of hippotherapy or THR can provide psychological benefits in the form increased levels of motivation and self-esteem related to opportunity for the participants to experience success in completing the tasks given to them, however no data was presented to support these suggestions.

2.8.3.1 Hippotherapy & the Impact on Psychosocial Factors

Similar to other physical activity interventions for children with CP, many hippotherapy studies have outcome measures focused on physical factors, mainly functional performance and posture, compared to a few that include psychological and social factors including QoL (Snider et al., 2007; Debusse et al., 2009; Sikwood-Sherer & McGibbon 2020). There is only a small evidence base for the psychosocial effects of hippotherapy interventions (Casady, & Nichols-Larsen, 2004; Frank et al., 2011; Deutz et al., 2018).

Jang and colleagues (2016) carried out a pilot study looking at the effects of hippotherapy on various psychosocial aspects in children with CP and their caregivers. Eight children with CP (GMFCS levels I to III) participated in the study and completed a 30-minute hippotherapy session once a week over a 10-week period with a physical therapist. For the evaluation of psychosocial and emotional factors, the subjects were asked to complete a number of questionnaires relating to depression, anxiety, self-esteem, and QoL. The results showed significant improvement in their physical aspects. On the other hand, for the psychosocial and emotional assessments, no statistically significant results were found in any of the outcome measures however there were slight improvements in scores relating to stress, anxiety, and self-esteem. The slight improvement in scores indicates that participation in hippotherapy does have the potential to improve the psychosocial aspects of children with CP, but as a pilot study there were no comparisons made with a control group or other form of exercise, but the study showed the way for future research.

In line with the findings from Jang et al. (2016), Detuz and colleagues (2018) found that hippotherapy significantly improved gross motor function dimension E relating to the ability to walk, run, and jump, while no significant increases in quality-of-life scores on the child health questionnaire. Detuz et al. (2018) had a large sample of 73 participants which were randomized into an early and late treatment group. Participants had hippotherapy treatment once or twice a week over a 16 to 20-week period. The participants ranged in the GMFCS classification from levels II to IV indicating a considerable range of CP severity range in the study. This is beneficial as it shows that the more impaired individuals are able to participate in hippotherapy and gain functional improvements in areas of walking, running, and jumping. Also, it provides evidence that there is another option for the more impaired individuals to be physically active and participate in an activity in a different environment.

2.8.3.2 From Physical to Psychological Benefits of Hippotherapy

A case study by Frank et al. (2011) showed a hippotherapy intervention to have a positive impact on the functional performance, and therefore to positively impact on the participant's participation and self-competence (age 6, GMFCS I). Post an 8-week hippotherapy intervention, the participant reported improvement in participation contributed to being more independent at home. Furthermore, they also reported to be able to walk further unassisted, participate in new activities, and play with friends more. The improvement in participation was in conjunction with a positive impact on the participant's perceived self-competence. Post-intervention the participant reported an increase in physical competence, peer acceptance, and a drop in their maternal acceptance score, furthered by the reporting of increased interactions and playing with friends. The case study is important because there is limited information reported regarding the effects of hippotherapy on factors of participation and self-competence. However, the participant was classified at GMFCS level I and therefore the functional improvements that may have led to improvement in participation aspects of self-competence may not be comparable to participants who are more severely disabled.

A trend from hippotherapy interventions for children and adolescents with CP appears to be that the physical benefits can lead to psychological and social benefits, which is important for the development of children and adolescents (Sikwood-Sherer & McGibbon, 2020). Debusse, Gibb, and Chandler (2009) conducted a qualitative study including ten children with CP (GMFCS I, IV and V), on the effects of hippotherapy to gain the perspective of the user and try to paint a more complete picture of the effects of hippotherapy for the CP population. Semi-structured interviews were conducted with the participants and some parents, to explore the "reality" of hippotherapy participation, and they identified five main themes; physical effects, psychological effects, movement experience, factors that influence

the experience, and responses to these effects. The physical effects are in line with previous research in terms of improved balance and motor function (e.g., Jang et al., 2016; Sterigou et al., 2017; Deutz et al., 2018), and for the purpose of this thesis, the focus will be on the reported psychological effects of hippotherapy.

The psychological effects identified by Debusse and colleagues (2009) included an increase in self-efficacy and self-esteem, sense of achievement, and a sense of wellbeing. In line with previous research, the improved motor function was reported to impact positively on psychological aspects of the participants' life. Participants felt that hippotherapy instilled a level of confidence in them which helped them to overcome the fear of completing certain motor tasks that may result in pain and/or failure (e.g., climbing the stairs unassisted). Furthermore, participants reported that their improved motor function allowed them to achieve a greater range of motor tasks, and they felt more confident while completing them. This reported increase in self-efficacy is important as it is thought to play an important role in influencing health behaviours. Therefore, an increased sense of self-efficacy may positively influence the choice of activities and the amount of effort individuals choose to put in (McAuley and Blissmer, 1999). This can lead to a positive cyclical chain of events with individuals having more confidence in their ability, and therefore choosing to participate in more activities, and in turn benefiting physically, psychologically and socially from those activities. The qualitative study by Debusse et al. (2009) suggests psychological benefits do not have to come from aerobic or high intensity exercise. The physical effects of hippotherapy can be enhanced by the psychological effects, which in turn can have a positive impact on an individual's perceived QoL (Sikwood-Sherer & McGibbon 2020).

Casady and Nichols-Larsen (2004) found in their study that four out of their six non-ambulant participants with CP showed a greater improvement in their overall GMFM scores after a hippotherapy intervention in comparison to the ambulant participants. This highlights the impact that participation can have on the more functionally impaired individuals with CP, and the potential to improve psychosocial aspects of the individual, similar to Frank et al. (2011) and Jang et al. (2016).

Research on hippotherapy thus suggests that psychological benefits, such as improved self-efficacy and self-perception, can be gained from participation in a low intensity activity which also provides some improved functional ability. The suggested link between motor function and subsequent psychological impact is important when considering frame running as previous frame running research has shown improvements in motor function (e.g., Bryant et al., 2015). Thus, the trend towards psychological benefits from frame running participation is also possible.

Although hippotherapy has been shown to improve motor function, the intensity of exercise can be low, and for the more severely impaired, there may not be many options to

exercise at a higher intensity, highlighting the necessity of an aerobic, higher intensity activity, such as frame running. If feasible, frame running can provide a valuable addition to hippotherapy, as they may be viewed as complementary activities, because they are both accessible to the population and can provide a more rounded physical programme (i.e., improved balance and trunk control via hippotherapy, and improved bone density via frame running). Furthermore, in addition to the physical benefits, the psychological impact of frame running would hopefully be identified, similar to hippotherapy. The importance of having opportunities for physical activities available for the more severely impaired individuals is clear, and another possibility for young people with CP, across all levels of severity is swimming, which is discussed in the following section.

2.8.4 Swimming

As previously discussed, there are a variety of perceived barriers that can prevent children with CP becoming physically active including pain, fatigue, injury risk, fear of social exclusion, beliefs that learning new skills is time consuming, and participation in physical activity is not enjoyable (Verschuren et al., 2012). Having a child perceive physical activity or exercise as fun, enjoyable, and safe can facilitate interest and engagement in the activity, emphasising the importance of offering opportunities to be physically active which are fun as well as safe to help counteract the fear of injury. Aquatic exercise and swimming interventions are amongst the more popular choices of treatment and exercise for children with neuromotor impairments, especially CP (Getz, et al., 2006), as it can offer low-impact and safe alternative to other more demanding forms of physical activity (Dimitrijević, et al., 2012), such as strength training (e.g., Verschuren et al., 2007) or high-intensity circuit training (e.g., Schranz et al., 2018).

As mentioned, swimming is a popular option for individuals with CP, and more so with children and young people with less severe mobility impairments (i.e., GMFCS I to III) as it provides the chance to experience moving in water and participate alongside their able-bodied peers. The competitive element of swimming can also be viewed as a facilitator to physical activity participation, as opportunities to compete for individuals with CP are restricted in comparison to their able-bodied peers. Moreover, with opportunities to compete at major swimming events at National and International level, including the Paralympics, and opportunity to progress and compete at increasing levels of competition is one of the advantages of swimming for the CP population. Frame running is similar to swimming in this context, with a growing competition schedule, frame running has featured at the World Para-Athletics Championships and is expected to feature at the 2024 Paralympics.

2.8.4.1 Impact of Swimming on Motor Functioning

A systematic review by Blohm (2011) highlighted eight studies investigating the impact of aquatic interventions for children with CP. Each of the studies included in the review reported functional benefits of participation for the participants including gross motor skills, leg muscle strength, and walking efficiency. The review by Blohm (2011) suggests aquatic interventions are beneficial and advantageous for children and young people with CP, and further benefits are discussed in the following sections.

The ease of movement, weight relief, and buoyancy from exercising in water can encourage safe movement, with reduced impact and forces on joints in comparison to exercising on land, making an aquatic environment more attractive to exercise in (Kelly, & Darrah, 2005). However, one perceived downside to the non-weight bearing aspect of swimming is that it may cause orthopaedic concerns for young people with CP who are susceptible to low bone density due to muscle weakness and motor impairments. Moreover, previous frame running research has shown children with CP to have increased bone density post a frame running intervention (Bryant et al., 2015), thus suggesting that frame running may be a viable alternative and/or complementary option for physical activity participation.

Exercises such as stretching and functional task orientated training have been shown to improve gross motor function in the children with CP, but some children and parents have reported these exercises to be difficult to carry out on land for a variety of reasons (e.g., the children are experiencing pain; Pin et al., 2006). Therefore, the buoyancy and anti-gravity positioning that is provided by being in water, providing an alternative environment, can help improve gross motor function of children with CP who were struggling on land. There are a number of studies showing the impact on gross motor function of swimming for children and young people with CP, and one study by Dimitrijević, Aleksandrović, Madić, Okičić, Radovanović, and Daly (2012) investigated the effects on aquatic skills and gross motor function in children with CP.

Dimitrijević et al. (2012), reported that children with CP who received the swimming intervention over a 6-week period, showed significant improvement in their gross motor functions and aquatic skills post intervention in comparison to a control group. Similar to Dimitrijević et al., (2012), Akinol, Gbiri, and Odebiyi (2019) examined the impact of a 10-week aquatic exercise training programme on the gross motor function of children with CP. Post training programme the treatment group reported a significant improvement in aspects of gross motor function in comparison to the control group. Akinola and colleagues (2019) reported the treatment group to have improved in aspects of gross motor function such as standing, sitting, rolling, and crawling, but not in walking, running or jumping, as has been documented in previous research (e.g., Declerck et al., 2016). This difference may be

attributed to the participants, as 60% of participants for Akinola et al. (2019) were nonambulatory (GMFCS IV or V), compared to other research in which all participants were ambulatory. Overall, the results provide further evidence of the physical benefits of aquatic training programmes, as well as highlighting it as an effective, safe, and feasible option for rehabilitation and physical activity for children and young people with CP.

As mentioned, Dimitrijević et al. (2012) and Akinola et al. (2019) included participants with CP of all severities, from GMFCS level I to V, documenting individuals with more severe CP are able to participate in aquatic exercise and swimming programmes, and improve aspects of gross motor function. Therefore, providing the opportunity to the more severely impaired population to participate in aquatic exercise, whether it is swimming or water orientation skills, should be considered as a feasible option for physical activity, therapy, and research environments. Furthermore, it is suggested that the improvement from aquatic exercise can have psychosocial benefits too, by improving QoL via motor function, and aspects social functioning (Aidar et al., 2007; Dimitrijević, et al., 2012). The effects of swimming and aquatic exercise training on psychosocial wellbeing are discussed in the following section.

2.8.4.2 Impact of Aquatic Exercise on Social Functioning & Enjoyment

An important facilitator to physical activity for children with CP is the enjoyment of the activity or exercise (Verschuren et al., 2012), and for this population it is particularly important that the enjoyable physical activity or exercise programme has limited risk of injury and does not push the capabilities of the child too much, making it one of the more popular types of physical therapy (Getz et al., 2006; Declerck et al., 2016). Moreover, if there are adapted facilities and there is individual support in the water, an aquatic exercise programme can be delivered to children with more severe levels of CP.

A RCT by Declerck and colleagues (2016) examined the impact of a 10-week swimming intervention on the perception of physical factors, functional ability, and enjoyment of ambulatory children with CP. The outcome measures looked at current levels of self-reported pain and pain in the previous week, self-perception of fatigue, and functional ability. To measure functional ability the participants completed a one-minute walking test measuring the distance at maximum speed, and the WOTA2 to measure their water orientation skills including function in the water. To measure enjoyment, participants from the swimming group rated on a 5-point scale how much they enjoyed each swimming session.

In line with previous research, the swimming group reported significant increase in their walking ability, as well as the aquatic functional skills. There were no significant changes reported in pain or fatigue between or within groups over the intervention and follow-up periods. This may be related to the fact that the participants were GMFCS levels I

to III, and the lack of fatigue that was reported before the intervention began was already high. The non-significant changes in fatigue and pain scores reported show engagement in a swimming intervention did not cause any adverse effects. As pain and fatigue are viewed as a barrier to participation in physical activity for young people with CP, these results provide positive evidence contrasting previous research indicating that pain and fatigue are results of exercise and physical activity for children and youth with CP (Verschuren et al., 2012).

The enjoyment levels for this study were high, with 86% of participants indicating they enjoyed the sessions “very much” (scoring 5 out of 5). High levels of enjoyment suggest that participation in a swimming programme can be fun and therefore can be considered as an option to offer to children and youth with CP, as it can overcome one of the barriers to participation. The high levels of enjoyment reported by Declerk et al. (2016) can be viewed as a facilitator to physical activity participation, and might be linked to the increased opportunity for social interaction during swimming sessions, an aspect of life that children and young people with CP may be restricted in.

Aidar et al. (2007) examined the effects of aquatic exercise intervention on the social function as well as manual ability of children with severe CP, and participants were included with associated disturbances such as mental retardation and sensory deficits. To measure effects on social function, the participants completed the “social function” subscale from the Pediatric Evaluation of Disability Inventory (PEDI). The results show a significant improvement in both manual ability and the social function subscale of the PEDI post-intervention. Significant improvements in the social function subscale suggest the positive impact that aquatic exercise can have on the social aspects of children with more severe CP, such as interactions with others, resolving problems, and community interactions. The aquatic exercise taking place in a more social environment will aid social inclusion and encourage more independence at home in domestic tasks which can improve other psychological factors like self-esteem and self-concept. The inclusion of the participants with more severe impairments is important for research as it provides evidence they can be considered as research participants in their own right, as well as it being the only way to gain a more direct insight into their lived experiences.

2.8.4.3 Impact of Swimming on Self-Perceptions

Influencing and improving an individual’s perception of their competence and efficacy can be important for children and young people with disabilities as they may struggle with positive perceptions due to their impairments, and some social stigmas surrounding the disability. The implementation of physical activity programmes can help focus an individual’s skill development and help them gain personal achievements, thus developing and improving self-perceptions (Hutzler et al., 1998). As mentioned earlier, swimming is a

popular option for therapeutic and physical activity programmes for children with CP due to the ease of movement, non-weight bearing, and low impact on joints associated with exercising in water (Akinola et al., 2019). With this in mind, Hutzler, Chachan, Bergman, and Reches (1998) examined the effects of swimming and aquatic exercises on the participants' perceptions of competence and independence, as well as motor function. There was a slight non-significant increase in self-concept scores for both the control and treatment group post-intervention. It was suggested that the water orientation programme did not elicit significant increases in self-concept because it did not provide the participants with the sense that they had mastered swimming, but more water related movements (e.g., entering the water, walking across the pool, gliding to and from the wall). The results from Hutzler and colleagues (1998) show positive impact on mainly physical factors. Furthermore, a benefit of swimming, for disabled and able-bodied children, is the social aspect as normally participation occurs in a community setting and therefore social inclusion can be seen as a facilitator to physical activity (Verschuren et al, 2016).

2.8.4.4 Swimming: A viable Option?

Swimming and aquatic exercise studies have shown that there can be significant physical benefits and skills gained from participation in the enjoyable activity, but the evidence is less clear for psychosocial factors. The results from the research discussed suggests that swimming and aquatic exercises should be offered to children and youth with CP. It is a suitable option for this population because it can be seen as an enjoyable and fun option to be physically activity which can help functionality, both in and out of the water, without causing significant pain or fatigue. As previously mentioned, the inclusion of more severely impaired individuals suggest that swimming is an appropriate option for physical activity and exercise as it provides the opportunity for a full body workout without putting any excessive strain on body parts and the warmer water can reduce muscle spasticity, allowing better movement in children and adolescents with high muscle tone (Getz et al., 2006). This makes swimming and aquatic exercise an attractive option for physical therapy and exercise in comparison to other options like strength-training or cycling where individuals will be unable to complete the programme, or not even be interested due to the fear of injury. However, as swimming is a non-weight bearing activity, it is unlikely to help increase bone density. Individuals with CP are susceptible to low bone density due to muscle weakness and inactivity which is an orthopaedic concern. Bryant et al. (2012) found an increase in bone density post a 12-week frame running intervention in children with more severe CP, showing frame running to be a viable option for physical activity that improves bone density in contrast to swimming. Moreover, swimming for individuals with CP is a relatively expensive activity as it requires a high level of individual support (one-to-one) in the case of more

severely impaired individuals. Furthermore, access to swimming can be difficult, as only some special schools have their own pools, and require additional equipment such as lifts and adapted changing facilities which are expensive. In comparison, placing some frame running bikes in a school can be relatively low-cost.

2.9 Identifying the Research/Knowledge Gap & Significance of the Study

As this chapter has shown, the importance of physical activity in children and youth with CP is well documented because of the physical and psychological benefits that can be gained from participation in a range of activities and exercise. The mobility and cognitive impairments associated with CP from the disturbance in the brain can limit the ability to be physically active, and the secondary health complications can increase the risk of inactivity and therefore the risk of developing diseases also increases. Of course, the physical and mental capacity of a child with CP has to be taken into account when trying to engage them in a new activity or exercise intervention.

As demonstrated in the previous sections, it is clear that there are some options available for participation in physical activity and exercise for children and youth with CP, including some for the more severely disabled (GMFCS levels IV and V), most notably swimming. However, one of the problems encountered when attempting to include GMFCS levels IV and V classified children in physical activity interventions, is that they can struggle to understand or follow instructions, and they may lack the physical capacity to complete the activity. Indeed, the evidence base for GMFCS levels IV and V is less well documented, despite the need for individuals with lower levels of functionality requiring more appropriate opportunities as they may have the most to gain from participation. Numerous studies cited in the previous sections indicated significant improvements in physical factors, including gross motor function, muscle strength, and water functionality, after a physical activity or exercise intervention. Less frequently researched was the potential for physical activity and exercise interventions to have an impact on psychosocial factors, such as self-esteem, self-efficacy, self-concept, and QoL. Often children and youth with CP are already at a disadvantage and do need to focus on stretching and loading their muscles, exercise their heart and lungs to maintain fitness levels, more regularly than children without CP. However, the potential impact on psychosocial factors from participation in physical activity should be investigated as improvement in these factors could assist in getting past some barriers to participation and in turn improve areas of physical functioning in a cyclical fashion.

Previous frame running studies have provided some evidence relating to the physical benefits of frame running, and some qualitative data regarding the enjoyment of frame running has been reported in studies aimed at measuring physical factors. However, there

has not been a study aimed at investigating the psychological effects of frame running on children with CP. This doctoral study will be the first known study to investigate the effects of frame running on psychological and social factors in individuals with CP using a mixed methods approach underpinned by the ICF model and a theoretical framework of behaviour change and psychological function in the context of physical activity. As previous research examining the impact of physical activity for children and young people with CP has suggested, and has been discussed in this chapter, an improvement in motor function can have a positive impact on psychological and social factors (e.g., self-efficacy, self-competence, and QoL) for children and adolescents with CP. Motor function improvements have been shown from participation in a range of physical activity and exercise, including frame running.

As seen throughout the literature review, there is a lack of inclusion for the more impaired individuals with CP. There is some evidence that frame running is suitable for the more impaired individuals with CP, however whether or not the overall influence of frame running participation is of benefit is not clear. Therefore, this is an important aspect to be explored in the current thesis.

Some aspects of frame running make it a suitable alternative for being physically active for individuals with motor impairments, and for some it may be the only viable option if they have very limited to no walking ability. This is important due to the necessity to be physically active for helping physical, psychological and social development throughout childhood and into adulthood. Furthermore, as there is currently no research on the effects of frame running on psychological and social factors, it is important to assess the potential for frame running participation to have a positive impact on these crucial factors. Thus, this thesis will explore the effects of frame running participation on psychological and social factors to add to the literature and research on physical activity for children with CP.

2.10 Research Questions

The following section lays out the research questions that were investigated throughout this thesis. These questions have been derived through consideration of the overall aims of the thesis, and the critical evaluation of existing literature and research in the area. In particular, the current gaps in knowledge and understanding have influenced the development of these research questions.

2.10.1 Research Question 1

Identifying domains that are important for parents, health care professionals, and frame running coaches when evaluating physical activity interventions for children with CP is

important because it will enable children with CP to gain as much as possible from the physical activity intervention and guide future physical activity interventions to ensure they are as effective as possible.

What domains and factors are viewed as most important for parents, health care professionals, and frame running coaches for evaluating physical activity interventions for children with cerebral palsy?

This research question will be addressed in the first study described in chapter 4.

2.10.2 Research Question 2

CP is a complex chronic illness, and with no cure, the focus of individuals with CP, parents, and medical professionals is to improve their QoL. Over the past decades QoL has transpired as an important domain to measure, but it is difficult to define with the World Health Organisation breaking it down into four main life domains; psychological, physical, environmental, and social relationships (McDougall et al., 2010). Furthermore, QoL should always be taken into account when assessing an individual's health and functioning to try and build a more complete view of the individual. Physical activity is thought to have a positive impact on the QoL of youth with CP (Verschuren et al., 2013), however, being physically active is not always possible for someone with more severe impairments. Frame running is an opportunity for youth with more severe CP to become physically active, however there is no current research investigating the impact of frame running on the QoL of youth with CP:

Does participation in a 12-week frame running training programme have an effect on the perceived QoL of children and young people with CP?

This will be addressed in study 2, described in Chapter 5, together with the next two research questions.

2.10.3 Research Question 3

The importance of psychological wellbeing is a widely discussed topic, and so are the psychological benefits that can be gained from being physically active. Psychological domains such as self-esteem, self-efficacy, self-concept, and psychological wellbeing have all been identified to be lower in youth with CP compared to their typically developing peers. Of particular note is self-efficacy, which appears to be a key factor in the adoption and

adherence of physical activity and health behaviours, which is furthered by the inclusion as a key construct in each of the three theoretical frameworks discussed earlier in this chapter. Therefore, frame running participation is an opportunity for youth with CP to be physically active and impact the psychological factors previously mentioned:

Does participation in a 12-week frame running training programme have an effect on the psychological factors: self-efficacy, self-esteem, self-concept, and psychological wellbeing children and young people with CP?

This question will be addressed together with the previous and next research questions in study 2, described in Chapter 5.

2.10.4 Research Question 4

Participation in daily and leisure activities are reported to be lower in children with CP in comparison to their age matched peers. Participation in activities of any description, (e.g., daily, leisure, and/or physical activity) is an opportunity for children with CP, just like everyone else, to develop social and communication skills, build friendships and relationships, and improve physical and mental health (King et al., 2004). However, there are many perceived barriers to physical activity participation for youth with CP and thinking that taking up a new activity is going to be boring or time consuming is a prominent barrier. Enjoyment is one of the main facilitators to physical activity participation, and therefore having an enjoyable activity is important to encourage engagement and adherence in physical activity. Furthermore, children with CP report less enjoyment from participation in activities (Whitney et al., 2019) and with less available and/or suitable opportunities the importance of having more options for this population is important. Therefore, the following question is investigated:

Does participation in a 12-week frame running training programme have an effect on the participation and enjoyment of other activities for young people with CP, and is frame running perceived as an enjoyable physical activity for young people with CP?

This question will be addressed together with the previous two research questions in study 2, described in Chapter 5.

2.10.5 Research Question 5

Previous research has shown participation in disability sport can have a positive impact on the development of psychosocial characteristics such as self-concept, self-esteem, and self-efficacy in both children with disabilities (Russo et al., 2008; Novak et al., 2013), and athletes with disabilities (e.g., van de Vliet et al., 2008; Adnem et al., 2010). However, there is limited qualitative research studying the influence of sport participation on psychosocial concepts and QoL. Therefore, it is important to employ qualitative research methods to gain more insight into the influence of disability sport participation on the psychosocial wellbeing and QoL of athletes.

What impact has frame running had on the psychosocial wellbeing and QoL of competitive frame running athletes with cerebral palsy?

This question will be addressed in study 3, which is described in Chapter 6.

Chapter 3 – Ethical & Methodological Challenges in Research with Individuals with Disabilities

3.1 Introduction

Developmental research clearly shows the differences between children and adults, as they are at different ages, have different experiences, and are at different stages of their development (i.e., their language ability; Harden et al., 2000). The differences between children and adults highlights they are separate entities, and have different views, opinions, and their own voice. In the past, much research has been ‘about’ or ‘on’ children, but researchers have recognised children’s competence to participate and express their own opinions. Therefore, research involving children and young people has grown and strengthened over recent years, with the realisation of children’s views actually being sought after, leading to a change of view where research involving children has gone from ‘on’ or ‘about’ to ‘with’ children.

Ethical issues and/or methodological considerations arise in all aspects of research, however they can hold more substance when involving children, and even more so when the research involves children and young people with disabilities. The presence of a disability, and any associated impairments may add complexity to any ethical considerations (e.g., gaining consent) or methodological implications (e.g., communication techniques within interviews). Nevertheless, it is important to include children with disabilities in research as they will hold their own views, experiences, perspectives and opinions, which may vary in comparison to children without disabilities. Furthermore, with reference to health and social research, children and adolescents with disabilities could be the largest beneficiaries of the research. However, children with disabilities are one group whose voices are still not heard as much as they should (Bailey et al., 2014). As previously mentioned, the inclusion of children with disabilities is important as they can be valuable research partners, and previous research has highlighted that all children should be able to participate when the correct methods are implemented, but the involvement of children with disabilities can present more challenges, ethically and methodologically, in comparison to children without disabilities, and these will be discussed throughout this chapter.

3.2 Chapter Overview

This chapter will briefly discuss the importance of involving children and young people, with and without disabilities in research and the special considerations to take note of, followed by the positive and negative implications of involving children with disabilities in research.

This is followed by a discussion of research practices, ethical issues, methodological considerations and implications involved in research with children with disabilities, and specifically cerebral palsy (CP). This will be done with reference to the current research involving individuals with CP conducted in the current thesis, with the problems encountered and solutions employed within the research discussed.

3.3 The Importance of Research Involving Children

In the past, there has been an abundance of research that concern children; however, the focus has been about them in comparison to more recent research which looks to involve children (Kirk, 2007). The focus of previous research indicates the lives of children have been examined, explored, and interpreted through the eyes and understandings of adults, either caretakers, guardians or members of their family (Punch, 2002). According to previous research, the two main issues which have contributed to children's lack of involvement in research are firstly, the belief that the data gathered from children was unreliable, and secondly, the concerns over children's vulnerability and the potential exploitation by researchers (Kirk, 2007).

Data gathered from children in previous research was viewed as invalid because the perception of children was that they were too immature to understand what they were saying, as well as the assumption that they lacked the capacity to express their opinions and/or experiences, therefore viewing the responses as unreliable (Docherty & Sandelowski, 1999). However, this stance has changed, and researchers have developed the view that children can and should participate in research and provide valid data. This is highlighted in previous research, where responses children gave were deemed to be valid and reliable because they were expressing their own views and opinions on how they perceive the world (Punch, 2002). Furthermore, the inclusion of children in research is important because, as participants to research, they are able to provide different insight, opinions, and/or experiences, ones that may not have been reported by adults in proxy reporting.

As researchers have changed the way of thinking about children's involvement, there is greater interest in children's childhood, interactions with their environments, and socialisation, each of which have been influenced by the development in children's rights (James, 2001). It has led to children being recognised as 'active agents' of research because of the differing opinions and experiences compared to adults, and it was recognised that this is a true representation of children's experiences which would not come from the proxy reports of adults. Additionally, this would help researchers learn about children at a fuller capacity, and not just assumptions or interpretations put across by the adults. It is clear that research with children is beneficial to provide a greater insight and

understanding into their development, however, it is accompanied by some ethical and methodological issues and considerations which are different to research involving adults. There are some similarities between the ethical and methodological considerations for children with and without disabilities in research, and so they will be discussed in reference to the current research, with differences for children with and without disabilities highlighted in the coming sections.

3.3.1 Children with Disabilities in Research

Even though the research practices, approaches, and methodologies involving children have strengthened over recent years, there are some groups whose voices are still unheard, and this includes children with disabilities (Stafford, 2017). Children with disabilities are overlooked more so than children without disabilities as legitimate participants of research, and this may be for a variety of reasons, including restrictive methodology or assumptions made on their capacity (Cocks, 2008). Furthermore, there are ethical considerations and issues to consider which are more salient when involving children with disabilities compared to children without disabilities. However, despite the potential difficulties associated with involving children with disabilities in research, it is key for them to be involved, as they are in prime position to report how they are, how things can be better, and the impact of rehabilitation, interventions and/or therapy.

As previously mentioned, the research involving children with disabilities is growing, however, the evidence base involving children with cerebral palsy is very limited, with research appearing to focus on children with autism (e.g., Lewis, 2009) and learning disabilities (e.g., Kelly, 2007). Furthermore, there is not much guidance on best practice for inclusion of children and young people with disabilities in research, and this was highlighted by a systematic review carried out by Bailey, Boddy, Briscoe, and Morris (2014). Bailey and colleagues (2014) reviewed research practices involving children and young people with disabilities and they reported that overall, the quality of evidence was low, with only a few well reported studies. On a positive note, the researchers identified positive aspects associated with involving children with disabilities in research for both the children as participants, and for research. For example, having children with disabilities involved in research has shown to have positive psychological impact on the child participants with improved feelings of confidence and self-esteem (Lightfoot & Sloper, 2003). Furthermore, having children with disabilities participate can also give them the feeling that their opinions are being understood, taken on board, and given some value, as well as the sense that their participation could be helping other children and young people with disabilities in the future (Lightfoot & Sloper, 2003).

As previously mentioned, involving children and young people with disabilities is important because they are an 'unheard' cohort, and they can stand to benefit the most from being included in research. However, their inclusion in research can cause ethical issues and practical challenges, and these are discussed in the following sections.

3.4 Ethical Issues

3.4.1 Gaining Access

As with every research study, gaining access to participants, and the correct sample size is important to the robustness and validity of the study. This can be a difficult and frustrating process even when the participants are adults but can be more so for children as there can be a more complex process involved when recruiting children (Kelly, 2007). Gaining access to children with a disability in research can be done in various ways including negotiating with families, schools, and service providers. Furthermore, the advertising of the research can also be done in a variety of ways including via the internet (i.e., social media and email), and through contact with gatekeepers. However, there is a risk of the researcher exploiting the relationship between the gatekeeper and the potential research participant, and this can be an ethical concern for the researcher (Flewitt, 2005). For example, parents of young children with CP may feel an obligation to participate in research in a therapeutic or rehabilitation setting in fear they may miss out on some beneficial treatment or be pushed further down the list for future therapies. Therefore, it is important for the researcher to clearly explain the research, the expected benefits (or lack thereof) and potential risks and provide opportunities for withdrawal from the research without the fear of consequences.

In the current research, access to participants was negotiated differently for each of the studies given each cohort was different. Access to participants for the first study was relatively straightforward as participants were adults without disabilities and able to provide their own consent. Gatekeepers were still used for this study, as initial contacts amongst physiotherapists, frame running coaches, and parents of children with CP were asked to advertise and share the research with friends, family, and colleagues, who they felt might be willing and able to participate. However, this may cause some ethical concern, as potential participants may feel obligated to participate because of the relationships between themselves and the gatekeepers. The most complex process occurred in the second and third study, as participants were individuals with cerebral palsy who presented a range of communication and cognitive abilities. Recruiting from this population raises a question regarding the participants ability to understand what they are being signed up to. Furthermore, the potential issue of the child not wanting to participate, but not having the confidence, or the ability in some cases, to tell the gatekeeper, possibly because of the

relationship dynamic (e.g., parent or teacher) they wish to withdraw from the research is an ethical concern. Therefore, providing opportunities for participants to withdraw at any time was important, and this ethical issue was considered throughout the research. The methodological implications for negotiating access to children and young people with CP are discussed later in this chapter (see section 3.5).

3.4.3 Assumption of Capacity

Despite the notion that the involvement of children and young people with disabilities in research is growing, they are sometimes judged prior to recruitment because of the way they look, communicate or move, and therefore excluded or not even considered for the research. These 'societal norms' have led to children with disabilities being overlooked as participants in their own right, and their views and/or opinions are expressed by proxy by an individual above them in the hierarchy such as a parent or teacher (Stafford, 2017). Due to the research with disabled children focusing on the views of adults and ignoring the voices of children, research involving children with disabilities has shown the individuals above them in a hierarchy (e.g., a parent or teacher) can question the child contributions towards the research (e.g., Davis, Watson, and Cunningham-Burley, 2008). The prejudice shown in research towards children with disabilities can be explained by two social constructs, 'adultism' and 'ableism'.

'Adultism' is a form of prejudice towards children which has contributed to their exclusion from research, and LeFrancois (2013) defined 'adultism' as the 'oppression experienced by children and young people at the hands of adults and adult-produced/adult-tailored systems' (p.47). Furthermore, an adultist view has the tendency to perceive children as incapable of providing accurate representations of their own experiences, and this can stem from how vulnerable children are thought to be (Stafford, 2017).

Another form of prejudice which can impact participation of children with disabilities in research is 'ableism'. Chiourad (1997) defined ableism as the 'ideas, practices, institutions and social relations that presume ablebodiedness, and by doing so construct persons with disabilities as marginalised, and largely invisible 'others' (p.380). Essentially, any attitudes or behaviours which are prejudicial and/or discriminatory towards an individual with a disability can be described as ableist (Wolbring, 2012). For example, assumptions of a child's capability can be made by prejudicial perceptions of their body shape and/or assumptions of their capacity can be made based on their speech/communication abilities. This can be dependent on an individual's perception of what 'normal' ability is. Assumptions for both capacity and capability can impact a child's potential for inclusion in research, particularly

the assumption surrounding capability because of the nature of CP and the mobility impairments associated with CP (e.g., muscle spasticity and the impact on walking ability).

Both 'ableism' and 'adultism' can be detrimental to the inclusion of children with disabilities in research in their own right, however, a combination of both social constructs heighten the potential for exclusion. Therefore, to overcome both ableist and adultist assumptions, a change in practice can begin with researchers recognising children with disabilities as legitimate research participants. This can help in guiding future research methods and practices towards the child with and without disabilities.

3.4.3.1 Capacity of Participants in Study 2

In study 2, the participants were not pre-judged on their ability to participate in frame running as the participants with CP were recruited through frame running clubs, and so it was assumed they possessed the capability to participate in frame running in some form which was confirmed by the coaches and physiotherapists present at the frame running clubs. Furthermore, the frame running research was focused on novice athletes and therefore a limit on ability was not set for participation, and inclusion was aimed at all those willing to participate given the accessibility of frame running to children and young people with more severe impairments.

Assumption of capacity is more complex and had to be considered throughout the second study as participants were asked to complete questionnaires regarding psychosocial aspects of their lives on numerous occasions. Considerations were two-fold, as cognitive impairments associated with CP may limit the ability for participation, and also what to do if the participants were affected by any of the questions raised throughout the intervention. Firstly, the potential participants and their parents/carers were consulted about their cognitive ability and made aware of what the research would entail. With a range of cognitive and communicative abilities across the participants, it provided an opportunity to examine the effects of frame running for children with CP with different cognitive abilities.

It was noted that some participants required assistance when reading and/or completing the questionnaires, which can have methodological implications and cause alterations to the protocol (e.g., change of research environment; see section 3.5.2). However, given the aim of providing valid results representative of individuals across impairment levels, including more severe levels of impairment, it was important to be as inclusive as possible in recruitment. Furthermore, the exclusion of individuals with more severe disabilities can also be viewed as unethical because they are the only ones able to provide an insight into their life experiences, and help with future care, research, policies and/or rehabilitation (Tuffrey-Wijne et al., 2008).

The assumption of capacity, including employing ableism and adultism, towards children with disabilities can clearly cause restrictions to participation and can lead to ethical considerations for researchers. However, as researchers come to the realisation that children with disabilities can provide a unique insight into their life and lived experiences, and the research does not rely solely on proxy reporting by 'other' adults, the approach to research methodology can focus more on the child. With a more child-centric approach to research, caution has to be applied for potential participants as participation could elicit negative emotions, and therefore hands ethical responsibility to the researcher (Kirk, 2007). Along with the assumption of capacity, another ethical consideration for research is the gaining of consent from participants, and this can be more salient for children with disabilities, and this is discussed in the following section.

3.4.4 Informed Consent

Informed consent is an important ethical matter for all research and means that participants voluntarily enter the research and have the understanding of the study including what they are required to do, and the potential dangers involved (Bogdan & Biklen, 1998). Gaining informed consent from children prior to participation can be more complex in comparison to gaining consent from adults, and this is furthered when the participants are children with disabilities. In all situations, researchers must provide enough information to potential participants in a way that is understandable to them and allows them to make an informed decision regarding participation (Einarsdóttir, 2007). The way the information is provided will vary across research studies but needs to be tailored appropriately to the children being recruited in terms of their age, developmental needs, and the presence of any disability to ensure it is in a language they are able to understand. It may be problematic ensuring children with and without disabilities understand fully what it means for them to take part, however it is important for them to comprehend the purpose of the research, what may be expected of them throughout the research, and how long for. Furthermore, the risks of participation should also be made clear, along with what will happen post participation in terms of the data and results.

Another important aspect of informed consent is ensuring that children understand their participation is voluntary, giving their consent is voluntary, and the understanding that withdrawal (at any time) from the research project is fine and holds no consequences (Davis, 1998). However, this can be difficult for the participants who have developmental disabilities (e.g., cognitive, communicative). Children with disabilities are under the legal guardianship of an adult who is able to provide consent for them, but this can cause some issues. Having an adult, whether it is a parent or third party (e.g., a teacher), provide consent for the child to

participate may mean the child is being volunteered as a research participant. Ethically, this is difficult for the researcher, as it contradicts the idea that all potential participants are to be able to comprehend the purpose of the research, what they are required to do, the potential risks, and their right to withdraw at any point. Furthermore, it provides a challenge to the researcher that seeking the informed consent is the obtained informed consent (Einarsdóttir, 2007). Even though it appears they have given their consent, some indicators such as hand gestures may indicate their real feelings (Alderson, & Marrow, 2004), and in this situation, the researcher can face an ethical dilemma as the consent obtained from an adult may not align with the wishes of the participant and opportunities to withdraw offered by the researcher may effectively be closed off to the participants.

In relation to children with disabilities that can impair their cognitive ability (e.g., cerebral palsy) thus impacting their capacity to give informed consent and/or express their views, different approaches to gaining consent need to be taken. In the past, children with learning disabilities were thought to not possess the ability to make decisions for themselves. However, this attitude has changed and developed, allowing children to be seen as competent not when they reach a certain age but when they possess the capacity to make decisions themselves and understand the consequences of decisions (Dunn et al., 2006). If one does not possess this capacity, one approach to gain informed consent is through 'consent networks' and this is the approach used in the current research. A consent network allows the researcher to obtain consent, and possibly assent, from the participant by involving individuals close to the child, who know the child well. Normally it would involve people who understand them, care about them, and recognise when they are unhappy or distressed, and these tend to be family members. Furthermore, viewing participation as an ongoing process, thus consent and assent (if required), can be continuously monitored for the participants, giving participants numerous opportunities to withdraw. Therefore, gaining proxy consent from a legal guardian or parent, along with assent, although not the most desirable outcome, is a satisfactory and necessary compromise for research, allowing for children with disabilities to participate in research.

3.4.4.1 Gaining Informed Consent for Individuals with CP

In study 2, the process for negotiating consent for the children with CP to participate in the intervention began with contact with the frame running clubs. Once contact had been established, the aims and scope of the study were outlined, including the participant inclusion criteria which specified that children with CP who were just starting, or lined up to start frame running were eligible for inclusion in the study. The gatekeepers provided contact details of parents whose children fitted the inclusion criteria and who agreed to be contacted, and these families were contacted with initial information about the research and what it

would entail. For this study, the children were given the opportunity to give informed consent, but a parent or guardian also provided informed consent to ensure legal consent was obtained. Given the participants ranged in their cognitive ability, as is the nature of CP, an individualised approach for gaining consent was employed, and it was a parent or guardian who introduced and explained the research to them.

This approach does leave participants open to being 'volunteered' for the research, and possibly having a more extended period of direct contact with the researcher, explaining the research and what it employs etc. may have given more meaning to the children. However, as part of the research involved attending frame running training sessions which they had planned to attend prior to giving consent, it was hoped that consent was genuine. Furthermore, the parents/guardians were made aware that some of the questions/topics included in the study may cause added distress, particularly for the older participants. This was combatted by providing contact details for mental health and disability charities if there was to be any negative impact on wellbeing from participation and the advice to stop if this was the case. Given the design of the study, with data collection occurring on multiple occasions, the opportunity to gain ongoing consent from participants was taken to ensure participants still wished to be part of the research throughout the process, and it was not just at the onset.

For study 3, the process of gaining consent began with establishing contact with potential participants (i.e., frame running athletes) and gatekeepers (i.e., frame running coaches), and the aims and scope of the research was outlined. As the recruitment and survey was online, gaining consent can be more difficult because there is a lack of interaction with the participants. In study 3, the survey was set up online to ensure participation was only possible by participants actively consenting to participate (i.e., they were not able to proceed without providing consent). This raises the risk of ableism affecting the study, as it can exclude the frame running athletes who are unable to read, understand, and answer the questions without assistance. Given the inclusivity of frame running and the severity of impairments some athletes might have, the risk of ableism may mean the views expressed are not representative of all athletes.

The negotiation of consent at the beginning of the data collection could be termed as 'provisional' consent as it was based on the agreement between researcher and parent of the participant and consent was ongoing based on the relationships between the researcher, participant and gatekeeper (Flewitt, 2005). The process of negotiating and gaining consent for both studies 2 and 3 stretched across several months, alongside the continued identification of potential participants. The longer period of time taken for identifying participants did allow for the participants to reflect on the research, the information given to them, and gives them the opportunity to ask questions, raise concerns or withdraw,

suggesting all participants consent was 'real'. Once consent is gained, and the data collection has begun, protecting the confidentiality of the participants is a key ethical principle in research (Nind, 2008).

3.4.5 Confidentiality & Anonymity

Ethical issues may be expected more within research involving children because of the way researchers and other adults can view children as vulnerable and incompetent (Kirk, 2007). Research has an ethical duty to protect identity of participants, unless stated otherwise, so the participants are not placed in any harm or are in fear of embarrassing themselves; this is particularly true in qualitative research where information gathered tends to be more personal, reflecting individual's thoughts and opinions (Bogdan & Bilken, 1998). To protect the confidentiality of participants, British educational research guidelines suggest that any details that could make the participant known should not be given, and pseudonyms should be given if a name is required throughout the results and discussion (British Educational Association, 2004). Confidentiality and anonymity are similar yet different concepts, which are both key ethical principles in all research (Wiles et al., 2006). Confidentiality means that any information gathered is not discussed with any other individual and that it is ensured participants cannot be identified when data is presented, which is mainly done through anonymisation (Wiles et al., 2008). The anonymity of participants means that it is not possible to identify a participant from the data presented in the research, and this is done through anonymisation of participants via identification (ID) numbers or pseudonyms.

In many studies, qualitative and quantitative, sensitive topics are involved, and confidentiality and/or anonymity are seen as particularly important in these cases. This does not solely relate to the processing and storage of data but also for data collection. For example, if conducting an interview with a child with CP in the family home, a separate space may not be feasible because the parent is there for communicative purposes, which may impact the data collected in terms of the interpretation of the participants views, feelings, and opinions, which they may not feel able to communicate fully. Confidentiality can be most problematic when dealing with sensitive data and vulnerable groups when it appears that an individual's health and/or life can be on the line (e.g., Valentine et al., 2001; Bradbury-Jones et al., 2018). Researchers may face an ethical dilemma if they encounter information that they feel may need to be passed onto authorities, for example an incidence of child abuse or violence, but this may be seen as exceptional circumstances and so researchers can be expected to pass on the information to higher authorities (Flewitt, 2005). Furthermore, it can be agreed with children prior to participation, if possible, that should the researcher discover information that can risk an individual's life, confidentiality can be

broken (Einarsdttir, 2007). However, this can also be difficult for children with disabilities who may not be able to comprehend the idea of confidentiality, presenting the researcher with further predicaments. If the information was viewed as something adults should know, but is in the opinion of the researcher solely, they have an ethical obligation to not betray the participants.

As previously mentioned, confidentiality and anonymity can be more difficult when carrying out qualitative research, where despite the use of pseudonyms or ID numbers, the participants may still be identified from data, for example if pictures, audio and/or video recordings or personal quotes are used which allow them to be recognised, thus putting them at risk (Flewitt, 2005). This can be particularly difficult when participants change their attitude towards the research post participation and consent may alter (Harcourt & Conroy, 2005). Furthermore, the readers of the research may interpret the data differently, making their own judgements on the data, and further putting participants at risk, hence the importance of both confidentiality and anonymity. However, anonymity has also been discussed as a negative aspect for participants involved as it can hide a person's story. For example, for children with disabilities inclusion in research is restricted, and the opportunity for them to participate, and share their story and /or achievements, may be something they want to do because they are proud. However, this can be dangerous as the removal of anonymity can incite a level of risk as the effects on others around them are unknown and unpredictable (Tuffrey-Wijne et al., 2005), highlighting the ethical importance of anonymity, and the ethical dilemmas researchers can face with regards to both anonymity and confidentiality.

3.4.5.1 Confidentiality & Anonymity in the Current Thesis

In the current research, across all three studies confidentiality and anonymity was maintained. For the qualitative studies, one and three, participants were given ID numbers were used to protect the identity of the participants. Study three involved frame running athletes (each participant was over the age of 18), and the frame running community is relatively small, especially at the elite end of the sport, and combined with a small sample size, protecting the confidentiality of participants can be more difficult. It has been stated that confidentiality of participants should not be promised by researchers to participants as the nature of research can be unpredictable (as discussed in the previous section) but protecting the confidentiality but protecting the confidentiality is the researcher's ethical responsibility (Einarsdttir, 2007), as well as a legal requirement. With this in mind, aspects discussed in the qualitative study did involve some sensitive topics about psychosocial wellbeing, hence protecting the confidentiality is important to not cause embarrassment, self-doubt, or anxiety for the participant. To ensure confidentiality, pseudonyms were given to participants, and

any details that could reveal the identity of the participant were not included in the reporting and presentation of the data.

With reference to the quantitative data, confidentiality was also important as the participants completed questionnaires which included some questions that are related to sensitive topics (e.g., psychological wellbeing). This was done by presenting data with ID numbers, and not including any information which may lead to the identification of the participant. In all three studies, as much confidentiality as possible was promised when negotiating consent, and to break this would be to betray both the participant's and parent's confidence.

3.4.5.1.2 Obtaining Ethical Approval in the Current Thesis

The ethical considerations for research can be complex, and they become more salient when involving children with disabilities. For each of the three studies in the current thesis, ethical approval was obtained from The School of Education Ethics Committee of the University of Edinburgh, but with the different study designs and populations in each study, each application had its own ethical considerations. Study 1, a two-stage online Delphi-survey, involved participants above 18 years of age, and did not ask about sensitive topics, presenting minimal risk to the participants, hence the ethics application was more straightforward in comparison to the subsequent studies.

Study 2, investigating the effects of a 12-week frame running training programme on the QoL and psychosocial wellbeing of children and adolescents with CP presented ethical considerations as the participants were children with CP and under 18 years. One of the ethical considerations highlighted by the ethics process was the concern regarding the cognitive load placed on participants when completing the questionnaires administered by the researcher, particularly given some of the questions were around sensitive topics such as psychological wellbeing, relationships, and perception of physical appearance. Participants and parents were explicitly made aware the research was voluntary and could withdraw at any point, along with available resources that could help (e.g., charity websites and phonelines). Furthermore, obtaining informed consent was highlighted as an important ethical consideration for this study as not all participants may have the capacity to give informed consent, therefore, it was ensured that parents or guardians provided informed consent for the participants prior to research. Study 3, an online survey investigating the effects of frame running participation on the QoL and psychosocial wellbeing of experienced frame running athletes, presented similar ethical considerations as study 2, regarding the inclusion of sensitive topics, and obtaining informed consent from participants. As an online survey, it was ensured that informed consent was obtained online, and it was possible for parents or guardians of children wishing to participate to provide consent, as well as adults.

The ethical issues which can revolve around children with disabilities inclusion in research such as assumption of capacity, gaining consent and confidentiality, can all have methodological implications for research, and the impact on the current methodology is discussed in the following sections.

3.5 Methodological Implications

In addition to the ethical consideration's researchers face for the inclusion of children with disabilities, as discussed above, researchers also encounter methodological challenges when carrying out qualitative and quantitative research. The methodological challenges faced in the current research, which will be discussed in the following sections, are gaining access to participants, data collection, and data analysis.

3.5.1 Gaining Access

As discussed in section 3.4.1, gaining access to participants with disabilities can be challenging ethically, but it can cause methodological issues too. Gaining access to participants for the current thesis was a long and complex process. Gaining access to participants for study one was the most straightforward process, with the use of gatekeepers, and as participants were all adults, there were no concerns about gaining consent.

Recruitment for the second study was the most complex process and extended across many months. The target population of eligible children is spread out across the country and difficult to reach directly. Moreover, frame running clubs are small and ran by volunteers who represent a variety of backgrounds, usually including physiotherapists, athletic coaches, and parents. Networking activities put the researcher in touch with various organisers of clubs. Although frame running is a relatively new sport, in the Scottish context there has been considerable promotion of the sport through Frame running Scotland and Scottish Disability Sport for over a decade. Clubs are steadily growing, but new members are added sporadically and unpredictably, usually as the result of word-of-mouth communication. When recruiting for the second study, if children were not already signed up to participate, identifying children who were willing and able to not only sign up to a new activity but also to a research study produced methodological implications. Furthermore, there was a high level of reliance on gatekeepers, both organisers of frame running clubs and parents, because even if the child did wish to participate a lack of time, finances, space, transport, the availability and storage of frame running bikes, access to athletics track and coaches could all be contributing factors to a lack of participation. These restrictions led to a narrowing of eligible participants, and initial consent did not always lead to participation, thus

recruitment was affected and was mainly based on convenience. Efforts were made outside of the frame running clubs to recruit participants, in the form of frame running come and try days across Scotland, but the children who attended were already participating in frame running, therefore giving a volunteer sample which may not represent the general cerebral palsy population. In retrospect, the cognitive load placed on the participants may have provided a negative view for parents, who may not want to inflict additional distress and anxiety on their child, therefore not moving forward past the stage of initial interest. Perhaps a more child friendly approach to the data collection would have encouraged more parents to have their child participate (i.e., less time spent filling out questionnaires). Given the importance of inclusion of children with disabilities in research, it will be important to have a suitable methodology to encourage engagement in research, such as a child-friendly approach. The importance of research methods and techniques for research involving children with disabilities is discussed in the following section.

3.5.2 The use of Research Methods & Techniques

As discussed earlier (see section 3.4.3), children with disabilities may be excluded from research because researchers assume they are unable to participate because of the way they look and/or speak (ableism), or they are believed to not possess the ability to be reliable or competent participants. Furthermore, children who have cognitive and communication impairments may be excluded from research because the methods used to gather the data, such as questionnaires and interviews are not suitable for the population (Morris, 2003). The main objective for researchers is to employ the most appropriate methods and tools which enable children to participate in research to the fullest extent and provide them with every opportunity to express their views and opinions (Davis, 1998). For all research it is important to use the most appropriate assessment tools, but for research involving children, more child-friendly or child-centric approaches have been considered (e.g., activity-based interviews; Stafford, 2017; Kirk, 2007). In the following sections the data collection methods used in the current thesis are discussed, and the methodological implications of the data collection methods for use with children with disabilities, specifically cerebral palsy.

3.5.3 Quantitative Research Methods

3.5.3.1 Questionnaires

The use of questionnaires is not a common method of data collection for young children, however quantitative data gathered from children has become recognised as an important method of data collection, so there is not sole reliance on qualitative data or proxy reported

quantitative data (Bell, 2007). As previously discussed, children being viewed as research participants in their own right has been an important development in research as the opinions and views children hold and which they can express through research can provide valuable data. In relation to qualitative data, children were thought to not possess the ability to express how they were truly feeling or separate the 'real' world from what adults see as their fantasy, and therefore they were thought not to be able to provide reliable data. In terms of gathering quantitative data, there is more of a concern surrounding children's cognitive and communicative capacity to understand the questions and provide good quality responses, as well as their ability to concentrate for a period of time to answer many questions (Bell, 2007). Furthermore, as Piaget's theory of cognitive development (1929) shows, as a child's functions develop through childhood and adolescence (e.g., language, and memory), this can impact on their ability to answer a question. Moreover, development will impact children to varying degrees and at different times suggesting gathering 'good quality' data from children can be a more difficult process in comparison to gathering such data from adults, but children's inclusion enriches the data collected across research and society (Scott, 2000).

The research methods involving children as participants have to be carefully considered given the range in cognitive and social development of children. For example, the use of standard questions is not appropriate for nursery aged children (Scott, 2000). Furthermore, decision and selection of research methods for children with disabilities can be more complex as it is dependent on the individual's disability and the severity of it. For example, the cognitive and communication impairments associated with cerebral palsy can make it more challenging to collect 'good quality' quantitative data from participants with CP. The use of questionnaires within research involving children with CP is a less common research method, but is done (e.g., King et al., 2004).

The use of questionnaires for children can be beneficial as it presents the researcher with an opportunity to collect data on a larger scale, in a shorter space of time (dependent on the questionnaire administered), and the data collected is more standardised in comparison to qualitative data, allowing for more comparisons. Furthermore, the participant may be more willing to complete a questionnaire compared to answering questions face to face and having to give more detailed and expansive answers. However, this can also have a negative effect as a child may not understand the question or know the answer but still respond, thus potentially introducing random error the results. Furthermore, not all participants have the communication, cognitive, or physical ability required to complete questionnaires which may discourage participation. To encourage engagement and improve the quality of the data collected, researchers can administer more 'child-friendly' questionnaires as they are more suitable and relatable to children. With reference to the

inclusion of children with CP, the use of questionnaires which are specific to the health condition can be beneficial as they are more relevant to the participants, able to detect more subtle changes, and an example of this is discussed in the following section.

3.5.3.2 Condition Specific Measures

One approach to improving data collection for children with disabilities is the use of condition specific measures (e.g., pediatric QoL inventory – cerebral palsy module; Varni et al., 2001). The use of standardised questionnaires designed for use across the general population can have a low validity when used in a population of individuals with a disability because some questions or items may not be relatable or relevant to those with disabilities. However, condition specific measures are designed so they can be applied to one group, such as individuals with CP, and are able to target the issues specific to the given condition (Gilson et al., 2014).

Using QoL as an example, it is one construct that gets a lot of attention, particularly for children with CP, as improving a child's QoL can be seen as the overall aim for a lot of research. Furthermore, measuring QoL can provide the researcher a more rounded picture of the impact CP is having on aspects of the individual's life (i.e., physical and social wellbeing; Gilson et al., 2014), thus, highlighting the importance of having condition specific measures. There can be additional difficulty when measuring the QoL of individuals with CP due to the complexity and unpredictability of the chronic condition (Makris, et al., 2019).

There are generic instruments for measuring QoL, and these can be administered across a range of populations and demographics to allow for comparison (e.g., Waters et al., 2009). However, the generic instruments carry potential issues as they may lack the sensitivity to detect more subtle changes in factors that are more specific to certain conditions (e.g., reduced muscle tightness in children with CP can lead to better mobility), and this may lead to information being lost or missed (Waters, 2009). Therefore, there are suggestions towards the use of condition-specific instruments, and even though some factors may overlap with the more generic instruments, the condition-specific measures are more sensitive to the changes in factors specific to that condition. For example, cerebral palsy measures of QoL will measure the pain and fatigue domains, as well as social and emotional well-being which are also measured in more generic scales (Waters, et al., 2009).

However, the completion of condition specific measures can remain problematic if the participants do not have the capacity to fully understand or answer the questions, leading to further methodological considerations for the researcher. Furthermore, condition specific measures are not available for each and every construct, therefore reliance is placed on previous research, and the use of the measure with specific populations, for

example, previous research has reported reliability and validity for the use of the Children's Assessment of Participation and Enjoyment for children with CP (e.g., Bult et al., 2010).

In the current research, a mixture of condition specific and generic measures were used with participants to measure the impact of frame running on psychosocial wellbeing of children with CP. As mentioned previously, the participants varied in age (4 to 18 years) and in cognitive and communicative ability making the process of outcome measure selection difficult. As participant take up was low, it was important to not exclude any potential participants by using measures that were not suitable or accessible for every participant. In a situation where the child is struggling to understand and/or complete the questionnaire, an adult 'close' to the participant (e.g., a parent, carer, or guardian) could be asked to complete a proxy-report, however, proxy reporting may also be more prevalent with this research population as researchers do not recognise children with disabilities in their own right. The proxy- versus self-report is briefly discussed in the following section.

3.5.3.3 Proxy-report versus Self-report

As previously mentioned, there are some instruments that are specifically applicable to children and young people with CP (Matza, et al., 2004) but there is a heavy reliance on proxy-report, which can lead to limitations in interpreting the results, especially if that is the only measure (Varni, et al., 2005). Parents may be able to report sufficiently and accurately on certain domains, such as physical functioning, but when it comes down to more internal domains, such as social and emotional well-being, a child self-report would be more accurate in comparison to a parent proxy-report (Varni et al., 2005). Of course, there are expected to be difficulties when children with developmental deficits (e.g., communication impairments) are asked to self-report how they feel.

A previous study by Varni et al. (2001) showed that typically developing children as young as 5 years old were able to reliably self-report when using the pediatric QoL inventory version 4.0 (PedsQL 4.0; Varni et al., 2001). Thus, Varni and colleagues (2005) investigated the ability of children with CP to self-report using the PedsQL 4.0, a generic measure of HRQOL. Participants ranged from ages 5 to 18 years, with 47% of participants with CP able to self-report. In comparison to their typically developing peers, the participants with CP reported a significantly lower overall HRQOL score. Specifically, in the CP groups, the less impaired children diagnosed with diplegia and hemiplegia self-reported higher HRQOL scores compared to the children with quadriplegia suggesting that the perceived HRQOL was related to the severity of their CP (Varni et al., 2005). The results may be as expected, with children with less severe impairments reporting better results, but importantly Varni & Colleagues (2005) reported almost half of the sample of children with CP were able to self-

report. Thus, suggesting proxy-reporting does not have to be the main method of quantitative data collection for research involving children with CP.

3.5.4 Qualitative Research Methods

3.5.4.1 Interviews

The gathering of qualitative data for children with and without disabilities is a more common method of data collection in comparison to quantitative research (Einarsdóttir, 2007). The nature of qualitative research is much more flexible as the researchers are not bound by standard methods but are able to be more creative with their methodology (Kelly, 2007). This can be particularly beneficial for research involving individuals with disabilities as it allows for a more flexible approach which may increase the levels of responsiveness from participants (Beresford, 1997). Furthermore, if individuals with disabilities struggle with cognitive and/or communication functioning, an interview approach can help to build a rapport with the participant and provide them with the opportunity to express their opinions, life experiences, and views (Kelly, 2007).

The presence of a cognitive or communication deficit can add to the methodological considerations facing researchers when involving individuals with disabilities in qualitative research, such as interviews. The methodological considerations for researchers involving individuals with disabilities include the authenticity of the opinions and views, validity of responses (i.e., whether responses are interpreted correctly and are in line with what the participant believes; Lewis, 2002). A standard approach to interviews, such as question and answer, can be restricting for participants in comparison to other methods which allow for more of a recall of experiences and knowledge (e.g., narrative methods; Atkinson et al., 2004). Furthermore, individuals with disabilities who require support to give their responses may have their responses distorted by the interpretation and phrasing from the support, which links back to the authenticity, reliability and validity of interviewing individuals with disabilities, particularly those with cognitive and communication difficulties (Lewis, 2004).

On the other hand, the use of open-ended questions in an interview involving individuals with more severe impairments can yield inadequate responses as they are unable to answer or can only provide a little information (Lloyd et al., 2006). Therefore, a more standard and structured approach (e.g., a yes-no set of questions) may provide the researcher with richer, and more reliable data, as well as the simplicity of the interview aiding the participants, and possibly encouraging participation (Coons & Watson, 2013). The two approaches mentioned in this section highlight the importance of selecting and modifying the approaches researchers apply when interviewing individuals with disabilities with varying levels of impairment (Lewis, 2009).

In an attempt to reduce methodological dilemmas, there are some participatory methods which can help the participants' voices to be heard authentically (Stafford, 2017). The use of more creative approaches has been emerging and popular methods for interviewing children with disabilities include the use of pictures, drawing, music, photographs, and art (Gillies & Robinson, 2012). The application of more creative-art methods is thought to help children express themselves in more than one way, thus aiding more abstract discussions and gaining access to more diverse experiences, and therefore more diverse data (Gillies & Robinson, 2012). Activity based interviews are one approach that can be beneficial towards children with disabilities because it can incorporate activities (e.g., drawing) to help participants explore and express their experience, as well as explore the meaning of them (Stafford, 2017). Furthermore, the application of creative, activity-based interviews presents the researcher with more flexibility and a non-threatening approach (i.e., fun), thus being able to accommodate for a wider range of physical, cognitive, and communication impairments. Although interviewing children with more severe CP, and associated impairments, can be more challenging and time consuming in comparison to those with mild or no impairments, it is possible with the selection of the most appropriate communication and methodology for the research population. More importantly, it may be the only way in which researchers are able to capture the lived experiences, opinions, and perspectives of the more impaired children (Coons & Watson, 2013).

3.5.4.2 Quantitative & Qualitative Methodology for Studies 2 & 3

With both quantitative and qualitative research methods discussed in the previous sections, there are advantages and disadvantages for each when involving individuals with disabilities in research.

In the current thesis, both quantitative and qualitative methods were applied, and in study 2 a quantitative approach was used with participants completing standardised questionnaires. The use of the selected questionnaires, which are designed for the population, and relevant to their life (e.g., paediatric QoL inventory – cerebral palsy module; Varni et al., 2001), allows for a lot of detail to be gathered through relatively straightforward methods (e.g., yes or no questions, and multiple-choice questions). The simpler question format and structured approach may provide richer data, particularly for individuals with more severe impairments, and children with parents' help can fill out the questionnaires reliably. Furthermore, the use of the questionnaires provides established validity and easier comparison between participants of the study, and with past and future research.

Quantitative methods were not applied in study 3 as the use of the same standardised questionnaires would provide a snapshot of how the participants were feeling at that time, but the aim was to gather athlete's opinions when reflecting on the impact of

frame running on their psychosocial development. Therefore, qualitative interview-style questions were constructed, with open questions inviting longer answers around the same psychosocial constructs as study 2. This was done in an online survey format to include as many athletes as possible from around the world, as well as those with communication impairments (for individuals with CP, communication problems are common, with prevalence estimated between 38% and 78%; Choi et al., 2018). The use of qualitative methods in study 3 meant there were no wrong answers, as the interest was around the participants' own feelings, reflections, lived experiences, and how frame running has affected them in various psychosocial domains. The aim was to elicit rich data regarding frame running participation and the effect on psychosocial wellbeing, which may not have been possible from the sole use of quantitative methods.

3.5.4.3 The Delphi Technique

Another form of data collection which was used in the current thesis was the Delphi technique, and this relates to the first study (see chapter 4). The Delphi technique was used to identify areas of interest when evaluating the effectiveness of a physical activity intervention for children with CP. This technique has shown to be useful for collecting information in a range of research domains and this brings a range of methodological interpretations (Donohone & Needham, 2009).

The Delphi technique is becoming a more popular methodology within social and health science (Ashmore et al., 2016). It typically consists of several rounds of questionnaires that are circulated to participants with the results from the previous round influencing the questions asked in the next round (Donohone & Needham, 2009). It is used for gathering opinions from a panel of experts or stakeholders in an area, often with the aim of building consensus or exploring a field beyond the existing knowledge (Iqbal, & Pison-Young, 2009). The interval between the rounds allows the participants to think independently about the questions, with a lower chance of being influenced by outside factors or via the power of persuasion and authority (as may be the case in focus group research, for example), and in turn should produce higher quality/less biased answers. One characteristic of the Delphi approach that is important to this study, is that it allows for the complexity of a topic to be addressed without there being any right or wrong answers; a Delphi study usually asks for opinions rather than facts. Participants of a Delphi study have the freedom to answer as they wish. Furthermore, the technique encourages honesty with no group pressures, and participants are able to challenge the topic with their own viewpoints (Donohone & Needham, 2009).

The Delphi technique can be used when it comes to decision making and inquiring into domains where there is little previous knowledge, as it provides a structured approach to

collecting data in a range of domains and situations (Hanafin et al., 2007; Ashmore et al., 2016). The reliability of the Delphi has been questioned as there are concerns over the strong dependence on the panel, (i.e., it has been questioned whether similar findings would be obtained if the same information was presented to a different panel; Ashmore, 2016). Furthermore, the validity has also been criticised as the researcher does not have any influence over the development of each stage (Keeney et al., 2001). However, it has also been stated that if the participants of the Delphi are representative of the topic of study, then the validity may be assumed (Goodman, 1987). Furthermore, previous research has shown the Delphi approach can be selected based on its merits and being a suitable approach for the aims of the research (e.g., Vargus-Adams & Martin, 2009; 2010).

The Delphi approach was selected for study 1 (see section 4.3.1). This method was deemed as suitable for study 1 as the aim of the study was to not to identify facts, but to identify opinions about what participants thought were most important to measure when evaluating physical activity interventions for children with CP. Moreover, the use of multiple rounds will allow the more general areas mentioned by the panel to be analysed, before focussing on gathering more precise feedback from each panel member on which factors, they thought to be most important to measure. Furthermore, it allowed for distribution across the UK, increasing the number of potential participants.

The results produced from a Delphi approach study represent a more opinion-based set of results compared to factual, and this is one of the issues with the interpretation of results from Delphi studies. Demonstrating and establishing rigour in research, which can be viewed as a marker of good quality research, is a complex process, and this is discussed in the following section.

3.6 Data Analysis and Interpretation: Establishing Rigour

3.6.1 Thematic Analysis

Within disability research, qualitative research is continuing to grow and be recognised as a vital tool to help with the understanding of the complexities of disabilities (O'Day, & Killeen, 2002). One of the strengths of qualitative research is that it can highlight interactions and/or attitudes within an individual's life and environment, which is important for disability research as the individuals with a disability are the only ones who are able to provide that insight, thus enhancing the understanding of disability. Given the importance of qualitative research in the disability field, it is important to demonstrate and establish rigour for good quality research. An important implication of this research is the reliability and validity of the data collected, both quantitative and qualitative, and this may be more challenging in research involving participants with more severe physical, cognitive, and/or communication impairments.

Through the analysis of qualitative data, researchers are able to have a more general overview of the data, and then focus more on themes and patterns that may emerge. Qualitative data analysis can be challenging and complex as it asks researchers to take a large amount of data, and identify significant words and phrases, and construct themes in a way that is suitable to communicate as results (Patton, 1990). Thematic analysis is one of the more widely used methods of analysis and is seen as a ‘foundational method’ of qualitative research (Braun, & Clarke, 2006). Other qualitative research approaches include grounded theory, discourse analysis, and interpretive phenomenological analysis, but as thematic analysis is applied in the current thesis, it is the focus of this section. Qualitative analytic methods can be complex, and one benefit of thematic analysis over others is the flexibility as it is independent of theory and can be applied across theoretical approaches (Braun & Clarke, 2006). The application of thematic analysis allows researchers to analyse the data in a systematic way to identify and report themes within the data which are relevant to the research, thus providing an answer to the research question(s). To do this, Braun and Clarke (2006) outline the process of thematic analysis through a six-step process and once themes are identified through familiarisation, coding, theme development, revision, defining of themes, it is important rigour is demonstrated to ensure the quality of the research (see Table 3.1).

Table 3.1 6-step process of thematic analysis from Braun & Clarke (2006)

	Phase	Description of Process
1	Familiarising yourself with the data	Transcribing data (if necessary), reading, re-reading the data, & noting down initial ideas
2	Generating initial codes	Coding interesting features of the data in a systematic fashion across the entire data set, collating data relevant to each code
3	Searching for themes	Collating codes into potential themes, gathering all data relevant to each potential theme.
4	Reviewing themes	Checking if the themes work in relation to the coded extracts and the entire data set, generating a thematic ‘map’ of the analysis
5	Defining and naming themes	Ongoing analysis to refine the specifics of each theme, and the overall story the analysis tells, generating clear definitions and names for each theme
6	Producing the report	The final opportunity for analysis. Selection of vivid, compelling extract examples, final analysis of selected extracts, relating back the analysis to the research question and literature, producing a scholarly report of the analysis.

3.6.2 Developing & Establishing Rigour

Irrespective of the participants' capacity and abilities, it is important for research to be rigorous, as rigour has been highlighted as a marker of quality (Smith & McGannon, 2018). In a general sense, rigour can be viewed as reliability and validity of research, and although the meaning of rigour can vary and mean different things between researchers, it is associated with the quality of the research process (Given, & Saumure, 2008) and the way in which rigour is evaluated must be appropriate to the research and the methodology (Davis, & Dodd, 2002).

In quantitative research, where processes are less flexible, rigour can be determined through the consistency of results, replication across time and contexts, and is based on the concepts of reliability and validity (O'Day & Killeen, 2002). Furthermore, with the aims of quantitative research, (i.e., to measure something through systematic examination of phenomena through hypothesis; McCusker & Gunaydin, 2015) the concept of reliability is important, for example the use of an outcome measure needs to be psychometrically sound for use in the selected research (Smith & McGowan, 2018). Furthermore, in quantitative research, common limitations such as a lack of a comparison or control group, or use of inadequate questionnaires (e.g., not psychometrically sound, or not suitable for the participants) can inhibit methodological rigour (Scrutton, & Beames, 2015). These limitations are found in quantitative research involving individuals with disability in the form of assumption of capacity, gaining access to participants, the use of condition specific measures, and the use of self- and proxy-reports, each of which were discussed earlier in this chapter. Thus, ensuring the correct methodology and psychometrically sound measures are used can help to evaluate and establish rigour in quantitative research as it rests on the concepts of reliability and validity.

3.6.2.1 Inter-rater Reliability

Though rigour has been viewed in a general sense as reliability and validity, these concepts are not appropriate in qualitative research as they reflect a positivist approach. One of the more common techniques to assess the analysis and establish rigour is inter-rater reliability. This method employs two or more, experienced and capable, researchers to code the data independently and come together to compare coding outcomes. Once any differing views/opinions over units of text are settled, and a high level of consensus has been reached, referred to as the discriminant capability, the data is deemed reliable (Campbell et al., 2013). Essentially, inter-rater reliability assumes there is one truth that can be identified.

Despite the popularity of inter-rater reliability, researchers have suggested it is an outdated and ineffective method to develop and establish rigour, and this can be for a

number of reasons (Smith & McGannon, 2018). Firstly, inter-rater reliability relies on the researchers to come to a 'high' level of agreement for the research to be deemed reliable, but there is no literature reporting a common and agreed upon threshold to what constitutes a suitable level of agreement for reliability to be accepted (Culver et al., 2012). This can leave the level of agreement up to interpretation of the researchers involved, thus making it difficult to know what is truly reliable.

To reach a level of agreement/consensus that may or may not be deemed reliable, discussions between researchers are to be had about the coding, and this can also cause some issues around inter-rater reliability. If there is a hierarchical nature between the researchers/coders, such as PhD students, research assistants and supervisors, then power differences between the coders may influence the final decisions made on the codes. Whether it is implicit or explicit, the demographics and experience can all influence agreement further highlighting the difficulty in accepting inter-rater reliability as a process to demonstrate rigour (Smith & McGannon, 2018).

Another, more fundamental concern regarding inter-rater reliability is the inability of humans to produce theory-free knowledge, as prior knowledge, experience of research and theoretical frameworks, mean that the coding process will be influenced (Guba & Lincoln, 2005). Different backgrounds, beliefs, and understandings can mean that however a researcher views the units of text is likely to be different to other researchers, with none of these views being wrong, thus coming to an agreement over meaning of text and codes is going to be unlikely (Campbell et al., 2013). For the reasons discussed, inter-rater reliability is viewed as a flawed method for demonstrating rigour in qualitative research, therefore alternative methods of establishing rigour to achieve high quality qualitative research should be identified.

3.6.2.2 Member Checking

Another method of demonstrating rigour in qualitative research is 'member checking' and involves the researchers asking the participants to review the data and/or results, followed by enquiring if the participants feel the data and/or results provide an accurate representation of their experiences (Lincoln & Guba, 1985). Thus, the research is deemed reliable and valid if the participant agrees the data and/or results are accurate. However, despite the value placed on member checking, previous research has suggested that employing it as a method does not enhance the quality of the qualitative research, hence there are issues with this as a method of developing rigour (Thomas, 2017). Firstly, it is not possible to establish if the participant is fully engaged in the verification process, and aspects such as a large data set, or uninteresting topic may encourage the participant to

agree with everything. Therefore, on paper, the research will be deemed credible and reliable, but the researcher is unaware if this is trustworthy or not.

Similar to inter-rater reliability, power relations may be an issue within the member checking process (Smith & McGannon, 2018). When a participant is asked to check the results and/or data, they may not understand fully what is being presented, thus feel inferior, but agrees with the results to appear equal. Moreover, the participants may feel disappointed or embarrassed by the researcher's interpretation of the data, and feel they are not being portrayed in the correct light (Sparkes & Smith, 2014). This can lead to ethical concerns for the researcher, as the participant may believe the researcher has exploited their power to expose a weakness or vulnerability, thus causing harm to the participants. Within disability research, participants with cognitive and/or communication impairments may also struggle to comprehend the findings when asked and agree with the researcher who they may view as the expert (Estroff, 1995). Individuals with disabilities can have lower self-esteem and self-confidence compared to those without disabilities, so may feel pressure to defer to the 'expert' and agree with the findings, leading to unclear reliability and credibility. Furthermore, qualitative research captures the views, opinions, and experiences of the participant at that point in time, and with a time gap between data collection and member checks (which is not normally disclosed; Smith & McGannon, 2018), the participants view etc. may have changed leading to disagreements about the interpretation of the data. The effectiveness of member checking can therefore be influenced by the time gap, inhibiting the accuracy of the initial data, thus the credibility of the data. The issues mentioned regarding member checking suggest it is not an effective method for establishing rigour in qualitative research.

Throughout the current thesis, member checking was not suitable for any of the three studies. In study 1, the use of the Delphi approach implements a form of checking within the methodology as participants are asked about areas of importance, and then asked to provide a rating of importance on those areas. In study 2, it was not relevant for the quantitative data collection. In study 3, the data was analysed within the frameworks and concepts discussed in chapter 2, and so was not appropriate to check with the respondent. The importance of demonstrating and establishing rigour in qualitative research is clear, as the process can act as a marker of the quality of the research, but for the reasons discussed it is a complex process for researchers.

3.6.2.3 Critical Friend

As discussed in previous sections 'member checking' and 'interrater reliability' were not suitable techniques for demonstrating rigour in the current thesis. One way to enhance rigour is through 'critical friend'. The process of 'critical friend' involves critical conversation

between researchers and other individuals who listen to the interpretation of the data and offer critical feedback (Smith & McGannon, 2018). It is important to note that the process involving 'critical friends is not to "agree" or achieve consensus but rather to encourage reflexivity by challenging each other's' construction of knowledge' (p. 508; Cowan & Taylor, 2016). Thus, the role of 'critical friend' is act as a theoretical sounding board for researchers to encourage development of interpretations and explanations originally identified by the researcher.

One positive aspect of the 'critical friend' process is a critical friend may be positioned differently to the researcher in their research and theoretical interests which can add another dimension to the development of findings as well as challenging the interpretations and explanations of the data. Within the critical friend process, it is important to note that if an agreement is reached with the researcher, it does not mean that the truth has been found or rigour has been achieved. It suggests a consensus or agreement between the researcher and critical friend has been reached that the constructed interpretation or explanation of the data is viewed as an informed and theoretically sound argument which can defend the case made by the researcher (Smith & Sparkes, 2006). Furthermore, if there is disagreement within the process, it does not suggest the original interpretations or explanations are wrong however can highlight additional resources for developing various interpretations of the data from the study (Smith & Sparkes, 2006).

Importantly, the dialogical approach between researcher and critical friend acknowledges that there can be additional interpretations or explanations of the data that exist, are defensible, but not being presented in a particular study (Smith & McGannon, 2018). As previously mentioned, inter-rater reliability was not a suitable option for the current thesis, thus in contrast to inter-rater reliability conversations, throughout the current thesis the use of the 'critical friend' process was implemented between the researcher and two colleagues to enhance rigour of qualitative analysis providing additional and different perspectives to the interpretations and explanations of the data. The variety of research experience and positioning of theoretical interests within the critical friend process lends itself to challenging and developing the findings, therefore a suitable process for the current thesis.

3.7 Summary

Throughout this chapter, the ethical issues and methodological considerations and implications that researchers encounter are discussed. The chapter refers to both previous literature and to the current research, highlighting the difficulties faced in carrying out

ethically and methodologically sound studies that include individuals with disabilities and specifically those with CP with varied cognitive and communication impairments.

The importance of including individuals with disabilities is clear, as they are a population whose voices are not heard as much as they should be, despite research being one of the only ways in which to gain insight into their lived experiences. Previous research has highlighted the prejudice that individuals with disabilities have faced, particularly through the bringing together of ableism and adultism (Stafford, 2017). Furthermore, the restrictive research methods used in previous research have also limited their participation (Davis, 2008), but the emerging use of more creative and appropriate research and communication practices (not used in the current research) highlight the possibilities for future research (e.g., activity-based interviews providing participants with more than one way to communicate and express their experiences; Lewis, 2001). Communication impairments can be common problems for individuals with CP (Choi et al., 2018). These individuals can be excluded from research as they are not able to express themselves fully, leaving their opinions and experiences unheard. The use of the online survey in study 3 allowed for the inclusion of individuals with communication impairments to express themselves and share their opinions and experiences.

Research involving individuals with disabilities can encourage good practice as researchers may need to be more careful in obtaining informed consent, and providing enough information about the research, in a way the participants can comprehend (Kirk, 2007). Therefore, to engage children with disabilities the research methods need to match the research questions as well as considering the specific needs and characteristics of the potential participants (e.g., physical or communication impairment). Furthermore, future research should recognise the value of involving children with disabilities, and the knowledge and experiences they can bring to research and literature, which applies to all levels of severity. Thus, the ethical and methodological considerations that need to be made for inclusion of children with disabilities can be complex, particularly for children with CP given the range of associated impairments of severity of their condition, but necessary to enrich our understanding of their experience, and to inform future research.

Chapter 4 – Delphi Study: Outcomes of Importance of Physical Activity interventions for children with Cerebral Palsy (CP) Identified by Family Members, Healthcare Professionals, Teachers, and Coaches.

4.1 Introduction

Parents, clinicians, health care professionals, and researchers are continually looking for simple, straightforward, and meaningful ways to treat patients, and to evaluate whether a certain treatment or intervention has been effective or not.

As introduced in Chapter 2, Cerebral palsy (CP) is a complex and chronic condition affecting an individual's movement, motor ability, and muscle tone, alongside associated deficits (including behaviour, cognitive, perception, and communication problems), to varying severities (Gulati & Sondhi, 2018). The heterogeneity of CP means there will be difficulties for healthcare professionals in selecting the most suitable treatment programme or intervention. Alongside selecting the most helpful interventions, determining whether or not the intervention(s) have been effective is another challenge health care professionals face. In order to evaluate the effectiveness of different interventions, there may be a broad focus (i.e., numerous outcome measures), as this will allow for a more rounded picture to be painted about the impact of that intervention on children with CP. Furthermore, a broad focus could assist all interested parties in understanding the changes that occur through development with CP in addition to the changes that may occur via participation in a specific intervention (Vargus-Adams & Martin, 2009).

For CP, standardised outcome measures have become more prominent in research and clinical settings in recent years. However, less research has been conducted to investigate the types of outcomes that are seen as 'most important' in relation to interventions for children and adolescents with CP (Vargus-Adams, 2010). Furthermore, views of what *is* most important will differ between different parties (i.e., a parent's view may be more focused on communication, a child's view may be more focused on being more functionally independent, and a health care professional could be more focused on decreasing pain). Therefore, before we measure outcomes related to a frame running intervention, we need to determine what is considered most important.

This chapter will provide a critical review of the previous literature investigating the establishment of important factors and selection of outcome measures for CP research. The aims of the current study will then be stated before proceeding to report the methods, results, and discussion of findings. The intention for this study is to add to the current literature by establishing the domains of most importance for evaluating the efficacy of a

physical activity intervention for children with CP from the viewpoint of parents and health care professionals.

4.2 Review of Previous Research Identifying Important Domains to Measure for Individuals with Cerebral Palsy

4.2.1 Identification of Areas of Interest

Cerebral palsy (CP) is one of the most common childhood physical disabilities, and as a lifelong condition, with no known cure, a focus on research in prevention, treatment, and effective interventions for optimising QoL and future potential is needed (Blair & Watson, 2006). Outcome measures and areas of interest for CP interventions have been evaluated previously (Oeffinger et al., 2008), however the evidence base for a more systematic approach is limited (Vargus-Adams & Martin, 2009; 2010; Tantilipikorn et al., 2012).

Prior identification of areas of interest and importance for CP research is important for future research. This can be particularly important with regards to the limited access to funding and high levels of competition in CP research. McIntyre, Novak, and Cusick (2010) conducted a three-part Delphi survey of different groups with vested interests in CP, including individuals with CP and their families (referred to as 'consumers'), CP intervention researchers and clinicians, and researchers of causes and prevention of CP. Using a Delphi survey, over three rounds, they aimed to identify research questions from each of the cohorts, and determine if a consensus could be reached between the groups, and subsequently which to prioritise for future research in CP.

In round one, the participants (50 consumers; 180 intervention researchers/clinicians; and 80 cause and prevention researchers) identified the problem areas related to CP which may be addressed by research. The returned responses allowed the investigators to identify the research problems for each group and these problems were reviewed and developed into research questions for the next rounds. Rounds two and three asked participants to give each question a priority rating on a 7-point Likert scale (1=very low to 7=very high), and once the questions were rated, the high priority consensus questions identified for each group were ranked and categorised into themes.

McIntyre and colleagues (2010) identified a large, and wide-ranging number of areas of interest, indicating possible directions of future research for individuals with CP. The results from their study indicate a desire to improve QoL and identify what the optimal treatments are for individuals with CP, and theoretically the latter will influence the former. Furthermore, the results suggest individuals with CP and their families are focussed on similar areas of interest. They are looking to prevent deterioration, co-morbidities, and are interested in the various ways in which the QoL of individuals with CP can be improved, be it

through intervention and/or community participation, or additional and higher quality service provision.

The inclusion of three cohorts led to the identification of a wide range of research areas and questions, and the results show that despite the large number of research questions raised, there can be agreement on the areas and questions that should be made a priority across the cohorts. As this was one of the first studies of its type, the use of the Delphi technique is a positive as it allows for the gathering of views of many individuals from different groups when trying to reach a consensus. The same method will be adopted for the current study. Previous literature has stated that a sample size of 30 participants per group should be seen as sufficient for a group consensus in a Delphi survey (e.g., Delbeq et al., 1975), although McIntyre et al. (2010) had a sample size of less than 30 in the consumer group in rounds 1, 2, and 3 (n=20), and in the cause and prevention researcher group in rounds 2 and 3 (n=19; n=18 respectively). However, a strength was the high response rate for the consumer group for round 2 and 3 (100% and 95% respectively) showing the high level of involvement from that group.

The results from McIntyre et al. (2010) have highlighted the areas of high priority for future research in CP. The effectiveness of interventions and outcomes was a minor theme to be identified, alongside identifying optimal treatments for CP. Given, the breadth of topics and research areas identified by the participants, it highlights there can be many directions for future research. Therefore, ensuring the efficacy of interventions, and measuring the most important domains using the most appropriate outcome measures is key to quality research. However, none of the participants' high-priority topics referred directly to physical activity, and with the benefits of physical activity for individuals with CP well documented, along with the reported influence on independence, and QoL, it would appear physical activity research was not a prominent thought. On this basis, the current study employed a similar methodology by using a Delphi approach to ascertain the most important domains to consider for evaluating physical activity interventions for children with CP, to provide some insight for future research.

4.2.2 Identification of Important Domains and Outcome Measures

Identifying the important domains and the most appropriate outcome measures for an intervention is important for the future of CP research and in turn, trying to positively impact on the QoL of individuals with CP. The heterogeneity of CP, along with the variability of change and development of CP, and lack of understanding of the functional and clinical changes in children with CP, can create difficulty when evaluating interventions and/or treatment in children and youth with CP. The change in a child with CP may be down to

inevitable growth and development, but it could also be a product of an intervention, or a response to treatment. Therefore, to help understand and address the change and development in children and youth with CP, measurement across domains is necessary (Vargus-Adams & Martin, 2009). Establishing the domains which are viewed as most important by individuals with CP and their families, along with the opinions of medical professionals, will help in the design of interventions by focusing on those key domains. Furthermore, to ensure the domains are evaluated appropriately for the CP populations, it is also important to establish the best outcome measures for these domains, potentially develop a core set of outcome measures to allow comparison between and within populations (Khan & Pallet, 2007).

Similar to the current study, but with a focus on evaluating effects of therapeutic programmes, Vargus-Adams and Martin (2009) conducted a Delphi study to establish the domains of importance, and the best outcome measures for evaluating those domains. Also, in line with McIntyre et al. (2010), the participants for the study of Vargus-Adams & Martin (2009) included youth with CP and their families, and medical professionals, to create as diverse a group as possible.

The survey consisted of five rounds overall, with the first three rounds establishing the domains of importance and rounds four and five establishing the best outcome measures. All three cohorts (youth, parents, and medical professionals) completed rounds one, two, and three, but only medical professionals completed rounds four and five. Survey one asked the participants the open-ended question, "What are the things you find most important to consider when you evaluate the effects of an intervention for yourself/your child/your patient with cerebral palsy?" (pp. 2090; Vargas-Adams, & Martin 2009), and they were asked to provide as many answers as they wanted. The responses were analysed using the ICF-CY coding and aggregated into domains by grouping responses of similar focus. The six domains that resulted from round one of the delphi survey were impairment, general health, gross motor skills, self-care and fine motor skills, speech and communication, integration and participation, caregiver issues, and QoL. These domains indicate the far-reaching impact of CP, and the wide range of areas of concern within the CP population, similar to McIntyre et al. (2010). However, none of the domains identified are unexpected because they are all common areas to CP research and a positive influence within these domains would potentially improve the overall QoL. Given these results, it is important that the current study focuses on identifying the important domains of physical activity interventions which will provide a more focused set of results relating to physical activity interventions for children with CP for future research.

Round two asked the participants to rank the listed domains in order of importance (medical professionals were asked to complete it twice, once thinking of ambulant children

with CP, and secondly for non-ambulant children with CP), and round three asked the participants to distribute 100 points between the eight domains. The results did not show a large difference in ranking between the domains, but QoL was ranked highest, and caregiver issues was ranked as least important. The results from these two rounds highlight how difficult it can be to identify what is most important in a general sense because each individual with CP is different, and their priorities will change throughout development, and so choosing a domain which is 'most important' is viewed as difficult, which can be a limitation of employing the Delphi technique.

The wide-reaching list of domains identified as important in round one is all well and good, but if a less effective outcome measure is used, or the most appropriate option is not administered, then it can undermine the reliability of the study. The following two rounds asked medical professionals to identify the most appropriate outcome measures for each domain identified previously with the aim to create a 'core set' of outcome measures for future research. There were many outcome measures identified (103), with each domain having between 10 and 27 measures, which were each ranked. The mean rankings highlighted a few that the medical professionals identified to be highly preferable (e.g., gross motor function measure; GMFM), but the majority had a low mean ranking. Though these results are not directly influencing the current study, the lack of agreement on which outcome measures are most appropriate for evaluating interventions for children and youth with CP, similar to the limited agreement on the domains, highlights the wide range of measures that could potentially be used. Therefore, there is a potential difficulty going forward to have a core set of measures for evaluating change in CP, and then comparing or generalising results across populations and/or interventions. The participants again indicated that the choice of outcome measures may be specific to an individual and their goals as the priorities can and will change throughout development. Such a range of domains and lack of consensus on how to best measure them highlights the difficulty in evaluating change and development from interventions in research with CP, and the uniqueness of researching CP populations. Therefore, it is important to carry out a Delphi study to gain insight into the best choice of outcome measures in the context of the physical activity intervention in this thesis, rather than assuming previous findings for each population.

Tantilipikorn, Watter, and Prasertsukdee (2012) and Vargus-Adams and Martin (2010), looked at the interventions, domains of interest, and outcome measures for children with CP, using the ICF-CY framework to try and further clarify the data. As previously mentioned, the ICF allows for the description of an individual's health condition under the following domains: body function and structure, activities and participation, environmental factors, and personal factors. Tantilipikorn et al. (2012) studied assessment measures and interventions reported for children with CP in central Thailand and referenced the ICF-CY to

ensure the necessary areas and domains measured. They found the outcome measures administered and interventions carried out could be classified across each of the components of the ICF-CY, but there was more bias towards body functions and structures, and less focus on activities and participation. The covering of all aspects of the ICF-CY for interventions and outcome measures highlights that measuring change and development in CP can be difficult, and the view of which are the most important domains or best measures for those domains is subjective, and this is in line with the findings of McIntyre et al. (2010) and Vargus-Adams & Martin (2009). Furthermore, the use of the ICF-CY allows for cross country and culture comparison which can be beneficial for addressing gaps and encouraging best practice, thus enhancing the QoL of children and youth with CP, therefore, the current study will employ the ICF-CY to help clarify the data.

Vargus-Adams and Martin (2010), using the data from Vargus-Adams and Martin's (2009) first survey round, analysed the responses to the open-ended question to youth with CP and their parents, and medical professionals, using the ICF-CY to assess the domains of importance. The responses were distributed across the domains of the ICF-CY, as well as some concepts that are not covered by the ICF-CY. Across the groups of participants, a focus was placed onto body functions and structures, however, minor differences between groups represented their views. For example, youth with CP showed more of a focus on specific motor skills such as hand/arm use, while parents indicated more of a focus towards motor function and communication. This highlights the difficulty in selecting best practice for CP measurement because choices will have to be made within families, and between researchers and clinicians about which areas to focus on for interventions. QoL was the most common non-ICF-CY domain reported as an important domain to evaluate in interventions, further highlighting the importance put on QoL by individuals with or affected by CP. Despite the lack of agreement between cohorts in establishing the most important domains, and the best measures to assess them, the importance of improving QoL is consistent across studies, whether it is being measured directly or as a means to an end, QoL is an important domain to assess in intervention effectiveness.

The research discussed in this section shows consistent messages and helps to set up the current study. The aim was to establish the domains parents of children with CP, health care professionals, and frame running coaches viewed as most important to measure when evaluating physical activity interventions for children with CP. Frame running coaches were included in the current study as their point of view is relevant to the physical activity intervention studied in this thesis, and one that has not been studied before in this context. The current study did not include children with CP as in previous research (e.g., Vargus-Adams & Martin, 2009), as the frame running coaches were a third cohort which was a lot of potential participants. The current study comprised of sequential online surveys, and is

adapted from previous research (e.g., Vargus-Adams & Martin, 2009; McIntyre et al., 2010). Despite the similarity to previous research, the current study puts focus on physical activity interventions, where other studies have focused on therapeutic programmes (Vargus-Adams & Martin, 2009) or outcome measures (Tantilipikorn, et al., 2012), or identifying research questions for future studies (McIntyre et al., 2010). Furthermore, the importance of physical activity for children with CP is well documented, and as their opportunities to participate may be further limited in comparison to children without CP, identifying factors which are important to measure may encourage future engagement in physical activity, and add to physical activity literature for individuals with CP.

4.3 Methodology

4.3.1 Study Design

The study used a two-stage Delphi technique. The Delphi approach was originally defined by Linstone and Turoff (1975) as “a method for structuring a group communication process, so that the process is effective in allowing a group of individuals, as a whole, to deal with the complex problem” (p3). A main purpose of the Delphi technique is for researchers to form a consensus or explore a field beyond the existing knowledge (Iqbal & Pison-Young, 2009). With the aim of the current study to provide data in an area of research where there is limited knowledge, the Delphi technique is selected as a suitable technique for the study.

There are numerous strengths associated with the Delphi technique (see section 3.5.4.3), and there are situations identified as being particularly suited to using the Delphi technique, which are also a feature of this study as characterised by the four methodological features of the Delphi technique highlighted in a systematic review by Jünger and colleagues (2017). For example, the relevant participants/specialists/experts from diverse backgrounds regardless of their geographical location, known as ‘panellists’ are questioned on the issue of interest; the delphi process remains anonymous which allows for every participant’s voice to be heard, avoiding conformity and social pressures from positions of power/authority; the procedure consists of numerous rounds; and the iterative nature of the process allows for the design of subsequent rounds to be informed from the results of the previous round. These conditions justify the use of a Delphi technique for the study - where participants are being recruited from throughout the United Kingdom, from a range of professions and with different levels of authority, so a face-to-face exchange would not be feasible to orchestrate and may have suppressed opinions of those with less authority (e.g., those without a medical background).

The current study consists of two rounds. The purpose of round one is to identify which factors the participants feel are most important to measure when evaluating the

effectiveness of physical activity interventions for children with CP, and this was done through the collecting of qualitative comments and responses. The purpose of the second round was for participants to rate the factors identified from round one, and how important they felt each factor was (out of 5) to provide an insight to which factors participants viewed as most important.

4.3.2 Ethics

Ethical approval for the current study was sought and obtained from The Moray House School of Education Ethics Committee of the University of Edinburgh. The information sheet explained research, and highlighted it was voluntary and participants would be anonymous throughout the study. The online questionnaire was set up, so participation was only possible if the participants actively consented to taking part (see Appendix A).

4.4 Delphi Study Round 1 – Online Survey: Identification of Important Factors for Measuring the Effectiveness of Physical Activity Interventions

4.4.1 Recruitment

Initial contact with potential participants was made at a Frame running training session with a small number of parents, physiotherapists, and frame running coaches. They were made aware of the research, and more specifically the current study, and these potential participants acted as gate keepers to a wider group of participants. Following distribution of the online survey link via email, further participants were recruited via the parents, health care professionals, and frame running coaches, by passing on the link to other individuals who they thought would be willing and able to complete the survey (i.e., snowball sampling). The second phase of recruitment included contact with other frame running clubs around the United Kingdom, and with CP focused charities (e.g., CPISRA, and The Bobath Centre), where information about the survey and the link were distributed via email and on social media (e.g., Twitter). The inclusion criteria were parents, family members and/or carers who had experience in caring for children with CP, health care professionals who had experience working with children with CP, and individuals who were involved in frame running (i.e., a coach or volunteer). A sample size of at least 30 participants was considered viable because it has been regarded as sufficient for contribution of ideas towards a group consensus (Delbecq et al., 1975). Furthermore, to have a broad representation, the aim was to have representation from all aforementioned groups (parents, health care professionals, coaches/volunteers).

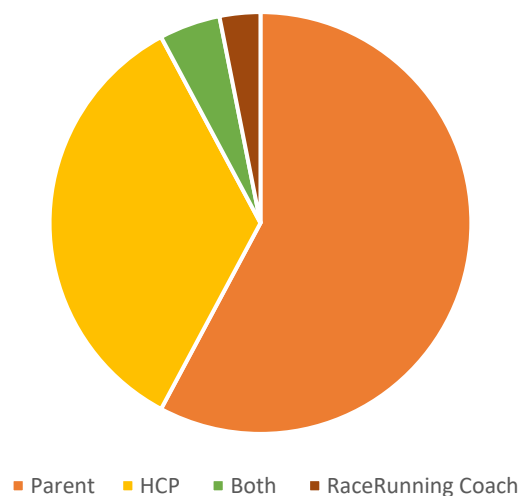
It is important to note that the selection of frame running coaches as participants over children as in previous research, such as Vargus-Adams and Martin (2009), was to add a

unique viewpoint from individuals involved in frame running as a relatively new sport. Furthermore, with communication and cognitive deficits associated with CP, children may require assistance in completing the survey and power relations may influence the responses provided by children not giving a true representation. Additionally, there was a high chance of overlap with parents and child participants, given the nature of recruitment, which may have influenced the responses given by the children thus not providing a true representation of the children's viewpoint.

4.4.2 Participants

There were 65 responses in total (one of which had to be excluded due to missing data), and the breakdown of the final 64 participants showed 57.81% of responses came from parents of children with CP (n=37), 34.37% came from health care professionals (HCP; n=22), 4.69% were from participants who were both a parent and a health care professional (n=3), and 3.13% of responses were from frame running coaches (n=2; see Figure 4.1). Furthermore, 68.18% of the health care professionals were physiotherapists (n=15), with other health care professions including medical doctors, paediatricians, and speech and language therapists. It was important to include participants from different populations because it allowed the perspectives from each population to be considered which can help to provide a more comprehensive view of which factors are most important when measuring the effectiveness of a physical activity intervention for children and youth with CP.

Figure 4.1 Distribution of Participants for Round 1 of the Delphi Study



Note. Both (parent & health care professional); HCP (Health Care Professional)

4.4.3 Procedure

The participants were asked to complete the online survey via the link that had been sent to their email address. For the first round of the Delphi study, the participants were asked, “Which factors do you consider to be the most important for evaluating the effectiveness of a physical activity intervention for children with cerebral palsy (CP) (e.g., physical, psychological, and/or social emotional)?”; this question was adapted from Vargus-Adams and Martin (2009).

4.4.4 Analysis

Responses from the first round of the survey were coded according to the domains of the ICF-CY, body functions, body structures, activities and participation, and environmental factors, with any factors out with these domains grouped as miscellaneous. Additionally, the six steps of thematic analysis (see section 3.6.1; Braun & Clarke, 2006) of the qualitative data were carried out to establish the domains and factors viewed as most important across the different populations.

4.4.5 Delphi Study: Round 1 Results

Over 100 items were identified by the 64 participants from the first round of the survey, and these were distributed across the ICF-CY domains. The responses were attributed to the ICF-CY chapters and second level categories to create the overall domains. The coding of chapters to second-level categories can provide more specificity compared to chapter level only, and this can be used depending on the users’ needs. Therefore, second-level categories were used in the current study to improve specificity. The factors selected for the following round of the study were determined by the researcher to reflect the views expressed by the participants.

For the current study the main domains identified were physical (n=25; such as mobility, pain, fatigue, appetite, sleep, and Cardiovascular (CV) fitness), psychological (n=10; such as self-efficacy, self-esteem, self-concept, and psychological wellbeing), and social-emotional (n=11; such as participation, enjoyment, sense of freedom, social inclusion and interaction, building friendships, sense of achievement, and QoL).

Some participants only stated if they thought if one, two, or three of the domains were important (e.g., psychological only). However, other responses were more specific (e.g., improvement in trunk control), and additional detail was sometimes added providing an explanation of why they thought a domain and/or factor was important to measure. The following sections will highlight some of the main factors expressed by the participants for each domain.

4.4.5.1 Social-emotional Results

Overall, the factor that was reported the most was social inclusion and interaction (n=18), highlighting the importance the participants were putting on the opportunity to for their child/patient to be part of a group, whether it is with children with or without disabilities. This was followed by enjoyment (n=12) - highlighting the importance of making a physical activity intervention enjoyable because that can encourage engagement and adherence, and in turn lead to additional benefits associated with physical activity participation.

“That the child is engaged and interested in the activity, that the child enjoys and is willing to take part in the activity. That they are able to achieve some success doing the activity, if they cannot participate can the activity be adapted for the individual. That they are included with their peers both those with a disability and those without.”
– Participant 13 (Parent)

“Fun and enjoyment and the engagement with the activity, then the physical aspects”
– Participant 22 (HCP – Physiotherapist)

The following responses were provided by physiotherapists, suggesting they put a precedence on social-emotional factors, which may not have been expected due to their profession, but also suggests the importance of social inclusion and interaction they see for children with CP.

“social – many children are isolated so the opportunity for social interaction/building relationships/emotional support element is valued. Access can be a challenge, and a physical activity intervention can not be effective if it is not accessible at all.” – Participant 2 (HCP – Physiotherapist)

“Social-emotional, when a child feels good about themselves in a social environment, they want to achieve more on all levels.” – Participant 59 (HCP - Physiotherapist)

The following responses from parents and health care professionals further highlight the importance placed on social inclusion for children with CP across different cohorts. Furthermore, the issue surrounding a lack of suitable activities, and therefore restricted opportunity for children with CP to be physically active was mentioned.

“Wellbeing – the opportunity for a child to feel included in an activity with peers and to feel they can achieve something meaningful and/or give them confidence in their own ability” – Participant 6 (HCP – Physiotherapist).

“Our biggest struggle is finding activities where there are others with a similar level and type of disability.” – Participant 21 (Parent)

“Social-emotional. To be part of an activity that my child wants to do with his peers.” – Participant 45 (Parent)

“social-emotional factors are most important as I think there are so few social activities that are accessible to disabled children where they can interact with their peers outside of an educational setting. I think this in turn can lead to them becoming isolated and withdrawn.” – Participant 61 (Parent)

The responses show how important the participants perceive peer interaction and social inclusion to be for children with CP, but also suggest the importance of participation in a group of individuals of similar ability or disability.

4.4.5.2 Psychological Results

With reference to the psychological factors, psychological wellbeing (n=8) was reported most frequently as the most important psychological factor, and this was followed by self-efficacy (n=5) and self-esteem (n=3) as important psychological factors to measure.

“Psychological: Increase in self-esteem and self-belief. Positive attitude and willingness to attempt new things” – Participant 7 (Frame running Coach)

“Psychological – in my experience with the young lady I work with, if she is unable to participate in sport alongside her peers, it has a detrimental effect on her self-esteem. Inclusion and equality are everything to her. She does not want to stand out or be different, but just to have an equal opportunity to participate and enjoy sport.” – Participant 38 (HCP)

4.4.5.3 Physical Results

With reference to physical factors, the responses were of a greater variety, and in some cases more specific, compared to both psychological factors and social-emotional factors. Cardiovascular (CV) fitness was reported as the most important (n=8) physical factor to measure, and this was followed by strength (n=4) and mobility (n=3) of children and young people with CP. The specificity of some of the responses highlights the broad view on the range of factors which can be measured from a physical activity intervention for children with CP, and this could be attributed to the heterogeneity of CP and the individual priorities of the individuals with CP.

“Physical – I want to see my son improving his overall fitness levels as he is otherwise quite sedentary. I would like for an activity to help him loosen off his tight

leg muscles in a similar way that the physio does. It is also important to me that a physical activity would give him a sense of achievement and success” – Participant 4 (Parent)

“Firstly physical, and in addition social inclusion, and participation in activities within peer groups. My daughter thrives in taking part in group activities and I am always aware of the importance of weight-bearing, muscle strengthening, and correct positioning for her postural care. Physical intervention enables me to do these in an enjoyable format for my daughter.” – Participant 41 (Parent)

“Physical, passive/active ROM (range of motion), strength, function, participation in activities and subsequent effect on mood.” – Participant 52 (HCP)

Although participants 4, 41, and 52, appear to be prioritising physical factors, their responses suggest they are also aware, and place an importance on the psychological factors (e.g., sense of achievement), and social-emotional factors (e.g., social inclusion) associated with physical activity participation.

As mentioned, there were many specific physical factors included in the responses including trunk and head control, posture, leg mobility, limb control, leg and pelvic strength, heart and lung function, and gait patterns, which are highly attributable to CP, but it shows there is a lack of consensus across the physical domains compared to the psychological and social-emotional domains.

4.4.5.4 Physical, Psychological & Social-Emotional Domains

There were responses which stated physical, social-emotional, and psychological outcomes are all important to measure. Some of these participants referred to the importance of physical activity for children and young people with CP, and the impact on numerous aspects of development, suggesting all should be measured, for example:

“Physical. A child with CP needs early intervention with physical activity. Physical activity leads to better health, able to cope with colds, viruses etc. without the possible need of hospitalisation. This leads to better psychological and emotional behaviour. Also, it leads to better posture and mobility which in turn leads to late or not at all surgical intervention. Physical activity is the most valuable therapy for children and young people with CP.” – Participant 20 (Parent)

“Physical activity intervention impacts every aspect of a child’s development; physical, psychological, social, emotional and also greatly improves the quality of a child’s social experience – neural pathways are created and strengthened through repetitive movement. Without the intervention a child would never be able to reach their full potential.” – Participant 27 (Parent)

Both participant 20 and 27 highlight how important they view physical activity and the impact they feel it can have on all aspects of a child’s development. Furthermore, participant 45 was

the only participant to suggest that being physically active and participating in an intervention is not always positive, and there can also be a negative impact from it:

“I feel this question is too broad as CP is the cause and not the outcome. Each child is different and what can be positive for one is a negative for another. In my daughters’ case, physical activity has been both.” – Participant 45 (Parent)

The comprehensiveness of responses varied between participants and population but their perception of what is most important to measure appeared to be similar because of the knowledge gained from different experiences with CP. For example, the following parent and health care professional both see all factors as important to measure:

“I actually think all factors are important, as doing as much activity as your disability will allow you is really important for your physical and emotional wellbeing, and also a social activity. Great to try new things as well.” – Participant 15 (Parent)

“Equal because they all effect each other. An individual with cerebral palsy has neurological damage which directly effects not just the physical domain (e.g., physical abilities), but effects directly as well as indirectly their cognition, behaviour, emotional regulation etc. It is important that all factors are looked at. Physical activity improves self-esteem, mental health, independence, musculoskeletal and general health.” – Participant 50 (HCP)

The view of participants encompassing the importance of all domains and factors indicates that it may be difficult to pinpoint what is most important, but also that there is an extremely broad range of outcome measures that can be measured in physical activity interventions for children and youth with CP. This is highlighted by the distribution across the ICF-CY (see Table 4.1), which also shows the responses provided were detailed. However, this can also be problematic for the design of future physical activity interventions as the focus of the interventions will vary between individuals with CP as they develop, and their goals and priorities also change.

“All of them...it depends on the individual which factor(s) will be most important at the time (and may also depend on the actual goals set/worked for).” – Participant 12 (HCP – Physiotherapist)

“This will vary depending on child’s current holistic assessment encompassing ICF framework – which will inform goal-setting and lead to identification of what domain the physical activity is currently aimed at.” – Participant 55 (HCP – Physiotherapist)

The extensive list of responses reflects the impact that physical activity can have on children and youth with CP (see Table 4.1), and shows that choosing the most important area, domain, and/or factor is not a simple task. In the design of the second round of the

survey, all of the responses from the first round were taken into consideration, with a combination of analysis of the factors across the ICF-CY framework and outside the framework. Based on the visual and qualitative analysis from the first round of the survey, the domains and factors were selected for the second round of the survey.

Table 4.1 Top 10 Factors participants perceive as most important to measure when evaluating the effectiveness of a physical activity intervention for children with CP, results from Survey round 1, and the ICF-CY coding

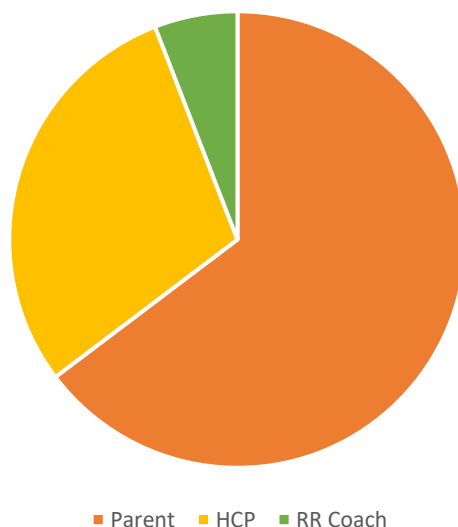
Participant Response	ICF-CY Distribution		Number of Responses
	Distribution	Chapter	
Social Inclusion & Interaction	Environmental Factors	3	18
Physical	NA	-	16
Enjoyment	NA	-	15
Social-Emotional	NA	-	14
Cardiovascular Fitness	Body Functions	4	8
Psychological Wellbeing	Body Functions	1	8
Access to suitable physical activity	Activities & Participation	9	7
Participation	Activities & Participation	9	6
Psychological	NA	-	6
Self-efficacy	Body Functions	1	5

4.5 Delphi Study Round 2: Online Survey – Rankings of Important Factors for Effectiveness of Physical Activity Interventions

4.5.1 Participants

From the 64 participants who took part in Round 1, there was a 53.13% response rate (n=34) for the second round of this study. The participants of the second round of the survey included 64.71% parents (n=22), 29.41% health care professionals (n=10), and 5.98% frame running coaches (n=2; see figure 4.2). Furthermore, 60% of the health care professionals responded were physiotherapists (n=6), 20% were medical doctors (n=2), and 20% were speech and language therapists (n=2).

Figure 4.2 Distribution of participants for round 2 of the Delphi Study



Note. Both (parent & health care professional); HCP (Health Care Professional); RR Coach (Frame running Coach)

4.5.2 Procedure

The design of Delphi round 2 was through a summary of qualitative comments and the inclusion of newly generated items, which has been identified as plausible strategy to design subsequent Delphi rounds in previous research (e.g., Bradley & Brasel, 2009; Jünger et al., 2017). Similar to previous research (e.g., Vargus-Adams & Martin, 2010), round two of the survey grouped the factors identified by participants in round 1 under either the psychological, social-emotional, or physical domain. Participants were asked to rate on a 5-point Likert scale (1 = 'not at all important' to 5 = 'very important'), how important they felt each factor was when evaluating the effectiveness of a physical activity intervention for children and young people with CP. Following the rating of factors, participants were given the opportunity to add any comments, in the free text box provided, about why they perceived the listed factors to be, or not to be important to measure.

4.5.3 Data Analysis

The individual responses to the Likert scale ratings for each factor were reported, and the descriptive statistics for each factor were calculated (mean and median rankings; see tables 4.2 to 4.5). Furthermore, non-parametric analysis, Wilcoxon signed rank test, was carried out between the domains (i.e., psychological, physical, and social-emotional) to identify any statistically significant difference in reported importance ($p < .05$). The qualitative data was

analysed through the 6-phases of thematic analysis (Braun & Clark, 2006), as well as applying the use of the ICF-CY framework, the same method as was used in round one.

4.5.4 Delphi Study Round 2 Results

All participants, apart from three, rated each of factors. The mean and median rating for each factor can be seen in the following sections, alongside stacked bar charts displaying the percentage of participants that rated each factor from 1 (not at all important) to 5 (very important). On visual inspection of analysis, the mean ratings for all 17 factors did not show any obvious difference between each individual factor, ranging from 3.06 for 'Appetite' to 4.85 for 'Enjoyment'. Furthermore, the median ratings showed no obvious difference from the mean ratings with 12 of the domains having a median rating of 5. Alongside the participants' ratings of importance for each factor, a number of participants expressed why they thought factors were important to measure in their opinion, and if there were other factors they thought should be measured, and these qualitative responses were analysed using thematic analysis.

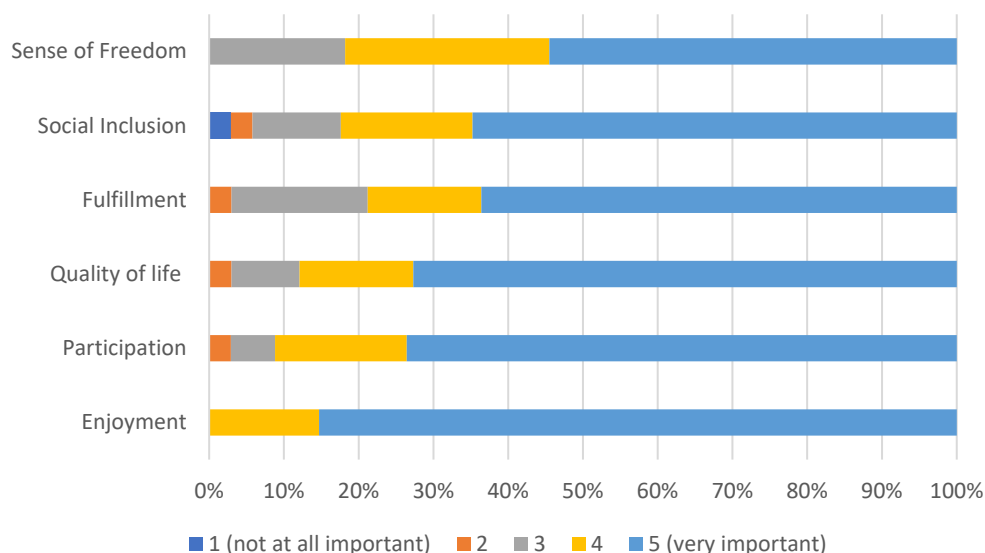
4.5.4.1 Social-emotional Factors

The mean ratings of the social-emotional factors are all reasonably high (see Table 4.2), with all six factors on average rated above 4. The highest rated social-emotion factor reported was 'enjoyment' ($m = 4.85 \pm .36$) and the lowest social-emotional factor reported was 'sense of freedom' ($m = 4.36 \pm .78$), although this was still considered high on the 5-point scale. The median rating for all social-emotional factors was 5 highlighting the importance that the participants put on each of the listed factors. Figure 4.3 displays the percentage of participants that rated each of the social-emotional factors from 1 (not at all important) to 5 (very important). Enjoyment was the highest rated factor with 85.3% of participants rating it very important, and the remaining 14.7% rating enjoyment as 4. 'Sense of freedom' was the lowest ranked with 54.5% of responses '5', however, it is the only social-emotional factor (apart from 'enjoyment') with no ranking below a 3, with 27.3% of responses '4' and 18.2% of responses '3', which suggests there is still a consensus across the participants at how important measuring 'sense of freedom' is.

Table 4.2 Mean, standard deviation (SD), and median ratings for the social-emotional factors

	Mean	SD	Median
Enjoyment	4.85	.36	5
Participation	4.62	.74	5
QoL	4.58	.79	5
Fulfillment	4.39	.90	5
Social Inclusion	4.38	1.02	5
Sense of Freedom	4.36	.78	5

Figure 4.3 Distribution of participants for round 2 of the Delphi Study



From thematic analysis of the qualitative data, one of the prominent themes is social inclusion, which is in line with the responses from the first round of the survey, despite having the fifth highest mean ($m = 4.38 \pm 1.02$). One participant indicated that because children with CP will always be physically limited the focus for physical activity interventions should be elsewhere:

“Disabled children find it hard to socialise due to a lack of suitable venues/groups/activities, leading to low mood and depression, and physical activity is known to lift mood...Physical effects will always be limited by the disability but there are no limits to psychological well-being. Disabled children are often isolated and if that can be alleviated via sport etc. then this is far more important than physical improvements being shown in my opinion.” – Participant 22 (Parent)

For some parents, the physical aspects may not be at the forefront of their minds because they place more focus on their child having a good experience and being given opportunities that children without disabilities have access to. For example:

“Having fun makes activities easier and more enjoyable and more likely the child will continue. Children love a sense of freedom, belonging and friendship, all this in turn gives fulfilment and a good QoL. It is like a circle, constant and all necessary to keep going.” – Participant 11 (Parent)

Having a suitable and accessible physical activity for children with CP may be important for their physical health and development but is also very important for their mental health and development, as children with CP (and other disabilities) want to have the same opportunities for participation as those without disabilities and being excluded for any reason can have detrimental effects of many aspects of their life and development.

“...main goal for me is for my son to feel like he is achieving and capable. It is also very important for him to be happy. Kids with disabilities often feel excluded. Any activity they participate in should be aimed at alleviating this.” – Participant 4 (Parent)

“...as a parent of a child with CP it breaks my heart when she cannot participate in activities and sports as the other children can. She wants too, but as she gets older, she does not want to hold the others back. Participating and achieving is a natural human activity. We strive to be part of our peer groups. When you are different (have a disability) this affects us massively until you find your own self-worth, but the journey is fraught, the physical disability creates the psychological and social-emotional stress.” – Participant 33 (Parent)

Overall, the qualitative data indicates the participants focus on the importance of having children with CP involved in physical activity, whether it be with children with or without disabilities, and the physical activity being enjoyable helps with engagement and adherence.

4.5.4.2 Psychological Factors

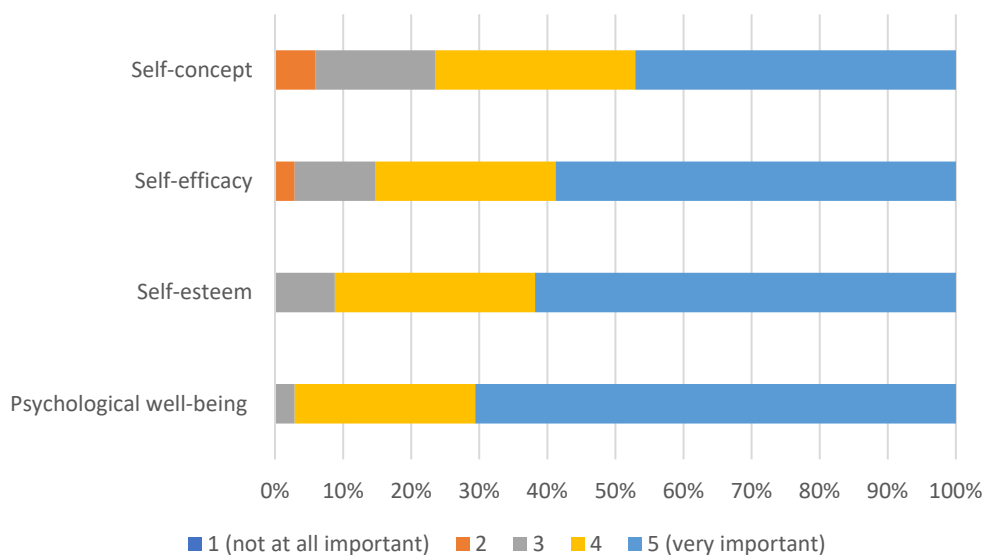
The mean ratings of the psychological factors are all above 4, similar to the social-emotional factors, indicating the participants viewed all four of the factors to be of high importance when measuring the effectiveness of physical activity interventions for children and youth with CP (see Table 4.3). The highest rated psychological factor was psychological wellbeing ($m = 4.62 \pm .61$), and the lowest rated was self-concept ($m = 4.12 \pm .95$). The small difference in mean rating between the factors is highlighted in the stacked bar chart (see Figure 4.4), with 70.6% of participants rating psychological wellbeing as a very important psychological factor to measure, compared to less than half of participants (47.1%) rating self-concept as a very important factor. This is highlighted by the median rankings for the

factors, with self-concept being the only psychological factor with a median factor of 4, compared to the others each having a median ranking of 5. Furthermore, more than 75% of participants rated all of the psychological factors either a 4 or a 5 level of importance.

Table 4.3 Mean, standard deviation (SD), and median rating for the psychological factors.

	Mean	SD	Median
Psychological Wellbeing	4.62	.61	5
Self-esteem	4.47	.71	5
Self-efficacy	4.41	.82	5
Self-concept	4.12	.95	4

Figure 4.4 Stacked bar chart indicating the perceived importance of psychological factors



Analysis of the qualitative data on the psychological domain indicated that a number of participants felt all psychological factors were important to measure, which may explain why the mean ratings were all within 0.5 of each other. As in previous sections, some participants would respond with a single statement, whilst others offered more detailed information, but in essence there was a consistency to the responses. For example:

“These are all recognised to be highly important in successful athletic performance. Clinical experiences show variable self-perception but often low, or no self-belief and/or unrealistic expectations.” – Participant 1 (HCP)

“These are fundamental to the general wellbeing of the child and their positive development.” – Participant 12 (Parent)

“All are important for the wellbeing of the child” – Participant 21 (Parent)

Self-esteem was one factor that was singled out by some participants as particularly important to measure. Previous research has suggested children with CP have lower self-esteem compared to their typically developing peers (Manuel et al., 2003). This was also suggested in the qualitative data:

“These children have low self-esteem to start” – Participant 17 (HCP – Physiotherapist)

“A child that is different feels very different. Psychologically it (physical activity) is very important for self-esteem.” – Participant 33 (Parent)

The participants recognised that psychological factors are important to measure, and this is clear in their mean ratings and qualitative data that suggest all are important. However, the potential difficulty in measuring the psychological factors successfully in comparison to the physical factors was also highlighted by the participants. This may be for a variety of reasons, for example, psychological wellbeing is not always ‘clear’ to see or always visible in comparison to physical attributes such as mobility or strength. Furthermore, psychological factors may be more susceptible to variables on the day of measurement, and the researcher is relying on the reporting of the participant and/or proxy report which may not be a true perception of the psychological aspect being measured.

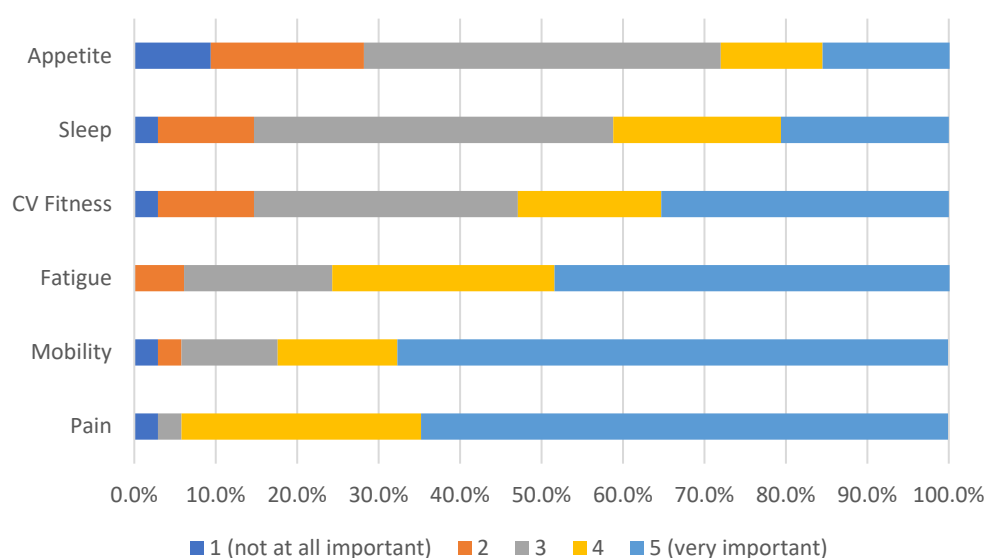
4.5.4.3 Physical Factors

The mean and median ratings for the physical factors showed the widest range across each set of factors (see Table 4.3). ‘Pain’ was rated the highest ($m = 4.53 \pm .83$), and ‘appetite’ was rated the lowest ($m = 3.06 \pm 1.16$). The distribution of ratings for physical factors was highlighted by the median scores ranging from 3 for ‘appetite’ and ‘sleep’, to 5 for ‘pain’ and ‘mobility’. Furthermore, the standard deviations of all factors apart from pain are .95 and above, underlining the wider distribution of the rankings for the physical factors. However, even though ‘pain’ had the highest mean ranking, ‘mobility’ had a higher percentage of participants rate it most important (67.6%; see Figure 4.5), compared to pain (64.7%). Having pain, mobility and fatigue rated the highest physical factors suggests the value the participants put on functional independence for children with CP, over the measurement of sleep, appetite, and cardiovascular fitness, which were only rated as very important factors by less than 40% of participants.

Table 4.4 Mean, standard deviation (SD), and median ranks for the physical factors.

	Mean	SD	Median
Pain	4.53	.83	5
Mobility	4.41	1.02	5
Fatigue	4.18	.95	4
CV Fitness	3.74	1.16	4
Sleep	3.41	1.02	3
Appetite	3.06	1.16	3

Figure 4.5 Stacked bar chart indicating the perceived importance of physical factors



The lower mean ratings of physical factors with a wider distribution across these factors show there was less consensus between participants about how important each of these physical factors are. This is emphasised by the qualitative data in which only 32% of participant responses suggested that all of the physical factors listed were important to measure. For example:

“They are all factors that may be relevant to performance, and potential indicators of the effects of exercise.” – Participant 1 (HCP)

“All have an effect on each other, overall ability and wellbeing of the child. Although they are all important holistically, the above list (the physical factors) can fluctuate in importance at specific times during the development.” – Participant 11 (Parent)

The above quote in particular addresses a wider point, in that physical factors can impact other areas of an individual’s life, but this can be different for each individual with CP given

the heterogeneity of the population of individuals with this condition. Furthermore, the priorities of individuals will change throughout their development and priorities of parents and/or health care professionals such as physiotherapists will also change and vary between individuals. Therefore, choosing an area to focus on for one intervention or physical activity interventions in general, whether it be physical, psychological, and/or social-emotional can then be difficult, as identified by the following responses.

“Physical abilities vary dramatically for a child with CP, so it is important to have a number of factors considered before embarking on any physical activity that could have a detrimental effect on a child’s wellbeing.” – Participant 12 (Parent)

“Physical effects will always be limited by their disability, but there are no limits to psychological wellbeing. Disabled children are often isolated, and if that can be alleviated via sport, then this is more important than the physical improvements shown.” – Participant 22 (Parent)

Additionally, participants highlighted the importance of measuring physical factors in reference to the impact they can have on the day-to-day activities and QoL of children and youth with CP:

“These (physical) aspects, they impact on QoL, and ability to take part in day-to-day activities.” – Participant 4 (Parent)

“Mobility can further lead to functional improvements in everyday life, and therefore improvements in mobility can be measured for effectiveness of a treatment. If pain and fatigue increase – compliance is low, and it usually has an impact on functionality and therefore can be detrimental to their day-to-day activities. This needs to be monitored as it will ascertain if the physical intervention has been effective or not” – Participant 31 (HCP)

The responses regarding the listed physical factors are similar to those of psychological and social-emotional factors in that each factor can be important but can be dependent on the priorities of the participants. Therefore, each factor may have an important role to play in evaluating the effectiveness of a physical activity intervention and in turn in the development of children and youth with CP.

4.5.4.4 Overall Domains

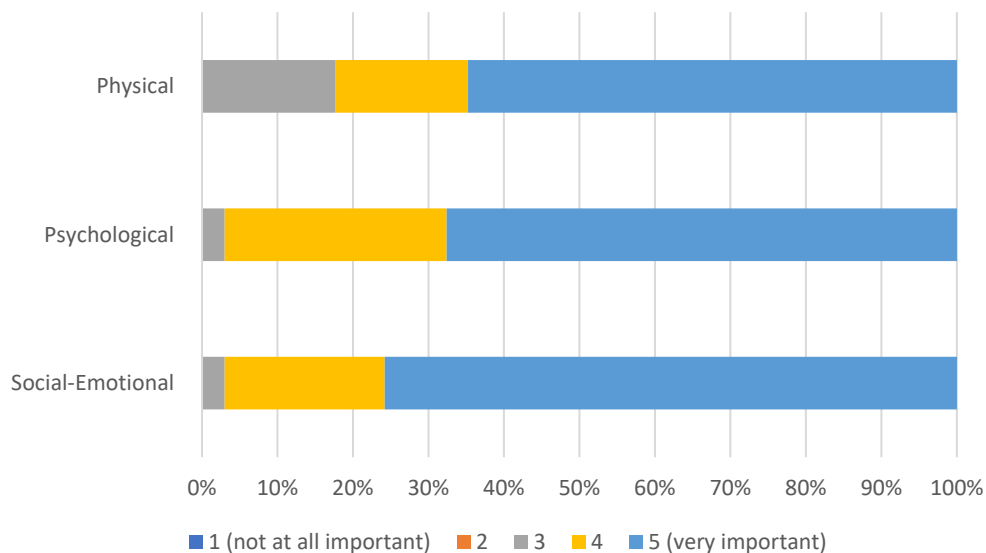
In addition to the individual factors, participants were asked to rate the importance of each domain overall. The mean and median ratings of the domains show the participants rated each highly with all median scores of 5 (see Table 4.5) and mean of social-emotional domain was highest ($m = 4.72 \pm .52$), followed by psychological ($m = 4.65 \pm .54$), and then physical ($m = 4.47 \pm .79$). These results are in line with the trend towards rating social-

emotional and psychological factors over physical factors, however the importance placed on all three domains is very evident with each domain having an importance rating of either 4 or 5 by over 80% of the participants (see Figure 4.6).

Table 4.5 Mean, standard deviation (SD), and median scores for the 3 domains

	Mean	SD	Median
Social-emotional	4.72	.52	5
Psychological	4.65	.54	5
Physical	4.47	.79	5

Figure 4.6 Stacked bar chart indicating the perceived importance of the 3 main domains.



A Wilcoxon signed ranks test indicated that there was no statistically significant preference between physical and psychological domains, $Z = 1.14$, $p = .253$, $r = .20$; no statistically significant preference between physical and social-emotional domains, $Z = 1.32$, $p = .187$, $r = .23$; and no statistically significant preference between psychological and social-emotional domains, $Z = 1.13$, $p = .26$, $r = .19$. Therefore, the differences in rating between the domains can be described as trends in the data. With the result of the Wilcoxon signed ranks tests showing no statistically significant difference between the three domains, this indicates the participants view that all three domains, physical, psychological, and social-emotional, are important to measure in physical activity interventions for children and youth with CP. This is further supported by the thematic analysis of the qualitative data

where overall wellbeing was identified as a main theme, with a trend towards the social-emotional and psychological factors over physical factors (as the descriptive statistics suggest). For example:

“Children with CP tend to thrive if they feel confident, valued, and part of a peer group. Their self-esteem is better, which in turn helps their general health and wellbeing.” – Participant 12 (Parent)

“Many physical barriers can be worked around, and the technology is making that easier. Feeling mentally healthy and socially engaged is more important to grow as a person and lead a healthy life.” – Participant 24 (Parent)

The parents and health care professionals have indicated the importance of measuring each of the domains and factors, however these can only be measured if there are suitable opportunities for children and youth with CP to participate, and this is echoed in participants' responses:

“Although I am a physiotherapist, kids with CP are often side lined and have difficulty making friends, so specialist physical activity intervention can be life enhancing.” – Participant 27 (HCP – Physiotherapist)

“There are so few sports suitable, that if you find one that provides enjoyment and a sense of belonging as well as providing physical benefits, it's gold dust.” – Participant 30 (Parent)

The responses highlight the importance of having an activity that is suitable, accessible, inclusive, and enjoyable because if the availability is not there, then it can be detrimental to the growth and development of the child in all aspects of their life. The next study in this thesis will determine whether frame running can be considered a viable physical activity for children and young people with CP which meets this remit.

4.7 Discussion

This study demonstrates the wide range of factors that parents, health care professionals, and frame running coaches view as important to measure for children with CP when evaluating the effectiveness of a physical activity intervention. It also indicates the numerous factors and areas of interest that need to be considered when designing physical activity interventions. The results from rounds one and two of this Delphi study are in line with previous research which investigated which factors, domains, outcome measures, and areas

of interest are most important in CP research (e.g., McIntyre et al. 2009; Vargus-Adams & Martin 2009, 2010; Tantilipkorn et al. 2011).

In round 1, the factors reported by the participants can be viewed as fairly comprehensive as they were distributed across the ICF-CY domains, body functions, body structures, activities and participation, and environmental factors (see table 4.1). The variety of responses show that a range of priorities is considered by families with children with CP and health care professionals, and these will vary between individuals with CP, and change across the stages of development and growth of the individual with CP. Interestingly, the factors in round 1 that were reported most frequently did not then have the highest mean ratings in the second round. For example, 'social interaction' was the most reported factor in round 1, however it ranked 5th out of the six social-emotional factors in round 2 ($m = 4.38 \pm 1.02$). This suggests that when presented with alternatives which participants may not have originally considered, and other factors have been brought to their attention via the Delphi approach, their opinion and/or priorities may have changed between rounds 1 and 2. Furthermore, with a drop of 46% of participants from round 1 to 2, this may have impacted the average rating of factors as the priorities and opinions of participants who did not respond may have been different from those who did. The following section aims to highlight the important factors as reported by the participants of the Delphi study and discuss their importance to children and young people with CP and the role they can play in promoting and engaging this population in physical activity.

4.7.1 Social-emotional Factors

The social-emotional domain was given the highest mean rating ($m = 4.72 \pm .52$), and three of the four top ranked factors were from the social-emotional domain, 'enjoyment', 'participation', and 'QoL' (with the other 'psychological wellbeing') which highlights the importance of this area to parents, health care professionals, and coaches.

4.7.1.1 Enjoyment

Enjoyment stood out as the most important factor, and with analysis of the subgroup ratings for parents and health care professionals, the parents' mean score for enjoyment at 4.96 was above the overall mean score of 4.72, and the health care professionals' mean score of 4.67 was slightly below the mean score. Regardless, this emphasises the importance of enjoyment as a key factor for physical activity interventions, which is in line with previous research which has shown the importance of enjoyment in relation to physical activity engagement and adherence (Majnemer et al., 2008; Declerk et al., 2016). DiLorenzo, Stucky-Ropp, Wal, and Gotham (1998) showed the importance of enjoyment to physical

activity participation, as enjoyment of physical activity was the only consistent predictor of physical activity participation across boys and girls aged 10 to 12 years out of a range of psychological and environmental factors.

Enjoyment can be viewed as an important facilitator to physical activity; thus, a fun and enjoyable activity can play an important role in encouraging engagement and adherence to physical activity. Furthermore, enjoyment can play an important role within theoretical frameworks when used to influence health behaviours, in this case, promoting physical activity participation. With reference to the Transtheoretical Model (TTM), enjoyment can be viewed as a facilitator to participation, thus encouraging children and young people to become physically active, or more consistent with their activity. For example, if a child with CP is physically active, but only exercises a little with the intention to be more consistent, they are deemed to be in the 'preparation' stage. As a stage-based model, which is thought to be more cyclical and not linear (Biddle et al., 2015), the child may be at risk of dropping their participation levels (i.e., encounter a barrier and return to the contemplation stage), but there is also the opportunity to promote participation and move into the 'active' stage. Enjoyment can have the ability to facilitate the progress and increase adherence in a physical activity, as finding an activity a child with CP perceives as enjoyable may help overcome other barriers faced previously (e.g., pain or fatigue), and increase motivation to participate. For example, a swimming intervention by Declerk et al., (2016) found high levels of enjoyment and adherence reported by the participants alongside improvement in walking and swimming skills with no adverse effect on pain or fatigue for children with CP (GMFCS levels I to III), and this is also reported by one participant *"...If something is not fun, is painful, tires you out, causes pain such that you can't sleep, or are unable to eat or drink properly, then it is a bad intervention."*

Furthermore, impacting levels of motivation is an important construct of the Self-determination theory (SDT), suggesting enjoyment can play a role in the SDT framework. For example, if an individual perceives the activity to be fun and enjoyable, they are more likely to be intrinsically motivated to participate and therefore more driven to overcome barriers to participate and less likely to be drop out. Majnemer, Shevell, Law, Birnbaum, Chilingaryan, Rosenbaum, and Poulin (2008) identified various determinants of enjoyment and participation in leisure activities for children with CP. The level of impairment and activity limitations were two factors that had little impact on enjoyment levels showing that irrespective of severity of CP, enjoyment is an important factor for engaging in leisure activities. These studies provide some support for why 'enjoyment' was the highest rated factor in the survey and is consistent with enjoyment being shown to have a strong correlation with physical activity participation (Sallis et al., 2000).

4.7.1.2 Participation

Participation had the second highest mean rating overall ($m = 4.62 \pm .74$) and is becoming recognised as a vital part of a child's development and wellbeing, and therefore the importance of participation as an outcome measure is growing (McIntyre et al, 2010; Schiariti, et al., 2014). Furthermore, as one of the main constructs of the ICF framework, the importance of measuring physical activity participation for children with CP is clear (Whiteneck & Dijkers, 2009). Anaby, Korner-Bitensky, Steven, et al. (2017) carried out a web-based survey across Canada to identify the focus of physiotherapists and occupational therapists' rehabilitation practices for children with CP. They found there was more focus on physical factors and impairments compared to the children's participation levels. Furthermore, the focus on participation was mainly about identifying problems, followed by assessments and interventions (e.g., gym participation), and there was very little focus on participation in community and leisure-based activities. In relation to the ICF framework, there is more focus on the 'body structures and functions' and 'activities' components than the 'participation' component (Anabey et al., 2017). The focus on these components over participation may be related to the difficulty in defining participation, and furthermore, the difficulty in measuring participation. The ICF framework's definition of participation is 'involvement in a life situation' (p4, WHO, 1980), which does not provide clarity to what specifically counts as participation or what a life situation is.

The lack of specificity in defining participation can therefore make it difficult to measure, and this is highlighted by the lack of a commonly accepted outcome measure. Furthermore, the measurement of participation can be subjective and rely on self-reporting followed by comparison with what are viewed as societal norms. Children and youth with CP may not be able to participate in the same number or variety of activities at the same frequency and/or intensity as their typically developing peers, and therefore comparing their participation levels to the participation of those without disabilities may be unhelpful (Brown et al., 2004). Moreover, this highlights the bigger issue for participation in children and youth with CP, in that there is a lack of suitable and accessible opportunities for this population to be physically active. Restricted participation, and a lack of appropriate opportunities for children and young people with CP was consistently mentioned by participants in the current study. The importance of having access to a suitable physical activity, that provides enjoyment, and a sense of belonging, along with the physical benefits was highlighted by reference to those activities as "*gold dust*" (participant 30).

The lack of suitable opportunities for children and adolescents with CP to participate, and be physically active, and the associated perceived detrimental impact on their physical, psychological, and social development, may be a contributing factor to the importance placed on participation in the current study. This is in line with previous research, with

Vargus-Adams and Martin (2009) also identifying participation as one of the main themes to measure for children with CP. However, similar to QoL, it is clear that participation is important, but when measuring participation as an outcome of an intervention, the construct will have to be clearly defined and the outcome measure carefully selected.

4.7.1.3 Quality of Life

'Quality of Life', which is not covered on the ICF-CY, was the fourth highest rated factor ($m = 4.58 \pm .79$); however, it was infrequently reported in round one. Similar to the current study, employing the Delphi approach, previous research by McIntyre and colleagues (2010) identified measuring QoL of children with CP as an important factor that can have a positive impact on improving lifestyle, and is highly recommended as a research priority.

Furthermore, some researchers have claimed that the majority of factors can be linked to, and impact on 'QoL', which has therefore been viewed as a 'holy grail' of intervention outcomes with emerging importance as the years have gone by (McDougall et al., 2010). Many of the other factors can be seen as impacting on QoL, thus improving one factor can in turn improve the overall QoL. This is also evident in the thematic analysis with participants simply saying all factors are important to measure because they can have "*big impact on quality of life*", or "*all aspects impact on QoL and ability to take part in day-to-day activities.*" Furthermore, as mentioned previously, impacting on any of the domains identified can be seen to eventually have an influence on the QoL, hence why QoL may be ranked most important to measure in previous research (e.g., Vargus-Adams, & Martin, 2009). Although rated as 'very important' by over 70% of the respondents in the current study, it only ranked 4th, and this may be related to the varying interpretations of QoL. It may be perceived as inclusion of all aspects of life overall, or have a narrower interpretation of psychological wellbeing, or somewhere in the middle.

The WHO defines QoL as "*an individual's perception of their position in life in the context of the culture and value systems in which they live, and in relation to their goals, expectations, standards, and concerns*" (WHO, 1995), highlighting that QoL is subjective and should be self-reported whenever possible. This is thought to be more difficult in children compared to adults, and even more difficult in children with disabilities, as they are sometimes viewed as unreliable research participants (Dickinson et al., 2007), thus selecting an outcome measure specifically based on an individual may be the best action, but not the most practical. One participant highlighted the difficulty in measuring the QoL despite the perceived importance:

“I think all of these (social-emotional) factors are important but difficult to measure and sometimes to define. For example, how are you intending to measure QoL? The isolation of children with CP is evident and it would be helpful to have illustration indicators employed in measuring levels and changes in these factors.” – Participant 1 (HCP)

Furthermore, as known by parents and health care professionals, at present, CP is a non-curable condition. Therefore, improving a child with CP's QoL, irrespective of how parents or health care professionals define the construct, will be a major focus for rehabilitation and therapeutic programmes, and future research, hence the high rating for QoL in the current study is not surprising. It is clear QoL is considered very important, yet there is awareness that it may mean different things to different people, and it might be hard to measure. Therefore, an intervention outcome measuring QoL will have to carefully define it as a construct, as well as justify the measure used (see chapter 5).

4.7.2 Psychological Factors

Psychological wellbeing ($m = 4.62 \pm .61$) had the highest rated mean score of the psychological factors, followed by self-esteem ($m = 4.47 \pm .71$), self-efficacy ($m = 4.41 \pm .82$), and self-concept ($m = 4.12 \pm .95$). The importance of measuring the psychological factors in the views of the participants was highlighted by the median scores of each factor being 5, apart from self-concept with a median score of 4. Previously, researchers trying to identify domains and areas of interest in CP research did not report the specific psychological factors identified by participants, as they were reported under the mental functions chapter of the ICF-CY (Vargus-Adams & Martin, 2010). Therefore, there is no comparison point between the results of the current study, and those of previous studies, which illustrates the large gap in investigating specific psychological factors related to physical activity interventions in young people with CP.

The benefits for an individual's psychological wellbeing from participation in physical activity are well documented for adults, but not as well documented for children (Parfitt, & Eston, 2005). However, if there are links between improving psychological wellbeing and being physically active for children, then encouraging participation should be a priority, particularly with the growing prevalence of mental health problems (e.g., anxiety, stress, and depression) in children. Given the importance of improving psychological wellbeing, and the potential benefits from being physically active, this can explain why participants rated psychological wellbeing as the most important psychological factor to measure. The impact of physical activity on psychological wellbeing, and the importance of it for children with CP is further discussed in chapter 5 (see section 5.1.4).

4.7.2.1 Self-esteem

Self-esteem was the second most reported psychological factor by participants in Round 1 (after psychological wellbeing), and this was compounded in Round 2 of the Delphi study with a high mean score of 4.47. Self-esteem refers to the extent to which one values themselves, and their sense of worth (Blasovich & Tomaka, 1991). Self-esteem is viewed as a vital part of an individual's physical and mental health and wellbeing (Fox, 2000). Furthermore, being physically active is thought to have a positive impact on an individual's self-esteem, highlighting why it is an important factor to measure.

In relation to CP, it is generally assumed that the self-esteem of children with CP is lower than those of children without disabilities, because of physical impairments and secondary health conditions related to CP. However, previous research has reported similar self-esteem scores between children with CP and without (Manuel et al., 2003; Riad et al., 2013), although this is not generalised across the CP population. Furthermore, it should be noted that physical activity participation can also have a negative impact on the self-esteem. The negative influence on self-esteem may be more relatable to the CP population due to the limited functionality associated with CP. For example, a child with CP participating alongside children without disabilities may try to compete at the same intensity but due to the limited functionality associated with CP, they are unable to. Therefore, those with CP may have reduced self-esteem when comparing themselves to their typically developing peers. A link between self-esteem and functionality in individuals with CP may be important when identifying individuals with low self-esteem. Previous research has shown levels of functionality can be a predictor of self-esteem for individuals with CP (e.g., low functional capacity can lead to lower self-esteem; Espín-Tello, et al., 2018). The varying impact that motor and cognitive impairments can have on functional capacity, can in turn negatively impact day to day activities including being physically active, thus having a negative impact on psychological factors including self-esteem (Horsman et al., 2010).

4.7.2.2 Self-efficacy

The importance of self-efficacy in physical activity is reflected in the high mean and median rating given by participants. Self-efficacy plays an important role in the encouragement, engagement, and adherence to physical activity participation in children with and without disabilities. Self-efficacy in the context of health behaviours, and particularly physical activity, can refer to an individual's perceived confidence to overcome barriers and become physically active, but also individuals can have varying levels of self-efficacy at different phases of physical activity (e.g., recovery or maintenance of specific activity; Schwarzer,

2008). This is highlighted by the role self-efficacy plays in the theoretical frameworks discussed in chapter 2, social cognitive theory (SCT) and transtheoretical model (TTM; see section 2.4.2 and 2.4.3). Within the transtheoretical model, self-efficacy is viewed as one of the more influential factors when predicting physical activity. Previous research in adults with disabilities showed that self-efficacy levels were a good predictor of physical activity when becoming physically active and making those first steps over the activity barriers. However, as time goes on and the TTM stages progress (i.e., maintenance of physical activity), the level of self-efficacy becomes less significant in predicting physical activity behaviours (i.e., self-efficacy may not be as important as time goes on for physical activity participation; Kosma et al., 2012), suggesting self-efficacy is most important early on in physical activity participation, and this is where most of the CP population is. As one of the most sedentary populations, and with physical activity participation opportunities restricted, initiating physical activity will be key for the physical and psychological development. Thus, the measurement of self-efficacy appears to be important for future research involving individuals with CP and physical activity participation, particularly at the early stages. For social cognitive theory, self-efficacy is viewed as pivotal because of the influence it may have on the other constructs within the model (goals and outcome expectations) and being physically active (Bandura, 2004). Perceived self-efficacy can impact the goals and the outcomes that are expected from their efforts (i.e., higher self-efficacy indicates that higher goals will be set, and more positive outcomes can be expected).

4.7.2.3 Self-Concept

Self-concept is a multidimensional construct and is recognised as a fundamental part of children's psychological functioning (Shields, et al., 2007). Although self-concept was generally regarded as important ($m = 4.12 \pm .95$), with 47.1% of participants rating it 5 (very important, it was the lowest rated psychological factor in this study. This may be due to the multidimensional nature of self-concept, and/or that it can sometimes be confused with self-esteem. It comprises of a child's perceived competence in various domains, for example, scholastic competence (how they view their ability to perform at school); social competence (how they view their ability to build and maintain friendships); and athletic competence (their ability to perform and do well in sports; Harter et al., 2012a). For children with CP, it is assumed that their global self-concept is lower compared to children without disabilities, and this may be down to motor impairments and associated conditions of CP. It can be very important to understand how children with CP view themselves, as the view they have, and the value they put on different areas of functioning can influence therapeutic and clinical decisions regarding next steps in managing their condition (e.g., if self-concept is 'normal' then focus can turn to improving physical aspects; Shields et al., 2007). Previous research

has indicated that children with CP do not report significantly lower global self-concept compared to those without disabilities (Shields, et al., 2006), but may feel less competent in certain aspects of self-concept such as athletic competence (King et al., 1993; Ziebell et al., 2009). Furthermore, different groups of children with CP might have lower self-concept in different areas (e.g., teenage girls vs. young boys), and therefore, interventions to improve self-concept may be most helpful if specifically targeted toward certain groups.

Exercise interventions have shown to contribute to improving self-concept for children with and without disabilities (Martin, & Whelan, 2012), and more specifically, children with CP (Darrach et al., 1999). On the other hand, some domains of self-concept have been reported to be inhibited by participation in exercise interventions. Dodd, Taylor, and Graham (2004) found the exercise group reported lower scholastic competence and social acceptance, after a 6-week training programme compared to the control group. The varying results from previous research suggest there is insufficient evidence to assume children with CP have lower self-concept compared to typically developing children. Furthermore, with self-concept identified as a predictor of mental well- and ill-being, as well as social functioning, QoL, and independence (Russo et al., 2008), this indicates why self-concept is an important factor to measure.

It is clear the participants put a strong emphasis on the importance of measuring psychological factors, and therefore the importance of these factors in relation to physical activity for individuals with CP in terms of motivation, engagement, and adherence is further discussed in chapter 5 (see section 5.1).

4.7.3 Physical Factors

The mean scores for the physical factors, although high, were lower and more widely distributed in comparison to the social-emotional and psychological factors, with 'pain' having the highest mean rating ($m = 4.53 \pm .83$), and 'appetite' having the lowest ($m = 3.06 \pm 1.16$). The wider distribution of ratings of the physical factors suggest that the participants were unable to reach a consensus about which physical factors were most important to measure. Additionally, the number of physical factors reported in Round 1 was greater than in any other domain, meaning the selection of physical factors can be more difficult, and this may be expected with the variable nature of CP. Furthermore, the social-emotional and psychological factors listed in the second round of the study all play an important role in the development of children and youth with CP (i.e., these are more universal), but this may not be the case for all physical factors as they are targeted more specifically. For example, the focus for one child may be on mobility, and the other on pain management, but for both, there may be an impact on psychological wellbeing. This highlights the issue that is

prominent in previous research and the current study, in that each individual may have a different physical focus or priority which will also change throughout development and growth. It is well documented that physical abilities can vary dramatically between children with CP, so numerous factors should be considered prior to engagement in a physical activity intervention to ensure there is no detrimental effect to the child's wellbeing. However, this can restrict recruitment of suitable participants for researchers leading to an ethical dilemma in not wishing to exclude individuals because of their abilities, but also methodological implications of smaller sample sizes.

One participant did suggest that the main priority of physical activity is the physical benefits, and that the social-emotional and psychological benefits will follow from being physically active. Additionally, two participants referred to the importance of all factors due to the potential cyclical impact of physical activity participation (i.e., becoming more physically active can positively impact your mobility), which in turn can provide more functional independence which can help to provide a sense of freedom boosting levels of self-efficacy and self-esteem, thus improving overall QoL. Furthermore, the impact each of the physical factors can have on each other can make it difficult to focus on one, and in turn make the designing of interventions, rehabilitation and training programmes more difficult due to the variability across individuals with CP and the factor important in their individual context. For example, pain is one of the most common barriers to physical activity (Riquelme et al., 2018), and an increased perception of pain can limit mobility and negatively impact sleep, which in turn can reduce physical fitness, and energy expenditure for day-to-day movement and activities increases, all of which contribute to increased levels of fatigue.

'Pain' was identified by participants as the most important physical factor to measure when evaluating the effectiveness of physical activity interventions. The importance of pain measurement is highlighted by participants in the current study, as 94.1% rated pain as 'important' or 'very important' on the Likert scale. This is not unexpected given the prevalence of pain experienced by individuals with CP from childhood. Ramstad et al. (2011) reported a prevalence of 62% for musculoskeletal pain in children and adolescents with CP (aged 8-18) and across all levels of severity (GMFCS levels I-V). Additionally, pain is influenced by the severity of the impairment (i.e., a more severe impairment suggests a higher perception of pain; Parkinson et al., 2010). As part of the daily life experience for individuals with CP, alleviating pain, and building pain coping and management strategies, should be an important goal for future therapy and rehabilitation programmes and interventions.

The daily presence of pain can influence lifestyle decisions including the limiting of physical activity participation, and in turn a loss of functional ability and locomotor skills (Tervo et al., 2006). The negative influence that pain can have on participation, QoL,

physical fitness (Verschuren et al., 2007), and psychological wellbeing (e.g., depression; Benrud-Larson, & Wegener, 2000) highlights the importance of coping with and managing pain, as it can improve various aspects of an individual's life (Findlay et al., 2016). Despite the limiting nature, and prevalence of pain in children with CP, Riquelme et al. (2018) reported higher light activity participation, less sedentary behaviour, and higher energy expenditure for children with CP compared to typically developing children with chronic pain. The results suggest the relationship between physical activity and pain is complex, and physical activity participation when experiencing pain can be down to the individual's ability to cope with it (Perruschoud et al., 2014). Given the high frequency of pain that children with CP experience on a day-to-day basis from childhood through development, this may cause them to develop coping strategies (e.g., activity perseverance) better than typically developing children with chronic pain, hence their ability to participate in some form of physical activity while experiencing pain.

The current study demonstrates the wide range of physical factors that parents and adults working with children with CP feel are important to measure, and each factor is going to be more important to one individual compared to another given the variable nature of CP. These individual differences can make it difficult for the design of future interventions/studies looking to improve physical aspects, but the current study shines some light on which physical factors the participants view as important.

4.7.4 Social-Emotional, Psychological, & Physical Domains

Of the three main domains, social-emotional was the highest rated ($m = 4.72 \pm .52$), with the psychological ($m = 4.65 \pm .54$), and physical ($m = 4.47 \pm .79$) domains also viewed as highly important aspects to measure. With over 80% of participants rating each domain as important (4) or very important (5), it highlights how important participants perceive each domain to be when evaluating the effectiveness of physical activity interventions for children with CP. The results emphasise the importance placed on each of these aspects of children's lives by the participants, and this may not be surprising given the influence the factors can have on QoL. In spite of the fact that all domains were considered important, the social-emotional and psychological factors appeared to be more universal. This, in combination with the gaps in research around psychosocial factors, and the complications of measuring individual and specific physical factors, plus restrictions on time and feasibility within the PhD study, meant psychosocial factors were selected as the focus for exploring in the frame running intervention. Thus, it is important to ensure the constructs are clearly defined, measured effectively, and implicated in the design of the intervention.

4.8 Limitations & Future Directions

The current study has several potential limitations. The design of the study was a Delphi style survey; however, it only consisted of two rounds. With additional rounds, the number of domains/factors could have potentially been filtered down further to a smaller number and therefore provided a more definitive rating of the more important domains; however, time pressures, and a diminishing response rate did not allow for additional rounds in the current study. Therefore, it is important to recognise that the data provides a 'snapshot' of the participants' views and opinions (limited by the constraints of the Delphi approach) and not a complete understanding of the most important areas of concern. This full picture will always be difficult to capture because of the nature of CP and the broad range of impairments related to CP; however, the Delphi technique offers a way to gather opinions.

Another limitation of the current study was the presence of the expert view from health care professionals, parents, and coaches, but not the inclusion of the views of children with CP as some other studies have been able to do such as Vargus-Adams and Martin (2009). Furthermore, the survey asked about evaluating the effectiveness of a physical activity intervention, and not about what is important to measure in relation to other types of interventions, or what the best outcome measures are when taking different stages in the development of individuals with CP into account. Thus, the results of the current study should not be generalised outside the settings of physical activity interventions and provides a view of important domains and factors across levels of development.

The current study raises the question about what the next best steps are to take in helping to understand the change and development in CP as there can be difficulty in focusing on one domain or group of factors that is suitable for an intervention with the ever-changing priorities of children and youth with CP. Because improving the QoL of children and young people with CP can be seen as the overall goal, there can be a desire to measure as many outcomes as possible, but this is ambitious and can be detrimental to the quality of the research. Therefore, it is important to accept that not every domain or factor can be measured, so focusing on one or two areas, as in the current thesis, may provide richer data.

4.9 Conclusion

This study highlights the wide range of factors considered to be important for physical activity interventions for children with cerebral palsy. The breadth of factors identified highlight a large number of aspects of life that can be impacted by being physically active, hence the importance of increasing participation in physical activity for children and adolescents with cerebral palsy.

The fear of a negative influence from physical activity participation can sometimes outweigh the potential positive benefits, thus a balance needs to be struck for physical activity interventions to ensure they are safe, enjoyable and fun. High levels of effort can sometimes be needed by parents and health care professionals to encourage children with CP to participate. As one respondent wrote, *“Children with CP are side lined and have difficulty making friends, so specialist physical activity interventions can be life enhancing”*, highlighting the importance of having suitable and accessible interventions, as it can encourage participation. Furthermore, the factors identified in the current study could contribute to the design of future physical activity interventions for children and adolescents with cerebral palsy, as it enables researchers to identify important aspects to measure. The qualitative and quantitative data gathered in the current study suggest there are many areas of focus for future research of physical activity interventions for children and young people with CP. However, for the children to participate, be physically active, and have the opportunity to gain the short- and long-term benefits of physical activity, they need to have access to a suitable activity, especially for those with more severe impairments whose opportunities are even more restricted. Frame running is one option that is accessible to individuals with more severe impairments (e.g., Bryant et al., 2015), but there is no research on the impact of frame running on the psychosocial wellbeing of children and young people with CP. Thus, the following chapter will explore the effect of a frame running training programme on the psychosocial wellbeing of children and young people with CP.

Chapter 5 – The effects of a 12-week frame running training programme on the quality of life and psychosocial wellbeing of children & adolescents with Cerebral Palsy

5.1 Introduction

The physical and psychological benefits of engaging in regular physical activity are well documented for children and adolescents with disabilities (Jaarsma & Smith, 2018). Despite this however, levels of physical activity among the CP population are shown to be lower in comparison to their typically developing peers (Bjornson, et al., 2007). Often children and adolescents with CP present an array of developmental difficulties, hence heightened interest in and importance of physical activity, particularly in the CP population. Furthermore, this is shown by a change in focus of rehabilitation programmes away from minimising deficits and towards enhancing physical activity participation (Capio, et al., 2010). Thus, providing opportunities for children and young people with CP to be physically active is important for their psychosocial and physical wellbeing and development. As discussed in chapter 2, the number of physical activity opportunities for this population are restricted, and even more so for those with more severe CP, therefore exploring the effects of frame running participation can provide insight into one of few opportunities for physical activity/sport participation for more severely affected individuals. Therefore, the aim of this study was to investigate the effects of participation in frame running at a recreational level, on various psychosocial factors.

As discussed in chapter 4, a two-stage Delphi study informed the factors selected and measured in the current study. The participants (parents, health care professionals, and frame running coaches) identified a broad variety of physical, psychological, and social-emotional factors which they perceived to be most important to measure for children and young people with CP participating in physical activity interventions. The results suggested that all factors were important, but participants placed more importance on psychological and social-emotional factors compared to physical factors, which may be due to the individualised nature of physical ability and development; hence it can be more difficult to focus on one area of physical development. Furthermore, there is a greater dearth of empirical research for physical factors/outcomes of physical activity interventions for children with CP in comparison to psychosocial outcomes and combined with the time and load constraints of the PhD project, this informed the focus of the frame running training programme, which was the participants' psychosocial wellbeing and quality of life. Informed by a combination of the results from study 1 and a lack of literature focussing on some the factors and how they may be impacted by physical activity participation, the focus of the current study involved exploring the impact on QoL, self-efficacy, self-esteem, self-concept,

and psychological wellbeing. Furthermore, with importance of enjoyment in promotion and adherence to participation in physical activity and exercise, the participants perception of frame running as an enjoyable adaptive activity was investigated. The following sub-sections will introduce each of the psychosocial factors that were included in the current study, and cover associated measurement considerations, with the details of the outcome measures selected discussed in section 5.2.4.

5.1.1 Quality of Life – An Introduction

Quality of Life (QoL) is emerging as an important construct in relation to childhood research, however, less is known about the QoL of children with CP, and more specifically, those with more severe impairments (Arnaud et al., 2008). CP affects approximately 2 to 3 per 1000 live births, and as a lifelong motor impairment with no known cure, there is an increasing importance placed on QoL as an assessment of treatment, rehabilitation, and intervention outcomes (Jonsson et al., 2019; Makris et al., 2019). Despite the growing popularity of QoL as an outcome measure, the definition for QoL can vary for children, adolescents, and adults, as the term is used commonly as a label for a variety of psychosocial and physical functioning variables (Smith et al., 1999). The World Health Organisation (WHO), while developing a measure for QoL, defined it as “an individuals’ perception of their position in life in the context of the culture and value systems in which they live in relation to their goals, expectations, standards, and concerns” (WHOQOL Group, 1993; pp. 153). More simply, QoL is a multidimensional construct and is an overall assessment of wellbeing across a variety of domains including the social and emotional functioning of an individual, and not just a focus on their physical health and functioning, such as what an individual can and cannot do (Bjorson & McLaughlin, 2001; Gilson et al., 2014). Overall, QoL is seen as a more global construct. Health-related QoL (HRQOL) is considered a sub-domain of QoL and includes the perceived psychological, physical, and social health and function (Maher, et al., 2015), thus health-related QoL will be the focus for the current study.

For children with CP, similar to their typically developing peers, QoL includes their perception of their physical, social, and emotional well-being and a measure of QoL needs to be sensitive to the changes children go through during development (Waters et al., 2009). There is contrasting evidence regarding severity of impairment and the relationship to a child’s QoL. The assumption was an individual’s level of functioning equates to their QoL, which is in line with research by Araud and colleagues (2008) who found parent-reported QoL to be strongly associated with impairment levels in children with CP across Europe (i.e., low level of impairment is associated with higher QoL). However, other research has reported weak relationships between the level of functioning and QoL, which suggests,

importantly, a good QoL can be achieved despite the presence of functional limitations (Livingston et al., 2007). Depending on the type and severity of CP, the clinical prognosis can range from being ambulant almost at a similar level to typically developing peers, to struggling with basic life functions like breathing (Park et al., 2016). Irrespective of the severity of CP, the best possible QoL should be an aim for the individual. The importance of QoL has been highlighted in previous research (e.g., Vargus-Adams & Martin, 2010), and in the previous study in this thesis participants gave QoL a high rating of importance as outcome measure in physical activity interventions. Furthermore, participants suggested QoL can be influenced by numerous factors (e.g., physical functions, environmental factors) and they can lead to an improved QoL in a linear and/or cyclical nature, so in attempts to improve the QoL of children with CP, the impact of these factors should be considered (Park et al., 2016). In sum, it is clear that improving the QoL is key, thus measuring QoL is important in the current study. However, it can be difficult to measure.

5.1.1.2 Measurement of Quality of Life

As mentioned, QoL is a multidimensional construct and can be difficult to measure due to the variety of factors encompassed in the measures, for example, daily functioning, school activities, pain, and movement (Ramstad et al., 2012). However, QoL measurements are becoming increasingly more common as a way to evaluate the effectiveness of rehabilitation and intervention programmes for children and young people with CP (Davis et al., 2006). Therefore, it can lead to difficulty in the comparison of QoL data across instruments and studies; however, it remains an important factor to measure to provide a rounded insight into the impact CP can have on their life, as well as the effects of interventions and/or therapeutic programmes on their QoL (Gilson et al., 2014).

To measure QoL, there are a number of instruments that can be administered, either general (e.g., Paediatric QoL inventory 4.0; Varni et al., 2001), or condition specific (e.g., Paediatric QoL Inventory – CP module; Varni et al., 2006) which can be completed by self- and/or proxy-report. The use of self-report measures may be the preferred option to measure QoL given the subjective nature of the construct. However, this is not always going to be possible given the range of impairments associated with CP. With the changing view and importance placed on child self-report measures for QoL (Davis et al., 2017), Varni and Colleagues (2006) developed a condition-specific measure of HRQOL for children and adolescents with CP, the Paediatric QoL Inventory – Cerebral Palsy Module (PedsQL-CP). The PedsQL-CP is the outcome measure selected for the current study to measure perceived QoL in the participants and will be discussed further in section 5.4.2.4. The correlations between parent and child report for the PEdsQL 4.0 were similar for the physical functioning, but when it came to the psychosocial scores, there were discrepancies between

the parent and child average scores, especially for the emotional functioning scores (Varni et al., 2006). These discrepancies highlight it can be difficult for parents to understand how their child is feeling and that proxy parent-report forms should only be used if the child is not capable of self-report (Varni et al., 2005). With reference to the current study, this is important to note given the targeted cohort, and the cognitive difficulties associated with more severe CP. Condition-specific measures and proxy vs. self-report measures are further discussed in Chapter 3 (see section 3.5.3.3). Irrespective of the measures used, it is important to measure the QoL of children with CP because it provides an insight to what aspects are affected when participating in interventions and/or rehabilitation, but also provides an insight to what can/should be targeted in future research aiming to improve their QoL (Gilson et al., 2014).

Previous research has explored the relationship between physical activity and QoL in children with CP, and Maher and colleagues (2015) used a generic measure, the PedsQL 4.0 to examine the relationships between physical activity, QOL, and happiness in children and youths with CP. Previous research has shown a positive relationship between physical activity and QoL in typically developing children, which can continue into adulthood (Bjornson et al., 2008; Brown et al., 2004), but this relationship is less well investigated in children with CP indicating a possible gap in the research. Results from Maher et al. (2015) showed that there was a significant positive association between physical activity and both physical and social functioning, and happiness. Furthermore, the results show that physical activity can benefit QOL, adding to the existing knowledge of potential benefits of being physically active for children with CP, and highlighting the potential impact frame running may have for children with CP.

Similar to Maher et al. (2015), Bjornson and colleagues (2008) investigated the influence of physical activity level on self-reported physical, behavioural, emotional health, and QoL in youth with CP aged 10 to 13 years. The results showed that level of activity did not influence QoL, but did impact their self-reported physical, emotional, and behavioural health. Despite the lack of impact physical activity had on QoL, it is still important to note the results on improving aspects of health by being physically active, and it adds to the knowledge that children with CP are able to self-report. In contrast to Bjornson et al. (2008), but in line with Maher et al. (2015), Verschuren et al., (2007) reported a significant improvement in QoL in children with CP when involved in an exercise programme compared to the control group. However, the participants were high functioning (GMFCS levels I and II only), and therefore the results may not generalise to the lower functioning children with CP (i.e., GMFCS levels IV and V). The results suggest participation in some form of exercise programme can significantly improve the QoL of individuals with CP, and therefore should

be considered when designing rehabilitation programmes and future interventions, hence why QoL is a selected outcome for the current research.

5.1.2 Self-efficacy – An Introduction

Self-efficacy is a key construct within two of the theories discussed in the literature review chapter (see section 2.4.3.3), the transtheoretical model (Prochaska & DiClemnte, 1982), and the social cognitive theory (Bandura, 1997). Self-efficacy was defined by Bandura (1986, pp. 391) as:

“people’s judgements of their capabilities to organize and execute courses of action required to attain designated types of performances. It is concerned not with the skills one has but with the judgements of what one can do with whatever skills one possesses. “

Self-efficacy beliefs are thought to have an important influence on behaviours because if an individual is lacking belief in their capabilities to perform or complete the chosen task or activity, then the incentive to perform is diminished (Suldo & Shaffer, 2007). The views an individual has on their capabilities will influence their choice of activities in which to participate, and the amount of effort and level of persistency to assign to a given activity (McAuley & Blissmer, 1999). Furthermore, the ability to complete an activity successfully will cause the individual to believe they have the ability to engage in the activity in the future, and this can be important in the maintenance of positive behaviours (e.g., being physically active).

Low levels of self-efficacy in individuals with CP are possible as they are more likely to suffer from a negative perception of their body and/or ability to complete activities due to a mobility or functional impairment, in turn leading to difficulties in social situations such as building friendships or group exercise (Kim et al., 2017). A study by van der Slot et al. (2010) examined the association of self-efficacy with HRQOL and participation in the life of adults with CP. The results indicated that the adults with CP had a low perceived HRQOL and difficulties in social participation in comparison to the reference population. From the results of the General Self-efficacy scale, the participants showed that there were no significant differences in self-efficacy scores between the adults with CP and reference population, apart from the persistence scale where the participants with CP scored significantly higher. The participants who scored highly on the effort subscale of the self-efficacy scale, were more likely to perceive a higher mental and physical HRQOL. This suggests an individual with higher self-efficacy, who is willing to expend more effort on a task, will in turn more likely

perceive a higher HRQOL, and this adds to the evidence that self-efficacy can improve social participation (van der Slot et al., 2010).

Self-efficacy is a prominent factor within the topic of physical activity, and it often correlates highly with physical activity behaviour, with research being conducted in both healthy and patient populations (Biddle et al., 2015). An individual's self-efficacy (the belief in their own capabilities) has been identified as a predictor of physical activity participation in healthy adults, and this includes the engagement in, and maintenance of physical activity (Ashford et al., 2010). Participation in acute bouts of physical activity can significantly impact the self-efficacy of the participant, and participation may have a greater impact on self-efficacy than change in physical fitness (McAuley, & Blissmer, 2000). Therefore, the participation in a physical activity intervention may have a greater impact on self-efficacy in comparison to smaller bouts of physical activity. The relationship between physical activity and self-efficacy is reciprocal, but it is also more complex than that. An individual's self-efficacy does not make it possible to predict physical activity behaviours, and not each physical activity will have the same impact on someone's self-efficacy (McAuley & Blissmer, 2000). Furthermore, increasing self-efficacy has been identified as a suitable method to increase levels of physical activity but the best method/technique/intervention to do so is not clear (Ashford et al., 2010).

An individual's perception of their self-efficacy does not suggest a single global personality trait, but it varies across various behavioural domains, for example, academic self-efficacy, physical self-efficacy, or emotional self-efficacy (Reid, 2002). Higher levels of self-efficacy in children can enhance their confidence, which will help them to take on more challenging tasks, and commit to the task for longer, and in turn help complete the task successfully (Suldo & Shaffer, 2007). For children with CP, self-efficacy has also been shown to impact participation (Bent, et al., 2001); however, the role of self-efficacy in children and adolescents with CP has not been thoroughly examined. Research in typically developing children and adolescents suggests that improving their self-efficacy should, in turn, improve their participation in physical activity. This is important because of the small amount of time children with CP report being physically active combined with the high amount of sedentary time reported, putting them at risk of an unhealthy lifestyle (Verschuren et al., 2016). Increasing their physical activity levels is important for their long-term health because of the link with high levels of sedentary time, low levels of physical activity, and increased risk to cardiovascular disease that can continue into adulthood.

As previously mentioned, an individual who is able to complete a task successfully will believe they can participate in the future, and therefore a positive experience with physical activity can help in developing self-efficacy. However, for children and adolescents with CP, the functional difficulties and associated impairments of CP that can limit their

ability to participate can be a reason for lower self-efficacy. The fear of failing, experiencing pain, or feeling humiliated in front of their friends, are all examples of why they may avoid being physically active, increase their sedentary behaviour, and in turn this is likely to negatively impact their self-efficacy. Conversely, these barriers to participation are more likely to be overcome by those who have a higher self-efficacy, which brings it back to the issue of identifying the best method to increase self-efficacy.

5.1.2.2 Measurement of Self-efficacy

High self-efficacy can help individuals build confidence in different environments, thus enabling them to take on new challenges, put in a sustained effort and hopefully succeed, with experiences of success continuing to enhance self-efficacy (e.g., doing better in school; Suldo & Shaffer, 2007). However, given the small amount of research on self-efficacy in children with CP (e.g., Reid, 2002), the measurement of self-efficacy for this population is difficult. Furthermore, measuring self-efficacy in children has traditionally been done using adult scales or adapting adult scales to use specifically with children but these scales were aimed at specific areas of functioning, like smoking and diabetes (Muris, 2001; Suldo & Shaffer, 2007). Bandura et al. (1999) developed a measure to assess children's general self-efficacy tapping into domain specific self-efficacy, covering social (capability to deal with social situations), academic (capability to cope with academic affairs), and self-regulatory (capability to deal/resist peer pressure) self-efficacy. Muris (2001) developed the Self-efficacy Questionnaire for Children (SEQ-C) to assess domain specific self-efficacy. Domain specific self-efficacy can be an indicator of an individual's mental health and functioning, for example, low social self-efficacy can be linked to depression, or low emotional self-efficacy (the capability of dealing with negative emotions) can be linked to high levels of anxiety (Bandura et al., 1999; Suldo & Schaffer, 2007). Each domain specific self-efficacy is important to the development of children; therefore, an improvement in any or all of these domains, including global self-efficacy, will be beneficial for improving the QoL of children and adolescents with CP, hence the inclusion of the SEQ-C in the current study.

5.1.3 Self-esteem & Self-concept

5.1.3.1 Self-esteem – An Introduction

Self-esteem is referred to as somebody's feelings of their own worthiness and competence (Muris et al., 2003). Self-esteem theory suggests that the overall view we have of ourselves (global self-esteem) is influenced by other perceptions of ourselves in more specific domains, for example, physical and social domains, and that each of these are developed from perceptions of further sub-domains (Biddle et al., 2015). This points to self-esteem

being a multidimensional construct with a hierarchical structure, with domains at the top (e.g., global self-esteem) being more difficult to influence and change compared to those lower down the hierarchy, where the more specific domains are open to change (e.g., physical and academic self-esteem; Biddle et al., 2015). It is frequently viewed as an important indicator of an individual's psychological well-being and in the past has been identified as the factor that can best indicate the influence of regular exercise on psychological well-being (Sonstroem & Morgan, 1988).

Self-esteem has been shown to be both directly and indirectly associated with physical activity (Sani et al., 2016), suggesting the promotion of regular physical activity should be aimed at those reporting low self-esteem. Also, with self-esteem being regarded as an important indicator of a child's psychological well-being (Muris et al., 2003), the potential of improving self-esteem via physical activity participation is a frequently used rationale when promoting health programmes (Biddle et al., 2015). Personal development, happiness and life satisfaction have been linked to high self-esteem, while poor physical health, depression and anxiety are linked to low self-esteem (Harter, 1993; Russo et al., 2008), highlighting the importance of being able to impact self-esteem in a positive manner.

Biddle et al. (2015) outlined two approaches, the 'motivational approach' and the 'skill development' approach, to describe how self-esteem can be a precursor of physical activity as well as an outcome. Self-esteem is seen as a motivational factor for physical activity in the 'motivational approach'. Here, individuals with high self-esteem are more inclined to participate in a physical activity because they see it as an opportunity to maintain or improve their competence and self-worth. The 'skill development' approach suggests that self-esteem is an outcome of physical activity participation and is influenced, in either a positive or negative way, through the development of skills and different experiences and this approach is used frequently in physical education programmes. The potential impact of physical activity participation on self-esteem in children and young people is important to acknowledge when investigating the impact on children with disabilities, and more specifically, CP.

Although some individuals may not experience difficulties in this area, children with CP are assumed to experience lower levels of self-esteem compared to their typically developing peers because of the mobility impairments and associated deficits linked with CP, which affect their performance in everyday activities (Shields et al., 2006). However, Manuel et al. (2003) found pre-adolescents and adolescents with CP showed similar scores on a measure of self-esteem when compared with healthy adolescents. Russo and colleagues (2008) carried out a study comparing self-esteem, self-concept and QoL between children with hemiplegic CP and typically developing children. Their results showed lower scores on the self-concept and QoL measures in the children with CP, but the measure of

self-esteem showed no difference in scores between the two groups, supporting the results from Manuel et al. (2003). A significant positive correlation between self-esteem and child report for QoL was also found, which is important because it can help when interventions aimed at improving QoL in children with CP, in turn, improve self-esteem, and vice versa. A possible explanation for self-esteem scores not being significantly different to those of typically developing peers is the high functional capacity of the participants in the study by Russo et al. (2008). With 97% of the participants with CP having a gross motor function classification (GMFCS) of level I or II, they had a reasonable level of functional capacity. Functional capacity is thought to impact self-esteem of individuals with CP because the limitations that are associated with CP can negatively affect participation and performance of activities of daily living, with greater functional capacity predicting greater self-esteem (Espín-Tello, et al., 2018).

5.1.3.2 Self-concept – An introduction

Another important construct in relation to a child's psychological functioning is self-concept (Shields et al., 2007). Moreover, self-concept is sometimes considered to be the same as self-esteem, but self-esteem is an extension of self-concept. Self-concept is considered an individual's perceptions of the self across different aspects of life (e.g., physical appearance, and scholastic competence), where self-esteem attaches a value to the perception (Biddle et al., 2015). Furthermore, self-esteem is viewed as a unidimensional construct compared to self-concept, which is viewed as a multidimensional construct (Martin & Whalen, 2012). The multidimensional nature of self-concept allows the benefits to be far reaching compared to self-esteem, and span across various and numerous outcomes (e.g., exercise performance, social, and academic).

Self-concept is thought to influence markers of ill- and well-being including social (e.g., social interactions) and cognitive (e.g., attention levels; Marsh & Craven, 2006) markers, and is known to develop through interactions with social experiences and cognitive processes (Cheong et al., 2018). Furthermore, poor self-concept is linked to poor academic performance, behavioural problems, and mental health problems including anxiety and depression, highlighting it as a predictor of psychological functioning. Due to the part self-concept can play in psychological functioning, a healthy self-concept for a child with CP is a goal of parents and health professionals (Gannotti et al., 2010). It is assumed that children with CP have a lower self-concept, and the levels of self-concept continue to diminish as the severity of CP increases. However, the understanding of self-concept in children with CP is lacking (Dunn, 2017). Self-concept is not that well researched a construct, with the previous research providing various, and sometimes contrasting results (e.g., Darah et al., 1999 and Soyupek et al., 2009). Furthermore, a systematic review by Shields and colleagues

determined there was no sufficient evidence to conclude children with CP had a lower global self-concept compared to their typically developing peers. The difficulty in generalising results and assuming a lower self-concept in children with CP compared to typically developing peers is the perception children have of their lives. Children with CP have different life experiences than typically developing children, so it cannot be assumed that children with CP will view their situation the same way. Of course, life experiences also vary between typically developing children, but the personal/environmental interactions for children with CP are different given the functional limitations and associated impairments of cerebral palsy. The self-concept measures used to study typically developing children may not capture the interactions and life experiences of children with CP.

A Delphi study by Cheong, Lang, Hemphill, and Johnston (2016) looked at what areas of self-concept in typically developing children would be identified as similar to those for children with CP. Health professionals, parents of children with CP, and children with CP participated in the Delphi study naming and selecting areas and factors they feel are relevant to self-concept and rating their importance to reach a consensus. The results showed that many of the items of self-concept identified for children with CP were similar to those of typically developing children. The items that reached high levels of consensus (meaning they were identified and agreed as relevant to self-concept in CP), were areas of more general performance, both personal and social (e.g., schoolwork and friendships). On the other hand, aspects that did not reach consensus, were viewed as higher level or more specific performance (e.g., algebra skills, or sports performance). Eighty percent of reported items were identified in self-concept measures for typically developing children. Alongside these areas of self-concept, 20% of the items were aspects of self-concept specific to the cerebral palsy population. These factors further highlighted the importance of participation and importantly showed the understanding of participation at a CP level. Furthermore, it shows an understanding of overcoming and adapting to difficult situations to participate (e.g., always trying their best). Additionally, they identified areas that are specific to having a cerebral palsy (e.g., difficulty in communication and mobility) which are important to their self-concept. The results indicate the competence of children with CP participating in everyday environments plays a more important role in their self-concept compared to higher/more specific skills and performance. This emphasises that self-concept instruments for typically developing children may not encapsulate the self-concept of children with CP. Furthermore, self-concept may vary within subgroups, and it is unclear whether self-concept is lower in children with CP, and continues to drop as severity of the condition, highlighting the benefits of a CP specific self-concept measure to assure the CP specific elements of self-concept are not missed.

5.1.3.3 Measurement of Self-concept & Self-esteem

Overall, it is clear that measuring self-concept is important for children with CP as it can be an indicator for QoL, social and psychological functioning (Russo et al., 2008; Cheong et al., 2016). Cheong, Lang, and Johnston (2018) investigated self-concept in children with CP using a CP specific measure, myTREEHOUSE Self-concept Assessment. The measure evaluates self-concept from personal, social, and perceived performance perspectives, and each of these are measured across eight domains of self-concept. The results showed 34% of participants reported low self-concept for social and perceived performance perspectives, highlighting that it is important to focus on their strengths and interests to positively impact self-concept. This was one of the first studies using a CP-specific measure for self-concept, therefore providing some initial insights into children with CP and what they think about themselves.

The foundations of self-esteem in children begin in childhood when they are able to compare themselves with their peers, and with this development they begin to understand where they are in different domains of functioning, for example, sport and academic performance (Muris et al., 2003). Furthermore, self-concept is thought to be a potential indicator of an individual's QoL, social and psychological functioning, and with self-belief constructs, including self-esteem and self-concept have consistently shown to impact on QoL and health outcomes (Harter, 2012), it is important to measure both in the current study. As the myTREEHOUSE self-concept assessment was not available for the current, so the Self-Perception Profile for Children and Adolescents (SPPC & A) was administered to measure self-esteem and self-concept (Harter, 1985).

5.1.4 Psychological Wellbeing – An Introduction

Psychological wellbeing is not just about one aspect, but a combination of a positive mental state and optimal functioning (Winefield et al., 2012), and was summarised by Huppert (2009) as follows: “psychological wellbeing is about lives going well. It is the combination of feeling good and functioning effectively.” (p. 137). In the current research, the focus on psychological wellbeing will be in reference to psychological problems and mental health. Mental health problems in children are becoming more prevalent, with approximately 10% of children in the UK, between the ages of 5 and 16 years, having a mental health disorder (Ahn et al., 2018). Children with disabilities are at an even greater risk of developing a mental health disorder and experiencing mental health problems in comparison to their typically developing peers (Goodman, 2002). Furthermore, children with chronic disorders, like CP, may be at greater risk of psychological problems for a variety of reasons, for example, at risk of experiencing negative social experiences compared to children without

disabilities, such as feeling left out, or being bullied because of their disability, which could lead to psychological maladjustment (Parkes et al., 2008). The mobility impairments associated with CP can lead to increased sedentary behaviour, and/or fewer opportunities to participate in physical activity, and in turn lead to missing out on opportunities for social development. These reasons individually, or interacting can potentially lead to psychological problems, but on the other hand, existing psychological problems may be a cause of low levels of physical activity participation, for example, low levels of motivation, or a perceived lack of ability to participate (Ahn, et al., 2018). Furthermore, the importance of measuring psychological wellbeing in children with CP was highlighted by the participants in the previous study, with participants rating psychological wellbeing as the most important psychological factor to measure. Moderate- and vigorous-intensity physical activity are both key for improving physical and mental health (Sani et al., 2016), however, the relationship between mental health and physical activity in children is less researched compared to the relationship in adults (Ahn, et al., 2018), and even less in children with disabilities (e.g., Matteucci et al., 2019). Whether it is formal physical activity (e.g., circuit and strength training), recreational physical activity (e.g., participation in sports), or leisure and unstructured physical activity, the body of evidence showing associations between participation and improvements in children's psychological health is growing (Ahn, & Fedewa, 2011). By contrast, there is evidence that increased levels of sedentary time and low levels of physical activity have been associated with poorer psychological well-being in children (Ahn, et al., 2018).

As previously mentioned, there is limited research exploring the psychological wellbeing of children and young people with CP, however it can be important to measure as good psychological wellbeing is associated with good physical health (Huppert, 2009). One study by Jiang and colleagues (2016) reported no association between the psychological wellbeing of young adults with CP and their gross motor function and body structure, however reported lower psychological wellbeing compared to able-bodied peers. The results suggest that the psychological wellbeing of young adults with CP may not be negatively influenced by their perception functional ability, however, the young adults have had more time to come to terms with their disability, compared to children and adolescents who are still developing, thus it is important to explore psychological wellbeing in children with CP.

5.1.4.2 Measurement of Psychological Wellbeing

Parkes and colleagues (2008) conducted a cross-sectional study investigating psychological problems in children with CP from ages 8 to 12 years and across all severities (GMFCS levels I to V). Using the strengths and difficulties questionnaire (SDQ) to measure psychological well-being, they found a significant number of the participants indicated they

were at high risk of mental health problems (e.g., anxiety, depression). The functional and intellectual impairments showed a positive correlation in the participants, i.e., the children with higher functional ability also had higher intellectual ability. However, the functional and intellectual abilities had different associations with the risk of high scores on the SDQ and therefore psychological problems. They found the participants with higher functional impairments were at lower risk of psychological problems. This may be down to the lack of sensitivity of the SDQ for children with more severe level of CP, but also, there is the suggestion that with higher functional impairment, there is less opportunity for them to participate in negative behaviours. Inversely, it was also suggested that the children with a milder form of CP, and thus a lower functional impairment, might struggle to accept their disabilities when they are more similar to their peers without disabilities (e.g., Jiang et al., 2016). There are a variety of factors (e.g., personal and environmental), and the interaction between those factors can cause psychological problems in children with CP, meaning the possibility of alleviating the psychological problems via treatment or incorporating physical activity participation should be a viable option for children with CP across all severities. Therefore, given the limited research into the psychological wellbeing of children with CP, it presents a gap in the knowledge that the current research will attempt to fill by exploring the impact of frame running participation on psychological wellbeing. However, due to the low number of existing studies, there is a limited number of validated instruments to measure psychological wellbeing. It is thought there is no 'gold standard' for measuring psychological wellbeing, with various outcome measures identified (e.g., Mental-Health Continuum-Short Scale; Keyes, 2005), but with measurement involving children with CP limited, the current study will employ the SDQ, similar to Parkes and colleagues (2008).

5.1.5 Participation – An Introduction

Participation is considered a vital outcome in health for children with and without disabilities (Imms, 2008), but can be difficult to define. A broad definition provided by the World Health Organisation (WHO) is "involvement in a life situation" (WHO, 2001). The importance of participation is highlighted by the fact that it was one of two new concepts introduced to the International Classification of Disability and Health (ICF) framework in 2001. 'Participation' replaced the 'handicap' concept, and this changed the direction of impact of disability from a negative to a positive, highlighting that interaction between an individual's physical and social environment can result in participation (Forsyth & Jarvis, 2002).

Participation is vitally important because the context of participation, whether it is social, leisure, or physical activity, can provide the basis for development in a variety of areas including physical and mental health (King et al., 2000). Participation in physical,

leisure and social activities can bring far-reaching benefits to children with CP, as well as those without disabilities, but those with CP are more at risk of lower levels of participation in daily activities due to mobility and cognitive deficits, and associated impairments (Lauruschkus et al., 2015). When children with CP do participate, the variety of leisure activities available to them is small, and there are fewer social activities, especially when social activities are spontaneous (King et al., 2006). As an individual with CP moves through childhood, adolescence, and into early adulthood, the levels of participation are thought to become more restricted (King et al., 2006). Therefore, it is important to be able to measure the types of activities, preferences for certain types of activities, and the dimensions of participation as it can help to guide future interventions aimed at increasing participation and giving the opportunity to individuals with CP to benefit from participation whether it be physically, socially, and/or psychologically.

A literature review of participation in children with cerebral palsy highlights that this population have a reduced level of participation, and the increase in impairments in turn decreases participation (Imms, 2008). The results also showed children with CP did participate in a wide range of activities but at a low intensity, with school settings as the most popular location for the participation research. This suggests that there is a gap for the current research to add to the participation in children with CP literature. This is because frame running can be viewed as a suitable, and different opportunity to participate in physical activity at a higher intensity, and in a different environment, (i.e., the community setting) for children with CP. Participation in physical activity and sport can potentially have a positive impact all the way to elite level sport. Groff et al. (2009) investigated the effects of participation in adapted sports on QoL and athletic identity for athletes with CP. The results showed that participation in sport can affect the athlete's QoL and athletic identity, suggesting that to encompass these benefits for individuals with CP, more opportunities to participate and compete in sport should be made available.

A qualitative study by Laurusckus, Nordmark, and Hallström (2015) looked at the experiences of children with CP with regards to what they enjoyed about participating in physical activities. The children responded with a mixture of positive and negative comments, highlighting the fact that participants were physically active because they wanted to be there, or they believed they had to be. Positive responses show an understanding that being physically active was good for them, they felt they had the capabilities to participate, they had a friend or family member to do it with, or simply just enjoyed the feeling (Laurusckus, et al., 2015). On the other hand, some opted out of participating in various activities due to various barriers including relating physical activity to feelings of pain and fatigue, feeling singled out when participating alongside children without disabilities, not feeling that they were able to participate, or not being offered the chance to participate

despite being willing and able to (Laurusckus et al., 2015). The results show there are barriers and facilitators to participation in physical activity, as discussed in chapter 2, and participation can be a key goal of interventions.

5.1.5.2 Measuring Participation

If you are able to understand the participation of an individual, when it elicits joy or promotes social engagement, and their preferences in activities, it can help to promote development and the short- and long-term benefits associated with physical activity participation (Imms, 2008; Murphy, 2008). Furthermore, it is important to understand the activities that children want to, and can participate in, across the environments that they enjoy (e.g., community and/or leisure), and this increased understanding of participation can help to enhance participation in day-to-day activities. Therefore, the importance of measuring participation of children and young people with CP is clear as it can enhance future participation and the all-important QoL, however there are a lack of instruments that fully measure participation across various life domains (Noreau, et al., 2007). On this basis, the Children's Assessment of Participation and Enjoyment (CAPE) was developed to measure participation. It can be used to help map out a child's participation across different activity types, how often they participate, with whom, and how much enjoyment they get from participation. Notably, it has been used in previous research involving children with CP (Majnemer, et al., 2008; Declerk et al., 2016), therefore this measure will be used in the current study to measure participation. One factor that is key to promoting participation in physical activity, is enjoyment, which is discussed in the following section.

5.1.6 Enjoyment – An Introduction

The perception of a specific activity being fun can be an important facilitator to participation in said activity; therefore, the importance of providing a fun and enjoyable option for physical activity can be key for encouraging and maintaining participation. As reported in the previous study (see section 4.5.4.1) enjoyment was identified as the most important factor to measure when evaluating the effectiveness of a physical activity intervention for children with CP. A better understanding of enjoyment, and how it can affect physical activity participation and health, can assist in the development of physical activity programmes.

The enjoyment of physical activity reflects the participant's feelings of pleasure and fun as a response to participation and can be a key component in helping to promote physical activity participation. Within self-determination theory (see section 2.4.1), enjoyment is thought of as an important component of intrinsic motivation that affects physical activity (Jin et al., 2018). An individual with high levels of intrinsic motivation for performing a

behaviour or activity will do so on their own terms (Ntounis, 2005). In contrast, if the performance is done under pressure or as an obligation and without enjoyment, the benefits of participation may be reduced, which in turn may lead to an unhealthy relationship with physical activity that can continue and develop into adolescence and adulthood (Kretchmar, 2008). Enjoyment, in the context of self-determination theory, can be a predictor of physical activity and in turn health, emphasising the importance of enjoyment in promoting physical activity programmes and health behaviours (Jin et al., 2018).

To enhance the benefits of being physically active for children with CP, rehabilitation programmes and interventions focussing on physical, psychological, social, and emotional outcomes need to incorporate engagement and enjoyment (Lai et al., 2015). Enjoyment has been shown to be an important predictor of whether or not children with coordination disorders will participate in physical education, and in turn if they choose to continue with their participation, particularly when it is no longer an obligation (Cairney, et al., 2007). With this in mind, the enjoyment of physical activity (e.g., frame running) can be a predictor of maintenance of the activity, and in turn, the continued yielding of the physical, psychological, and social benefits associated with physical activity participation. Therefore, measuring enjoyment could provide an indication of continued participation and the possible engagement in future physical activity and/or health behaviours.

5.1.6.2 Measuring Enjoyment

Enjoyment of physical activity is suggested to have a positive impact on levels of participation but choosing a single measure of enjoyment is not straightforward for the chosen population given the subjective nature of the construct (Moore et al., 2009). The CAPE asks participants to recall their enjoyment over the past four months, but the current study aims to measure enjoyment of each frame running training sessions. Therefore, the participants were asked to rate their level of enjoyment after each training session on a 5-point Likert style scale using faces as pictorial representation instead of numerical (see Figure 5.2). There were no psychometric properties for this measure, however, it is similar in format to the CAPE, a validated scale, and previous research has shown it to be a simple yet effective tool to measure enjoyment in children with CP (e.g., Declerk et al., 2016). Furthermore, it is important to include as a measure as it allows for better understanding of the enjoyment levels that are related to the frame running training sessions specifically.

5.1.7 Research Questions

This study is one of the first studies to look at frame running and the impact participation can have on psychosocial wellbeing. Each of the constructs discussed in this chapter have been

shown to impact the overall wellbeing of children with CP, with some more researched than others (e.g., QoL is better understood than self-concept or self-efficacy). Furthermore, the constructs are thought to be influenced by the engagement in and adherence to physical activity, however, opportunities for individuals with CP are restricted and they can struggle to find suitable opportunities to be physically active. Thus, this can increase the difficulty for children with CP to break the sedentary behaviour patterns, leading to an unhealthy and inactive lifestyle which can be carried through from childhood, adolescence, and into adulthood. Therefore, the current research exploring the effects of a 12-week frame running training programme on the psychosocial wellbeing of children and adolescence with CP will provide initial insight into the impact of frame running participation on the selected constructs. On this basis, the current study will aim to answer the following research questions:

2. Does participation in a 12-week frame running training programme have an effect on the perceived QoL of young people with CP?

3. Does participation in a 12-week frame running training programme have an effect on the psychological aspects: self-efficacy, self-esteem, self-concept, and psychological wellbeing of young people with CP?

4. Does participation in a 12-week frame running training programme have an effect on the participation and enjoyment of other activities for young people with CP, and is frame running perceived as an enjoyable physical activity for young people with CP?

5.2 Methodology

5.2.1 Study Design

The design for this study was a single-case quasi-experimental pre- and post-test design, with a baseline phase (from baseline to pre-test) and a midpoint (between pre- and post-test) for additional data collection and assessed the changes from pre-test to post-test by analysing the responses of five single subject-cases. Single-case research methods have been used to facilitate an understanding of interventions and their effectiveness in numerous disciplines (Barker et al., 2013; Forrest-Lawrence 2018).

A review of single-case research in sports psychology by Barker, Mellalieu, McCarthy, Jones, and Moran (2013) found that exploring the effect of interventions on psychological constructs was a viable study design. A feature that is unique to this study

design is it provides the researcher with the capacity to carry out investigations with one or very few cases, as in the current study. The design lends itself to the current study as it allows a rigorous evaluation of the effects of the intervention. Furthermore, as a new research area (i.e., impact of frame running on the psychosocial wellbeing and QoL of children with CP), and working with unique populations (i.e., children with CP), the current design is viewed as valuable (Barker et al., 2013).

Additionally, a review by Beckers, Stal, Smeets, Onghena, and Bastianenen (2020) reviews evidence of single case study design involving children and adolescents with CP and concludes that it is a suitable design for the selected participants. Case study allows for detailed analysis of individual data and thus provides detail that otherwise may be missed (e.g., due to averaging). Especially with novel interventions and a heterogeneous sample: an intervention may benefit one person but not the other, and the benefit may not be clear at group level. Additionally, as previously mentioned, a case study design is useful when exploring a new area of research, assessing feasibility before embarking on a large-scale RCT. Furthermore, the use of a randomised controlled design can lead to difficulties in the generalisability due to the narrow selection of participants with CP who have a high diversity of impairments thus leading to smaller sample sizes for research (Beckers et al., 2020).

Given the practical constraints, which included participant numbers, equipment and group availability, and no experimental control, the use of a single case quasi-experimental pre- and post-test design was chosen over a randomised experimental design. As discussed earlier in this section, the current study lends itself to this design, and provides the ability to track the participants thoroughly at multiple points, allowing for more in-depth analysis in a new area of research. However, if sufficient numbers were available, it is acknowledged that using a randomised experimental design would produce more robust and credible results (Reichardt, 2009). Therefore, the effects of the frame running programme were assessed by comparing the scores from the measures completed by the single-case participants across the four time points: baseline (BL), pre-test (T1), mid-point (T2), and post-test (T3).

5.2.2 Participants

5.2.2.1 Participant Demographics

Six participants signed up to participate in the study, however one participant dropped out following the baseline measures. The five racerunners (2 males, 3 females; M age = 13.6 years, SD 5.85 years, range 5 – 18 years) attended one of two frame running groups in Scotland and had range of frame running experience. The demographic information for the participants is shown in Table 5.1.

Table 5.1 Participant Demographics of the 5 participants

Participant #	GMFCS Level	Education System	Frame running Experience (months)
1	II	Mainstream	0
2	IV	Special Needs	0
3	II	Mainstream	6
4	V	Mainstream	0
5	III	Special Needs	11

5.2.2.2 Inclusion & Exclusion Criteria

Participants for this study were children and adolescents diagnosed with CP aged between 4 and 18 years. Inclusion criteria were the ability to participate in frame running sessions and classified as GMFCS levels II to V. Exclusion criteria were cardiac or respiratory conditions that could have a negative impact on their ability to participate in the exercise, and/or any involvement in orthopaedic surgery 6 months prior to the training programme and during the allotted time period.

5.2.2.3 Recruitment

Participants were recruited through numerous channels via frame running clubs, special schools, and cerebral palsy centres and charities around Scotland. The recruitment of the participants lasted around 12 months prior to the first data collection point at baseline. The length of time for recruitment was much longer than anticipated due to a variety of reasons including the voluntary nature of frame running groups, and barriers to participation that were discussed in Chapter 2 (see section 2.5) and 3 (see section 3.4.1).

A frame running “come and try” day was organised at Craighalbert centre, Cumbernauld, Scotland, with frame running bikes available for children to come and ride for the first time. Due to the weather and time of year that this was held, December, the children were only able to try the bikes in a hall, so there was not as much freedom to move around on the bike compared to being outside. Parents were given an information sheet about frame running and the current research (Appendix C). Further visits to school “come and try” days, along with visits to frame running clubs, allowed the information to be spread about the current research. Visiting training sessions, the coaches, teachers, and parents at the sessions were told about the current research and handed or sent an information sheet via email. The coaches of the frame running groups also agreed to hand out information sheets to the parents at training sessions when the main investigator was not present. Potential

participants were referred to the main investigator, and then contacted via email and provided with further information about the research and what it would entail (see Appendix D).

5.2.4 Ethical Approval

Ethical approval for this study was obtained from the Moray House School of Education and Sport Ethics Committee at the University of Edinburgh. Before the start of the study, full consent was obtained from the parent(s) or guardian(s) of the participant, and consent was obtained from participants in the form of written consent via signatures or assent if they were physically unable to provide written consent. See Appendices E & F for consent forms for parents/guardians and children respectively; ethical consent is further discussed in chapter 3.

5.2.5 Outcome Measures

5.2.5.1. QoL: Pediatric Quality of Life Inventory – Cerebral Palsy Module (PedsQL-CP; Varni et al., 2006).

The PedsQL-CP is designed for toddlers right through to adolescents, 2 to 18 years, and is a 35-item questionnaire covering seven scales: (1) Daily Activities; (2) School Activities; (3) Movement and Balance; (4) Pain and Hurt; (5) Fatigue; (6) Eating Activities; and (7) Speech and Communications. The scales are split into age groups, and each group has a parallel child self-report and parent-proxy report measure, apart from ages 2 to 4 years (toddler), which only has the parent proxy-report. The child self-report has three categories, 5 to 7 years (young child), 8 to 12 years (child), and 13-18 years (adolescent), and the parent proxy-report covers all four age ranges. The children are asked how much of a problem each item has been over the previous month, and the parents are asked the same question in relation to their perceptions of the child's HRQOL. For all parent proxy-reports and child self-report from ages 8 to 18 years, items are rate on a 5-point scale from 0 (never a problem) to 4 (always a problem). For the child self-report scale for children aged 5 to 7 years, the items are reworded, and the response scale is reduced to a 3-point scale including 0 (not at all a problem), 2 (sometimes a problem), and 4 (a lot of a problem), so it is simplified for the younger participants. Scores are transformed onto a 0 to 100 scale, and each item is reverse scored meaning that the higher the individual's score is the better their perceived HRQOL. The PedsQL-CP has also shown that it can account for sensitivity within types of CP as children with hemiplegia and diplegia perceived a higher HRQOL compared to those with quadriplegia (Varni et al., 2006). Also, the use of the PedsQL-CP versus the generic Peds-QL 4.0 was able to distinguish between the HRQOL of children with CP and

the HRQOL of typically developing children. The PedsQL-CP has reasonable psychometric properties, as internal consistency reliability coefficients range from 0.63 to 0.93 for the self-report, and 0.88 to 0.96 for the parent proxy-report. Given the psychometric properties, and the PedsQL-CP being condition specific, with child self-report scales (which not all HRQOL measures have; Waters et al., 2009), there is good evidence for this outcome measure to be used in the current research.

5.2.5.2 Self-Efficacy: Self-efficacy Questionnaire for Children (SEQ-C; Muris, 2001)

To measure self-efficacy in a more general sense, Muris (2001) constructed and published the Self-Efficacy Questionnaire for Children (SEQ-C). The SEQ-C measures three domains of self-efficacy, academic self-efficacy, social self-efficacy, and emotional self-efficacy across 24 items that represent these three domains. Academic self-efficacy is the perceptions of the child or adolescent's ability to manage their own learning behaviour and doing well at school. Social self-efficacy is the perceived ability to get along with peers and deal with assertiveness. Emotional self-efficacy is the perceived ability to cope with and regulate negative emotions. Each item is scored on a 5-point Likert scale from 1 (not at all) to 5 (very well). The reliability of the SEQ-C is reasonably high with Cronbach's alpha of 0.82 for social self-efficacy, 0.84 for academic self-efficacy, and 0.86 for emotional self-efficacy (Suldo & Shaffer, 2007).

With reference to the current study, given the lack of a CP specific self-efficacy tool, the SEQ-C was chosen as the outcome measure because of its established use in children and adolescents (Muris, 2001). The link between global and domain specific self-efficacy and mental health, and the impact an improvement in these self-efficacy domains may have on physical activity participation.

5.2.5.3 Self-esteem & Self-concept: Self-Perception Profile for Children & Adolescents (SPPC & A; Harter, 1985).

The current research set out to investigate the effects of participation in frame running on psychosocial factors, including self-esteem and self-concept. A commonly used tool to measures these two factors is the self-perception profile. There are two versions of the one for children (8 to 12 years; SPPC) and one for adolescents (13 to 18 years; SPPA). The SPPC is a 36 item self-report measure of a child's self-esteem and self-concept and has been used in research with children with CP (Russo, et al., 2008; van Wely, et al., 2014).

The 36 items evaluate specific perceptions of the individual's perceived competence across six subscales, five of which are specific domains of self-esteem, scholastic

competence, social acceptance, athletic competence, physical appearance, behavioural conduct, and a global perception of self-worth, global self-worth. The global self-worth domain was included to investigate how much an individual actually likes themselves as a person because of the different weight they place on different aspects of self-esteem (Wichstrøm, 1995). The SPPC consists of six subscales: scholastic competence; social competence, athletic competence, behavioural conduct, and global self-worth (Harter, 2012a). The SPPA consists of the same six subscales as the SPPC with three additional subscales: job competence; romantic appeal; and close friendships (Harter, 2012b). The three additional subscales of the SPPA were added as the domains are viewed as areas of life relevant to adolescences.

Each item on the SPPC and SPPA is presented with two opposite descriptions, for example Figure 5.1 below from the SPPA (Harter, 2012b), and the same format is used on each questionnaire:

Figure 5.1 Example question from SPPA (from Harter, 2012b)

Really True for me	Sort of True for me				Sort of True for me	Really True for me
<input type="checkbox"/>	<input type="checkbox"/>	Some teenagers do very well at their classwork	BUT	Other teenagers don't do very well at their classwork	<input type="checkbox"/>	<input type="checkbox"/>

The participant is to select the statement that they relate to the most and indicate whether it is really true or sort of true. Each item is then scored on a 4-point scale (1-4) with higher scores indicating a more positive view of themselves. Harter (2012a; 2012b) used this question format, the structured alternative format, because it can reduce the tendency for socially desirable responses and offer a wider range of options. However, this format does add the potential of extra complexity for the younger participants (Biddle et al, 2015), and is of note for the current research due to the cognitive limitations of some of the participants. Both the SPPC and SPPA are found to have satisfactory reliability, internal consistency, and test-retest reliability (Wichstrøm, 1995; Muris, et al., 2003), however these results are based on research with typically developing children.

5.2.5.4 Psychological Wellbeing: Strengths & Difficulties Questionnaire (SDQ; Goodman, 1997)

The strengths and difficulties questionnaire (SDQ; Goodman, 1997) is a self-report and parent-report behavioural screening test and a measure of general psychological well-being.

The SDQ is for children and adolescents between the ages of 4 and 17 years. The child self-report SDQ can be used by participants aged 11 to 17 years, and the parent proxy-report is for their child between 4 and 17 years old.

The SDQ has 25 items and asks about 25 attributes, some of which are positive and the others negative (Goodman et al., 1998). The 25 items are split between five subscales, four of which measure difficulties frequently experienced by children and adolescents and one that measures a strength. The subscales of conduct problems, hyperactivity, emotional symptoms, and peer problems measure the difficulties, and prosocial behaviour measures strength (Goodman et al., 1998). Each subscale consists of five items, and each item is scored on a 3-point Likert scale (0=not true, 1=somewhat true, or 2=certainly true). Each subscale is scored separately by the summing of each item in that subscale giving a score between 0 and 10 with the higher scores suggesting more difficulties. Furthermore, each subscale can be considered separately, or combined into a total difficulties score.

The total difficulty score is calculated by summing the scores of each subscale, apart from prosocial behaviour, giving a total difficulty score between 0 and 40. The prosocial subscale is not included in the total difficulties score because as prosocial behaviour is a social behaviour it will benefit a person or society (e.g., sharing or donating), and therefore the absence of prosocial behaviour does not represent psychological difficulties but a strength (Goodman, et al., 1998). The total difficulties score will provide an idea of psychological wellbeing by comparison to established cut-off scores (Goodman, et al., 1998; Parkes, et al., 2008).

The scoring was grouped in bands for total difficulties scores into 'low difficulties (0 to 15)', 'medium difficulties (16 to 19)' and 'high difficulties (20 to 40)'. However, the higher the score for the prosocial scale (0-10) indicates a strength in the individual.

The psychometric properties of the SDQ are defined as satisfactory, with the reliability and validity making it a quick and useful measure for psychological wellbeing in children and adolescents (Goodman, 2001; Muris et al., 2003). The internal consistency for the total difficulties score is acceptable with a Cronbach's alpha of 0.73, which has been replicated across other countries and a test-retest reliability of 0.62 (Goodman et al., 2001; Essau et al., 2012). Furthermore, the subscales have shown a range of coefficients, from acceptable for prosocial (.59 - .87), emotional symptoms (.63 - .78), and hyperactivity (.66 - .78). However, the conduct problems and peer problems subscales have reported low coefficients .41 - .67 and .27 - .52 respectively.

As previously mentioned, the SDQ has been used in research with children with CP (Parkes et al., 2008) and also to investigate associations between physical activity and mental health (Ahn et al., 2018), supporting the use of the SDQ in the current research.

5.2.5.5 Participation and Enjoyment: Children's Assessment of Participation & Enjoyment (CAPE)

The Children's Assessment of Participation and Enjoyment (CAPE) is a 55-item child self-report measure that is designed to document the participation in everyday activities of children and adolescents (King et al., 2000). The CAPE is suitable for ages 6 to 21 years, and for children and adolescents with or without disabilities. Furthermore, the CAPE has been translated into various languages including Dutch, Spanish, and German, providing evidence of the cross-cultural validity and utility of the CAPE (Brown & Thyer, 2020).

It examines the diversity, intensity, and enjoyment of an individual's participation, as well as where and with whom they participated in the activity. The 'diversity', which activities the child does, is simply a yes or no answer, and if the child has not participated in the named activity in the past four months they move to the next item. 'Intensity', which is how often they participate, is rated on a 7-point Likert Scale from 1 (one time in the past 4 months) to 7 (one time a day or more). 'Enjoyment', how much they enjoy the activity, is measured on a 5-point Likert scale from 1 (not at all) to 5 (love it). The 'who' asks participants with whom they partake in the activity most often on a 5-point scale; 1 (alone), 2 (family), 3 (other relatives), 4 (friends), or 5 (with others). 'Where' asks the most frequent location of the activity on a 6-point Likert scale from 1 (at home) to 6 (beyond the community). Each question was asked in relation to five activity types: active physical, recreational, social, self-improvement, and skill-based, providing an overall participation score, as well as the option to obtain a participation score for each activity type, and a score for formal versus informal activity. The CAPE can be used to investigate the effectiveness of interventions that are designed to affect the participant's participation levels (King et al., 2000).

The CAPE can provide an assessment of the different dimensions of participation in different types of activities; it also allows researchers to see the difference in participation between formal and informal activities. Formal activities are more structured compared to informal activities, and have a formally chosen instructor or coach, for example, organised sports. On the other hand, informal activities are usually unplanned and led by the child, for example, play (King et al., 2000). The distinction between participation in formal and informal activities is important as engagement in formal activities is assumed to be more enjoyable due to the structured nature (King et al., 2006). Furthermore, being involved in formal activities is thought to be advantageous in the development of intrinsic motivation, and in turn, the level of competence perceived by the child (Larson, 2000). This can be linked to self-determination theory with developing intrinsic motivation, thus increasing participation (Jin et al., 2018). The distinction between these two types of activities is also important

because there is the possibility of social, environmental, and personal factors affecting the participation of formal and informal activities in different ways (King et al., 2000).

The validity and reliability of the CAPE has been supported in previous research for children and adolescents with and without disabilities (Bult, 2010). The internal consistency of the five activity types of the CAPE measured with Cronbach's alpha ranged from 0.30 to 0.62, and the test-retest reliability of the CAPE was 0.72 to 0.81 (King et al., 2004). Furthermore, the CAPE has been used in previous research with children and adolescents with CP (e.g., Bult et al., 2010; van Wely et al., 2014). In line with the previous research, the current study administered the CAPE to participants to provide an insight into the level of participation in different types of activity, and whether frame running participation affected those other types of activity (e.g., more social activity, or skill-based activities). Furthermore, the CAPE tries to avoid the negative side of participation by not asking what participants find difficult or are not able to participate in, but measures their involvement and participation, which can be important for children with CP as they may struggle with what they can and cannot participate in (Ullenhag et al., 2012).

Note: the measuring of enjoyment for frame running specifically is described in section 5.1.6.2.

5.2.6 Frame Running Training Programme

5.2.6.1 Programme Design

The training programme consisted of 12 weeks of frame running, with participants attending one session a week, and each session lasting approximately 45 to 60 minutes. The length of the intervention was based on evidence suggesting that significant changes in physical and psychosocial factors can only be seen after an 8-week physical activity intervention. An intervention lasting more than 8-weeks is preferential as it allows more opportunity for maintenance of effects and potential long-term benefits. Bryant and colleagues (2015) carried out a frame running intervention and found, over a 12-week period, there was a significant improvement in bone density of children with severe CP, as well as positive comments from interviews regarding enjoyment of frame running. Therefore, the positive results found by Bryant et al., (2015) provide a basis for the length of the current training programme.

5.2.6.2 Delivery of the programme

Each participant completed the frame running sessions at their local frame running group. One of the frame running clubs, where one participant took part in the sessions, is outdoor only, and the sessions would take place on an athletics track. The training sessions at the


other frame running club, where the remaining participants took part in the sessions, also took place on an outdoor athletics track, but during the winter months the sessions were able to move to the indoor space. The attendance for the participants was logged by the participants or their parent/guardian by marking the date of each session they attended on their enjoyment measure sheet (see Figure 5.2).

The frame running training sessions were led by qualified frame running coaches, but parents were also present to assist participants or help with safety aspects if needed, for example, transferring from wheelchair to frame running bike, or attaching protective tubing to the bikes. The training programme was based on basic athletics principles and was standardised across the frame running clubs (i.e., warm-up, stretching, skills/distance, cool down). Given the nature of cerebral palsy with varying functional and cognitive capacity between athletes, sessions can be tailored or individualised to each participant depending on their ability and severity of disability. Observations of sessions along with speaking to coaches and athletes, generated the following example of a training session:

“2 laps warm-up, stretching, frame running skills (e.g., starts, stops, lane steering, cone slalom), recovery, distance run (between 200m to 1500m), recovery, short distance runs (e.g., 4x100m), 2 laps warm-down.” (Frame running Athlete)

The frame running skills can be carried out by participants of all abilities, but sometimes the groups would be split by ability allowing the more experienced racerunners to train at a higher intensity and skill level. The participants also continued to receive their physiotherapy and any other therapy and/or continued to participate in other usual physical activity.

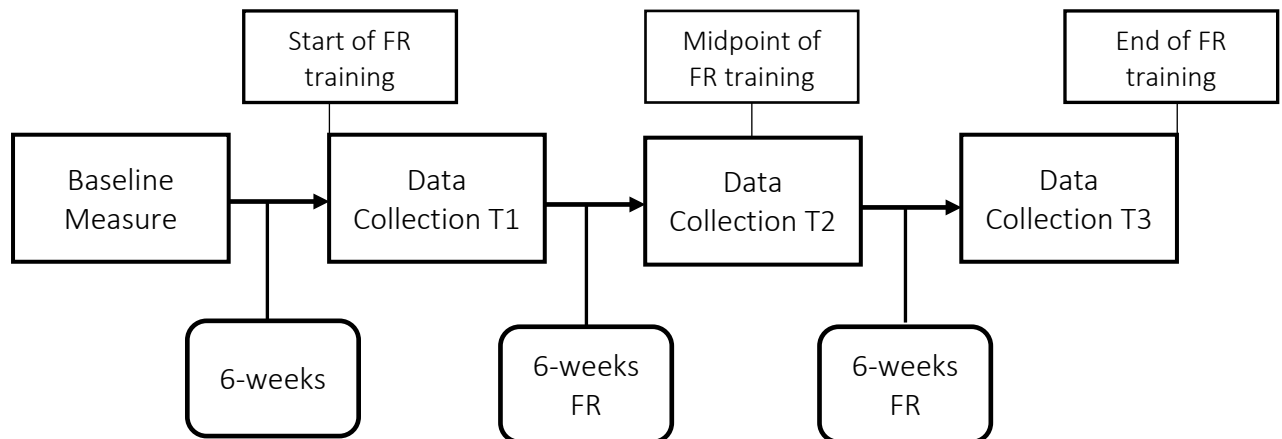
Figure 5.2. Enjoyment measure of frame running sessions example

DATE OF SESSION	HOW MUCH DID YOU ENJOY THE LAST FRAME RUNNING SESSION?
	
Example: 1/05/2018	X

5.2.6 Procedure

A flowchart showing the timeline for the participants is shown below (see Figure 5.3).

Figure 5.3. Flow diagram representing the timeline of the participants frame running (FR) training programme.



Note: FR = Frame running

The selected outcome measures were piloted with an independent participant (child with CP and parent) who did not take part in the intervention study. The pilot participant's gross motor function (GMFCS) was classed as level 5, and the comments made regarding the testing were that the tests could not be done in one sitting, as it was too much of a mental strain. Furthermore, the parent could not be 100% sure if the participant could understand everything, but it was all answered to the best of their knowledge. Following the pilot test, the participants were made aware that they were to take their time when completing the questionnaires and if they were unable to do them in one sitting, that was ok. The questionnaire pack consisting of the information sheets, consent forms, instruction sheets, questionnaires, and final checklist were put together and posted out. Furthermore, having participants complete the outcome measures in their home environment allowed for the participants to use their usual communication strategies and devices, making it as least stressful an experience as possible. Regular contact was kept with parents to ensure they and the participants were managing with the outcome measures and to allow them to communicate any problems they may have had.

The questionnaire packs were sent out via post and not posted online as not all of the selected outcome measures (e.g., the CAPE) were able to be transferred to an online format. Each participant and parent/carer were given the opportunity to ask questions about their individualised pack as they were tailored to the age of the participant. Each of the

outcome measures apart from the enjoyment measure was administered on four separate occasions, with a gap of 6-weeks of frame running between each collection point (T1, T2, and T3), apart from the CAPE, which was only administered on two occasions, at baseline and at T3. This is due to the CAPE asking about participation in the four months prior to completing the questions. At each data collection point, the questionnaires were sent to the participants via post with an addressed and stamped envelope enclosed to return the completed questionnaires. The enjoyment measure was distributed at T1 only and returned at T3. After each frame running session, the participants dated the form and scored their enjoyment of the session. They returned it after they completed the intervention at T3.

5.2.7 Data Analysis

Due to the age differences between the participants, the number of measurements completed varied for each participant, for example, participants 1, 2, and 5 completed the self-perception profile for adolescents which contained three additional scales compared to the self-perception profile for children, which was completed by participants 3 and 4. Also, participants 3 and 4 did not complete a strengths and difficulties self-report measure (measure of psychological wellbeing) because they were under the age recommended for the measure, therefore there are only parent strength and difficulty scores for these participants. Furthermore, some measures were missing a significant percentage of data meaning that they were unable to be interpreted sufficiently, for example, the self-efficacy questionnaire for children had missing data for participant 4 at all time points meaning it was not included in the data analysis for participant 4.

All participants completed the baseline measurements, however participants 1 and 2 missed one round of measures (T1), and therefore only show three data collection points on their analysis (baseline, T2, and T3). Participants 1 and 2 did not complete and return T1 measures within the deadline and continued their frame running. In single case study design, participant data are displayed together for each outcome measure, however, give the large number of data points, and different versions of outcomes (e.g., SPPC and SPPA), participants' data will be analysed on separate graphs where necessary to provide a clearer visual representation of the participant data. Furthermore, for analysis of social validation questions (see section 5.3.7), a mean rating of agreement of the statements and thematic analysis was carried out, following the six steps of thematic analysis, to identify any themes present in the comments (Braun & Clarke, 2006).

As a multiple-case study design, all the data collected was tabulated, and the analysis of the data was performed through descriptive statistics and visual inspection, and each participant's data was considered longitudinally. The use of visual inspection as

analysis tool is similar to previous single-case design studies, for example, Mardon, Richards, and Martindale (2016), and Neil, Hanton, and Mellalieu (2013).

5.3 Results

5.3.1 Quality of Life

The Paediatric Quality of Life CP module (PedsQoL-CP) self-report forms measured the participants' perceived QoL across seven subscales; daily activities, school activities, movement and balance, pain and hurt, fatigue, eating activities, and speech and communication. The participants are asked how much of a problem each item has been over the previous month on a 4-point scale from 0 (never) to 4 (almost always). The items are reverse scored and linearly transformed to a 0 to 100 scale (Varni et al., 2006). Therefore, a higher score indicates that item is less of a problem, and thus, a higher QoL. There is currently no evidence of minimal importance of difference for the PedsQoL-CP to indicate whether significant improvement is made from one data collection to another. One participant (participant 4) did not complete a self-report form as there was not one suitable for their age.

5.3.1.1 Participant 1

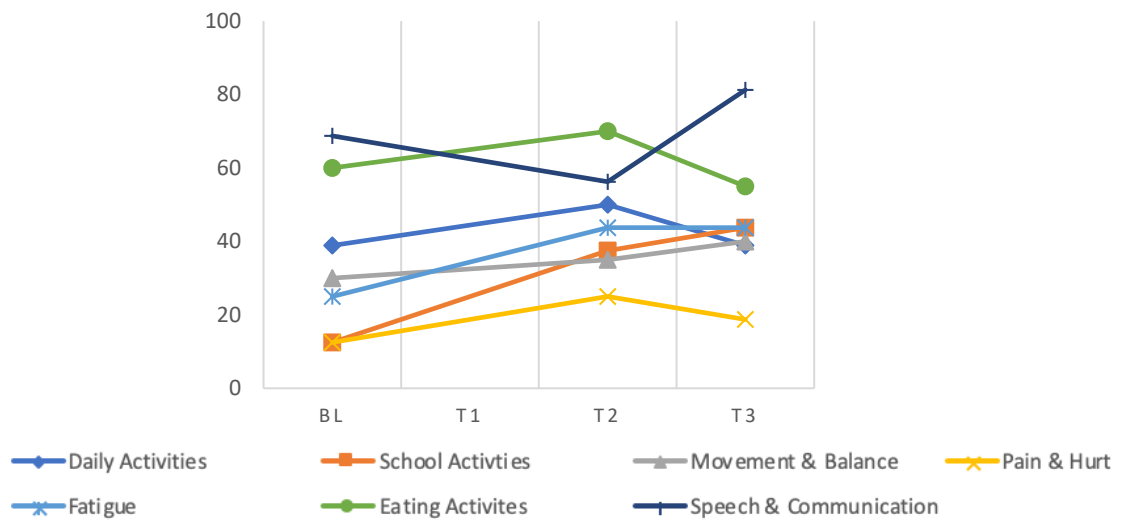
From visual analysis of the self-report, participant one showed improvement from baseline to T3 in school activities, movement and balance, pain and hurt, fatigue, and speech and communication subscales. They reported no change in their daily activities, and a drop in their eating activities (see Table 5.2).

Table 5.2 Participant 1 results from 12-week RR training programme from PedsQL-CP self-report form

P1	D.A	S.A	M&B	P&H	Fatigue	E.A	S&C
Baseline	38.89	12.5	30	12.5	25	60	68.75
T1	NA	NA	NA	NA	NA	NA	NA
T2	50	37.5	35	25	43.75	70	56.25
T3	38.89	43.75	40	18.75	43.75	55	81.25

Note. Mean scores for each subscale from 3 data collection points, Baseline, T2, & T3 (D.A – Daily activities; SA – School Activities; M&B – Movement & Balance; P&H – pain & hurt; E.A – eating activities; S&C – speech and communication).

Figure 5.4 Changes in QoL subscales - Participant 1

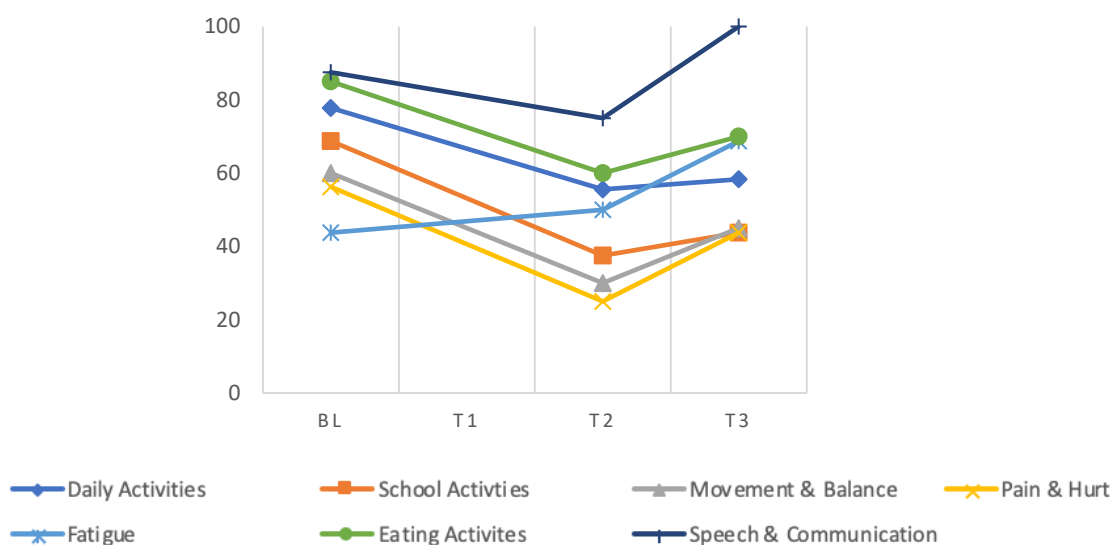


From baseline to T2, there was improvement in all subscales apart from speech and communication, which showed a 12.5% drop, indicating an overall trend towards improvement in QoL. The largest improvement reported was in school activities subscale, a 31.25% improvement (BL 12.5 to T3 43.75), suggesting frame running participation may have been a contributing factor to improved perception of their school activities. Furthermore, the highest scores were reported in speech and communication, T2 reported 81.25%, showing speech and communication was ‘almost never’ a problem (see Figure 5.4).

Table 5.3 Participant 1 parent-report form results from 12-week RR training programme from PedsQL-CP

Parent 1	D.A	S.A	M&B	P&H	Fatigue	E.A	S&C
Baseline	77.78	68.75	60	56.25	43.75	85	87.5
T1	NA	NA	NA	NA	NA	NA	NA
T2	55.56	37.5	30	25	50	60	75
T3	58.33	43.75	45	43.75	68.75	70	100

Figure 5.5 Changes in QoL subscales – Participant 1 Parent-report



The PedsQL- CP parent report-form was also completed for the current study to compare the perception of QoL between the participant and parent (see Table 5.3). Parent-report 1 reported a drop in seven subscales from baseline to T3 suggesting they perceived a lower QoL in these subscales, and only reported an improvement in their perception of fatigue, and speech and communication subscales from baseline to T3 (see Figure 5.5). The speech and communication subscale was the highest scoring subscale on both the self- and parent-report form, showing agreement of the speech and communication items to not be major problems for participant one. Furthermore, at baseline, the parent-report indicated a higher mean score for each of the seven subscales in comparison to the self-report scores, highlighting the perception of QoL was higher in the view of the parent compared to the participant. This may also account for the drop in scores for the parent-report versus the increase in scores of the self-report between baseline and T3, as the scores at T3 are more in line, with the biggest discrepancy shown in the ‘pain and hurt’ subscale.

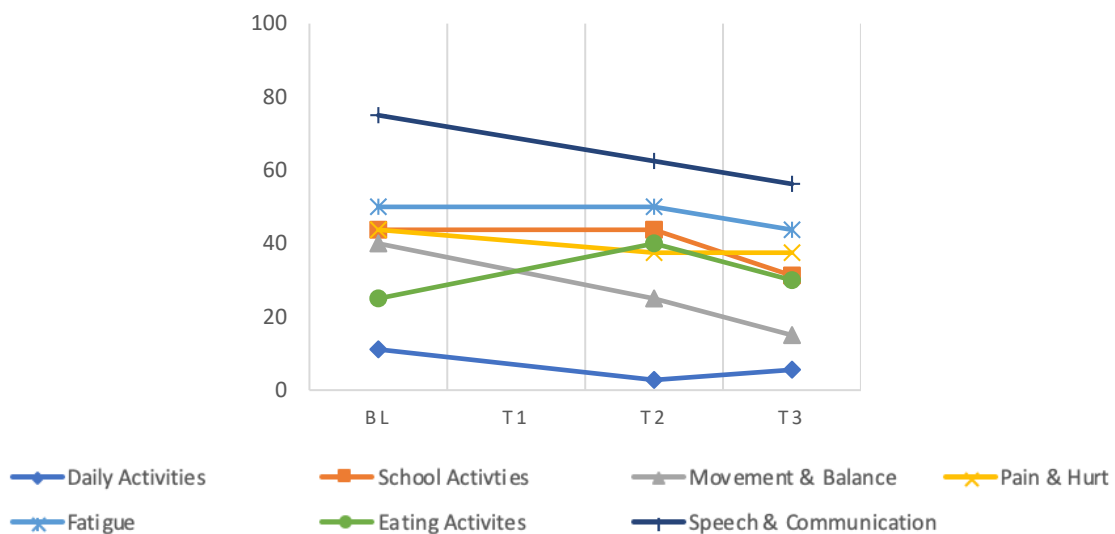
5.3.1.2 Participant 2

Table 5.4 Participant 2 results from 12-week RR training programme from PedsQL-CP self-report form.

P2	D.A	S.A	M&B	P&H	Fatigue	E.A	S&C
Baseline	11.11	43.75	40	43.75	50	25	75
T1	NA	NA	NA	NA	NA	NA	NA
T2	2.78	43.75	25	37.5	50	40	62.5
T3	5.56	31.25	15	37.5	43.75	30	56.25

Note: Mean scores for each subscale from 3 data collection points, Baseline, T2, & T3 (D.A – Daily activities; SA – School Activities; M&B – Movement & Balance; P&H – pain & hurt; E.A – eating activities; S&C – speech and communication).

Figure 5.6 Changes in QoL subscales – Participant 1 Parent-report

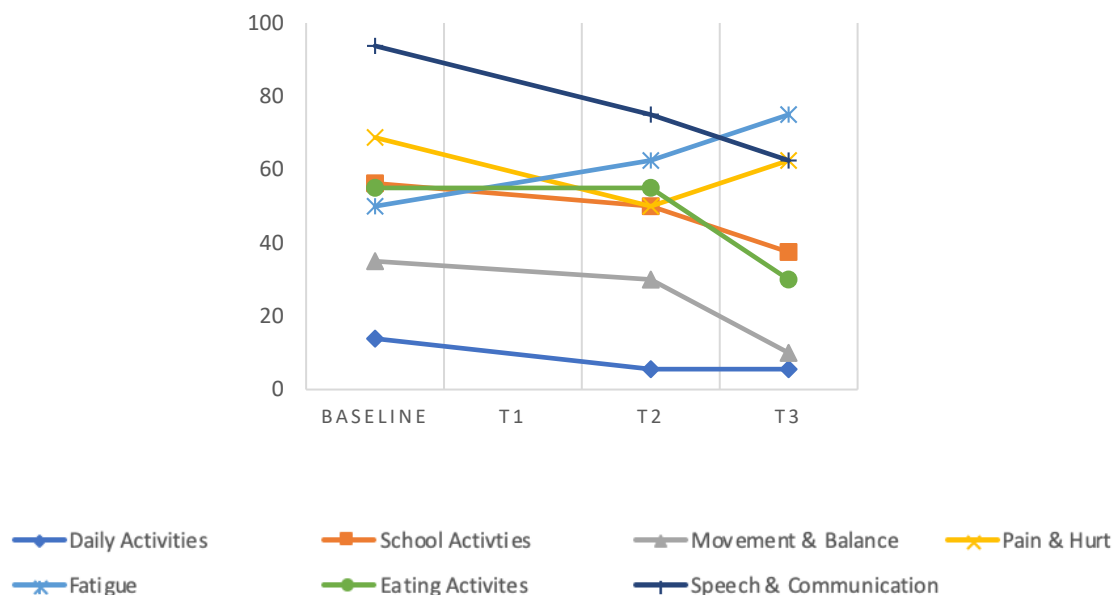


From visual analysis of the participant 2 data, the self-report QoL showed a decrease in all subscales bar 'eating activities' from baseline to T3 (see Table 5.4). The drop in mean scores indicates the participant finding these areas of life to be more difficult since beginning the frame running programme. From T2 to T3, after 12-weeks of frame running, there was a very small increase in 'daily activities' score, no change in the 'pain and hurt' score, and a decrease in the other five subscales (see Figure 5.6).

Table 5.1 Participant 2 parent-report form results from 12-week RR training programme from PedsQL-CP

Parent 2	D.A	S.A	M&B	P&H	Fatigue	E.A	S&C
Baseline	13.89	56.25	35	68.75	50	55	93.75
T1	NA	NA	NA	NA	NA	NA	NA
T2	5.56	50	30	50	62.5	55	75
T3	5.56	37.5	10	62.5	75	30	62.5

Figure 5.7. Changes in QoL subscales – Participant 2 parent-report form



The parent-report for participant 2 showed similar results to the self-report results. From baseline to T3, improved perception (i.e., higher scores) was reported on only the ‘fatigue’ subscale, and all other scores dropped across that time period (see Table 5.5). In line with the self-report scoring, the ‘daily activities’ score was very low (5.56) at T2 and T3, highlighting the difficulty participant 2 has with this aspect of life (see Figure 5.7).

Participant 2 reported small decreases in all subscales, from baseline to T3, apart from eating activities, and the parent-report scores reported similar results with a drop in 6 of 7 subscales from fatigue (improved scores from baseline to T3). The results suggest frame running may have had a negative influence on the participants perceived QoL, but it is not possible to attribute the drop in scores solely to frame running participation.

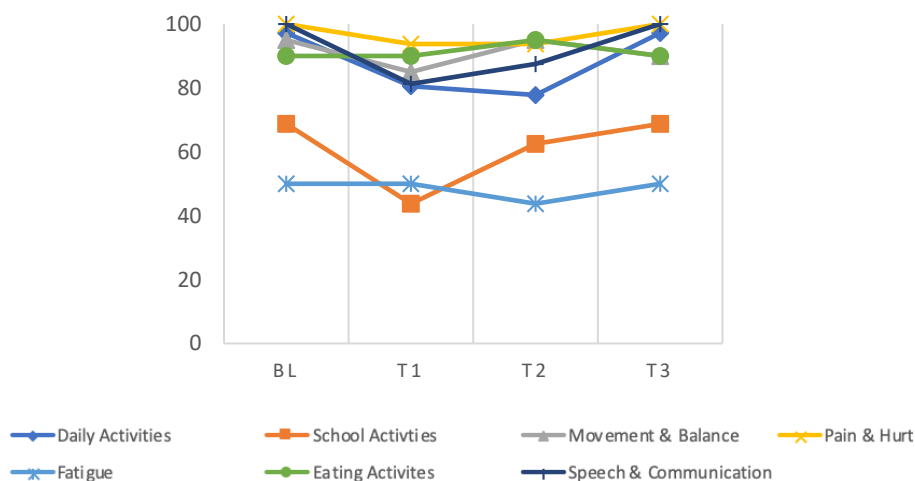
5.3.1.3 Participant 3

Table 5.2 Participant 3 results from 12-week RR training programme from PedsQL-CP self-report form

P3	D.A	S.A	M&B	P&H	Fatigue	E.A	S&C
Baseline	97.22	68.75	95	100	50	90	100
T1	80.56	43.75	85	93.75	50	90	81.25
T2	77.78	62.5	95	93.75	43.75	95	87.5
T3	97.22	68.75	90	100	50	95	100

Note. Mean scores for each subscale from 4 data collection points, Baseline, T1, T2 & T3 (D.A – Daily activities; SA – School Activities; M&B – Movement & Balance; P&H – pain & hurt; E.A – eating activities; S&C – speech and communication).

Figure 5.8 Changes in QoL subscales - Participant 3



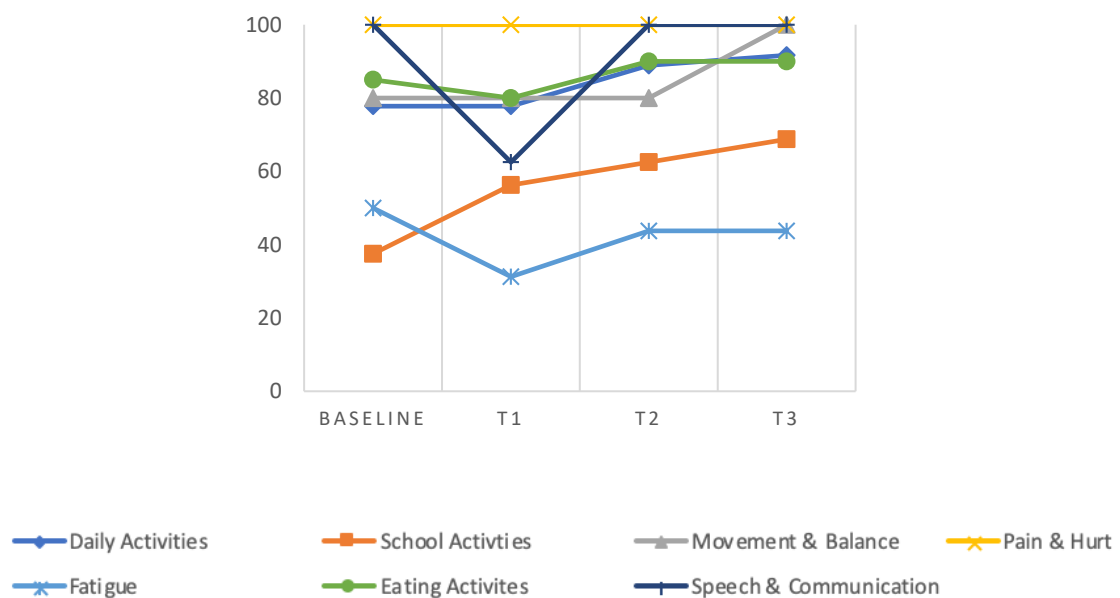
Visual analysis of the PedsQL-CP module self-report for participant 3 shows high scores at baseline (≥ 90) in all but two subscales, 'school activities and 'fatigue' which reported satisfactory scores of 68.75 and 50 respectively (see Table 5.6), which is also clear to see in Figure 5.8. This suggests, at baseline, participant 3 had a good perception of their QoL. At baseline and T3, scores of the subscales are almost identical indicating the perception of QoL was very similar to prior to participating in frame running training sessions. From baseline to T1, there was no change or a drop in subscales, however, from T1 to T3 there

was an improvement in all subscales (see Figure 5.8). The results suggest the 12-week frame running programme may have had a positive impact on the QoL for participant 3, but it is not possible to attribute reported improvements solely to frame running participation.

Table 5.7 Participant 3 parent-report form results from 12-week RR training programme from PedsQL-CP

Parent 3	D.A	S.A	M&B	P&H	Fatigue	E.A	S&C
Baseline	77.78	37.5	80	100	50	85	100
T1	77.78	56.25	80	100	31.25	80	62.5
T2	88.89	62.5	80	100	43.75	90	100
T3	91.67	68.75	100	100	43.75	90	100

Figure 5.9. Changes in QoL subscales – Participant 3 parent-report form



Parent-report scores of the PedsQL-CP module for participant 3 were similar to the self-report, reporting high scores (≥ 75) for all subscales apart from 'school activities' and 'fatigue' at baseline (see Figure 5.9). The parent-report scores were not as high for some subscales (e.g., daily activities, school activities) compared to the self-report scoring suggesting participant 3 felt they were doing better regarding these aspects.

At baseline to T3, 'pain and hurt' and 'speech and communication' subscales remained at the maximum score of 100, indicating participant 3 was perceived to have no problems with the items presented in the subscales. From T1 to T3, there was an improvement across all subscales (see Table 5.7), suggesting that the 12-week frame running participation may have had a positive impact on the perception of their child's QoL, at the time they were completing the outcome measures, which was reciprocated in the self-report scoring.

From baseline to T3, participant 3 reported no improvements in subscales, with the parent-report scores showing an improvement in daily activities, school activities, movement and balance, and eating activities subscales. The results suggest that the parent of participant 3 perceived aspects of QoL to improve across the frame running programme, but it is not possible to say whether the frame running participation is the sole cause of the improvement in perceived QoL.

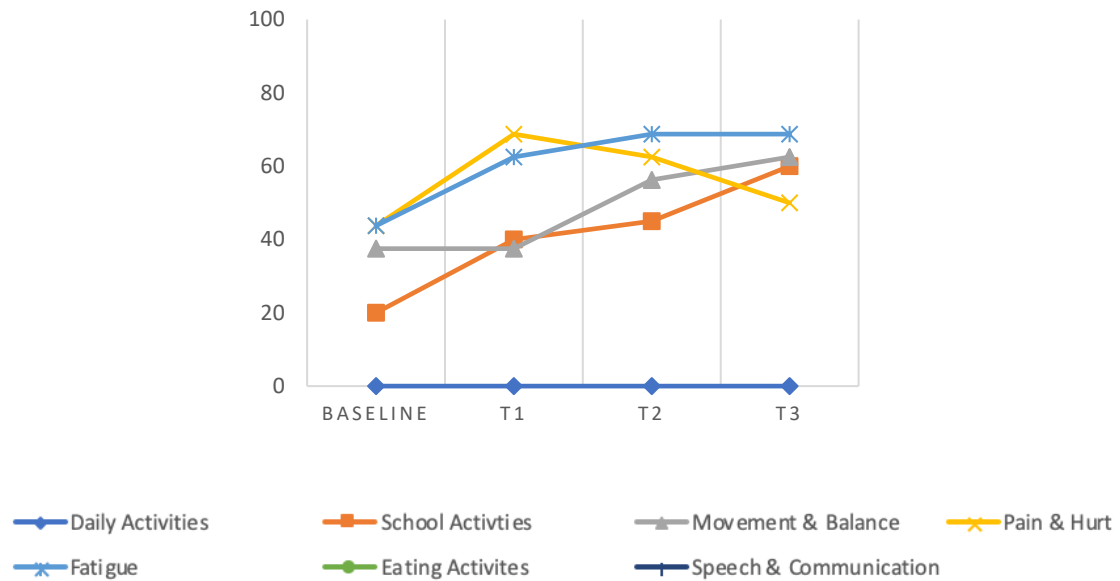
5.3.1.4. Participant 4

Table 5.8 Participant 4 results from 12-week RR training programme from PedsQL-CP parent-report form.

Parent 4	D.A	S.A	M&B	P&H	Fatigue	E.A	S&C
Baseline	0	NA	20	37.5	43.75	43.75	NA
T1	0	NA	40	37.5	68.75	62.5	NA
T2	0	NA	45	56.25	62.5	68.75	NA
T3	0	NA	60	62.5	50	68.75	NA

Note: Mean scores for each subscale from 4 data collection points, Baseline, T1, T2 & T3 (D.A – Daily activities; SA – School Activities; M&B – Movement & Balance; P&H – pain & hurt; E.A – eating activities; S&C – speech and communication).

Figure 5.10. Changes in QoL subscales – Participant 4 parent-report form



Data for participant 4 was only presented from the parent-report form given the age of participant at baseline. Furthermore, there were no 'school activities' or 'speech and communication' subscales (see Table 5.8). From baseline to T3, 'daily activities' scoring was at zero indicating that parent of participant 4 perceived them to have severe difficulties in their daily activities which may be expected given the severity of the CP (GMFCS level V) and the age of the participant (4 years; see figure 5.10). From baseline to T3, scores in movement and balance, pain and hurt, eating activities, and fatigue subscales increased suggesting these aspects, perceived by the parent, were less of a problem for the child, thus possible improvement in perceived QoL. However, attributing the improved scores in the subscales to frame running is not possible.

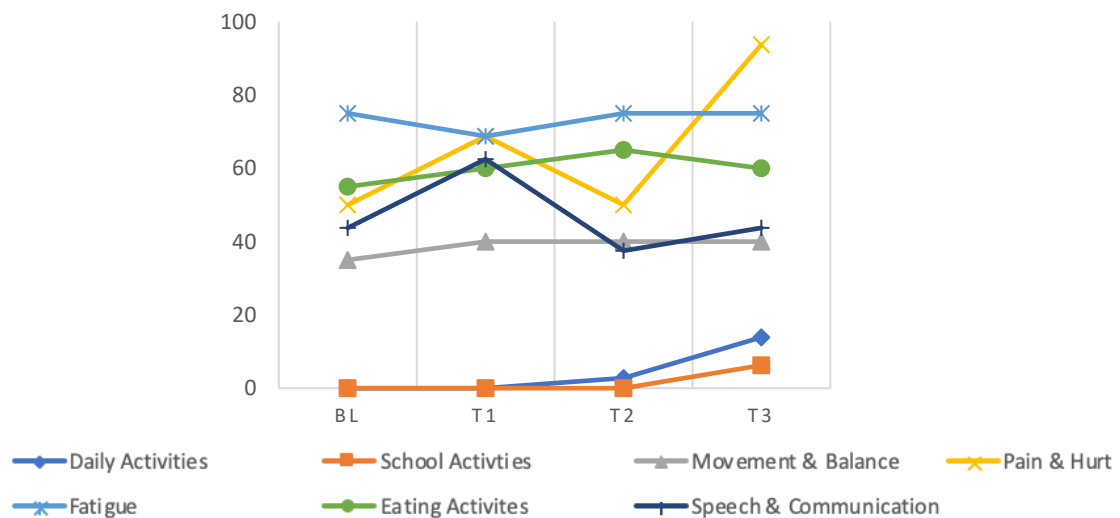
5.3.1.5. Participant 5

Table 5.9 Participant 5 Results from 12-week RR training programme from PedsQL-CP.

P5	D.A	S.A	M&B	P&H	Fatigue	E.A	S&C
Baseline	0	0	35	50	75	55	43.75
T1	0	0	40	68.75	68.75	60	62.5
T2	2.78	0	40	50	75	65	37.5
T3	13.89	6.25	40	93.75	75	60	43.75

Note: Mean scores for each subscale from 4 data collection points, Baseline, T1, T2, & T3 (D.A – Daily activities; SA – School Activities; M&B – Movement & Balance; P&H – pain & hurt; E.A – eating activities; S&C – speech and communication).

Figure 5.11 Changes in QoL subscales - Participant 5



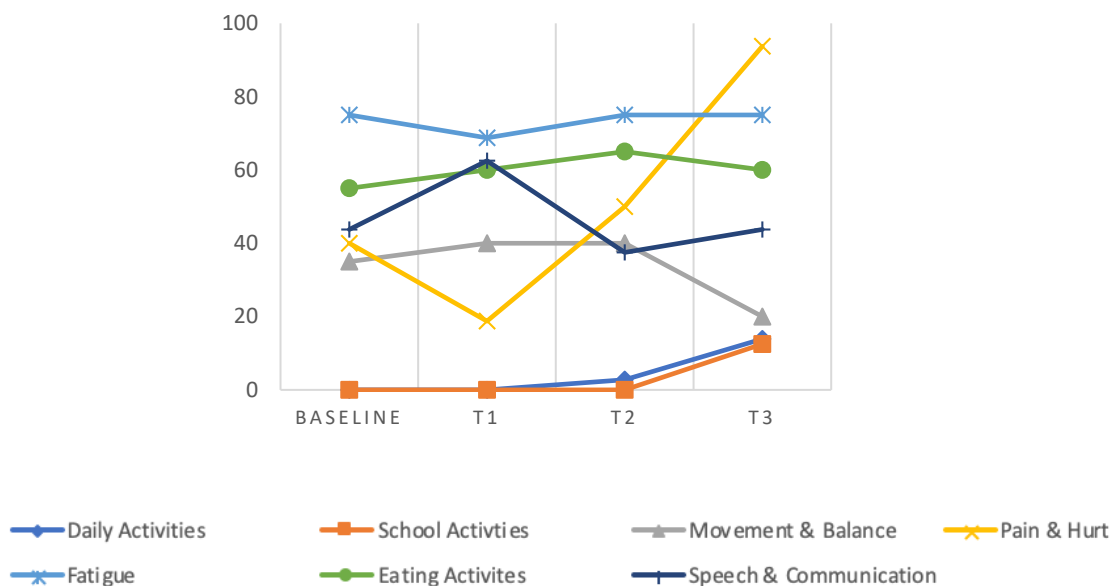
The visual analysis of the data for participant 5 shows a mixed picture across the subscales. At baseline 'daily activities' and 'school activities' are both reported at zero indicating these aspects of life are always a problem for the participant (see Table 5.9). From baseline to T3, 'fatigue' and 'speech and communication' subscales reported no change; however, the other five subscales show a trend towards improvement, and predominately for the 'pain and hurt' subscale (baseline = 50 to T3 = 93.75; see Figure 5.11). This suggests that frame running

participation may have had a positive impact on the perception of ‘pain and hurt’, which can be a barrier to participation particularly for the CP population

Table 5.10 Participant 5 parent-report form results from 12-week RR training programme from PedsQL-CP

Parent 5	D.A	S.A	M&B	P&H	Fatigue	E.A	S&C
Baseline	0	0	35	40	75	55	43.75
T1	0	0	40	18.75	68.75	60	62.5
T2	2.78	0	40	50	75	65	37.5
T3	13.89	12.5	20	93.75	75	60	43.75

Figure 5.12. Changes in QoL subscales – Participant 5 parent-report form



The scoring for the parent-report form was almost identical for participant 5 (see Table 5.10 & Figure 5.12), suggesting the participant may have required some assistance when completing the outcome measure causing some recency bias for the parent-report form.

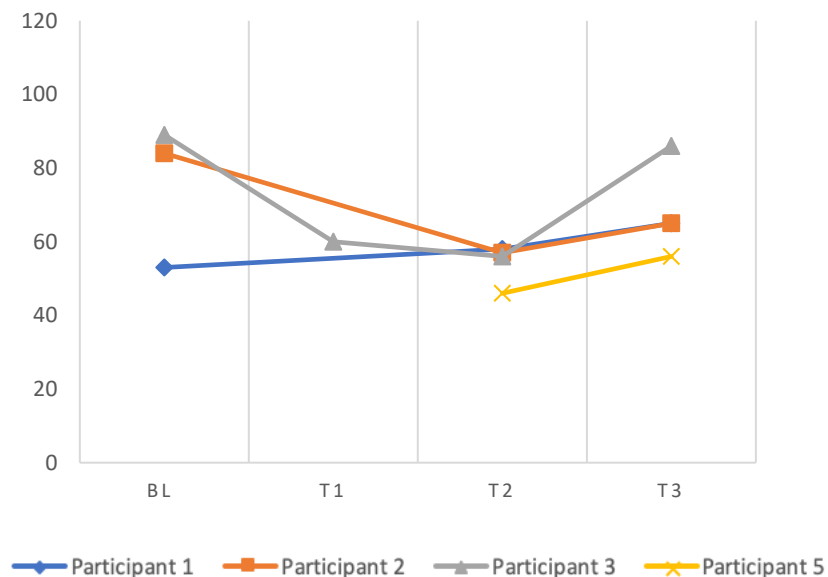
5.3.1.6 Overall Quality of Life

Across both the self-report and parent-report forms, small positive and negative impacts from a 12-week frame running training programme were seen on different aspects related to the perceived QoL for children with CP. Overall, here was no clear pattern across the

participants of which aspects would be positively or negatively impacted, and the results produced a slightly mixed picture, this may have been expected given the range in functionality of the participants. For example, participant 2, GMFCS level IV, reported a drop in all scores from baseline to T3 suggesting frame running participation may be more difficult, and in turn cause negative views on their QoL. On the other hand, participant 3, GMFCS level II, reported no change from baseline to T3 in five of the subscales, which were all scores of 90 or above, suggesting with their level of functionality, frame running participation did not have a significant impact on their perceived QoL. Furthermore, there was not one subscale that showed uniform results across the participants. However, over 50% of participants reported an improvement in ‘fatigue’, ‘movement and balance’ and ‘pain and hurt’ subscales from baseline to final data collection point. These are important areas of focus for children and adolescents with CP as they have been shown to have a positive impact on the development of functionality, and in turn, belief for future independence. This highlights the impact from participation in a 12-week frame running programme is dependent on the individual participant and how they perceive their QoL.

5.3.5 Self-efficacy: Self-Efficacy Questionnaire for Children (SEQ-C).

Figure 5.13. Changes in total self-efficacy, as measured by the summing across all items, from the Self-Efficacy Questionnaire for Children (SEQ-C) for participants 1, 2, 3, and 5



Note: participants 1 & 2 had missing data at T1, and participant 5 had missing data at baseline and T1 (insufficient items completed for data analysis).

To investigate the effects of a 12-week frame running programme on self-efficacy, the self-efficacy questionnaire for children (SEQ-C) was administered to participants 1, 2, 3 and 5 (participant 4 was out with the age band). Each subscale is scored from 0-40, and the total self-efficacy score is scored from 0 to 120, with higher scores equalling higher perceived self-efficacy.

At baseline, visual analysis of the total self-efficacy scores shows participants 2 and 3 perceived high self-efficacy prior to participation in the frame running training programme, reporting scores over 70%. From baseline to T3, participant 1 total self-efficacy scores increased suggesting a greater perception of their self-efficacy across the 12-week frame running training programme. On the other hand, participant 2 and 3 reported a drop in their total self-efficacy scores from baseline to T3. However, for participant 3 there was a lowering of self-efficacy from baseline to T2, but a sharp increase from T2 to T3, suggesting an occurrence of an event to improve their perception of self-efficacy, but it is not clear. The 6-week frame running period from T2 to T3 showed a trend towards improvement in each participant's perception of their self-efficacy. As seen in Figure 5.13, there is not a clear pattern regarding the impact of frame running participation on participants' self-efficacy, and as mentioned each participant reported an increase in scoring from T2 to T3, however it is not possible to attribute the results to frame running participation solely. Moreover, this is highlighted by the fluctuating scores reported by participants across other data collection points. Furthermore, visual analysis of the subscales of the SEQ-C (academic, social, and emotional self-efficacy), showed no clear pattern of frame running impact on any subscales across each of the participants, further highlighting the difficulty to determine any effects of frame running on the perceived self-efficacy of children and young people with CP.

5.3.3 Self-perception profile for children and adolescents (SPPC & SPPA).

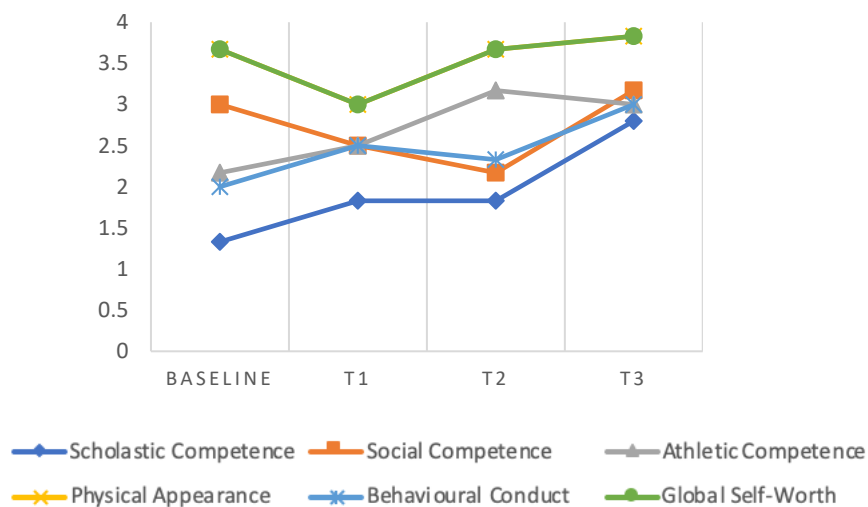
As the self-perception questionnaires were split into children and adolescent versions, they are presented separately. Note there are no results reported for participant four who was outside the age bracket for the SPPC, however their parent did help to complete throughout the study, but there is no precedence for parent-report SPPC of younger participants, so data is available regarding the participant's self-esteem and self-concept, but not reported.

For each subscale of the SPPC and SPPA, the mean score is between 0 and 4, with the higher score indicating better self-concept and self-esteem. The SPPC consists of five self-concept scales, and a global self-worth scale, interpreted as the self-esteem score. The SPPA consists of the same subscales, plus three additional subscales of 'job competence', 'romantic appeal', and 'close friendship.'

5.3.3.1 Self-perception profile for children

Participant 3 was the only child participant to complete the SPPC, as participant 4 was outside the recommended minimum age. The visual analysis of the data shows from baseline to T3, there was improvement reported in all aspects of self-concept, and the self-esteem (see Figure 5.14). Furthermore, from T1 to T2, scores in the subscales for 'physical appearance', 'athletic competence', 'scholastic competence' and 'global self-worth' all increased, with a decrease in 'social competence' scores, and no change for 'scholastic competence' subscale. From T2 to T3, visual analysis shows further increase in 'physical appearance' and 'global self-worth' subscales, and an increase in 'scholastic competence', 'social competence', and 'behavioural conduct' subscales. The only subscale to show a decrease in scores from T2 to T3 is 'athletic competence'. The results for participant 3 suggest that the 12-weeks of frame running may have had a positive impact on the self-concept and self-esteem but attributing the small trends in improvement to frame running is not possible.

Figure 5.14 Changes in self-perception for participants 3, as measured by mean scores from the subscales of the Self-Perception Profile for Children (SPPC)



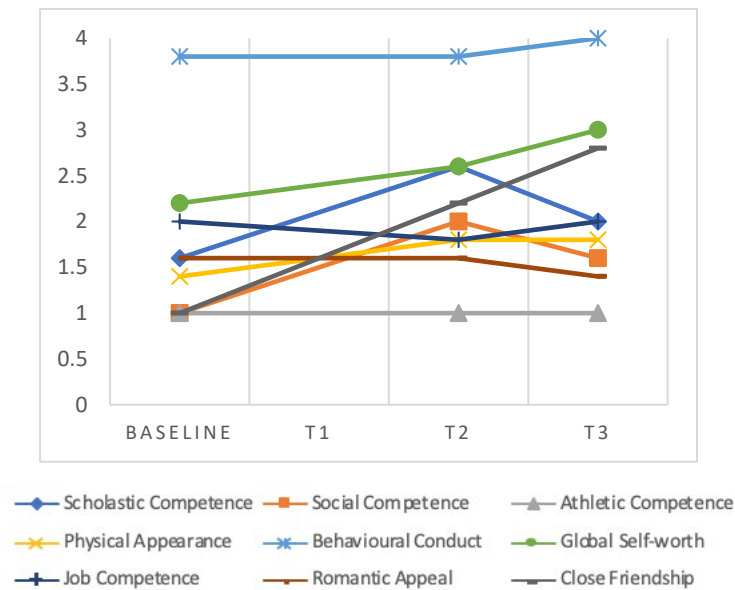
5.3.3.2 Self-Perception Profile for Adolescents

5.3.3.2.1 Participant 1

Visual analysis of participant one showed from baseline to T3 an improvement in all subscales, apart from job competence which reported no change, and a decrease in perceived 'romantic appeal' (Figure 5.15). The 'behavioural conduct' subscale was the highest scoring subscale at each data collection point, and reporting maximum 4 at T2, suggesting participant one has a good behavioural self-concept. Furthermore, the consistent

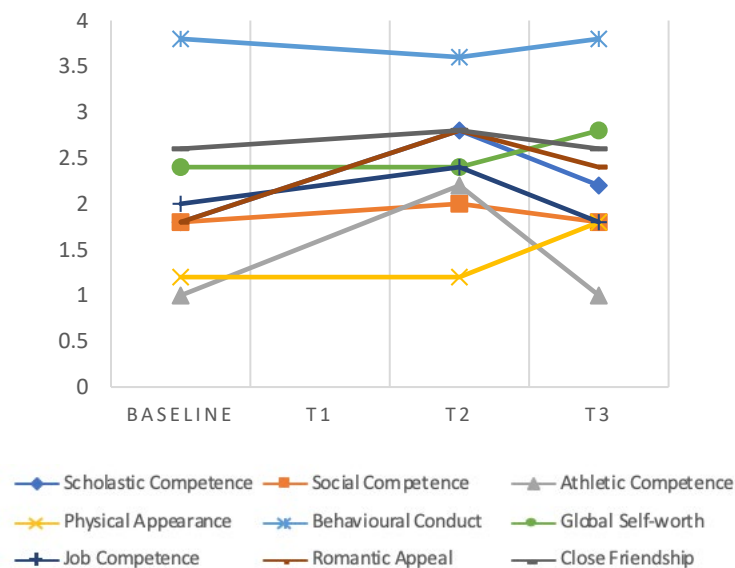
increase in self-esteem scores across the frame running sessions, combined with the improving scores of the majority of aspects of self-concept suggests there may have been a positive impact from frame running on self-concept and self-esteem. An improved self-concept and self-esteem can be important for psychological development and future participation in physical activity.

Figure 5.15 Changes in self-perception for participants 1, as measured by mean scores from the subscales of the Self-Perception Profile for Adolescents (SPPA)



5.3.3.2.2 Participant 2

Figure 5.16 Changes in self-perception for participants 2, as measured by mean scores from the subscales of the Self-Perception Profile for Adolescents (SPPA)

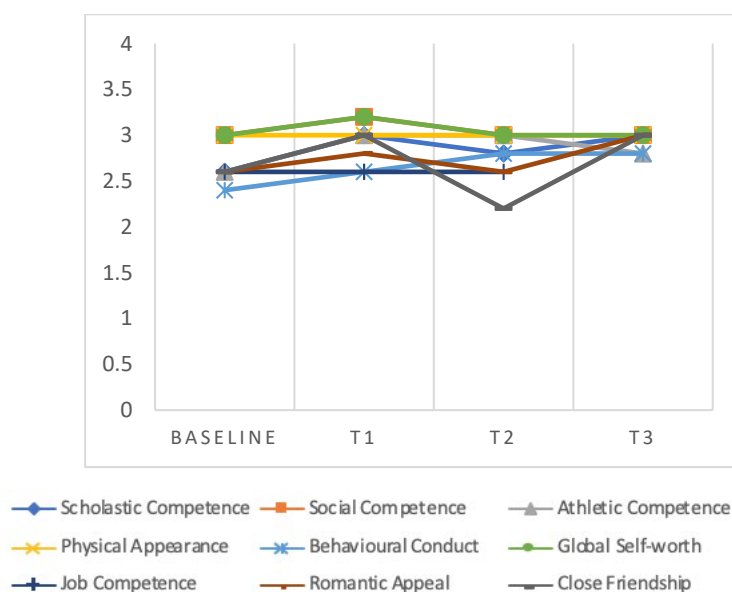


From baseline to T3, participant 2 reported an increase in ‘romantic appeal’, ‘scholastic competence’, ‘physical appearance’, and ‘self-esteem’, alongside a drop in in ‘job competence’, and ‘social competence’ (see Figure 5.16). Furthermore, similar to participant 1, the ‘behavioural conduct’ subscale is reporting high scores from baseline (3.8) and across the data collection points (T2 = 3.6; T3 =3.8) suggesting a good behavioural self-concept for participant 2, but not impacted by frame running participation. Figure 5.16 suggests that at T2, there was the biggest impact on self-concept with the clustering of scores bar the ‘physical appearance’ and ‘behavioural conduct’ subscales. Furthermore, from T2 to T3, there is a reported improvement in self-esteem and ‘physical appearance’ suggesting the frame running participation may have impacted positively on their perceived levels of self-esteem, and the physical appearance aspect of self-concept, but it is not possible to attribute it solely frame running participation.

5.3.3.2.3 Participant 5

From baseline to T3 there is an improvement reported in some aspects of self-concept and self-esteem. Perceived ‘scholastic competence’, ‘physical appearance’, and ‘self-esteem’ increased, and there was a decrease in perception of ‘social competence’ subscale only. The visual analysis of the data indicates the measured aspects of self-concept, and self-esteem, are consistently good as the mean scores are grouped close together and relatively high in comparison to the other participants (see Figure 5.17).

Figure 5.17 Changes in self-perception for participants 5, as measured by mean scores from the subscales of the Self-Perception Profile for Adolescents (SPPA)



From T1 to T2, participant 5 showed no increase in any subscale scores, no change in 'physical appearance' and 'athletic competence' subscales, and a decrease in the other subscales, including self-esteem. The results suggest an environmental or personal interaction causing a drop in self-concept and self-esteem. Across the next 6-week frame running training, from T2 to T3, participant 5 increased scores in 'scholastic competence', 'romantic appeal', and 'close friendships' subscales, and a decrease in 'athletic competence' subscale. The remaining subscales, 'social competence', 'physical appearance', 'behavioural conduct' and 'global self-worth' scores remained the same from T2 to T3. Overall, from baseline to T3, participant five reported consistent scoring suggesting participation in frame running did not appear to have a positive or negative impact on their reported self-concept and self-esteem.

5.3.3.2.4 Conclusion

The main finding from the SPPC and SPPA, was a reported improved level of self-esteem for each participant post frame running training programme, which is important given the evidence of the positive impact good self-esteem can have on personal development, health outcomes, and QoL (Russo et al., 2008; Harter, 2012). On the other hand, the impact of frame running participation on self-concept is unclear, as there is no defined pattern across the self-concept constructs or between the participants. However, frame running may have affected some aspects of self-concept related to engaging in physical activity, such as physical appearance (see Figure 5.16), social competence (see Figure 5.15), and athletic competence (see Figure 5.14).

5.3.4 Psychological Well-being: Strengths & Difficulties Questionnaire (SDQ)

Each of the five subscales from the strengths and difficulties questionnaire are scored between 0 to 10, and a total score of 0 to 40, with the lower the score the less difficulties with mental health disorders and a better psychological wellbeing. The total score, and scores for each of the subscale can be categorised in 'low', 'medium', or 'high' difficulties, which had been defined based on a survey of the population of the USA and United Kingdom (Essau et al., 2012; Goodman, 2016).

Table 5.11 SDQ total scores (TS) categories for self-report and parent-report forms.

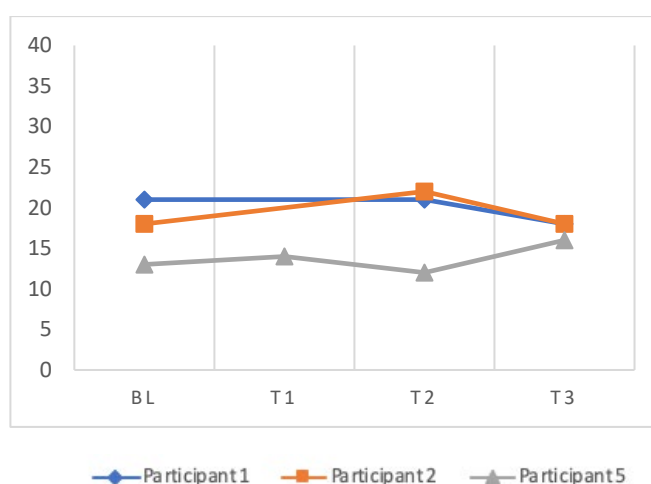
	3-band Categorisation		
	Low	Medium	High
Self-report TS	0-15	16-19	20-40
Parent-report TS	0-13	14-16	17-40

5.3.4.1 Self-report SDQ Total Scores

The total score categorisation for the SDQ child self-report form is 'low' (0-15), 'medium' (16-19), 'high' (20-40; see Table 5.11). Participants 1, 2, and 5 completed the SDQ self-report form (see figure 5.18), and results for all participants 1 to 5 had a parent-report form (see Figure 5.19).

The visual analysis of the total difficulties score for the self-report form of SDQ suggests it is not possible to attribute frame running participation to changes on the psychological wellbeing for participants 1, 2, and 5. Participant 1 did report a trend towards improvement in psychological wellbeing going from 'high' to 'medium' difficulties. The total difficulties scores of participant 5 fluctuated slightly from baseline to T3, but an overall small decrease in psychological wellbeing was reported (i.e., a higher score; baseline = 13 to T3 = 16), but it is not possible to attribute the higher scores to frame running participation only.

Figure 5.18. Total scores of SDQ self-report form for participants 1, 2, & 5, across 12-week frame running training programme



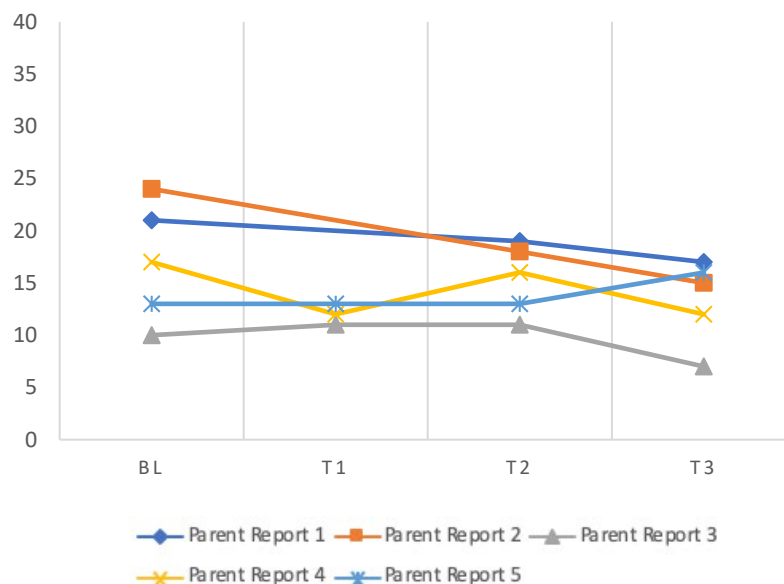
5.3.4.2 Parent-report SDQ Total Scores

The parent-report form of the SDQ investigated the parent's perception of the participant's psychological wellbeing, and the total difficulties category scores are slightly altered from the self-report (see Table 5.12). From baseline to T3, participants 1 to 4 reported an improvement in psychological wellbeing (i.e., lower scores), with participant 5 reporting a small decrease in their psychological wellbeing (i.e., higher scores; see Figure 5.19).

In reference to the 3-band categorisation of scoring for the total difficulties score of parent-report SDQ, participant 1 remained in the 'high difficulties' despite the small downward trend in the scoring. Participant 2 went from 'high' to 'medium' difficulties suggesting a positive impact on psychological wellbeing. Participant 3 did report a drop in scoring (i.e., improvement in psychological wellbeing), but consistently scored in the 'low' difficulties category suggesting a good psychological wellbeing prior to frame running participation. Participant 4 reported a drop from 'high' to 'low' difficulties from baseline to T3, again suggesting a trend towards an improvement in psychological wellbeing.

The comparison between the self-report and parent-report forms for participants 1, 2, and 5 did not show any major discrepancies. One difference of note was seen for participant 2, who reported 'medium' difficulties at T2, and parent-report at 'high' difficulties, suggesting the participant felt they were in a better mental state compared to the view of the parent.

Figure 5.19. Total scores of SDQ parent-report forms for all participants across 12-week frame running training programme.



Overall, the total difficulties score for the self-report SDQ does not show a clear picture about the impact of frame running on their psychological wellbeing. However, the total difficulties score for the parent-report SDQ show a downward trend (i.e., better

psychological wellbeing), suggesting they see an improvement in the child’s psychological wellbeing across the time of the frame running participation. Therefore, the impact on the psychological wellbeing from frame running participation is unclear across the participants. However, there are positive signs in the downward trending of the total difficulties scores, however it is not possible to link frame running participation to the improved perception of participant’s psychological wellbeing.

5.3.5 Participation and Enjoyment: Children’s Assessment of Participation and Enjoyment (CAPE)

The Children’s Assessment of Participation and Enjoyment (CAPE) scoring provides information about the participant’s participation and the characteristics related to participation. There is no minimum difference reported for the CAPE and PAC, however, a 1.00 reflects a different option in each subscale (e.g., ‘enjoyment’: 1=not at all, and 2=somewhat, sort of). As previously mentioned, CAPE explores participation of 4 months prior to completing the measure, thus data at baseline and T3 were analysed given the time period between the two data collection points.

5.3.5.1 Overall scoring

The overall diversity scores from the CAPE indicates the number and range of activities that the respondent participated in out of the 55 listed activities, and the higher the score out of 55 shows a greater diversity of participation (see Table 5.12). It is important to note that the activities included were not solely based on being physically active, and alongside physical activities, participants were asked about their participation within recreational activities, social activities, skill-based activities, and self-improvement activities.

Table 5.12 Diversity scores from CAPE (/55)

	Ppt1	Ppt2	Ppt3	Ppt4	Ppt5
Baseline	26	23	35	16	20
T3	21	21	30	21	25

At baseline, participant 3 was the only participant to report participating in over 50% of the activities listed (63.64%), with the other participants scoring between 29% and 48% (see table 5.13). Visual analysis from baseline to T3 shows participants 1, 2, and 3 reported

a drop in number of activities they participated in, while participants 4 and 5 reported an increase in the number of activities.

5.3.5.2 CAPE Activity Type Scores

5.3.5.2.1 Diversity Scores across CAPE Activity Types

The activity type scores show the participants participation across informal (i.e., unstructured and spontaneous) and formal (i.e., structured and pre-planned) activities but can also be analysed as five different types of activities, which are recreational, physical, social, skill-based, and self-improvement. The scores across the different activity types can provide an insight into the participants activity patterns, and if there is a balance to the children's participation which can impact overall well-being and health of a child (King et al., 2004). Interpretation of the activity type scores for each participant has to be carried out with caution, particularly for the current population, for example, opportunities and ability to participate in physical activities may be low providing a low diversity score but there is a high enjoyment score indicating they enjoy participating in those activities. For the current study, diversity and enjoyment of the activity types were the focus of analysis as the research was exploring the effects of frame running on participation and enjoyment of other activities.

Table 5.13 Diversity scores of participants across CAPE Activity types

Participant	1		2		3		4		5	
Data collection point	BL	T3	BL	T3	BL	T3	BL	T3	BL	T3
Activity Type										
Informal (/40)	20	13	17	19	23	24	15	18	16	18
Formal (/15)	6	4	6	4	8	6	1	3	4	7
Recreational (/12)	7	5	4	6	7	8	7	8	8	6
Physical (/13)	1	1	1	1	4	6	1	3	4	3
Social (/10)	8	8	11	7	7	7	4	4	3	8
Skill-based (/10)	6	4	5	4	5	4	1	3	3	6
Self-improvement (/10)	4	3	5	4	8	6	3	3	2	2

The visual analysis of the activity type scores does not provide any clear evidence of a positive or negative influence on participation across the activity types that can be attributed to frame running participation (see table 5.13). However, of note was the low diversity of physical activity participation ranging (7.7% to 46%), and only participants 3 and 4 reporting an increase in their physical activity diversity from baseline to T3. As previously mentioned, low diversity of physical activity may be expected for children and adolescents with CP due to the barriers of participation to physical activity for this population (e.g., lack of suitable opportunities; functional impairment; or fear of failure). Furthermore, the participants have reported high enjoyment across the activity types, with the exception of 'self-improvement' activities, which was the only activity type to have a reported enjoyment level of below 60% (participant 3 and 5 only). Overall, the influence of a frame running training programme on the participation on other types of activity is not clear, and the data from the CAPE suggests that children and adolescents with CP do not participate in a high number of activities, particularly, physical activity, however, they do report high levels of enjoyment for

the activities that they do participate in. Therefore, enjoyment scores are important to note along with the 'diversity' score.

5.3.5.2.1 Enjoyment Scores across CAPE Activity Types

'Enjoyment' is another dimension of the CAPE to focus on given the role enjoyment can play in the promotion and adherence of the activities, and in turn, ensuring the gain of benefits of participation whether it be physical, psychological, social, and/or emotional. Visual analysis of the enjoyment scores across the activity types presents a mixed bag of results, with no clear picture on the effect of frame running on the enjoyment of different activity types.

Looking at informal (spontaneous) versus formal (with rules) activities, participants 1, 3, and 4, reported improved levels of enjoyment (out of 5) from baseline to T3, however, participants 2 and 5 reported drop in enjoyment of both informal and formal activities (see Table 5.15). Furthermore, the high enjoyment scores of all participants at T3 for physical activity, combined with the low diversity score for physical activity highlight the importance of enjoyment can be key to participation. Overall, there is no clear effect of frame running on participation enjoyment levels across different types of activity.

Tale 5.15 Enjoyment scores of participants across different activity type

Participant	1		2		3		4		5	
Data collection point	BL	T3	BL	T3	BL	T3	BL	T3	BL	T3
Activity Type (/5)										
Informal	3.75	4.17	4.65	3.95	3.48	4.17	4.6	5	3.13	2.83
Formal	3.83	4.5	4.16	3.75	3.13	4.17	4.78	5	4.5	4
Recreational	3.43	4.4	4.75	4.33	3.71	4.13	4.43	5	3.13	2.67
Physical	4	4	5	4	3	4.33	5	5	4.5	4.33
Social	4.38	3.75	4.36	4.29	4.57	4.71	4.75	4.75	2.33	2.86
Skill-based	3.67	4.5	4	3.75	3	4.5	5	5	4.33	3.83
Self-improvement	4.25	5	3.67	4	2.13	3	4.67	3	2	2

5.3.6 Level of Enjoyment

Participants 1, 2, 4, and 5 completed their enjoyment sheet which had asked them to rate how much they had enjoyed each frame running session from 1 to 5 but using smiley face to represent each enjoyment level (e.g., 1 = not at all, 3 = a little bit, and 5 = very much).

Table 5.16 Levels of enjoyment of the frame running training

	Mean	Median	Range
Participant 1	3.75	3.5	2-5
Participant 2	4.08	4.5	2-5
Participant 3	<i>No enjoyment data returned</i>		
Participant 4	5	5	5
Participant 5	5	5	5

The data from Table 5.16 shows that participants 4 and 5 had a mean and median score of 5, the maximum score, signifying the frame running training sessions were enjoyed as much as possible, and participant 2 also reported high enjoyment scores with a median of 4.5 out of 5. However, participant 1 reported the lowest median enjoyment scores of 3.5 out of 5 indicating that the training sessions were enjoyed 'a little bit' which is low in comparison to the other three participants.

5.3.7. Social Validation

The four social validation questions asked parents to rate their level of agreement to the statements and they were given the opportunity to comment how they felt regarding the frame running training programme (see Appendix F). Each response was ranked on a 7-point Likert scale (1=not at all, to 7=extremely), and thematic analysis (Braun, & Clarke, 2006) was carried out on the qualitative data.

5.3.7.1 – Q1: 'The frame running training programme has been important to me/my child'

The mean rating for question 1 was 5.6 (range 4-7), suggesting the participants thought the frame running training programme was quite important to them and/or their child. The thematic analysis of the qualitative data indicated the identification of two themes: the physical and social benefits of frame running participation.

5.3.7.1.1 Physical Benefits

The physical impact was not an aspect measured in the current study, but it is clear the participants and parents believe that the frame running participation has provided them with physical benefits. Frame running was described as '*good exercise*', and participants reported how they felt frame running had impacted them physically both on and off the track, suggesting the perception of frame running benefits are not restricted to the track. For example, one participant recognises the impact it has on their walking ability, '*it allows me to walk better, but I wish I had started younger*', which is an important reference as previous research has shown walking ability to be directly associated with QoL of children with CP (Dickinson et al., 2007). Furthermore, parents reported the physical benefits they saw in their children, highlighting the importance of having the opportunity to be physically active, particularly for children with CP whose opportunities are restricted.

'Leg exercising has kept the brain aware of the left and strengthened both legs. The prognosis 5 years ago was that a wheelchair would be required all the time within a

couple of years. Regular frame running and swimming have maintained the ability to weight bare and walk unaided (on level ground) for short distances.’ – Participant 5
As well as helping with walking ability, frame running was perceived to have helped with disability and lower limbs.

‘It helps my daughter with her disability, keeps her active, and helps her leg muscles as she grows, and she enjoys it a lot.’ – Participant 4

From the small sample of the current study, the recognition of a positive physical impact of frame running is clear, even though the current quotes are subjective.

5.3.7.1.2 Social Benefits

As well as physical benefits, social benefits of frame running were identified by both participants, *‘I like meeting new people and interacting with them’*, and parents, *‘... helping my son meet new people and interacting with them’*. Social interaction is important for the development of children, hence any opportunity for children to interact with new people and build friendships will be beneficial. The importance of social interaction for children with CP was highlighted in chapter 4, with parents of children with CP, frame running coaches, and health care professionals reporting social inclusion and interaction as one of the most important factors of a physical activity intervention, and this is furthered in the current study with the recognition of social interaction by both parents and participants.

5.3.7.2 – Q2: ‘The procedures used throughout the frame running intervention were appropriate’

The average agreement rating for the above statement was 5.2 (range 4-7) indicating the participants’ agreement levels were satisfactory. Two participants reported their enjoyment for the frame running training sessions and were described as ‘enjoyable’ and ‘efficient’. Furthermore, the data collection aspect of the intervention received mixed responses. The questionnaires used were referred to as ‘straightforward’ to use, ‘easy to answer’, and ‘all questionnaires used were relevant to helping my daughter with her disability’. On the other hand, the data collection methodology was thought to be ‘too time consuming and repetitive’ suggesting further revisions to future methodology and outcome measure selection should be considered.

5.3.7.3 – Q3: ‘I am satisfied with the frame running training programme.’

The average agreement rating of the above statement was 5.4 (range 4-6) indicating a satisfactory agreement level. One theme identified from the thematic analysis of responses was the ‘satisfaction of the staff’ involved in the frame running training sessions. Parents

reported the sessions as *'well organised'*, and the layout of the sessions were able to be tailored to an individual's ability, so they are able to include each of the participants and not have any feeling or be excluded. This is important given the diversity of the CP condition, and the wide-ranging mobility and cognitive impairments associated with CP, which may restrict participation in other activities. Furthermore, appropriately trained and knowledgeable staff have been reported as facilitators to participation for children and adolescents with CP, by both children and parents as they feel more comfortable having their children under the supervision of properly trained individuals (Shimmer et al., 2013).

'Well organised programme, people are very friendly and go out of their way to help the children. Always enough volunteers to help, who motivate and use positive reinforcement to encourage the children.' – Participant 3

The above comment from a participant further highlights the important role of the coaches including their assistance and support which can be a positive influence on the enjoyment and maintenance of frame running participation.

5.3.7.4 – Q4: 'I would recommend frame running to a friend'

The average agreement rating of the above statement was 6.2 (range 4-7), which is the highest of all the statements, suggesting the participants feel strongly about recommending frame running to a friend. Furthermore, the thematic analysis of the responses identified the main theme of 'first time experiences'. The high statement rating combined with the qualitative data suggests the importance participants and parents place on frame running, and the recognition of first-time experiences for children with CP highlights the potential of frame running. An important aspect of frame running is the experience it offers to be physically and socially active for individuals who may be restricted by their disability, particularly for those with more severe limitations:

'I would recommend frame running to anyone who feels they like to be involved in activity but can't because of their disability' – Participant 4

Furthermore, some of the first-time experiences for this population may act as a facilitator to promote participation in frame running, whether it is physical:

'Once they are out of their wheelchairs and onto their frame running bikes, they have freedom of movement which they cannot otherwise experience.' – Participant 5

Or social:

'The club is very socially inclusive, and the runners feel they are the centre of attention, which is unusual for them.' - Participant 5

The recognition of frame running providing opportunities for first time experiences, like running, or being the centre of attention, provides some insight to the parents and participants views of why they feel frame running is important and would recommend it to others. Given the mixed set of results from the quantitative data, the qualitative data results are important to understand the role that frame running can play in lives of children and adolescents with CP, despite the small convenience sample. The social validation of the training programme reported was satisfactory, ranging from 5.2 to 6.2, and with the inclusion of the qualitative data the potential short- and long-term impact of frame running participation was referenced, providing a positive outlook for frame running.

5.4 Discussion

This is one of the first known studies to investigate the effects of frame running participation on QoL, and psychosocial wellbeing of children and young people with CP. The following section will discuss the data in relation to the existing literature and explore the impact frame running has had on the psychosocial wellbeing of the participants.

5.4.1. Quality of Life

As previously mentioned, QoL can be considered as the 'holy grail' of outcome measures, as improving the QoL of an individual or a population is a sought-after outcome across research. QoL has been identified as an important outcome measure for children and young people with CP in previous research (e.g., Vargus-Adams & Martin, 2009), as well as in the current thesis (see chapter 4). Furthermore, with previous research showing QoL of children and young people with CP to be lower in comparison to their typically developing peers (Livingston et al., 2007), exploring ways to facilitate and promote a better QoL (e.g., through being physically active; Maher et al., 2015) is important for future therapeutic and rehabilitation programmes.

The PedsQL-CP module measured each participant perceived QoL across the seven subscales; daily activities, school activities, movement and balance, pain and hurt, fatigue, eating activities, and speech and communication. It is important to note there was no total score the PedsQL-CP module, which would allow for comparison of the perception of overall QoL. The differing scores across the seven subscales are reflected in the participants' gross motor classification, varying in severity from level II to V (Table 5.1). The scores for each participant will be discussed to identify the effects of a frame running training programme on

perceived QoL (*Note: the higher the reported scores indicate that the participant perceives that area as less of a problem in their life*).

The measures of QoL for children with CP were developed over 12 years ago, including the PedsQL-CP module used in the current study, and there are concerns whether the important domains for measuring QoL in children and adolescents with CP remain the same (Davis et al., 2017). A study by Davis et al. (2017) investigated which domains related to QoL are most important for children with CP as they can be different in comparison to children without disabilities, and vary across the severity spectrum of CP. Through conducting interviews with parents of children with CP, identified eleven factors that are seen as important to QoL in this population, including physical health, behaviour and emotion, movement and physical activity, independence and autonomy, social connectedness and relationships, variety of activity, nature and the outdoors, and access to services. Of the eleven identified factors by Davis et al. (2017), the PedsQL-CP module only covers the following four factors; body comfort, communication, movement and physical activity, and variety of activity.

5.4.1.1 Daily Activities

The nature of CP affects the development of movement in posture, which can cause activity limitations in day-to-day activities as well as physical activities. The 'daily activities' subscale refers to basic tasks such as getting dressed, putting on shoes, items that require some independence, and therefore the higher the score would indicate a trend towards being more independent.

Only two participants, 3 and 5, reported an improvement in their 'daily activity' scores from baseline to T3 suggesting they are having less problems and experiencing more independence across the 12-week frame running programme. Furthermore, a positive association between physical activity participation and the ability to do more basic activities (Shkaiko-Thomas et al., 2012) suggests that frame running participation may have a positive influence on the ability of children and adolescents with CP to perform the basic, daily activities, and in turn experience more independence.

The low scores reported for participants 2, 4, and 5 (scored between 0% and 13.89%), indicate difficulty with the 'daily activity' items, and this may be related to the functional level of the participants. Participants 2, 4, and 5, are classified at level IV and V of the GMFCS indicating they have moderate to severe mobility issues and therefore limitations to their functional independence. On the other hand, participants 1 and 3 are both classed at GMFCS level II, and they reported higher scores on the 'daily activities' subscale suggesting the more functional mobility, the more ability an individual can have to participate and complete basic day to day activities. Similar to the current study, Omura, Fuentes, and

Bjornson (2018) found the level of physical impairment of children with CP had a negative influence on the participation on daily life. Furthermore, a link between higher functional dependence and a lower perception of QoL is a common correlation for children and adolescents with CP (e.g., Tarasulu & Livaneligon, 2010; Chulliyil et al., 2014; Omura et al., 2018).

Overall, the influence of frame running participation on daily activities is not clear, with a trend towards improvement in two participants, and a decrease in the remaining participants. However, given previous research (e.g., Shiako-Thomas et al., 2012), further research may identify a positive association between being physically active in the form of frame running and improved ability and participation in daily activities.

5.4.1.2 School Activities

School activities are an important subscale as school can influence the psychological and social development of children and adolescents. These can be more prominent in the CP population as children and adolescents with CP may attend either mainstream or special education.

In the current study, two participants attended special needs education (participants 2 and 5), and the other three attended mainstream education (participants 1, 3, and 4). The school activities subscales only contain four items so there cannot be an assumption that an improvement in the school activities indicates they are enjoying school more or performing better in school. However, the perception of improvement in these activities may lead to an increase in self-confidence and self-esteem at school which in turn a positive impact in enjoyment and activity participation and performance within a school environment.

Participants 1 and 5 reported improvements in school activities, participant 3 reported no change, and participant 2 reported a small decrease in school activities score from baseline. The parent-report forms of participants 1, 2, 3, and 5 reported the same perception of school activities from baseline showing the parents view of their child's ability in certain school activities is in line with the child. The education system the participants attend may also have an effect on the perception of school abilities because special needs education may be more tailored to the individual compared to mainstream education and therefore, they may perform better and find school more enjoyable (Boldyreva et al., 2020). Furthermore, schooling system can affect participation and activity performance. For example, Schnecker, Coster, and Parush (2005a) found within a mainstream school environment, children with CP's activity performance and participation showed a positive association between their mobility levels, and evidence of a neuroimpairment (i.e., a learning disability, or a speech and language deficit). Similar to day-to-day activities, functional ability can be an important predictor for participation and performance in children and adolescents

with CP within a school environment. School participation is important because it provides the opportunity to acquire and develop skills and build competence, which leads to improved life satisfaction and QoL, which is important for psychological and social development (Schnecker et al., (2005b).

Similar to the daily activities' subscale, with only two of the participants reporting a perceived improvement in their school activities, there is no consistent pattern regarding the effects of frame running participation on school activities. Furthermore, there was no clear pattern with regards to the effect of mobility and education system of the participants on their school activities. Moreover, the limited number of items, and the lack variety of the items in this subscale highlights the necessity for school activities to be investigated separately or in a more robust manor as education, participation, and activity performance within the school setting can be very impactful for youth with CP.

5.4.1.3 Movement & balance

The movement and balance subscale items relate to having problems when moving arms, legs, and keeping their balance while sitting and standing, all aspects of life that are disturbed by the presence of CP. This aspect of QoL is important in relation to frame running as previous research has given evidence for participation in physical activity to improve mobility and balance in youth with CP (e.g., Lauruschkus et al., 2017). Impairment of movement and balance are some of the primary deficits associated with CP (Drubicki et al., 2010), and the relationship with performing daily tasks emphasises the importance of maintaining and improving both movement and balance but this can be difficult for children with CP. Furthermore, the perception of good movement and balance, especially by the individual is important because it can add a feeling of independence, has the potential for improving an individual's self-esteem in performing daily tasks and confidence in being physically active, and motivating factor when thinking about engaging in new physical activities or to push themselves further in the activity they are already participating in.

The self-report scores for participants 1, 3, and 5 showed small improvements from baseline suggesting the problems with movement and balance occurred less often across the participation in frame running. In comparison to the self-report scores, the parents of participants 2 and 5 perceived the ability for movement and balance to have decreased. A positive for frame running is the results suggest a trend towards improving this aspect of life in some participants, because frame running provides another alternative for physical activity participation, interventions, rehabilitation and/or training programmes. Numerous interventions aimed at individuals with CP aim to improve mobility, strengthen weak muscles, improve functional status, and functional independence in home, school, and community environments (Papavasilou, 2009). Interventions, therapeutic, and rehabilitation

programmes can be in different forms, including fitness training (aerobic and anaerobic; Verschuren et al., 2007), static cycling (Williams & Pountney, 2007), hippotherapy is a good option for non-ambulant children for improving balance and postural control (Zadnikar & Kastrin, 2011), and strength training (Dodd et al., 2003). However, many of the interventions, rehabilitation, and/or therapeutic programmes that have been shown to have a positive impact on functional performance and balance for children and adolescents with CP are not accessible for the more severely impaired. Williams and Pountney (2007) found functional improvement in non-ambulant children with CP (GMFCS level IV and V) after a 6-week intervention on a static bike.

The results show if a suitable and accessible option is available for the more severely disabled, they are also able to improve their functional status, and frame running is accessible to all levels of mobility. Therefore, as the results of the current study indicate a trend towards improvement, a future study with more of a focus on the impact frame running can have on movement and balance would be beneficial.

5.4.1.4 Pain & Hurt

The items for this subscale cover aching and stiffness of their muscles and joints, and overall hurt and effects on their sleep, which are frequent problems in children and adolescents with CP (Wiat et al., 2008). Within the CP population there is a high prevalence of pain, over 60% reported by those who can self-report, and higher prevalence of pain from 33% to 75% in comparison to the general population which can have a negative influence on daily activities participation and performance (Dorlap & Bartlett, 2010). Furthermore, the severity of the impairment can impact on pain (Parkinson et al., 2010), and intensity of pain has been shown to have a significant association with the way it impacts daily activities in children and adolescents with CP (Penner et al., 2013; Terro et al., 2008).

Pain is one of the more common barriers to participation in physical activity, particularly in children and adolescents with CP, with the fear of pain from engaging in a new activity being a demotivating factor (Yamaguchi et al., 2014). Being physically active can reduce the perception of pain but can also cause additional pain. The fear of experiencing more pain can be a barrier to participation of physical activity with the CP population, and they regularly report higher pain, which can interfere with participation and performance in physical and daily activities (Martin, 2013; Whitey et al., 2018).

Participant 3 showed a small improvement in their pain and hurt subscale and reported a score of 100 indicating at T3 suggesting they did not experience pain related to these items at all, and limited issues with their muscles and joints. Participant 3, classified GMFCS level II, does not require a mobility aid and with limited mobility impairment, less reported problems of pain might be expected. Participant 4 and 5 also reported an increase

in their pain score from baseline to T3 (i.e., an improvement in their perception of pain; bl 37.5 to T3 62.5; bl 50 to T3 93.75 respectively), which suggests the introduction of frame running may have a positive impact on their perceived levels of pain by alleviating some of their pain. Both participants 4 and 5 are more functionally impaired (GMFCS V and IV respectively) and given the association between the severity of impairment and impact of pain (Parkinson et al., 2010), the improvement shows a positive trend towards helping with pain in more severely impaired individuals, and in turn influence their participation and performance in day-to-day activities and QoL.

Participants 1 and 2 reported a drop in the scores, and the increase in the frequency of problems with pain and hurt may be related to starting a new activity, in this case, frame running where the participants have engaged and used muscles that they do not frequently use, or they experience pain from sitting on the saddle. Therefore, there is the possibility that over a longer period of assessment the participants may become used to the training and more physically active and the perception of pain and hurt is reduced.

Overall, the results show a trend towards improvement in the participants' perceived pain and hurt from frame running participation with three of the participants reporting an increase. Furthermore, two of the participants being more functionally impaired suggests the positive effect of frame running and highlights the accessibility of frame running to all levels of functionality.

5.4.1.5 Fatigue

Along with muscle and joint pain, fatigue is another common barrier to participation in physical activity for children and adolescents with CP due to the extra energy expenditure related to CP impairments (Houlian, 2009). Furthermore, fatigue becomes an important consideration when wanting to move for those with CP and can come from general day to day activities with mobility and musculoskeletal problems causing the experience of fatigue (Brunton, & Rice, 2011).

The fatigue subscale scores for participants 1 and 4, reported an improvement from baseline, participant 2 reported a decrease, and participants 3 and 5 reported no change in scoring from baseline. The parent-report scores for fatigue of participant 1 and 2 reported higher fatigue scores which may show an underestimation of how the participants are struggling with fatigue and problems related to fatigue. Parent 3 reported a slight increase in their child's fatigue scores from baseline to T3, which does add positively to the potential impact of participation in frame running for children with CP.

The items for fatigue ask the participants about their perception of tiredness and energy levels, but a low score (indicating a high level of perceived fatigue problems) does not mean the individual is always active, or, inversely a high score (indicating a low level of

perceived fatigue problems) does not mean the individual is inactive and has energy to burn. Individuals with CP have shown to expend higher levels of energy to maintain their functional capacity compared to those without disabilities, in turn increasing their level of fatigue (Berrin et al., 2007). Furthermore, an individual with CP who is not physically active may have a lower efficiency while moving and therefore general day to day movements can lead to higher energy costs and potentially higher fatigue levels. This higher level of fatigue can interfere with participation in physical activity and lead to a negative relationship between levels of physical activity and fatigue in the CP population, and continued fatigue can even cause cessation of physical activity (Eken et al., 2013; McPhee et al., 2016). Therefore, the average to slightly above average scores for the fatigue subscale in the current study shows the participants sometimes encounter problems with fatigue (i.e., feeling tired and/or no energy) is a positive outcome in respect to frame running as participation has not had an adverse effect on the participants. Similar to pain, fatigue can be a limiting factor for daily living participation and performance and school functioning (Varni et al., 2006), and therefore the results of the current study suggest a trend towards improvement in this aspect. However, within this subscale, muscle fatigue is not measured, which can also be a common impairment for individuals with CP (Ratel et al., 2006), and future research can study the effects of frame running on muscle fatigue.

5.4.1.5 Speech & Communication

The speech and communication subscale looked at the problem's individuals had with communicating with their family and other people, which can have a big influence of their perceived QoL. However, speech and communication are not solely down to using words, as communication involves sending and receiving messages, which can be done in various methods including speech generating devices, pictures, and speech (Himmelmann et al., 2013).

From baseline participant 1 reported an improvement, participant 2 reported a decrease, and participants 3 and 5 reported no change in their subscale scores, and the pattern of scoring was replicated in the parent-report form. Participant 3 reported a maximum score of 100% suggesting they encountered no problems with speech and communication. High speech and communication scores may be because parents predominately see them in a home setting where the child feels more comfortable to communicate in comparison to a group training session.

Frame running participation is not going to directly impact the participants speech and communication abilities; however, it may indirectly affect their speech and communication ability as individuals are part of a new environment and forced to

communicate with unfamiliar people, and this may be a cause of the results in the current study.

5.4.1.6 Quality of Life conclusion

To conclude, this is one of the first studies to look at the impact of frame running participation on the perceived QoL in children and adolescents with CP, and there is some indication that participation in frame running can have a positive impact on each aspect of the quality-of-life measure (e.g., improvements in perception of movement, fatigue, and pain). However, there are many factors that can affect QoL and pinpointing the cause for the improvements and decreases in scores for each participant is difficult to do. Furthermore, as previously mentioned, there is no overall score for the PedsQL-CP module to give a more complete picture of the participants perceived QoL, and some of the subscales included only have a small number of items relating to one aspect of that topic, for example, both the fatigue, and the pain and hurt subscales only contain four items. Therefore, to look at whether frame running participation has had an effect on the factors related to QoL, more specific outcome measures can be administered to participants, for example, faces pain scale or the GMFM-66. Additionally, the self-report technique is viewed as one of the better methods for assessing QoL in children, and there may be reasons that a self-report is not possible, for example, cognitive or language impairments, but a parent-report form should not be seen as a substitute for the self-report (Theunissen et al., 1998).

Self-report can be viewed as the 'gold standard' for assessing some areas, for example pain, but children with CP may not be able to provide a clear perception of their pain level (Hadden & von Baeyer, 2002), therefore, sometimes a parent-report is required. The current study looked at the results from a combination of the self- and parent-report forms for the PedsQoL-CP module, and there is evidence of large differences between results at the child-parent level (Theunissen et al., 1998), and therefore results should be interoperated with caution. Finally, previous research has shown that participation in sports and/or recreational activities has been important for QoL and had a positive impact in children with CP (Feitosa, et al., 2017), indicating that further studies to assess the effects of frame running on QoL should be considered.

5.4.2 Self-efficacy

The current study is one of the first studies to look at the effect of frame running on self-efficacy in children with CP, and this was done by administering the Self-Efficacy Questionnaire for children (SEQ-C) to four out of the five participants, as one participant was outside the age bracket for the measure. The SEQ-C assesses self-efficacy in three

subscales, which relate to a child's ability to manage different social situations and challenges: social self-efficacy, academic self-efficacy, and emotional self-efficacy. Each subscale is scored from 0 to 40 and the total self-efficacy score is scored from 0 to 120, with the higher the score equalling higher perceived self-efficacy.

As can be seen from the results, section 5.3.5, the impact of participation of frame running on the total self-efficacy scores for participants one, two, three, and five is unclear, despite the increases shown for participant 1, 2, 3, and 5 from T2 to T3. The identification of an improvement the total self-efficacy scores for each participant at one data collection point does indicate the possibility that frame running participation can have a positive impact on levels of perceived self-efficacy. The range of total self-efficacy scores across the participants (38.33% to 74.17%) indicates the differing levels of self-efficacy within this study, and with all the scores apart from three recorded as under 55% it is in line with previous research suggesting that children and adolescents with CP report similar or lower levels of self-efficacy in comparison to children and adolescents without disabilities (Wickman, et al., 2018). Furthermore, the SEQ-C consists of the three subscales, investigating emotional, social, and academic self-efficacy, and not just general self-efficacy as they are domains specific to children and adolescents.

Academic self-efficacy relates to the perceived capability to master academic subjects, manage learning behaviour, and fulfil academic expectations (Muris, 2001), and these behaviours may be difficult for the participants of this study due to potential presence of cognitive impairments and low IQ found in children with CP (Stadskliev, 2020). The participant one reported a small increase in academic self-efficacy from T1 to T3, however participant two and three reported decrease in academic self-efficacy. Participant three, then showed an increase in perceived academic self-efficacy from T2 to T3, as did participant 5. The fluctuation in scores across participants suggests that the participants' academic self-efficacy may not be solely influenced by participation in racrerunning. Social self-efficacy scale refers to the participants perceived capability for assertiveness and peer relationships (Muris, 2001), and this subscale can be important as there has been an association with low social self-efficacy and an affective disorder, social phobia identified (Muris, 2002; Minter & Pritzker, 2017). From T1 to T2 participants one, two, and three all showed an improvement in their perceived social self-efficacy, and from T2 to T3, participant three showed further improvement, alongside participant five.

The improvement in all participants for perception of their social self-efficacy scores, suggest frame running participation may have a positive impact. Furthermore, this is an important benefit to note for frame running as peer relationships can be an important facilitator to continuous participation in physical activity, which can lead to the additional

physical and psychosocial benefits associated with physical activity (e.g., Martin & Smith 2002).

The emotional self-efficacy subscale relates to the participant's perceived ability to cope with negative emotions (Muris, 2001), and low emotional self-efficacy showed an association with panic and anxiety (Muris, 2002; Minter & Pritzker, 2017). Participant one reported the same scores from T1 to T2, but participant two, three and five again showed improvement in their perceived emotional self-efficacy scores. Similar to social self-efficacy, these scores for emotional self-efficacy are positive for frame running participation as the additional peer contact in a community setting may improve these aspects of self-efficacy.

It is well established that high self-efficacy is linked to enhanced levels of confidence, and low self-efficacy is linked to anxiety and depression (Suldo & Schaffer, 2007), but self-efficacy is also important for promoting health behaviours. Becker and Shaller (1995) investigated general self-efficacy and health attitudes among adults (18-49 years) with CP versus adults in their community without CP. The results showed that the adults with CP perceived their ability to perform health-promoting behaviours as lower than the adults without CP. Furthermore, in relation to general self-efficacy, the adults with CP reported their perception of their ability to perform exercises was much lower than their perceived ability to perform health-promoting behaviours. Results from Becker and Shaller (1995) reported a lower self-efficacy of the participant's ability carrying out exercise compared to carrying out other health promoting behaviours, and it highlights the need for more feasible and accessible opportunities for the CP population to exercise to enhance self-efficacy and frame running has the potential to do so. Furthermore, they highlight the relevance and importance of self-efficacy for exercise and health promotion for adults with CP, and therefore the importance of self-efficacy in children with CP and exercise should also be a priority for research moving forward.

The current literature investigating self-efficacy in children with CP is scarce. for example, Reid (2002), and Hellwig, Buschmann, and Burmann, (2020), who studied self-efficacy in children with CP but in the context of the benefits of virtual reality rehabilitation, and the designing of digital games for the CP population, respectively. However, there are a couple of studies investigating the self-efficacy in adults with CP, for example, Becker and Shaller (1995), and van der Slot (2010). This indicates a gap in the literature which the current study can add to, as it is one of the first study to investigate the effects of frame running on the self-efficacy for children with CP. Therefore, despite the effects being unclear, with no previous literature in relation to frame running, it can add to the literature regarding the effects of physical activity participation on self-efficacy in children and adolescents with CP in a positive manner.

5.4.3 Self-esteem & Self-concept

The current study is one of the first studies to look at the effect of frame running on self-esteem and self-concept in children and adolescents with CP, and the self-perception profile for children and adolescents is one of the most commonly used questionnaires for assessing self-esteem and self-concept in children and adolescents (Harter, 1985; Muris et al., 2003).

There are usually assumptions that children and adolescents with CP normally have lower levels of self-esteem (person's sense of worth; Russo et al., 2008; Espin-Tello, 2018), and low self-concept (what a child thinks about themselves; Shields et al., 2006) in comparison to children and adolescents without disabilities. This is thought to be because the motor impairments associated with CP (e.g., muscle weakness, and muscle spasticity) make everyday activities (e.g., walking) difficult, which may negatively affect their self-concept (Shields, et al., 2007). Furthermore, as self-concept is seen to be a determinant of self-esteem (Russo et al., 2008), a low self-concept in children with CP can be assumed to negatively affect self-esteem. However, in the current study, participants 1, 2, and 3 showed improvements, and participant 5 showed no change, in the global self-worth subscale (i.e., self-esteem), suggesting that participation in frame running has a positive impact on the participants self-esteem. In comparison to the subscale means produced by Harter (1985) for children without disabilities, participant 1, 2, and 5 scored similarly to the subscale means for their age and showed no clear differences suggesting that self-esteem in adolescents with CP can be similar to those without disabilities. Participant 3 also scored similar to the subscale means for their age and reported slightly higher scores at T2 and T3 compared to the averages reported by Harter (1985).

These results are supported up by previous studies investigating self-esteem in children with CP compared to children without disabilities, where the results have shown children with CP to report a similar level of self-esteem to children without disabilities suggesting CP may not necessarily be associated with reported low self-esteem in children and adolescents (Manuel, et al., 2003; Schuengel et al., 2006; Russo et al., 2008). The results of the current study are positive with regards to self-esteem as a high self-esteem can be associated with positive mental health and a positive influence on an individual's behaviours (Fox and Lindwall, 2014). Participation in physical activity can also have a positive or negative impact on self-esteem, and the improvements reported for self-esteem of three of the four participants in the current study, across their frame running training sessions, alongside the self-esteem scores reported being similar to mean scores of non-disabled populations is a positive outcome for frame running participation.

In relation to self-concept, Harter (1985) referred to it as a multidimensional psychological construct which endeavours to discover what people think about themselves by investigating the awareness of an individual's characteristics and attributes, for example,

athletic competence, physical appearance, and behavioural conduct (Harter, 1985). Furthermore, self-concept is seen as a very important part of psychological health, and so if some domains are impaired in children with CP there may be a negative impact on the psychological health on the individuals (Bandura, 1986, Reid, et al., 2006).

Previous research has shown that some aspects of self-concept in children and adolescents with CP are reported lower in comparison to children without disabilities (Reid, et al., 2006, 2007). Participants 1 and 2, both female adolescents, reported lower scores in social competence, athletic competence, physical appearance, and job competence compared to the subscale means of adolescent females without disabilities (Harter, 1985). These results are in line with a systematic review by Reid, Murdoch, Loy, Dodd, and Taylor (2006) where they found evidence across six studies that adolescent females may be an at-risk group because the scores for the domains of social acceptance, physical appearance, scholastic competence, and athletic competence were all lower than the group without disabilities. On the other hand, participant 5, an adolescent male scored in line with subscale means of adolescents without disabilities for each domain of the SPPA. Given that previous research has found lower scores for athletic and scholastic competence in male adolescents with CP compared to male adolescents without CP (Reid, et al., 2007), the results from the current study may be an outlier as a single case study design. Participant 3 reported improvements in all self-concept domains, and at T3, the scores for each self-concept domain had increased to be in line with the means for children without disabilities. The results recorded by participant three suggest that participation in frame running may have a positive impact on different aspects of self-concept.

Overall, the results of the current study suggest that children and adolescents do not exhibit low self-esteem even when they may feel less competent in some domains related to self-concept, for example, athletic competence and physical appearance of participants 1 and 2. However, the improvements in aspects of self-concept that may be attributed to being physically active, such as, physical appearance, social, and athletic competence, suggest frame running may have a positive influence on these aspects. Furthermore, improvement in physical appearance scores have been shown to correlate with self-esteem (Russo et al., 2008), which can also be related to engaging in, and maintaining physically active behaviours. In relation to self-esteem, the improvements reported for participants 1, 2, and 3, and no change for participant 5, of the 'global self-worth' scale suggests that frame running participation may have a positive impact on their self-esteem.

5.4.4 Psychological Well-being

The current study is one of the first studies to investigate the effect of frame running on psychological well-being in children and adolescents with CP, and administration of the Strength and Difficulties Questionnaire (SDQ; Goodman, 1997) as one of the most widely used questionnaires related to mental health in children and adolescents (Vostanis, 2006), allows any potential effects of frame running on psychological wellbeing to be investigated. The prevalence of mental health disorders in children and adolescents is increasing, and approximately 10% of children and adolescents aged between 5 and 16 years have significant mental health problems (McGinnity et al., 2005). Therefore, it is important to look at possible effects of physical activity, frame running on the psychological wellbeing of children and adolescents with CP, who are at a higher risk of developing mental health disorders than their typically developing peers (Whitney et al., 2008a).

For the discussion of the results for psychological wellbeing, the total score (TS) for the strengths and difficulties score will be viewed as either borderline/normal (TS < 16) and abnormal (TS > 16), which is in line with previous research investigating psychological wellbeing in children and adolescents with CP by Parkes et al., (2008). With participants 1 and 2 reporting in the 'abnormal' category it is an indication of being at risk of mental health problems, and with the other participants in the borderline/normal category, suggests that the level of risk of psychological problems can change across individuals and time, which may be expected, particularly within this population (Whitney, et al., 2018).

The current study shows the children and adolescents with CP may be at a greater risk of mental health problems compared to children and adolescents with CP, in line with Parkes et al., (2008). The results of the current study do not indicate that frame running participation can have significant impact on the psychological wellbeing in children and adolescents with CP, but there are small, yet encouraging improvements in self- and parent-reported psychological wellbeing. The mean TDS for children aged 8-12 with CP across Europe was 12.4 (Parkes, et al., 2008), and all participants, self- and parent-report, reported higher TDS across the frame running training apart from participant 3 who reported below 11 from baseline to T3, and participant 5 self-report of 12 at T2. For comparison to children and adolescents without disabilities, the TDS normative data from the United Kingdom was 10.3 for self-report and 8.4 for parent-report (SDQinfor.org/norms). With the very small number of studies administering the SDQ to a CP population, the results from this study were used as a reference point for the participants in the current study; however, the participants are aged 8-12 years only.

There may be a variety of factors that can link a heightened risk of mental health problems in children and adolescents with CP, including a low-level physical activity, as a physical risk factor for the development of mental health disorders. Furthermore, the CP

population are also at susceptible to other risk factors including the presence of an intellectual impairment (Parkes et al., 2008), and the experience of pain and sleep disorders (Whitney, et al., 2018). Alongside low levels of physical activity, lower enjoyment and participation in recreational and social activities in youth with CP has been reported in comparison to their typically developing peers (Michelsen et al., 2014). Less time participating in social and recreational activities can lead to less social interaction, and the opportunity to build and maintain friendships is also lower than the general youth population. Friendships are important for youth with and without disabilities but can be of high importance to the CP populations with studies showing that a lack of friendships and bullying also be a risk factor for developing mental health problems (Whitney, et al., 2019). A systematic review on cerebral palsy by Novak, Hines, Goldsmith, and Barclay (2012) found from the papers included in the review that 75% of children with CP were in pain, 50% of children with had an intellectual disability, 33% of children with CP were non-ambulant, and 20% of children with CP suffered from a sleep disorder. These statistics show the prevalence of potential risk factors to mental health disorders in youth with CP, and as they are comorbid with CP, the need for successful intervention to address the risk during early phases of development are important for future studies.

The results from this study may not show any significant improvements in solely the psychological wellbeing scores of the participants from frame running participation, however this may not be expected considering the evidence indicating that children and adolescents with CP are at higher risk of developing mental health disorders and the risk factors are associated with CP. However, the opportunities that come along with participation in frame running have the potential to attenuate the risk factors, for example, the chance to build and maintain friendships with other youth with CP while becoming more physically active, thus the physical and psychological benefits from then being more physically active.

5.4.5 Participation & Enjoyment

The current study is one of the first studies to investigate the effect of frame running on the participation and enjoyment in children and adolescents with CP. There is a large evidence base indicating that participation in regular physical activity benefits physical well-being in youth with and without disabilities, and the research exploring the impact on psychosocial wellbeing of youth with disabilities is growing (Trost & Loprinzi, 2008; Sienko, 2019). However, low levels of physical activity are a common issue for children with CP in comparison to their typically developing peers, and low levels of physical activity are exacerbated by high rates of sedentary behaviours (Maher, et al., 2007). Furthermore, when children and adolescents with CP are physically active, the overall variety of physical

activities are smaller and the activities are of lower intensity compared to their typically developing peers (Maher et al., 2007; Verschuren et al., 2016).

5.4.5.1 Influence on Participation of Different Activity Types and their Enjoyment

The results of the current study were in line with previous research, with the participants reporting participation in a variety of activities across the five different activity types (Imms, 2007). However, whether frame running participation had a positive impact on the participation of new activities or increased participation in current activities is unclear. It is important to note, there was no control group for the current study, and so was not possible to compare changes in activity participation from baseline to T3 to participants who did not participate in frame running. Furthermore, the winter months occurred across the middle of the frame running training programme, and this may have had a detrimental impact on the participation of some activity participation, thus for children and adolescents with CP to participate in the reported array of activities is a positive.

Participation is important to measure as it is the context in which children can develop skills, build and develop friendships, and enhance their physical and psychosocial wellbeing (King et al., 2009). The different types of activity listed in the CAPE allow a more rounded picture of the participants participation, as the different interests and variety of skills that may be necessary for participation in the different types of activity will affect the way scores interpreted. The scores across the different activity types can provide an insight into the participants activity patterns and if there is a balance to the children's participation which can impact overall well-being and health of a child (King et al., 2004).

5.4.5.1.1 Formal & Informal Activity

The difference between formal and informal activities is thought to be important as each type of activity may affect the development of children differently (Beauvis, 2001). As seen from the results (see Table 5.15), there is no clear or consistent pattern regarding the influence of frame running participation on the diversity of formal or informal activities. Each participant did report participation in both types of activities which is important because of the benefits associated with each type of activity. Given the structured and organised nature of formal activities, which normally involve a coach or a leader (e.g., frame running), can lead to the development of social skills, self-belief concepts (Beauvis, 2001), and physical and psychosocial development (Mahoney et al., 2006). Informal activities are normally initiated by the child and less structured, which can assist in learning and developing exploration, however, are thought to have less benefits in comparison to formal activity participation

(Engel-Yeger et al., 2009). Previous research has shown children with disabilities participated in significantly less formal and informal activities (King et al., 2009). Thus, less participation can affect the building and development of the benefits associated with formal and informal activities.

The diversity of participation in formal activities in the current study was quite low, with only one participant reporting over 50% diversity at baseline, and zero at T3. With greater benefits associated with formal activities, participation might be key for development, but for children with disabilities, their opportunity to participate can be limited, or taken out of their hands. Children may not have a choice about what formal activities they participate in because of limited opportunities, impaired functional ability and/or understanding, thus parents make their choices. If a child with disabilities take part in an activity that they haven't chosen themselves, then there can be a negative influence on enjoyment and intrinsic motivation, and with the link between enjoyment and intrinsic motivation, it can impact the child's ability to develop skills and enhance wellbeing (Hunter & Csikszentmihalyi, 2003; King et al., 2009). Furthermore, in the current study there is no clear pattern of frame running participation influencing the enjoyment of formal or informal activities, with enjoyment in both formal and informal activities similar at baseline and T3. There is contrasting evidence regarding enjoyment in formal and informal activities for children with and without disabilities. Mahoney and colleagues (2006) reported for typically developing children, participation in formal activities was more enjoyable in comparison to informal activity participation. On the other hand, King and colleagues (2009) found children with disabilities reported higher enjoyment for informal activities over formal activities, thus children with disabilities may not gain the psychosocial benefits associated with the more structured formal activities. Furthermore, the lower enjoyment might be related to the lack of motivation and choice the children have to participate in the activity, hence the importance of enjoyment and having more suitable opportunities for children with disabilities to participate, to encourage engagement and adherence.

5.4.5.1.2 Recreational, Physical, Social, Skill-based, & Self-Improvement Activities

The 'recreational', 'self-improvement', and 'social' activities include more skills that can be used in day-to-day life, transferred across activities, and importantly can be used through development and into adulthood. These activities are important to highlight as participation and performance in day-to-day activities is lower in children and adolescents with CP compared to those without disabilities, and an improvement in these activities would in turn help their functional status, and future independence (Park et al., 2016). All participants

reported good diversity of recreational (42% to 75%), social (30% to 100%), and self-improvement (20% to 80%), but there was no clear influence of frame running on the participation of these types of activities. Furthermore, the enjoyment reported for recreational, and social activities was high for all participants from baseline to T3 (53% to 100%) indicating the participants enjoy participating in the activities selected. The good diversity and enjoyment reported for these activities suggest the participants may have the opportunity to gain and develop skills, which can influence their ability in day-to-day activities.

The 'physical' and 'skill-based' activities which require the participants to be active, also requires more specific skills in comparison to the three previously mentioned activity types, and skill-based can also include being physically active (e.g., swimming, horse riding, and gymnastics). The diversity of 'physical' (8% to 50%), and 'skill-based' (30% to 60%) was notably lower compared to the other activity types, however the enjoyment reported for 'physical' (60% to 100%) and 'skill-based' activity (60% to 100%) was similar to the other activity types. Again, there was no clear pattern or influence of frame running participation on other activities.

The low diversity of physical activities reported by participants in the current study, combined with the reasonably high enjoyment scores, highlights the limited opportunities, and availability of suitable options or alternatives for the participants, hence a suitable activity would theoretically elicit high levels of enjoyment. This is similar to results reported by Mainemer and colleagues (2008) who found school-aged children with CP reported good diversity in participation of activities with high levels of enjoyment, but participation in physical activity and skill-based activities was lower in comparison to other types. Furthermore, this highlights the necessity for more appropriate opportunities to be available for individuals with CP to be physically active, such as frame running, as it provides the opportunity to break up a sedentary lifestyle (which is a problem in the CP population; Maher et al., 2007), exercise at a higher intensity (particularly those who have more severe mobility impairments) and develop social skills meeting new people in similar situations.

5.4.5.2 Frame running Participation & Enjoyment

The high enjoyment scores reported across the 12-weeks of frame running in the current study (mean rating of 3.75-5 out of 5; see Table 5.17), suggests frame running is perceived as an enjoyable activity for children with CP. Furthermore, these results are in line with previous research by van Schie et al. (2015) and Bryant et al., (2015), who both found frame running to be an enjoyable activity. Enjoyment can be a key facilitator in the promotion and engagement of frame running participation, hence the current results are a positive finding for future promotion of frame running participation. Furthermore, given the more structured

nature of the frame running training sessions (i.e., a formal activity), it would suggest frame running participation might provide the opportunity for participants to gain the benefits associated with formal activity participation, such as development of self-concept and self-esteem, building friendships, and enhancing physical and psychosocial wellbeing (King et al., 2009). Moreover, the struggles for children and adolescents with CP to meet the physical activity guidelines are well documented (Verschuren et al., 2016), thus are at increased risk of secondary conditions including obesity, pain, and skeletal fragility (Fowler et al., 2017), and with enjoyment thought to facilitate motivation in physical activity, the high enjoyment scores for frame running in the current study are significant in suggesting frame running can be a viable option for increasing physical activity in children and adolescents with CP.

5.4.5.3 Further reflection

The validity and reliability of assessments of individuals with CP is important and are critical to consider for research and practice. As discussed in the next section, the selection of outcome measures is thought to be a limitation of the current study as not all the measures work in harmony, for example, some participants reporting self-report scores, some only having proxy-report scores because of age. Of the outcome measures selected only one measure, the Peds-QoL CP module, was condition specific and this has been shown to account for sensitivity between types of CP. Furthermore, with a lack of a total score it does not allow for an overall comparison of the QoL, and there is no clear evidence of a minimal difference needed to show influence. Each of the SDQ, CAPE, SPPC, and SPPA have been shown to work with children with CP, with the SDQ and CAPE highlighted as popular measures for children with CP (Schiarti et al., 2014). To my knowledge there is no research to indicate that the SDQ, CAPE, SPPC, and SPPA, are sensitive to change in CP. The responsiveness of a measure is important because it indicates the measures' ability to detect minimal but clinically relevant changes, which can be vital involving research with CP population as the changes may be more subtle compared to able-bodied individuals. The measures selected were informed by previous research, which indicated their feasibility within the CP population. Therefore, to help provide a clearer picture of the sensitivity of change and responsiveness of measures, the calculation of a stable baseline may have been beneficial. In the current study, there was only one baseline measurement taken from participants leaving the next data point (T1) to show instability in the measures selected, highlighting the importance of a stable baseline. It is important to note that collecting a stable baseline can be difficult due to the variability of human performance (Lobo et al., 2017).

An intervention period of 12-weeks has been shown in previous physical activity research to be an adequate length of time for effects to show in participants, however, with the lack of knowledge around the measures selected being sensitive to change within the

CP population, it may not be a long enough time period for the for changes to be detected. Therefore, a longitudinal study may provide a clearer reflection of the outcome. The data collection in a single case design can be a double edge sword with both have both limited data points as a limitation, but also the difficulty in collecting upwards of five data points in a study phase (Lobo et al., 2017).

In the current study, visual inspection of the data was carried out as data analysis with the data considered longitudinally. This form of analysis has been shown in previous single-case studies as the traditional method for evaluating treatment effects in single case research (e.g., Mardon et al., 2016). The statistical analysis in the current study could have been taken further to explore the magnitude of the intervention effect. Had the visual analysis of each participant suggested a functional relationship through three trends of effectiveness, calculating the effect sizes for each participant using cohen's *d* or hedge's *g* (an extension of cohen's *d*, and can correct for smaller sample sizes; Lobo et al., 2017) with a bigger effect size suggesting a stronger relationship between frame running participation and the impact on the psychosocial construct. The points discussed in this section suggest some methodological issues in the study, and that moving forward feasibility research may be the next best logical step (see section 7.7.2). However as novel research, the results from the current study do provide some initial insight to the impact of frame running.

5.5 Limitations & Future Directions

There are several potential limitations to the current study. First, as this was not a controlled study design, it is not clear whether the changes reported by the participants are down solely to participation in frame running training programme, external factors, or a combination of both. For example, better self-efficacy could be attributed to feeling more confident in school; however, it could be contributed to by the uniqueness of frame running.

Secondly, as a single-case design, the generalisability of the results can be a limitation, as it is not possible to generalise the effects of frame running training sessions on psychosocial wellbeing and QoL to the wider CP population. Furthermore, with the small sample size, and participants showing an interest in frame running prior to signing up (i.e., a convenience sample) the design lacks representativeness. Therefore, the high adherence and enjoyment across the current study may be higher compared to a group of participants who were not previously invested in frame running. However, given the current study is novel research, it can provide some initial evidence on the impact of frame running on psychosocial factors and QoL, for children and adolescents with CP. Additionally, despite the small sample size, age (5-18 years) and severity of CP (GMFCS levels II-V) of the participants is well covered in the current study, indicating frame running is a suitable

alternative to be physically active for children and adolescents with moderate to severe mobility impairments.

A further limitation to the study was a lack of a stable baseline which is common within single case design, as it was not deemed possible due to time constraints of the study, not feasible for the participants to complete the additional load that would have been necessary for a stable baseline to be measured. Therefore, it must be noted that any changes recorded from BL to T1 are not an effect of frame running but reflect the instability of the constructs and measures.

Another limitation were the outcome measures selected. Each of the outcome measures administered were self- and/or parent-report, which can cause various problems when trying to generalise or interpret the results or identify causal factors and/or relationships. The variables that can have an effect on the participants while they were completing the outcome measures each time, could have had a bearing on the results. For example, cerebral palsy can be an unpredictable condition, and one participant might be having a bad day, and feel negatively about themselves, and look back on how they have been with a more negative outlook, in turn, impacting the results for that data collection point. Furthermore, the understanding of some constructs can be difficult for children without disabilities (e.g., self-concept), and so any cognitive impairment associated with CP, can increase the difficulty in the perception of some constructs (e.g., self-concept, or pain). The intellectual functioning (i.e., the IQ) of the participants was not measured for the current study, and therefore the impact of an intellectual deficit on the understanding or completion was unclear. Furthermore, a deficit in intellectual functioning may influence the participants' ability to understand and complete the self-report forms, and therefore require some form of assistance to complete the outcome measures, and in turn may not show a true representation of how the participant was feeling. The use of parent-report forms can then also be compromised by this as there may be a recency bias, when completing the outcome measure. Parents may also have extra belief in frame running, and the benefits their child could gain, and therefore overestimate how their child is feeling/doing. Additionally, some of the outcome measures used were not specific to cerebral palsy, and therefore may not have been sensitive enough to pick up more subtle changes in the participants (e.g., self-efficacy and psychological wellbeing), however, the majority of the measures selected had previously been used within the CP population (e.g., SPPC and SPPA in Darrah et al., 1999). Finally, the number of outcome measures used was quite high (6), covering a broad range of constructs, and the load may have caused mental fatigue for the participants.

It will be important to continue the studying of frame running, and the potential impact it can have on the psychosocial wellbeing of children and adolescents with CP given the limited opportunities in physical activity, sport, and exercise for the cerebral palsy population.

The inclusion of a control group, more CP specific measures (e.g., myTREEHOUSE Self-concept Assessment; Cheong et al., 2018), and a narrower/focussed outcome (e.g., QoL, or psychological wellbeing only), in future studies may provide a clearer picture of the impact frame running may have on the important psychosocial aspects of life.

5.6 Conclusion

In conclusion, the current study is one of the first studies to look at the effects of 12-weeks of frame running training on children and adolescents with CP, and therefore can add to the growing literature regarding frame running and adds to the current literature regarding physical activity participation and the effects the youth CP population.

The main takeaway, and major positive, from the current study was a reported improvement in the self-esteem in 80% of participants across the 12-weeks of frame running, however, whether the improvement in their self-esteem is solely down to frame running is unclear. Additionally, the visual analysis of the data did indicate there was a trend towards improvement for some participants in some constructs, for example, psychological wellbeing for 80% of participants improved as shown by the parent-report form, and two participants reported an improvement in aspects of self-concept including athletic competence and perceived physical appearance. The results from the other constructs measured, QoL, self-efficacy, participation, self-concept, and psychological wellbeing did not show any clear effects for each participant but results for the domains were in line with previous research regarding this population. Furthermore, the lack of a control group makes it difficult to make any clear statements. However, each participant reported 100% adherence (each attending 12 weeks of training), high levels of enjoyment for the training sessions (mean rating of 3.75-5 out of 5), and each participant has continued to attend frame running training sessions post the study. Each of these points are a positive, and important for frame running to be recognised as an alternative and enjoyable opportunity for individuals with CP to be physically active, and have experience the freedom of running, and high intensity exercise, things they may not have had the opportunity to experience without frame running.

The literature on the physical, social, and psychological benefits that can be gained from being physically active (Martin and Whalen, 2012) are well documented, and therefore, theoretically, frame running can provide those benefits to the participants also. The qualitative data from the social validation questions indicate that parents and children involved in frame running recognise the physical and social benefits of participation in frame running, and the gap that frame running can fill for the cerebral palsy population, highlighted by the high agreement (6.2/7) that they would recommend frame running to a friend.

The small number of studies that have already investigated frame running (e.g., Bryant et al., 2015) have shown the positive impact on a variety of domains for individuals with CP, and therefore the results from the current study should be interpreted with caution but seen a positive step towards future studies investigating similar topics. Given the importance of the psychosocial constructs to all individuals with CP, providing and developing additional opportunities, such as frame running, to participate and compete from grassroots level through to international level will be very important for the future of the adapted sport and CP population.

Chapter 6: Impact of Frame running on Psychosocial Wellbeing: The view of competitive athletes

6.1 Introduction

6.1.1 Importance of Physical Activity from Childhood to Adulthood

The current study investigates the perceived impact frame running participation has had on competitive athletes' psychosocial wellbeing and QoL. An athlete's journey to high level participation (e.g., International competition) will normally begin as a child, thus during childhood is when decisions can be made regarding adherence and motivation to continue participating. Hence it is important to gather information on the perceived impact of frame running on psychosocial wellbeing across the athlete's career, in turn helping to facilitate and promote frame running as a suitable and accessibility adaptive sport.

The importance of physical activity for children with and without disabilities is well documented and has been discussed earlier in the thesis (see section 2.2). The continuation of physical activity participation through childhood, adolescence, and into adulthood is very important for a healthy lifestyle, with a direct link between physical activity during childhood and adulthood suggested in previous research (Dwyer et al., 2009). An active lifestyle and lifestyle choices throughout childhood can directly or indirectly improve health in adulthood as active children become active adults (Wilson & Clayton, 2010).

Previous research has shown that as children with CP progress through adolescence and into adulthood, the frequency and intensity of the individuals being physically active drops off for a variety of reasons (e.g., lack of suitable opportunities, lack of funding, or lack of social support). On top of environmental and/or personal factors restricting participation, being physically inactive can lead children with CP to experience a deterioration in their muscles, and a drop in their functional capacity, further limiting their ability and/or desire to engage in physical activity, exercise, or sport. This can lead to a 'cycle of deterioration' (Debusse et al., 2009), as individuals with CP are less likely, or unable, to be physically active, leading to further mobility impairment, and increased possibility of additional health risks associated with being inactive (e.g., loss of muscle strength, or cardiovascular fitness).

Furthermore, the risk of cardiovascular disease, obesity, and type 2 diabetes are higher for inactive individuals, as well as the negative impact on psychological factors (e.g., self-efficacy), and social development (e.g., social exclusion). Therefore, it is vital to identify opportunities to break the cycle of deterioration for individuals with CP (e.g., hippotherapy; Debusse et al., 2009), hence the importance of exploring the impact of frame running on psychosocial wellbeing from experienced athletes to provide data on how frame running participation can be another opportunity to break the vicious cycle.

Sport has power as a tool for rehabilitation and improving the sense of wellbeing in community and rehabilitation settings (Blauwet & Willick, 2012). Some would argue there is a sport for everyone, and sports inclusion for children with disabilities can be more difficult, especially because the development of their cognitive ability, coordination, and mobility will vary across children, and across disabilities. This can cause children to reach milestones at different times and can impact their ability to participate in suitable and adaptive physical activity and sports (e.g., children with CP can have verbal and motor impairments which need to be considered for participation). There are opportunities for individuals with more severe disabilities to participate, and the scope for performing at a higher, even elite, level is growing (e.g., swimming, boccia, and frame running). However, the evidence base for the impact of sports on individuals with more severe disabilities is limited (Wilson & Clayton, 2010), and with an increased focus on CP, swimming is a good example, as it can be performed on a recreational basis (Declerk et al., 2016), and on a competitive basis (Dutia et al., 2020) by individuals with mild to severe levels of impairment (e.g., spastic quadriplegia).

The health benefits of regular physical activity and exercise for individuals with disabilities are well documented, and improvements in equipment and opportunities has led to steady growth of participation in adaptive sports (Spörner, et al., 2009). However, those with disabilities still face a variety of barriers to participation, and these are continued to be reported through all levels of competition, and the barriers encountered by athletes, along with the facilitators to promote participation, are considered in the following section.

6.1.2 Barriers and Facilitators for Athletes with disabilities

As previously mentioned, sport is often used in a rehabilitation setting for those with disabilities to improve their QoL, and familiarise them with being physically active, as well as presenting them with sporting opportunities (Bragaru et al., 2011). However, post rehabilitation, the number of individuals with disabilities that continue to be physically active is small (van Der Ploeg et al., 2007). Previous research has reported individuals with disabilities identify additional barriers to being physically active compared to those without disabilities (e.g., unsuitable facilities, or unsuitable supervision/support; Rimmer et al., 2000). On the other hand, facilitators to physical activity for those with and without disabilities have been reported to be in line with each other, with both groups identifying factors such as health and social benefits, and enjoyment as facilitators (Rimmer et al., 2000).

Elite level athletes with disabilities have also experienced facilitators and barriers to participation in sport, though barriers may have been more at the beginning of their sporting journey. Jaarsma, Geertzen, de Jong, Dijkstra, and Dekker (2014) identified barriers and facilitators of sports for Dutch Paralympians and included all athletes with physical

disabilities to provide a broader insight. The results of the research showed that wheelchair athletes reported more barriers to participation compared to ambulant athletes, and 37% of athletes did not report any barriers at all. Barriers for wheelchair athletes may be expected to be higher as they may require additional help, or adapted facilities compared to ambulant athletes. For example, wheelchair athletes experienced 'being dependent on others to exercise' and 'transport' barriers more frequently in comparison to ambulant athletes.

Overall, the majority of barriers identified by the athletes were environmental (e.g., small number of suitable facilities; transportation; and qualified supervision was limited or non-existent), and the majority of facilitators were personal (e.g., fun; health and fitness; social experiences; competitive element; and acceptance of disability). The environmental barriers suggest Paralympic athletes experience (or have experienced) similar barriers to physically inactive individuals with disabilities. Furthermore, the results suggest future studies and policies can aim to overcome these barriers, and these barriers may be ones similar to those encountered throughout athletes' frame running careers. Therefore, the current study will explore if any barriers, past or present, have impacted the psychosocial wellbeing of the athletes.

As Jaarsma and colleagues (2014) reported, wheelchair athletes identified more barriers, and reported experiencing barriers to participation more frequently compared to the ambulant athletes. This indicates an area of focus for future research to improve participation and opportunities to participate for this population. Furthermore, it highlights the necessity for suitable opportunities for wheelchair athletes to participate in sport from recreational to Paralympic levels. Participation in organised or adaptive sports by athletes with disabilities may offer similar physical benefits to being involved in regular physical activity (e.g., increased muscle strength, physical fitness, or disease prevention; Lape et al., 2018). Previous research has shown the psychological benefits of sport participation for athletes with disabilities, which can play important roles in continued participation, positive psychosocial wellbeing, and QoL (Blauwet & Willick, 2010). However, there is no research exploring the impact of frame running on the psychological wellbeing of frame running athletes, therefore the current study aims to provide insight into this aspect of adaptive sport literature.

6.1.3 Athletes with Disabilities and the Impact of Sport Participation on their Quality of Life and Psychosocial Wellbeing

Previous research has reported improved psychosocial functioning of athletes with disabilities attributed to participation in their selected sports (e.g., Muraki et al., 2000). However, there is currently no research exploring the impact of frame running on the QoL

and psychosocial wellbeing of competitive frame running athletes, and the current study aims to provide initial insight into this aspect of adaptive sport. The psychosocial constructs selected for the current study are in line with constructs identified by participants in the Delphi survey (see chapter 4), and the constructs examined in the novice frame running athletes in study 2 (see chapter 5) to allow consistency throughout the thesis. In the following section, existing research into QoL and psychosocial constructs will be discussed with reference to athletes with disabilities. This conceptual understanding is how it is important for gauging the influence that frame running participation may have had on these athletes.

6.1.3.1 Quality of Life

Sproner, Fitzgerald, Dicianno, Collins, Teodorski, Pasquina, and Cooper (2009) reported anecdotal evidence of wheelchair athletes' perceptions of their QoL having improved because of participation in sporting events, and this was accompanied by greater levels of confidence and motivation. Groff, Lundberg, and Zabriskie (2009) also examined the influence of adapted sports participation for athletes with disabilities, specifically, athletes with cerebral palsy. Groff and colleagues administered QoL measures with 73 athletes with CP who were competing in the 2005 Cerebral Palsy World Games. To measure QoL, participants completed the Influence on Quality-of-Life Scale (IQLS; Zabriskie et al., 2005), measuring the influence participation in a particular sport or programme has had on their perceived QoL. Results showed participation in adaptive sport can have a positive influence on the CP athlete's QoL. Results for individual items of the IQLS further highlighted athlete's perception that participation was beneficial with over 80% of participants either agreeing or strongly agreeing adaptive sport has positively influenced their QoL and overall health. Furthermore, a significant negative correlation was identified between perceived influence on QoL, and severity of disability of athletes.

The results suggest athletes with more severe disabilities perceive participation to have less of an influence on their QoL, and given the importance of QoL, particularly for those with more severe disabilities, the severity of disability should be considered for future research. Additionally, the results highlight that the athlete's participation in sport is a significant factor in the influence on their QoL and health. Therefore, given the importance of QoL and health, suitable opportunities for participation in adaptive sports need to be developed and made available at all levels of competition, hence why the impact of frame running on an athlete's QoL is explored in the current study. Furthermore, it adds to the evidence of positive impact on QoL from participation in physical activity and sport for individuals with CP, and the benefits are still there to be gained from consistent and regular

sport participation at an international level of competition which is important to note for frame running as an adaptive sport. However, this has not been studied for frame running and there is a gap in the adaptive sport research with no current research involving frame running and QoL in experienced athletes.

6.1.3.2 Psychological Wellbeing

Psychological wellbeing is another important factor that can be enhanced by sports participation for athletes with disabilities. Athletes with disabilities may be expected to have poorer psychological wellbeing (e.g., higher risk of depression, anxiety, and negative moods). This may be because living with disabilities can make daily tasks and activities stressful, in turn tasks and activities are avoided which can lead to further risk of stress, anxiety and depression (Bandura, 1986). Previous research has reported participation in competitive sport can lower the risk of depression, and anxiety, and have a positive influence on mood (Ströhle, 2009), however there is no research regarding the influence of competitive frame running on psychological wellbeing, hence the importance of inclusion in the current study.

Fiorlli and colleagues (2013) examined the psychological wellbeing in wheelchair basketball players and non-basketball playing wheelchair users. Results of the wheelchair basketball group showed better psychological wellbeing and social skills in comparison to the wheelchair users. Furthermore, the psychological wellbeing scale scores suggest that the competitive profile of the basketball players influenced the significantly better psychological profiles compared to the non-basketball players. More specifically, the athletes reported lower depression, anxiety, and sleep disorders highlighting participation in wheelchair basketball (adaptive sport) can have a positive influence on psychological wellbeing. Therefore, as importance of improving and maintaining a good psychological wellbeing is highlighted in previous research, it is important to for the current research to add to the literature of adaptive sport by examining the effects of frame running on psychological wellbeing. Furthermore, individuals restricted to wheelchairs who also have an upper body impairment affecting trunk and arm movement may not have the functional ability to participate in sports such as wheelchair basketball (or wheelchair rugby), thus heightening the risk of poor psychological wellbeing. However, frame running can provide the opportunity to individuals with more severe impairments to be physically active (Bryant et al., 2015), and compete, highlighting the importance of investigating psychological wellbeing in the current study.

Campbell and Jones (1995) also examined the effects of sport participation on psychological wellbeing of wheelchair athletes, and non-participating wheelchair users.

Similar to Fiorlli et al. (2013), the wheelchair athletes reported better psychological wellbeing with lower levels of depression and anger, and a more positive perception of their overall health and wellbeing compared to the non-participating group. Furthermore, Campbell and Jones (1995) reported a difference in wheelchair athletes based on their level of competition. The results showed that the athletes competing at international level reported higher scores for psychological well-being, self-esteem, and lower scores in depression comparison to the athletes competing at either regional or recreational level. Results suggest competing at a higher level (i.e., International compared against regional competition) can have a positive influence on psychological wellbeing, including lower anxiety and higher self-esteem for wheelchair athletes.

Further to competition level being identified as having an influence on the psychological wellbeing of wheelchair athletes, Campbell (1995), examined the impact of sports participation on the psychological wellbeing of wheelchair athletes with acquired disability (i.e., amputee) and congenital disability (i.e. cerebral palsy). The results suggest athletes with acquired disabilities perceive themselves to have a better psychological wellbeing compared to those with congenital disabilities. The acquired disability group reported significantly higher self-esteem, and lower scores on depression, anger, and anxiety compared to the congenital disability athletes. With congenital athletes reporting lower psychological wellbeing, it suggests being disabled from early life may contribute to a lack of independence and/or coping skills which athletes with acquired disabilities may have developed prior to becoming disabled. Campbell (1995) adds another factor which can influence psychological wellbeing in wheelchair athletes, however, they did not investigate why the athletes with acquired disabilities may report a more positive psychological wellbeing, and this may be of note for future research. Furthermore, it highlights the importance of having individuals with congenital disabilities, such as cerebral palsy, be physically active and participate in sport, to reduce the discrepancy between themselves and individuals with acquired disabilities, further highlighting the importance of investigating the impact of frame running on psychological wellbeing in the current study, thus adding to the adaptive sport literature.

6.1.3.3 Self-efficacy

The importance of self-efficacy within sports is well documented and thought to be one of the more influential psychological constructs (Feltz, et al., 2008). Bandura (1997) notes that efficacy beliefs are not the judgments about a set of skills which produce an action, but more about the belief of what can be accomplished/goals that can be reached with the skills each individual has (i.e., self-efficacy judgements are about what one thinks they can do with their own skills, not about what one has). Furthermore, self-efficacy judgements can also

influence an athlete's effort in training and their levels of persistence (Bandura, 1997). Despite the importance of self-efficacy in sport, the research within disability sport is limited compared to able-bodied sport, however the evidence base is growing (Martin, 2008).

Greenwood and colleagues (1990) investigated the importance of self-efficacy, as an influence on a wheelchair tennis athlete's psychological wellbeing compared to non-tennis wheelchair users. Self-efficacy of performing both tennis and day to day wheelchair mobility tasks were measured along with psychological wellbeing. They found tennis athletes reported significantly higher self-efficacy for both tennis and day to day mobility compared to a non-tennis wheelchair group. Additionally, the tennis group reported a more positive psychological wellbeing compared to non-tennis wheelchair participants highlighting sports participation can have a positive impact on psychological wellbeing for disabled athletes. Furthermore, Greenwood et al. (1990) show tennis athletes are more confident in tennis related mobility (which may be expected) but also in more general wheelchair mobility tasks which may lead to improved independence and in turn, a better QoL.

Adnem and colleagues (2010) reported similar results within a group of wheelchair quad rugby athletes who scored higher on self-efficacy items relating to rugby skills and self-efficacy of day-to-day activities compared to the wheelchair users with no quad rugby experience. Furthermore, Martin (2002) showed wheelchair athletes with a strong self-efficacy to perform and train better in comparison to those athletes exhibiting a weak self-efficacy. The research mentioned highlights participation in adaptive sport can improve self-efficacy of athletes with disabilities in specific areas of performance as well as in general day to day activities. This indicates the potential for frame running to have an impact on self-efficacy for frame running tasks (e.g., bike mobility), but also in day-to-day situations as the self-efficacy beliefs can be specific to distinct domains of functioning (Feltz et al., 2008), hence it is important for self-efficacy to be included in the current study.

6.1.3.4 Self-concept

The multidimensional nature of self-concept is thought to influence a multitude of life markers (e.g., sport performance) given it is a profile built on the perceptions one has for oneself (Fox, 1998). Furthermore, it is important to understand each person is not going to perceive an equal level of competence across domains, and individuals will have varied self-concepts across domains (e.g., social, physical, academic; Shapiro & Martin, 2010). This is highlighted by the Self-Perception Profile Scales for Children, Adolescents, and Adults, with each scale including additional domains which would not be relatable to the younger population, while emphasising the developing structure of self-concept through maturity (Harter, 1982).

Self-concept for athletes with disabilities may be more complex in comparison to able-bodied athletes due to physical and/or cognitive limitations. However, the relationship between self-concept and physical activity, sport, and exercise, may enhance the self-concept of athletes with disabilities compared to individuals with disabilities who are not physically active. Sherrill, Hinson, Gench, Kennedy, and Low (1990) was one of the first studies to examine self-concept of youth athletes with disabilities (including cerebral palsy). The results across eight self-concept domains (Harter, 1988) suggested patterns in the self-concept data from the athletes with disabilities were similar to those in the mean scores of youth athletes without disabilities. However, the use of a general self-concept scale may not have captured the true perception of each domain of the athletes with disabilities. For example, the majority of athletes with CP were wheelchair bound, and when asked about 'physical appearance', are the scores reflective of acceptance of their disability and appearance, or as a tool of denial and not accepting their disability? Future research with more condition specific and domain specific outcome measures, such as the myTREEHOUSE Self-Concept Assessment (Cheong et al., 2017) would be welcome to capture the impact on aspects of self-concept more relevant to the population, such as athletes with CP.

Physical self-concept is one aspect of self-concept that is widely studied within the world of exercise and sport and has been identified as a predictor of physical activity, and in turn, can be a means of improving levels of physical activity (Marsh et al., 2006). Ferriera and Fox (2008) examined the difference in perception of the physical self between elite basketball athletes with and without disabilities. The basketball players without disabilities reported higher scores across the subscales for perceptions of the physical self compared to the wheelchair athletes, suggesting, as previously mentioned, the physical self-concept of athletes with disabilities may be more complex with different characteristics to the general population. However, the wheelchair basketball athletes reported higher physical self-concept scores compared to sedentary individuals, suggesting the positive impact sports participation can have on the development of disabled athlete's physical self-concept. It shows there can be a benefit to self-concept development from being physically active for athletes with disabilities and furthermore, physical self-concept is thought to be an important variable in relation to reaching desired goals and outcomes, which can include physical activity/exercise behaviour and adherence (Marsh et al., 2006).

Motivation towards participation in physical activity is associated with an individual's perceptions of their competence, and therefore, youths with disabilities will require a positive self-concept to encourage them to participate (Shapiro & Martin, 2010). Youth with disabilities may have mobility limitations, and this can lead to the expectation of a lower physical self-concept, reducing confidence in movement, and then avoidance of physical

activity/exercise as it could be viewed as a risk. Even though it is assumed that children with CP have a lower self-concept in comparison to children without disabilities, a systematic review by Reid and colleagues (2006) did not find sufficient evidence for this to be a generalised statement. The multidimensional nature of self-concept can also be identified within athletes with CP. For example, an athlete with CP may have a strong perception of their sport competence, but a low flexibility competence because of tight muscles associated with CP (Martin & Whalen, 2012). However, with no research on self-concept and frame running, and self-concept in athletes with CP also not well researched - despite the evidence of self-concept as a predictor of physical activity for individuals with disabilities, it indicates a gap in the research for future studies (Rimmer et al., 1996). Furthermore, a trend towards improvement in aspects of self-concept, such as physical appearance and athletic competence, was reported in the current thesis after a 12-week frame running training programme (see section 5.4.3), therefore identifying any impact on the athlete's self-concept in the current study would provide further insight to self-concept and frame running.

6.1.3.5 Self-esteem

Self-esteem is another important construct to consider for athletes with disabilities as it has been identified as an important factor of functioning and performance which is related to both psychological and general wellbeing (Buckworth et al., 2013). For example, high self-esteem is associated positively with mental and social health improving academic achievement, happiness, and life satisfaction (Qasim et al., 2018).

Previous research has shown individuals with disabilities are less physically active than those without disabilities and they face more personal and/or social challenges limiting their participation and it is the limiting of participation which can have a negative impact on their psychological wellbeing, including their self-esteem (Dishman et al., 2004). General improvements in self-esteem have been reported for athletes with disabilities across various levels of competition (van der Vliet et al., 2008), and the athletes with disabilities reported significantly higher self-esteem than their inactive peers (Campbell & Jones, 1994). Furthermore, these results were similar for wheelchair basketball athletes, who reported higher self-esteem in comparison to sedentary individuals with disabilities, highlighting the influence participation in sport can have on self-esteem, and in turn, mental health (Ferreira & Fox, 2008). Van de Vliet, van Biesen, and Vanlandewijck (2008) examined the self-esteem of Flemish athletes with disabilities and found the athletes reported similar levels of self-esteem in comparison to their able-bodied peers, adding evidence of the potential for sport participation to positively develop psychosocial constructs for individuals and athletes with disabilities.

Of course, it may be expected that international level athletes would report higher self-esteem compared to inactive individuals without a disability, because they have reached the pinnacle of their sport, and competing at that level will have a positive impact (Campbell, 1995; Roux, 2012), but these results show that low self-esteem is not an inevitable characteristic of disability. The relationship between physical activity and self-esteem for individuals and athletes with disabilities is what is important to note. Scarpa (2011) reported individuals with disabilities who exercised on a regular basis reported higher self-esteem in comparison to their peers who did not exercise regularly, which again highlights the positive relationship between self-esteem and physical activity. However, at the high level of competition, research showing enhanced self-esteem for athletes with disabilities is mainly focused on wheelchair sports (e.g., Richardson et al., 2017; Qasim et al., 2018). This highlights a gap in the literature, to assess the impact of sports participation on self-esteem for athletes with disabilities who are not involved in a wheelchair-based sport, such as frame running, hence self-esteem will be included in the current study on the impact of frame running athletes.

6.1.3.6 A Lack of Qualitative Research for Athletes with Disabilities & their Psychosocial Wellbeing

As discussed in previous sections, participation in disability sport can have a positive impact on the development of psychosocial characteristics including psychological wellbeing, self-efficacy, self-esteem, self-concept, and QoL. Participation provides athletes with disabilities the opportunity to overcome physical, psychological, and social limitations associated with disabilities (e.g., impaired mobility; poor psychological wellbeing; at risk of anxiety and depression; social exclusion). Previous research has examined the effects of sports participation on psychosocial constructs, but the results focus mainly on quantitative data, which may only provide a snapshot of an athlete's situation. Furthermore, the use of non-condition specific outcome measures may not detect the more subtle or specific changes and/or development for athletes with disabilities. To gain a better understanding and uncover more meaning behind the reported results on the influence of sports participation for athletes with disabilities on psychosocial constructs, qualitative research is a suitable and valuable option (Lape et al., 2018).

Qualitative research on athletes with disabilities and psychosocial constructs is limited, with more of the focus of previous qualitative research on identifying barriers and facilitators to physical activity for individuals with various disabilities, including CP (Conchar et al., 2016), spinal cord injuries (Stephens et al., 2012), and lower limb amputees (Bragaru et al., 2013).

Richardson, Paphomas, Smith, and Goosy-Tolfrey (2017) conducted semi-structured interviews with wheelchair tennis athletes to research the psychosocial impact of participation. The wheelchair athletes reported that they perceived that participation enhanced their psychosocial wellbeing. Athletes reported how participation gave them improved self-confidence with reference to how sport changed their life, and how they felt they were able to do everything, which is in line with previous research with wheelchair sport participation reported to improve self-confidence (Sproner et al., 2009). Furthermore, athletes emphasised how participation had enhanced their physical functioning with reference to feeling fit and strong, and not feeling dependent on anyone, which is also important for future functional independence and enhancing their QoL.

Resilience was another aspect the athletes identified to be enhanced by participation. Enhanced resilience suggests the individual is able to cope and adapt more in adverse and stressful situations, which can help individuals who may experience poor psychological wellbeing (e.g., depression or anxiety) or social isolation overcome the psychosocial problems they may face. Furthermore, the tennis athletes perceived personal growth in relation to the development of their abilities which can enhance their QoL and self-esteem on and off the court. The social aspect of disability sport was also enhanced as reported by the athletes as they gained new life experiences (e.g., travelling on a plane to new countries) that allowed them to meet new people and build friendships through participation in wheelchair sports. The qualitative data from Richardson and colleagues (2017) indicate that participation in disability sport can enhance psychosocial wellbeing and be a positive experience, and therefore it can be beneficial to examine psychosocial impact of disability sport for athletes. However, the impact of frame running on the psychosocial wellbeing of athletes is not known, therefore the current study aims to provide an insight.

Based on previous quantitative research, and the lack of qualitative research examining the psychosocial impact of disability sport, the aim of the current study is to address the gap in the literature and investigate the impact of frame running on psychosocial wellbeing for athletes with cerebral palsy. The current study is novel research and aims to add new insight by examining the effects of frame running participation for individuals with CP, a population who may be faced with restricted opportunities and/or expected to be inactive due to impairments associated with their disability (e.g., moderate or severe CP). Therefore, the study aims to consider QoL and each of the psychosocial constructs introduced in order to answer research question five.

Research Question 5: What impact does frame running have on the psychosocial wellbeing of competitive frame running athletes with cerebral palsy?

6.2 Methodology

6.2.1 Study Design

This study applied a cross sectional qualitative research design to provide a broader understanding of how frame running has impacted on psychosocial factors of frame running athletes. Open-ended questions aimed to explore the individuals' views of how frame running has impacted psychosocial aspects of their lives.

The use of a web-based survey in current study allowed for the participants to express their opinions without being influenced by the researcher. Open-ended questions have the advantage that they may elicit a response from the participant which is more spontaneous compared to answering a close-ended question. Moreover, answering open-ended questions provides the participants with the opportunity to produce a set of more diverse answers, and give more detailed information relating to their personal views, experiences, and views compared to the answering close-ended questions (Reja et al., 2003). Furthermore, the design of the online survey, including the appearance of the overall survey (i.e., any multimedia used, and the introduction page), are important areas to consider due to the self-administration of the survey and ensuring the survey was as far-reaching and as accessible as possible to the frame running athletes around the world (Reja et al., 2003).

The design of the current survey was important for the particular population as it allowed for the participants to complete the questions in their own time, giving them more time to think about their responses, and importantly not feel overwhelmed or under pressure with the questions, some of which explored sensitive topics (e.g., psychological wellbeing). Furthermore, the online survey allowed participants to complete in their own home environments, thus avoiding environmental distractions (e.g., staff interruptions; Easton et al., 2000), but may also allow the participant to feel more comfortable and relaxed compared to being in a more social environment, where there may be more pressure felt about what how they feel. Additionally, the design allows for the participants to complete the questions using their own communication system, if required, given the possibility of impaired communication and speech associated with CP. Therefore, the use of online surveys and open-ended questions was thought to yield a richer data set compared to verbal interview responses due to the possibility of verbal communication impairments. For these reasons, the use of an online survey was selected over the use of online interviews; however, the study design in terms of participant sample size, question design/ piloting, and qualitative data analysis was comparable to an interview study.

6.2.2 Participants

6.2.2.1 Inclusion Criteria

The inclusion criteria for the current study were frame running athletes, aged 14 and over, with a good level of English, and access to a computer. A frame running athlete was defined as an individual who participates in frame running competitions at a local level and above (i.e., to international competitions such as the World Para Athletics Championships). With the diversity disabilities and CP severity, individuals with severe cognitive impairments, restricting their ability to understand and/or respond to the questions without major assistance were advised not to take part.

6.2.2.2 Recruitment

The participants for the current study were recruited online via a number of channels. Initial contact was made with frame running athletes around Europe, and they were sent a participant information sheet with details about the study alongside a link for the survey, where participants provided informed consent. Snowball sampling (Biernaki & Waldorf, 1981) was applied to the current study given the small community of frame running athletes. Participants were asked to share the link with others who they thought might be willing and able to complete the survey, whether they were from the same club or had met them competing around the world. Next, contact was made with heads of frame running organisations and countries that participate/compete at the International Frame running Camp and Cup, and the information and survey link was passed on directly to their athletes. Finally, information about the study and a link to the survey was shared on social media for athletes to access and share. The recruitment process lasted five months.

6.2.2.3 Participant Demographics

Ten participants (male $n = 4$; female $n = 6$) completed the online survey, all aged over 18 years (range 19 to 44; $m = 25$ years). Nine participants reported having CP, and one participant reported having a rare genetic condition affecting skeletal development. The participants varied in nationality (Scotland $n=7$; England $n=1$; Denmark $n=1$; Australia $n=1$), CP type and distribution, GMFCS level, frame running classification, level of competition, and training frequency (see Table 6.1). Furthermore, the participants represent a varying level of frame running experience and competition level (see Table 6.1), providing the potential for comparison of impact on psychosocial wellbeing across different levels of frame running participation and ability.

All participants reported using a mobility aid to varying degrees, 8 out of 10 used a wheelchair, and 2 reported that the use of a mobility aid was dependent on their

environment (e.g., walking unaided around home but uses mobility aids when going out). In comparison to the high number of participants reporting the use of a mobility device, just one participant reported using a communication device to assist them with their communication.

Additionally, information was recorded in relation to the athletes' frame running training in terms of their training frequency. The frequency of training ranged from 1 session per week to 6-7 sessions per week (see Table 6.1). Furthermore, the type of training they participated in varied, with participants reporting speed work (100% of participants reported participating in this), mobility work (88.9%), weight training (77.8%), and flexibility (55.6%). The lower percentage of participants reporting flexibility training, may be due to participants having separate physiotherapy sessions away from frame running where the focus is on flexibility. Participants were also asked about the diversity of their physical activity, and 50% of participants identified frame running as their only form of exercise. The other 50% reported participating in range of activities including skiing, paracycling, swimming and boccia.

Table 6.1. Demographic information for participants

Disability		GMFCS Level		FR Classification		Participation Level	
Spastic Bilateral CP	(n=4)	Level II	(n=3)	RR1	(n=1)	International	(n=4)
Ataxic CP	(n=2)	Level III	(n=4)	RR2	(n=3)	National	(n=4)
Dyskinetic CP	(n=1)	Level IV	(n=3)	RR3	(n=5)	Local	(n=2)
Mixed CP	(n=1)			NC	(n=1)		
CP Type Unknown	(n=1)						
Rare genetic condition	(n=1)						

Note. CP=cerebral palsy; GMFCS=gross motor function classification system; FR=frame running; FR = Frame Running; NC= Not Currently Classified

In reference to frame running classification there are three classes, RR1, RR2, and RR3. The classification mainly involves assessing impairment (e.g., spasticity and motor control), but only those aspects that are believed to be relevant to frame running. One participant is RR1 classified, meaning this athlete had the most severe difficulties in limb and trunk control, and co-ordination, all of which affect the functionality of the athlete while on the racerunner. Three participants are classified as RR2 meaning athletes will have moderate

involvement of upper and lower limbs, and the trunk. Furthermore, the stride pattern is more effective than the stride pattern of the RR1 classified athletes. Five of the participants are classified as RR3 meaning athletes are viewed as those with the highest functional capacity on the racerunner. Athletes with RR3 classification can have moderate lower limb coordination difficulties, good trunk control, and mostly symmetrical leg movements, but they are unable to run without the frame running bike (racerunning.org). Each frame running class is represented in the current study, showing the range of functional capacity of the participants in the current study (see Table 6.1).

6.2.3 Ethics

Ethical approval was obtained from the Moray House School of Education and Sport Ethics Committee at the University of Edinburgh. The participant information sheet provided before the survey questions explained that participation was completely voluntary, and participants were able to withdraw from the study at any point without any consequences. The participants completed the online consent form and were informed that the information provided would be used in the current study. Ethical considerations included the fact that some questions cover topics that some may find difficult to talk about (e.g., psychological health; links to resources for support were provided). Ethical considerations of working with children and adolescents with CP are discussed in chapter 3.

6.2.4 Online Survey Question Design

For the current study, the survey questions were designed to answer the research question. The use of surveys has been reported as an appropriate methodology, particularly as this area of research is still relatively unknown (Creswell, 2009). The design of the questions was influenced by previous research, which has shown athletes with disabilities have perceived improvement in the psychosocial wellbeing to participation in their sport (e.g., Sproner et al., 2009; see section 6.1.3). However, the impact of participation on the psychosocial wellbeing of athletes with CP is still a growing area of research (Martin, 2006), with previous research showing more of a focus towards the physical effects, thus the current questions look to explore the effects of frame running participation on the psychosocial wellbeing of competitive frame running athletes. The psychosocial constructs included in the current study have been influenced by the research carried out in study 1 and 2 of the current thesis (see chapters 4 and 5 respectively). Furthermore, previous research has shown constructs such as self-efficacy, self-concept, and self-esteem can play important roles in the promotion of health behaviours, including physical activity (Bandura, 1999), hence exploring the perceived impact of frame running on the selected constructs may provide valuable insight

for frame running. Furthermore, a gap in the literature regarding the psychosocial wellbeing of athletes with CP has been identified, and the current research aims to provide insight into the impact of frame running on psychosocial wellbeing in competitive frame running athletes.

The questions asked in the main part of the survey (i.e., after the questions about the athlete's age, gender, nationality, impairment, and participation) are shown in table 6.2. Question 1 examined the participants' first impressions and experiences of frame running, including how they got into it. This is important because individuals with disabilities are less likely to participate in physical activity and sport, and therefore can miss out on physical and psychosocial benefits of regular physical activity (Lape et al., 2018). Furthermore, the limited opportunities available to youth with CP, particularly those with more severe impairment (e.g., GMFCS level IV or V) are a major contributor to their inactivity, and so the information from competitive athletes may provide valuable insight for future promotion of the sport.

Question 2 asked the athletes why they have continued to participate in frame running, and if enjoyment has played an important role in their continued participation. The fear of an activity not being fun or enjoyable is a prominent barrier to physical activity participation and can negatively impact future motivation to continue participating (Verschuren et al., 2016). Therefore, the importance of physical activity and sport being enjoyable can be key for initial engagement in sport, and this was highlighted by Dutch Paralympians who reported fun and enjoyment as a key facilitator to their participation (Jaarsma et al., 2014). Furthermore, the current research (see section 5.4.5.2) and previous research has reported frame running to be an enjoyable activity for children and adolescents with CP (e.g., van Schie et al., 2015; Bryant et al., 2015), thus exploring if current athletes have a similar perception, and if enjoyment plays an important role in their participation can provide further insight into frame running and the perception as an enjoyable opportunity to be physically active.

Question 3 asked participants about the influence of frame running on social aspects of the athlete's lives. The restriction of opportunities youth with disabilities have to participate in physical activity (due to personal and/or environmental barriers), limits their social interaction and opportunity to develop socially, hence it is important to examine the impact of frame running in the current study. Furthermore, increased social interactions can help with improved social integration in community, school, and work environments which can be important for social development and perceived life satisfaction (Lape et al., 2018).

Question 4 focussed on participants self-efficacy, and the influence of frame running on the participants' ability to cope with day-to-day activities, social and emotional situations. The importance of self-efficacy in a day to day and sporting environment is well documented, and the decisions made by individuals about their capabilities will influence

their choice of activity and level of effort they put in (McAuley & Blissmer, 1999). Furthermore, self-efficacy has been reported as a predictor of physical activity behaviour, and as youth with CP may be assumed to have lower self-efficacy than their typically developing peers, improving self-efficacy is important for improving social participation and QoL (Kim et al., 2017; van der Slot et al., 2010).

Question 5 focussed on the multidimensional construct of self-concept, and asked participants how they felt frame running may have influenced aspects of self-concept (e.g., physical appearance, athletic identity, or social acceptance). Given the multidimensional nature of self-concept, it can be complex to understand but it is thought to be important for a variety of social and behavioural factors (e.g., peer interactions, or sporting performance; Harter, 1985). On this basis, it is important to examine self-concept as it can provide an insight in how the individuals think about themselves across the awareness of their own characteristics and attributes (Martin & Whalen, 2012).

Question 6 asked the athletes if they perceived frame running participation to have impacted their self-esteem. Based on findings it is assumed that individuals with CP have reduced levels of self-esteem (sense of self-worth) because of the associated impairments (e.g., difficulty walking), and the impact it can have on their self-concept (Manuel et al., 2003; Russo et al., 2008). However, previous research has shown little or no difference in perceived self-esteem between youths with and without disabilities (Lindwall, 2014). Therefore, it is important to examine the athlete's self-esteem as high self-esteem can be associated with positive mental health (Muris et al., 2003), and improving self-esteem via physical activity participation is a frequently used rationale for health programmes (Biddle et al., 2015).

Question 7 focussed on the psychological wellbeing of athletes and the influence frame running may have had on their mental health. Individuals with CP are at a higher risk of mental health disorders, and negative psychological wellbeing compared to individuals without disabilities (Parkes et al., 2008). Being physically active can positively impact on factors that negatively affect mental health (e.g., pain; Whitney, 2018). Furthermore, with low levels of physical activity also seen as a risk factor of psychological wellbeing (Whitney, 2019), current athletes can have a more positive outlook on their psychological wellbeing compared to inactive individuals with disabilities (Fiorilli et al., 2013).

Question 8 asked participants to describe the impact they perceived frame running to have had on their QoL. Previous research has shown that participation in adaptive sports has positively impacted the QoL of athletes with CP (Groff et al., 2009; Yazicioglu et al., 2012). Furthermore, improved QoL can be accompanied by increased levels of confidence and motivation highlighting the impact QoL can have on psychological and physical development, hence, the importance of QoL to the research (Sproner et al., 2009).

6.2.4 Procedure

The questions were piloted by the researcher with an international athlete with CP, who was a para sport athlete but not involved in frame running, so as to avoid the loss of a potential participant. This allowed for the pilot test to be applicable - as the athlete's participation in CP specific sport at an international level provided the opportunity to assess whether the questions posed generated appropriate responses. Post pilot testing, the ten finalised questions were entered into an online survey format using secure web-based survey software (JISC Online Surveys) and distributed to potential participants via the methods described in recruitment (see section 6.2.3). Participants read the information sheet and completed the informed consent process before answering the survey questions. The online survey consisted of questions that collected demographic information (see Table 6.2) and 8 open-ended questions focussing on psychosocial factors and the potential impact that frame running participation has had on those factors.

6.2.4.1 Qualitative Data Analysis

The data from the participants was anonymised, and the participants were given identification numbers for reference. A hybrid of qualitative analysis methods were used in the current study, with each response analysed following the step-by-step guide of the six phases of reflexive thematic analysis (Braun & Clarke, 2006; Braun et al., 2018). The study incorporates an inductive approach (Boyatzis, 1998), and then a deductive approach (Crabtree & Miller, 1999), using the International Classification of Functioning, Disability and Health framework (ICF; WHO, 2001).

The sample size of 10 participants in the current study may be viewed as small, with previous research suggesting that 15 to 30 participants is common within qualitative research (Braun, 2013). However, selecting the correct sample size in qualitative research can be difficult as there are no standard rules of magic formulas for determining an adequate sample size (Braun et al., 2018). A common concept that has been used to try and determine the correct sample size for qualitative studies is 'saturation', and this refers to a point when no new information is generated from the data collected (Morse, 1995). The use of 'saturation' as a concept has been highlighted by previous research as it has been suggested saturation can be reached through relatively small sample sizes 6-12, therefore it is referenced for justification (e.g., Ando et al., 2014). Though a smaller sample size is present in the current study, 'saturation' is not an appropriate method as the area of research is new, so there is little to no prior evidence to indicate if the additional data being generated is new or not. Furthermore, thematic analysis is about the identification of new patterns of meaning, and as this occurs after data collection and analysis is required, it is

difficult to judge if the data generated is new or not (Braun et al., 2018). The current sample size was informed by a variety of contextual and pragmatic considerations, including the small frame running community, the breadth of the research question, the absence of previous research, and the diversity within the population, such as the potential for communication and cognitive deficits associated with CP.

The process of reflexive thematic analysis offers flexibility and allows for search of themes which are seen as important to the concept of the research question, which in the current study is the impact of frame running on psychosocial wellbeing. Following the 6 phases of reflexive thematic analysis (Braun & Clarke, 2006; Braun et al., 2018), the iterative process allows for the familiarisation of the data through the process of “carefully reading and re-reading the data” (p. 258; Rice & Ezzy, 1999 *in* Fereday & Muir-Cochrane, 2006), while noting interesting individual comments, significant words and phrases identified from the participant’s responses, thus producing initial codes (see Table 6.3). Furthermore, as a new area of research familiarisation of is important to provide the researcher an entry point to the data (Braun et al., 2018). Previous research has noted that the coding of texts has been recognised to be difficult given the interpretation of the questions by participants in previous research (Saldaña, 2013; Belotto, 2018). Following the generation of codes, the grouping and further analysis of similar codes, akin to building blocks, allows for the construction of themes. Through the process of revising and defining the identified themes, the final themes and sub-themes become clearer with reference to how the data is related to the research question, and what is meaningful about the data (see Table 6.4). To enhance rigour of the qualitative analysis of the current study, the process of ‘critical friend’ was carried out. The process of ‘critical friend’ allows for critical dialogue between the researcher and additional people, with the researcher allowed to explain their interpretations of the data while receive critical feedback (Smith & McGannon, 2018). Important to note that the role of the ‘critical friend’ is not solely to reach a consensus or agree with the researcher, but to challenge the researcher’s construction of knowledge, and the possibility of alternative interpretations or explanations. This was an important process to include in the research as it is a new area of research, so there is no prior information regarding the inclusion of additional voices may enhance the quality of the data (see section 3.6.2.3 for discussion on ‘critical friend’ process).

In the second step of the analysis, a deductive approach used the ICF framework. As the ICF framework is used to describe the health status of an individual, the health, disability and function of an individual are all seen as inter-related, and further influenced by contextual factors (i.e., environmental and personal factors). Based on the high inter-relatedness, the current study looked to use the ICF framework to categorise the impact that frame running has had on athletes’ psychosocial wellbeing. Keywords, phrases, themes, and

subthemes were reviewed and analysed to identify any reference relating to the ICF framework components and to identify the distribution of responses across the framework (i.e., body structures and functions, activities and participation, and environmental factors).

Within thematic analysis, Braun and colleagues (2018) mapped out three broad schools of thematic analysis: coding reliability approaches; reflexive thematic analysis approaches, and codebook. Within the current study, the thematic analysis can come under predominately the reflexive thematic analysis approach. There is little relation to coding reliability approach as there is no 'consensus' coding or use of predetermined themes. Reflexive thematic analysis is the most prominent school in the current study as it aims "to provide a coherent and compelling interpretation of the data, grounded in the data." (p6. Braun et al., 2018). Furthermore, given the subjective nature of the reflexive approach, it allows for the researcher to engage with the data and the analytical process (Braun & Clarke, 2019), as well as provide a voice to an underrepresented group (Braun et al., 2018), in this case frame running athletes.

Table 6.2 Questions for Experienced Frame running Athletes

	Question	Rationale	Reference
1	<p>a. How did you get into frame running?</p> <p>b. How did you feel about frame running the first time?</p> <p>c. How much does frame running mean to you?</p>	<p>Individuals with disabilities are less likely to participate in physical activity and sports, and therefore can miss out on the physical and psychosocial benefits associated with regular physical activity. Therefore, having background information on their journey into and continued frame running could be beneficial for future promotion of the sport.</p>	<p>Lape et al., (2018)</p>
2	<p>Can you describe why you continue to participate in frame running? Is enjoyment important?</p>	<p>The fear of an activity not being fun or enjoyable is a prominent barrier to physical activity participation, and then the maintenance of being physical active. Therefore, with enjoyment also linked to intrinsic motivation, the importance of a physical activity/sport being perceived as enjoyable can be key to initial engagement in the sport and continued participation.</p>	<p>King et al., (2009); Verschuren et al., (2016)</p>
3	<p>Has frame running influenced social aspects of your life? If so, in what way?</p>	<p>Participation in adaptive sports provide the opportunity for individuals with a disability to be part of social interactions in sporting environments which they may not have had access to before participation. Increased social interactions can help in improved social integration in community, school, and work environments, and potentially impact on perceived life satisfaction.</p>	<p>Lape et al., (2018)</p>
4	<p>Do you feel frame running has influenced your self-efficacy for day-to-day activities, and your ability to cope with these activities, social, and emotional situations? If so, why?</p>	<p>An increase in the belief for an individual to participate in activities, complete tasks, and deal with situations, is important to the development of an individual's physical and social self. Self-efficacy has been shown to be a factor for increasing physical activity in youths and can also be an important factor in the adoption of other health behaviours. A low self-efficacy can be an indication that there may be a likelihood of quitting the sport/physical activity.</p>	<p>Bandura (1997); Biddle et al. (2015)</p>

5	<p>Across the time you have been frame running, do you feel your view of yourself changing and/or developing? This can be reference to how you feel on and off the bike, including how you view your physical, athletic, and social ability, and physical appearance.</p>	<p>Self-concept investigates what people think about themselves across their awareness of their own characteristics and attributes. It is also thought to influence a variety of social and behavioural factors (e.g., peer interactions, and sport performance respectively), and as a multidimensional construct it can be important to psychological health also.</p>	<p>Harter (1985); Martin & Whalen (2012)</p>
6	<p>Do you believe your self-esteem (sense of self-worth) has changed since you began frame running? If so, please say how and why you think it has changed, and how it has impacted your life.</p>	<p>It is assumed that individuals with CP have reduced levels of self-esteem (sense of self-worth) because of the associated impairments (e.g., difficulty walking), and the impact it can have on their self-concept. Some studies have shown little or no difference in perceived self-esteem between youths with and without disabilities which is important as high self-esteem can be associated with positive impacts on mental health and behaviours.</p>	<p>Manuel et al., (2003); Russo et al., (2008); Fox & Lindwall (2014)</p>
7	<p>Do you feel that participating in frame running has affected your psychological wellbeing? Do you believe that it has affected your behaviour and mood (including levels of anxiety and depression)? If so, how?</p>	<p>Individuals with CP are at higher risk of mental health disorders compared to individuals without disabilities. Being physically active can positively impact factors that negatively affect mental health (e.g., pain), and as low levels of physical activity are seen as a risk factor to poor psychological wellbeing, athletes, theoretically could have a more positive outlook on mental health.</p>	<p>Whitney (2018; 2019); Parkes et al., (2008)</p>
8	<p>Can you describe what impact you think frame running has had on your quality of life? In other words, has it had an effect on your standard of life, health, comfort, and happiness?</p>	<p>QoL can be viewed as the 'holy grail' of outcome measures, and adapted sports have shown to impact QoL in athletes with CP. Improved QoL can be accompanied by increased levels of confidence and motivation highlighting the impact QoL can have on psychological, social, and physical development, hence, the importance of QoL to the research.</p>	<p>Groff et al., (2009); Sproner et al., (2009); Yazicioglu, et al. (2012)</p>

Table 6.3 Example of Thematic Analysis Process

Original Text	Identification of relevant phrases	Initial Coding	Extraction of keywords and phrases	Identified Theme	Components according to ICF framework
<p><i>“I have achieved things in frame running that I never thought I could or would be able to achieve (e.g., competing in sport at the highest level), and I guess it has shown me I can achieve my goals & ambitions if the environment is right for me. I feel that has been important to remind myself of that in other aspects of my life where things are more challenging.”</i></p>	<p>Achieved things in frame running I never thought I could or would; achieve goals and ambitions</p>	<p>Achieving goals</p>	<p>Achieved; achieve goals and ambitions;</p>	<p>Sense of achievement</p>	<p>‘Activities and participation’</p>

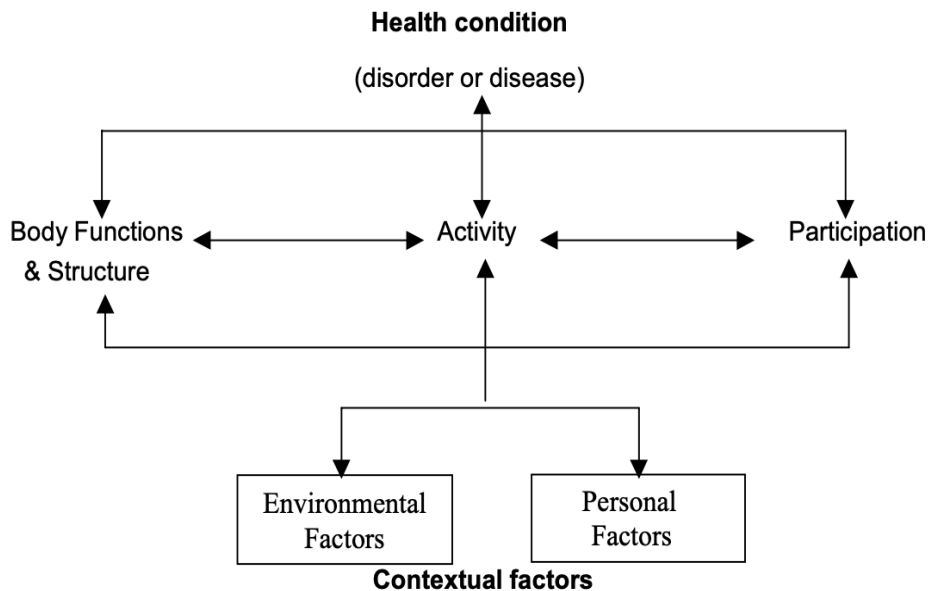
6.3 Results & Discussion:

The following section will report and discuss the themes and sub-themes identified through the thematic analysis process (see Table 6.3 for an example). A summary of the themes and sub-themes extracted from the participants’ responses, along with the relevant components of the ICF framework, is shown in Table 6.4. The themes and sub-themes will form the structure of this combined results and discussion section and will consider how these factors affect the psychosocial wellbeing and QoL of the frame running athletes. Results will be interpreted in relation to existing research.

Note: although one frame running athlete did not have CP, the aim of the study was to look at the perceived impact on frame running athletes, and as a frame running athlete it was thought to be unfair to exclude them from the study. Furthermore, the sample size of the study was small, and as it does not clearly state in the inclusion criteria athletes must have CP, the participant remained part of the study. As the focus of the thesis is CP, thus the main focus of the results and discussion will be CP.

6.3.1 ICF Framework

Figure 6.1 International Classification of Functioning and Disability (ICF) Framework (WHO, 2001)



The ICF framework (introduced in chapter 4) is important to CP because of its ability to offer a common language and a global approach to thinking about health and describing functioning and disability (Rosenbaum & Stewart, 2004). The inclusion of the ICF framework in this study was important to show the way athletes have perceived frame running to have impacted their lives, and how the results can be distributed across the ICF framework, whether it is 'social engagement' ('participation' component) or 'boosting of self-esteem' (mental functions in body function component). As a tool which can provide guidance to help overcome issues, for example, performance issues are inhibited by the environment, mobility interventions become a viable option (Rosenbaum & Stewart, 2004). Therefore, the inclusion of the ICF provides additional perspective of how frame running can impact aspects of disability and health, as well as showing the complex interactions of frame running. The results can help promote frame running as a viable intervention to reduce the gap between the individual's current capacity and the desired outcome.

With the aim of this study to evaluate the impact of frame running participation on the psychosocial wellbeing of competitive frame running athletes, the ICF framework was adopted to consider the impact and identify any future areas of need. The interaction between each of the components of the ICF framework: body function, body structures,

activities and participation, and contextual factors are used to discuss health and disability (see Figure 6.1). However, it is important to note for the current study that the ICF framework does not include QoL.

Overall, as can be seen in Table 6.4, the responses of the frame running athletes encompass the three main components of the ICF, body functions and structure, activities and participation, and environmental factors. Personal factors are a component of contextual factors, however, are not classified in the ICF due to the variation across societies and cultures (WHO, 2002). Therefore, based on the assumption that each component of the ICF framework is linked to the others (WHO, 2007), competitive athletes report that frame running participation can impact on all aspects of disability and health. With responses distributed across the ICF framework components, this suggests that frame running participation has an effect on each component. However, it is worth noting that the impact of frame running on psychosocial wellbeing is not the same for each athlete, as will be shown in the following sections. Furthermore, no themes encompassing the 'body structures' component, but this may be expected given the focus of the questions was on psychosocial wellbeing.

Table 6.4 Main themes, sub-themes, and relation to ICF framework component identified through thematic analysis

Main Theme	Sub-theme	ICF Framework Component
Enhanced Life Experiences	<i>Life changing experience</i>	Activities & Participation
	<i>A growing sense of independence</i>	Activities & Participation
Enjoyment of new experiences	<i>Joy of meeting new people</i>	Activities & Participation
	<i>Joy of being involved in a competitive environment</i>	Activities & Participation
		Environmental factors
Opportunity to build and develop friendships		Environmental factors
	<i>Sense of belonging</i>	Activities & Participation
	<i>A feeling of inclusion</i>	Activities & Participation
Improved self-confidence	<i>Enabling social confidence</i>	Body Function
		Body Function
	<i>More positive future outlook</i>	Body Function
Development of self-concept	<i>Increased self-belief</i>	Body Function
		Body Function
	<i>Improvement of perceived athletic identity</i>	Body Function
Boosting of self-esteem	<i>Improvement of perceived athletic ability</i>	Body Function
		Body Function
	<i>Increased perceived competence</i>	Body Function
Positive impact on psychological wellbeing	<i>Sense of achievement</i>	Activities and Participation
		Body Function
	<i>Access to a suitable physical activity</i>	Body Function
Positive influence on QoL	<i>Mood Boosting activity</i>	Environmental Factors
	<i>Importance of Social Interaction</i>	Environmental Factors
		NA
	<i>Perceived increase in standard of living</i>	NA
	<i>Improved academic life</i>	Activities and participation

Note. ICF = International Classification of Functioning and Disability Framework; NA = Not Applicable

6.3.2 Background to Frame running Participation & Enhanced Life experiences

The responses to question ‘1a. *How did you get into Frame running?*’ (see Table 6.2) provided some narrative background about the various ways in which the athletes were introduced to frame running. Six of the participants were recommended frame running by various channels, including family and friends, physiotherapists, and CP charities (e.g., Bobath Scotland); three participants were introduced to frame running through advertised “come and try” days; and one participant, already aware of frame running, viewed it as an opportunity to improve their psychological and physical health:

“I felt isolated and wanted to improve my fitness in a group” – Participant 10

Questions ‘1b. How did you feel about frame running the first time?’ and ‘1c. How much does frame running mean to you?’ were also used to provide some background information as well as to gauge the participants’ initial thoughts and perceptions on frame running. It was anticipated that frame running would mean a great deal to the participants as for most it has been a big part of their lives for a significant period of time. The majority of responses clearly indicated frame running to be a vital part of the participants’ lives, not just for what it gives them in the present, but also for what impact it has had on their past and is likely to have on their future aspirations. Furthermore, the importance and significance of frame running was highlighted by 60% of participants reporting that frame running ‘was everything’ to them, and this is emphasised by the main themes identified within the current study (see Table 6.4).

The main theme identified from the analysis of question 1 responses focuses on enhanced life experiences, with two sub-themes identified of life changing experiences, and a growing sense of independence. Participants saw frame running as a chance to change their lives, and experience things that they had not thought were possible:

“I just thought frame running was the coolest thing ever! I loved the sense of freedom and speed it gave me. It was the first time I had been able to run independently, so that was pretty special. Growing up, all I wanted to do was do sport, but it was difficult for me to participate in so discovering frame running was a real ‘light-bulb’ moment for me – finally I was able to participate in a sport independently.” – Participant 3

Participation in sport is one aspect of life that individuals without disabilities may take for granted, but for children with CP it is not so simple, and the reference to simply wishing to participate in sport, and frame running being able to provide that experience, shows a simple yet life changing impact that frame running has had for the athlete. Furthermore, the experience of being able to run for the first time was echoed by other participants:

“Amazing. It was the first time I had ever experienced running” – Participant 5

“I felt free, it was the first time I had ever ran” – Participant 10

One participant compared the experience to what they thought running without disabilities would feel like:

“It felt to me what able bodied movement would feel like” – Participant 4

It is clear the experience of running was an important recognition by the participants from when they began frame running, and this can further enhance life experiences by helping them to overcome barriers to participation by simply providing an opportunity to be physically active, thus providing the opportunity to experience the physical, psychological, and social benefits associated with being physically active (e.g., improved CV fitness).

Restricted mobility is a common impairment associated with CP (Rosenbaum, 2006), and with restricted mobility, individuals are restricted in their opportunities to participate and be independent. A low level of independence can impact negatively on the perceived QoL as individuals can feel like they are always reliant on someone, which can discourage participation, not only in physical activity but in day-to-day activities as individuals do not want to feel like a burden leading to increased risk of inactivity and social isolation. Participants reported frame running as their first experience of feeling free and independent:

“It was literally like I had found my freedom as I didn’t need to rely on anyone else to take part as I normally do, it means everything as it enables me to be independent” – Participant 1

Furthermore, one participant referred to frame running as a political movement to enhance the lives of individuals with CP from complete novices to the elite racers, highlighting the awareness of the current athletes that there is a shortage of suitable opportunities for individuals with CP to be physically active, and recognise that frame running has the potential to fill this gap:

“It is a recreation, a sport, a way to relax or to distract but is also a global, political movement which showcases the abilities of significantly affected individuals with cerebral palsy who are continually underrepresented in elite sport.” – Participant 4

The theme of enhanced life experiences is further reported by participants in the recognition of the impact frame running participation had on their physical and psychological health:

“Means a lot to me, not only physically but also mentally.” – Participant 6

“It means a lot as I tend not to go out too much, so it gives me a regular exercise slot” – Participant 8

As previously mentioned, individuals with CP can be restricted in their mobility, thus can be limited in their participation in not only physical activity but in day-to-day activities, and this

can lead to being stuck in a cycle of deterioration, and frame running participation can break this cycle.

However, not every participant initially had a positive experience of frame running as reported by the other participants. One participant felt that frame running was '*strange and tiring*', and another reported not feeling confident in themselves to take on frame running because they perceived their walking ability to not be good enough. Despite the negative first experiences of the participants, it suggests there might be aspects of frame running that encouraged engagement (e.g., enjoyment, or social inclusion) and helped to overcome the initial physical barriers encountered.

6.3.3 Enjoying New Experiences:

Question 2 explored why participants continued with their participation in frame running, and if enjoyment was an important factor that they considered while continuing with their frame running. Thematic analysis identified the main theme of 'enjoying new experiences' and this came across in two subthemes, through the joy of meeting new people, and of being involved in a competitive environment.

The majority of participants referenced that enjoyment was important to their participation, and when the athletes felt that they were engaged in the activity, it brought them a sense of joy. However, the feeling of joy does not just purely from being physically active but also from meeting up with friends and having that connection of meeting a similar group of individuals. Prior to frame running their opportunity to do this was restricted, and frame running gives the participants the enjoyment and freedom to meet with people that they may not have been able to before:

"Enjoyment is definitely important, as well as the social aspect and the independence it gives me. I love to go a run to clear my head, and it is very important for my wellbeing." – Participant 5

"I continue to participate in frame running because it still gives me so much joy in my life" – Participant 6

The enjoyment of new experiences, including frame running, is an important recognition from current athletes due to the role the concept of enjoyment can play in promoting participation in physical activity. Enjoyment is an important factor in motivation, and motivation is one of the most important personal factors in physical activity (Saebu & Sørensen, 2011). Previous research has shown enjoyment to be vital to adherence in disability sport (Martin, 2006). The personal factor of enjoyment of an activity can lead to intrinsic motivation, and based on the self-determination theory, intrinsic motivation can be

key for the commitment to physical activity (Chatzisaraantis, & Hagger, 2008). Furthermore, the enjoyment of frame running was highlighted in the previous study (see section 5.4.5.2) with each participant reporting a mean enjoyment score between 3.75 to 5 (out of 5), suggesting frame running is perceived as an enjoyable physical activity across each of the levels of participation.

The subtheme of 'enjoyment of being involved in a competitive environment' shows frame running is able to provide a new experience, and a suitable, adaptive opportunity to be physically active for a group of individuals that are underrepresented across all levels of para-athletic competition. Therefore, it provides an outlet for the competitive side of the athletes that they may not have been able to express previously.

"I think what motivates me the most is the desire to keep improving my times and see how far frame running can take me. From a young age, it has always been my goal to compete at the Paralympics in frame running, and whilst frame running is not at the Paralympics, that will be my main source of motivation. Even if I didn't compete, I would probably do frame running for the health and social benefits but it's the competition opportunities I enjoy the most." – Participant 3

A feeling of competitiveness for the participants also acts as a motivational tool as it drives them to continue participating and improving their health and fitness. Furthermore, as they are surrounded by like-minded people, the competition will continue to push them forward with their participation as they strive to be the best.

"I like the competition, and I believe it has helped me improve my fitness and health in general" – Participant 1

"I enjoy the feeling of when you are running it feels like time stops and you are the only person in the world. I am also really competitive." – Participant 9

"I love to compete; my fitness levels are important for my mobility." – Participant 10

The comments exemplify the need to provide opportunities to compete for individuals with CP, who are an underrepresented population at World Para Athletics and Paralympic competitions. Furthermore, the sense of competition, and competitive feeling is a factor that motivated the athletes to continue participating, and previous research has shown that elite athletes with disabilities' psychological wellbeing can be associated with the level of competition they are participating in (Campbell & Jones, 1995). Campbell and Jones (1995) reported athletes participating at international level having a better psychological wellbeing with lower levels of anxiety in comparison to athletes competing at regional level, suggesting once athletes begin competing there may be added pressures which can negatively impact

psychological wellbeing, and this is then improved on as experience grows. One athlete from the current study highlights this possibility, and how the impact of frame running participation has varied as they progressed through levels of competition:

“...I especially enjoyed beating people and I relished no longer coming last! Now as I’m internationally, there is quite a lot of pressure to perform and constantly improve as you would expect so I’d say the impact has changed over time but still remains overall positive as I’m doing what I love.” – Participant 3

The quote shows that the joy of being involved in competition stems from the beginning of their frame running participation, and from racing (and beating) peers, bringing participants enjoyment and highlighting frame running can enhance life experiences from the beginning right through to international competition.

6.3.4 The Opportunity to Build and Develop Friendships

The third question explored the perceived impact that frame running participation has had on the social aspects for the participants. The main theme identified was ‘the opportunity to build and develop friendships’ and identified the subthemes of ‘sense of belonging’, ‘feeling of inclusion’, and ‘enabling self-confidence’. The themes identified from the current question are important given the social challenges that children and young people with CP can face (e.g., social isolation), thus frame running may help to facilitate the development of friendships which are important for psychosocial development (e.g., motivation). Each participant expressed in their own words how frame running had provided them with the opportunity to make new friends locally and/or internationally while providing them with a sense of belonging which can be very important for maintaining participation.

“Frame running is probably one of the most inclusive sports out there. I have made so many great friends who I can chat with regularly (even during lockdown), and I definitely feel included” – Participant 5

“I have made friends worldwide due to this (frame running), and I am always included.” – Participant 2

Furthermore, participants alluded to the view that being part of the frame running community had enabled them to be more confident in themselves and around other people, including outside of the frame running community, which is an aspect of their lives that they may not have thought was an option for them.

“I really struggled at school to make friends and fit in with my classmates. So, getting introduced to the frame running community at competitions and through training

allowed me to make friendships with people in a similar situation to me which I needed growing up. I am also a lot more confident than I was.” – Participant 3

“Frame running has certainly helped me to make friends not only in the frame running community but also in my local community.” – Participant 7

Prior to frame running, the athletes may have suffered from social isolation (Barkley et al., 2012) and/or social stigma as they can be viewed negatively in the wider society which may negatively impact on their psychosocial wellbeing (Read et al., 2014). Like other disabilities CP can be highly stigmatised, and individuals with CP can face negative attitudes from the general community, thus acting as a barrier to further participation and support (Read et al., 2014). Previous research has shown that disability sport and physical activity are associated with the development and maintenance of friendships (Weiss et al., 1996), and participation provides the opportunity for the individuals to interact which can bring self-enhancing benefits, such as the development of psychosocial wellbeing (Martin & Smith, 2002; Richardson et al., 2017). Furthermore, given ‘the development of friendship’ was the main theme within the current study it sheds light on the positive and friendly environment that frame running can provide to individuals. Furthermore, this can help individuals to feel more comfortable in themselves and add to their confidence. Additionally, the experience of belonging to a group can give the individuals with CP the experience of feeling cared for and supported which can also help in breaking the feeling of being stigmatised (Conchar et al., 2016). The findings from the current study are in line with this, suggesting that frame running can offer the opportunities to develop friendships and this has a positive effect on the psychosocial wellbeing.

6.3.5 Improved self-confidence

The fourth question explored the impact that frame running has had on the participants self-efficacy and their ability to cope with day-to-day activities, social, and emotional situations. Two of the participants responses simply indicated that they felt frame running had not influenced their self-efficacy by stating ‘no’ and ‘not really’. However, from the remaining responses, thematic analysis identified the main theme of ‘improved self-confidence’, and subthemes of ‘more positive future outlook’ and ‘increased self-belief’. When the participants first got into frame running, it was a completely new experience given the uniqueness of the sport and the population that it is aimed at (i.e., individuals with limited to no walking ability). Through their continued participation an athlete’s self-confidence has helped them to have the opportunity to learn new skills and improve on previously learned ones. The participants described that frame running participation gave them the belief that they were able to do things that they would not have thought were possible before beginning frame running.

“frame running has given me more self-confidence, and I believe that I can do anything if I set my mind to it.” – Participant 6

“it’s improved my confidence in many situations, I have so much more self-belief than before I found frame running.” – Participant 10

An individual’s self-confidence refers to the perception of their own ability and judgement, and previous research has shown disability sport to increase athletes’ self-confidence (Spornier et al., 2009). In the current study the athletes identified self-confidence to have improved in various situations including school environments, making friends, and for day-to-day tasks. A number of responses indicated that frame running participation had improved their general self-confidence (see quotes above). Other participants also felt the self-confidence in their own ability had increased since participating in frame running, which had led them to have *‘confidence in my ability to do things that I wouldn’t have done previously’* and importantly belief for their future, *‘it’s given me belief for my future independence’*. Furthermore, frame running appeared to have encouraged one participant to take on new physical activities, and in turn they discovered the benefits of exercise on their self-efficacy:

“I think exercise in general does this for me, and that can be anything. In the beginning, frame running was my only form of exercise and so it did (improve self-efficacy), but I have learnt how to access more forms now and it is not unique to frame running.” – Participant 4

The above quote indicates that participation in frame running might have provided the belief to search for, and try other, new forms of physical activity. Moreover, participants expressed that frame running participation has helped them with aspects of their psychosocial wellbeing, such as self-esteem, and provided them with the increased self-belief to take part in activities that they may not have thought as possible prior to participating in frame running.

“I believe it (frame running) has helped me with self-esteem and self-belief as now I know that I can do things that I wouldn’t have done previously because of my training.” – Participant 1

Self-esteem is an important component of psychosocial wellbeing, and previous research has shown that participation in physical activity and sport can enhance the self-esteem of athletes with disabilities (e.g., Greenwood et al., 1990), and the current research suggests frame running may be able to provide an avenue to develop the self-esteem of athletes with CP. Another important component of psychosocial wellbeing is self-efficacy and two

participants suggested that frame running has improved their self-efficacy but has also helped them be more prepared for the future and taught them life lessons, again, that may not have seemed plausible prior to frame running participation.

“I have achieved things in frame running that I never thought I could or would be able to achieve (e.g., competing in sport at the highest level), and I guess it has shown me I can achieve my goals/ambitions if the environment is right for me. I feel that has been important to remind myself of that in other aspects of my life where things are more challenging. For example, the exam system at school isn’t setup for people like me so I really struggled through my exams. Also, performance sport in general teaches you so much (i.e., how to perform under pressure and how to best prepare yourself etc.) and frame running has given me that opportunity to go into performance sport that I otherwise wouldn’t have had and hence I have developed valuable life skills.” – Participant 3

“Yes. Frame running has made me physically AND mentally stronger. It has taught me life lessons that I use in everyday life and tasks.” – Participant 5

The above responses highlight the far-reaching impact that frame running has had on these individual athletes, and their self-belief and ability to participate in additional physical activity, day to day activities, and the belief they can participate and complete other tasks outside of frame running, thus indicating the importance of their frame running participation on impacting psychological factors. Furthermore, previous research has shown sports participation to increase self-efficacy and self-esteem in athletes with disabilities after the intervention, and bring self-efficacy scores up to a comparable level as individuals without disabilities (Wickman et al., 2018). Self-efficacy is also important in promoting participation, as it can help individuals with the adjustment of failure. For example, someone with a high self-efficacy can see failure as an opportunity to change and move forward, therefore making it more likely for them to continue participating, however, someone with low self-efficacy may attribute failure to something unchangeable and give up (Bandura, 1997). Self-efficacy cannot always be generalised across activities and tasks (Samson & Solmon, 2011), and with two participants stating they perceived frame running to have no influence on their self-efficacy the idea of participation having a positive impact on self-efficacy cannot be generalised. The current study is in line with previous research reporting athletes show self-confidence and self-efficacy from participation of disability sport (e.g., Sproner et al., 2009; Richardson et al., 2017; Wickman et al., 2018). Furthermore, the results are contributing a new body of knowledge regarding the psychosocial wellbeing of frame running athletes.

6.3.6 Development of Self-concept

Question five explored the impact that frame running may have had on the participants self-concept and referred to how the view of themselves may have changed and/or developed, whether it be on and/or off the frame running bike. Two participants did not respond to this question, and one participant simply responded 'no' to the question.

The main theme identified was the 'development of self-concept, with two subthemes of 'improvement of perceived athletic identity' and 'improvement of perceived athletic ability'. One participant referred to frame running directly impacting their athletic identity, however they felt a ceiling has been reached because of level of competition available for the sport.

"In the beginning yes, it allowed me to develop the identity and physique of an athlete. As I've progressed in my Paralympic career this has changed. Frame running is still not as respected in the Paralympic community as other sports and does not have a medal event, or now any dedicated coaches. It will take some elite sport infrastructure changes before frame running can deliver that for me." – Participant 4

The above response does show that the introduction to frame running may have a positive impact on the athletic identity of the participants. Considering this may be one of the first and only opportunities this population may have had to be physically active in this manner (i.e., experiencing the sensation of running and/or participate in competitive sports), the discovery of an athletic identity may be expected. This is an important response as individuals with CP may struggle to construct their identity, but frame running can provide them with an identity as an athlete, such as *'it allowed me to develop the identity and physique of an athlete'*, or as simple as being seen as equal: *'On my bike I feel free and just like everyone else'*. Individuals with disabilities can struggle to develop a sense of identity because of stigmas and/or constraints of society, and this can further limit opportunities to participate or try out activities that may help to form and develop the self (e.g., sports; Groff et al., 2001).

Athletic identity can be an important aspect of self-concept, and a strong athletic identity can help to develop a strong self-identity, something that youths with disabilities may find difficult. Furthermore, a strong athletic identity has been associated with enhanced psychosocial wellbeing, confidence, and improved levels of participation in physical activity (Tasiemki et al., 2004). Previous research has shown that participation in elite disability sports has enhanced an athlete's athletic identity (e.g., wheelchair tennis; Richardson et al., 2017). This may not relate to all athletes in the current study because not every athlete competed at the highest level, and their perception of what determines an 'athlete' may vary and be dependent on the level of competition. Furthermore, previous research has shown Paralympic athletes report higher athletic identity compared to other athletes with disabilities

(van der Fliet et al., 2008). Additionally, Paralympic athletes reported similar athletic identity to able-bodied athletes highlighting the influence disability sport can have on individuals with disabilities, and how it may lead to athletes breaking a stigma within the wider community. The current study supports previous research by suggesting that participation in disability sport, in this case frame running, can help individuals develop an aspect of their self-identity, which in turn can help them to feel more accepted within society, and further break a societal stigma so as to not be viewed solely as someone with a disability.

An improvement in the perception of the participants' athletic ability was also identified as a subtheme, as the frame running athletes referred to improved levels of general fitness and balance, which they believe to translate into promise for the future of their everyday lives.

"...My fitness and balance improved significantly which helped in my everyday life. I was able to do more in everyday life without feeling tired." – Participant 3

"I have more of a can-do attitude, it's given me a belief in my future independence. I now believe I can live independently in the future." – Participant 10

Furthermore, one participant identified frame running as their 'escape' and explained that it gives them the opportunity to be seen as 'normal':

"On my bike I feel free and just like everyone else. I'm not confined to my wheelchair and don't need to worry about falling over. Frame running is my escape in a way." - Participant 5

The idea of participants' identifying themselves as 'normal' when frame running is important to note as the feeling of being 'different' or isolated compared to those without disabilities can negatively impact psychosocial wellbeing. Self-concept is an important but complex and multidimensional construct, and the responses from the current study suggest frame running participation may help in the development of aspects of self-concept, and more specifically 'athletic identity'. Furthermore, an athlete's athletic identity has been reported to increase with time spent participating in physical activity and sport, as well as increasing in higher levels of competition (Groff et al., 2009). The responses from the participants in the current study are in line with this finding, as the athlete's referring to their athletic identity have many years of experience and compete at international level, Therefore, responses show that frame running participation can impact some aspects related to self-concept, such as athletic identity and ability, but the multifaceted concept is complex and may cause confusion among the participants.

6.3.7 Boosting Self-esteem

Question six explored the impact that frame running has had on the self-esteem of the athletes, which is important given an individual's sense of self-worth can be an important indicator of their psychological wellbeing. Two participants did not respond to this question, and two participants felt that frame running has not had an impact on their self-esteem, as they have always been confident.

The main theme identified from thematic analysis was the 'boosting of self-esteem' with the subtheme of a 'sense of achievement'. Participants felt that participation in frame running has boosted their self-esteem and helped them to believe they are capable of doing and achieving more than they thought was possible. Furthermore, they felt they were able to see their hard work pay off in training, competing, and outside of frame running.

"I have definitely grown in confidence. I train really hard and definitely get a boost of self-esteem each time I see that hard work paying off." – Participant 5

The sense of achievement gained from frame running has given participants the feeling that not only are they able to participate, but the '*belief in the fact that I am good at things.*' The boost in self-esteem along with the belief that the participants are not just able to participate but are good at tasks and/or activities provides participants with a better outlook on life.

"Yes definitely. I think this goes back to the sense of achievement I've felt doing frame running. Each year, I have gotten better and better – whether that is lowering my times or improving fitness/strength. Doing frame running and seeing the rewards by working hard has encouraged me to get involved in other things such as charity work and helping with the development side of frame running in turn boosting my self-esteem as I have realised by becoming more involved in this work, I can help make a difference to people's lives." – Participant 3

The above response shows, for this participant, participation in frame running has given them a sense of achievement in various dimensions. Furthermore, a 'boost' in self-esteem has led them onto trying new things and in turn passing on their experience to others, thus recognising the impact that frame running has had on their self-esteem. The suggestion of a boost in self-esteem from frame running participation is important to note because of the assumption in previous research that individuals with CP have lower levels of self-esteem compared to those without disabilities. Furthermore, with a perceived improvement in self-esteem, it may be plausible to assume that the overall confidence of the athlete has also increased, which may further enhance their psychosocial wellbeing.

6.3.8 Positive Impact on Psychological Wellbeing

Question seven looked at the impact of frame running on individual's psychological wellbeing, including any perceived impact on the individual's mood, behaviour, and mental health factors, including stress, anxiety, and depression. One participant did not respond to this question. The main theme identified was 'positive impact on psychological wellbeing' and this was highlighted with all nine responses indicating that frame running has had a positive influence on their psychological wellbeing. The thematic analysis identified the subthemes of 'access to suitable physical activity', 'mood boosting activity', and 'importance of social interaction'. The participants had differing responses, showing a varying impact but overall, it was positive.

Participants highlighted the importance of accessibility to suitable physical activity opportunities is vital for positive psychological wellbeing by referencing the use of the frame running bike as an escape when feeling down.

"My mood levels certainly improve after I have been out for a run or walk" – Participant 8

"Yes...if I'm feeling down before I get on my racerunner, afterwards I feel 100% better and more positive." – Participant 6

Access to a suitable activity can be one of the main barriers to physical activity and sport participation in individuals with CP, especially those with more severe CP, as opportunities are more restricted for those with lower functional ability. Poor accessibility can lead to inactivity and the associated health risks, including poor psychological wellbeing, therefore it is important to identify a suitable opportunity to break a cycle of deterioration. Furthermore, individuals with CP are at a higher risk of mental health disorders such as anxiety and depression (Whitney, 2018), thus promoting an activity to help alleviate the risk, such as frame running, is important for future rehabilitation programmes.

"It is a mood booster and helps with my anxiety in daily life and my routine." – Participant 1

This participant indicates the help that frame running gives in dealing with their anxiety in daily life which is important for moving forward and improving QoL, providing further indication that frame running can have a positive impact on psychological wellbeing. Furthermore, some of the athletes were more general in their responses noting that frame running had a positive influence on their mood or was an escape for them when they were feeling down, which are all positive influences on psychological wellbeing. Furthermore, the way in which participants view frame running, and how it can impact psychological wellbeing

is important given the emphasis on mental health and psychological benefits of physical activity and exercise. The recognition of frame running as '*amazing mental health tool, more than any other sport*', alongside '*moods are definitely better after exercise*' and '*it's a mood booster*', indicates the importance participants place on frame running for positively impacting their psychological wellbeing.

Previous research has shown that social interaction through sport participation can also help with the development of psychosocial wellbeing (Martin and & Smith, 2002) and the importance of social interaction with the group environment of frame running was also referenced.

"Keeps me going when I get down, however before I joined the club, I was training on my own which I did not like and began to miss friends." – Participant 2

The feeling of loneliness while training on their own highlights the importance of social interaction for positive psychological wellbeing, and that the physical aspect of frame running solely is not enough for developing a good psychological health. Furthermore, it highlights there are various factors that can/need to interact for a positive psychological outcome (e.g., the environment, social group, and physical activity).

The ups and downs of progressing in frame running through different levels of competition (i.e., local to national to international), were expressed by one participant.

"It has had a massive impact on my psychological wellbeing. One of the things I remember the most from my childhood was my inability to participate in sports my peers did. I used to get so frustrated, and I was fed up coming last at everything. However, when I discovered frame running, it gave me something positive to put my negative energy into. I especially enjoyed beating people and I relished no longer coming last! Now as I'm internationally, there is quite a lot of pressure to perform and constantly improve as you would expect so I'd say the impact has changed over time but still remains overall positive as I'm doing what I love." – Participant 3

The above participant highlights the importance of frame running on their psychological wellbeing from the very beginning and the impact of frame running participation has varied as they progressed through levels of competition. Previous research has shown that the psychological wellbeing of elite athletes with disabilities can be associated with the level of competition they are participating in (Campbell & Jones, 1995). The athletes participating at international level reported a better psychological wellbeing with lower levels of anxiety in comparison to athletes competing at regional level, suggesting once athletes begin competing there may be added pressures which can negatively impact psychological wellbeing, and this is then improved on as experience grows. The negative aspect of elite level frame running on psychological wellbeing is also referenced with strains on mental

health coming from pressure to constantly improve in training and competitions. As the only reference to the varying pressures on psychological wellbeing of competing at international level, the above quote shows the difference between the initial positive impact from participation with friends etc. and the pressures when competing at the highest possible level. The responses for the current study are in line with previous research showing disability sport participation to improve the psychological wellbeing of athletes (e.g., Firoilli, et al., 2013), thus suggesting frame running participation can have a positive impact on the psychological wellbeing of participants.

6.3.9 Positive Influence on QoL

Question eight explored the impact frame running participation has had on the athlete's perceived QoL including their standard of living, health, and comfort. The main theme identified through thematic analysis was the 'positive influence on QoL', and thematic analysis also identified two subthemes 'improved academic life' and 'perceived increase in standard of living'. One participant did not respond to the question.

All nine responses referred to the way in which frame running participation has impacted on physical aspects and in turn improving their standard of living. Participants referred to '*improved muscle tone and tightness*', '*keeping fit*', '*being their most mobile*', and '*improved strength*.' Further to the physical aspects, participants referred to the impact frame running has had on social and emotional aspects, with responses referring to '*being happiest*' when on the racerunner and '*feeling more confident*'. Furthermore, the combination of making friends and being able to participate and compete in a physical activity are aspects which were also previously mentioned to other questions, highlighting the importance placed on these areas of life by the frame running athletes. This may be down to the lack of opportunity to experience these things as an individual with CP, and something that this population may not have had the opportunity to do without frame running. Participants referred to how frame running has impacted their QoL on a macro-level.

"My health has improved, before my attendance at school was always in 80% range, leaving school my attendance was 98%. It's also opened so many doors for me, I'm now an ambassador for a charity and [X]'s sport personality." – Participant 10 (edited to protect anonymity)

The impact of frame running on various aspects of life to improve aspects of academic life (e.g., mobility, pain relief, self-esteem, or self-efficacy) indicates the wide-ranging impact that frame running can have. Furthermore, participants referred to the opportunities that frame

running has given them away from the track also, and this is highlighted by the response below.

“Frame running completely changed my life. It has given me a purpose in life and always a goal to work towards. I was very lucky to discover frame running when I did and at a young age and has been a significant part of my life. Frame running has unlocked so many avenues for me on and off the track. It seems as if everything I do is because of frame running in one way or another.” – Participant 3

Frame running has clearly improved the standard of living of the participants in numerous ways, which has been recognised by the participants, and so they wish to use that to pass on to others whether it is as a ‘charity ambassador’ or to ‘help motivate them to take up frame running despite being disabled’. The impact from frame running on the QoL of the participants provided a wide range of responses, and this may be down to various interpretations of the definition of ‘QoL’ by the participants. Previous research has suggested that participation in physical activity can improve performance and participation in day-to-day activities for individuals with CP, and an increased level of participation and performance has also been linked to a higher level of satisfaction (Mesterman et al., 2009), and this study has shown that the same can be said for Frame running participation.

6.3.10 Athlete’s Final Thoughts

A final opportunity was given to the participants at the end of the survey to comment on their perception of the influence of frame running on their lives, whether it was positive or negative, and three of the ten participants responded to the question. A comment regarding the accessibility of frame running was made in reference to the cost of a frame running frame as they are expensive pieces of equipment (currently approximately £1300 per bike in Northern Europe). The accessibility to frame running is important because having suitable opportunities for the CP population to be physically active at all levels is a necessity to overcome the barrier of physical inactivity. One participant reiterated the importance of the social interaction with friends they have made as a result of frame running because there may not have had that opportunity to make friends in a similar environment if frame running was not accessible. Finally, one participant highlighted the major impact that frame running has had on their life by stating, *‘overall frame running has changed my life for the better and opened up endless possibilities for me that wouldn’t exist otherwise’*. This response, despite being individual and not generalised to all frame running athletes, indicates the potential that frame running holds for individuals with CP (and related conditions) to impact their life in a variety of ways, whether it is by improving mobility, psychological wellbeing, social interaction, community engagement, or another way, the influence can be life changing.

6.4 Limitations & Future Directions

There are several potential limitations of the current study. Firstly, the use of an online survey means there was no interviewer/researcher present, and if there was a misunderstanding in relation to a question or an instruction, there is not the same opportunity for clarification, and this can lead to a dilution of the data quality. The fact that some respondents left some questions unanswered may indicate that they were not sure about what those questions were asking them. Additionally, the interviewer/researcher not being present could influence the motivation of the participant to complete the survey online due to a lack of interaction, and this may lead to selection bias (Reja et al., 2003). In addition to a possible lack of motivation, or inability to clarify any misunderstanding, the lack of presence of an interviewer/researcher means that 'probing' is not possible. The use of 'probing' as a research technique can be beneficial as it can elicit additional and possibly more extensive information, which is beyond the participant's initial answers (Brantley, & Wogalter, 1999). Each of these points mentioned highlight the importance of the design of the questions and the overall design of the online survey. Given these constraints, the piloting of the survey as part of this study was especially important.

Furthermore, a limitation in relation to the thematic analysis process, the presence of only one researcher analysing and interpreting the data may have led to some bias in the identification and interpretation of the themes. However, in the context of qualitative analysis, confirmability and dependability are more important and these are established through detailed description of the method, with transparency about each step, and the researcher being open to audits of the data by another researcher.

Further to the limits of the survey design, was the potential for language barriers, as English was not the first language of all the participants. This may have led to some misunderstandings of the questions, or athletes not being able to fully express their point of view and experiences.

A further limitation to the current study was the recruitment of participants (see section 3.5). Firstly, the sample comprised of athletes who are already frame running, as the recruitment came through the frame running community, this is likely to introduce a bias towards positive responses about frame running. Furthermore, the recruitment process coincided with the beginning of the COVID-19 pandemic meaning access to potential participants at competitions was no longer possible. Therefore, the study was reliant on snowball sampling, and with the unknown situation individuals were placed in (e.g., lockdowns and/or shielding) potential participants may not have felt inclined to participate and answer questions about their QoL and psychosocial wellbeing during unprecedented times.

With reference to future research, given the study was solely qualitative, a quantitative study with domain specific outcome measures, such as myTREEHOUSE Self Concept Assessment (Cheong et al., 2017) with a more focussed sample (e.g., international athletes only) would provide additional information on the impact of frame running participation on psychosocial wellbeing for athletes with CP. Furthermore, similar to previous research (e.g., Ferriera & Fox, 2008), research comparing the relationship between variables such as competitive level, and length of career and the effect on psychosocial wellbeing (e.g., self-esteem and self-concept) of frame running athletes and athletes without disabilities would provide an insight into the athlete's psychosocial wellbeing in both adaptive and able-bodied sport. With the previous research suggesting self-esteem is lower for individuals with CP, and the little research involving CP and self-concept, it would provide a valuable comparison with able-bodied athletes and the impact of adaptive sport on psychosocial wellbeing of athletes. Additionally, a longitudinal study studying the impact of participation on psychosocial wellbeing, tracking athletes from when they first start frame running, and follow them as they remain or dropout of the sport. This will add another viewpoint, capturing experiences of athletes as they have progressed and dopped out, however such projects are expensive because of the duration.

6.6 Conclusions

The aim of the current study was to explore the perceived impact of frame running participation on the QoL and psychosocial wellbeing of competitive athletes. Previous research has shown participation in sport at high-level competition can have a positive influence on QoL (e.g., Sproner et al., 2009; Groff et al., 2009), and psychosocial constructs such as self-concept (e.g., Ferriera & Fox, 2008), self-esteem (e.g., van de Vliet et al., 2008), self-efficacy (e.g., Adhem et al., 2010), and psychological wellbeing (e.g., Fiorlli et al., 2013). However, the evidence base is still relatively small, both from quantitative and qualitative research, and there was no existing research exploring the impact on psychosocial wellbeing from frame running participation, thus the current study is novel research.

The results from the current study provide an insight into frame running as a growing sport, but also add to the literature regarding the QoL and psychosocial wellbeing of athletes with disabilities. Through thematic analysis, athletes identified frame running to have had a positive influence on their social development through the opportunity to build and develop friendships, by meeting people around the world, while having a sense of belonging and inclusion in a sporting environment, which are all social aspects which they may not have thought were possible prior to frame running participation. Furthermore, athletes suggested

frame running to have had a positive influence on their psychological development and wellbeing by developing aspects of their self-concept (e.g., athletic identity), and improving their self-confidence and self-esteem. Frame running participation enabled athletes with a more positive outlook on life, sense of achievement, and provided them with access to a physical activity/sport that allowed them to manage their psychological wellbeing (e.g., anxiety).

Frame running participation is reported to have an influence of the athlete's QoL, by providing them the opportunity to experience new things in life, such as competing at international competitions, but also by enhancing the athlete's sense of independence thus improving their perceived standard of living and QoL. Hence the results regarding frame running are important for the design of future interventions, therapeutic and rehabilitation programmes. Furthermore, the thematic analysis, and use of the ICF framework, contributed to the understanding that frame running participation can influence the body function, and activity and participation components of the ICF framework highlighting the broad impact frame running can have. The constructs focused on, and the detailed answers given. The results from the current study are from current athletes with CP who compete at a local level and above in frame running, thus cannot be generalised to the wider CP and frame running population. However, some of the athlete's responses were anecdotal and made reference back to the early stages of frame running, suggesting frame running may enhance QoL and psychosocial wellbeing across all levels of participation.

Chapter 7: General Discussion

7.1 Recap of research – Aims and Research Questions answered

Overall, the aim of the thesis was to provide insight and baseline evidence for frame running, and the effects on the psychosocial wellbeing of children, young people, and competitive frame running athletes with CP.

7.1.1 Research Question 1 – What domains & factors are viewed as the most important for parents, health care professionals, and frame running coaches for evaluating physical activity interventions?

Prior to examining the effects of frame running, study 1 aimed to answer the question of which domains and factors were viewed as most important to measure when evaluating the effectiveness of physical activity interventions for children and young people with CP, as perceived by parents of children with CP, health care professionals and frame running coaches.

Over the two round Delphi study, enjoyment (4.85), participation (4.62), and psychological wellbeing (4.62) were reported with the highest mean ratings and were thus viewed as the most important factors to measure. However, the results show the participants did not reach a consensus regarding one factor being the most important, with ratings of 4 and above (out of 5) for 82% of the factors, suggesting participants viewed the majority of factors as important, which is in line with previous research (e.g., Vargus-Adams & Martin, 2009). The results suggest the responses may be dependent on the individual and/or the situation when trying to decide which factor is the most important to measure. Furthermore, this may provide reasoning for why participants placed less importance on physical factors than psychological and social-emotional factors, with the four lowest rated factors being physical.

The physical abilities and development of children and young people with CP can vary quite drastically, particularly as severity of CP increases (e.g., GMFCS I compared with GMFCS III). Therefore, focussing on physical factors can be individualised (e.g., lower limb strength; hand mobility; gait pattern etc.) in comparison to psychological and social-emotional factors which can be viewed as more universal. For example, most parents and health care professionals can recognise the importance of good psychological wellbeing or being involved in a fun and enjoyable activity, but the child's leg strength may not be as much of a priority if they are ambulatory. With the view of psychological and social-emotional factors being more universal, alongside having higher rating of importance compared to the physical factors, and a gap in research regarding physical activity and psychosocial

wellbeing in children and young people with CP, these factors were chosen as the focus of measurement for the frame running training programme.

7.1.2 Research Question 2 – Does participation in a 12-week frame running training programme affect the perceived QoL of children & young people with CP?

The aim of the 12-week frame running training programme was to provide baseline information on the impact of frame running on the psychosocial wellbeing of novice racerunners with CP. One of the important factors to measure as an outcome, as perceived by participants in study 1 and evident in previous research, was QoL. Across the five participants, it was unclear what effect frame running had on the aspects of quality life measured in the current study. Participants showed no real pattern for daily and school activities subscales. However, the three physical subscales: movement and balance, pain and hurt, and fatigue, did appear to show a trend toward improvement post frame running training programme. However, it can be difficult to pinpoint factors that may have contributed to an improvement in an aspect of, or overall QoL. Furthermore, the definition of QoL can vary between individuals, and so making a generalised assumption on whether QoL has improved or not can be difficult. Therefore, the current research suggests frame running participation may have a positive influence on the physical aspects measured, but it is not possible to definitively say the frame running participation was solely responsible for the trend towards improvement, as there are many other factors which may have contributed to the results.

7.1.3 Research Question 3 – Does participation in a 12-week frame running training programme effect the self-efficacy, self-esteem, self-concept, & psychological wellbeing of children & young people with CP?

There is no previous research exploring the effects of frame running on the psychological factors of self-efficacy, self-esteem, self-concept, or the psychological wellbeing of children and young people with CP, thus the current research is offering novel insight. In comparison to physical factors, psychological factors can be more difficult to measure as the researcher is reliant on the participant to report how they truly feel. Furthermore, this may be more difficult for individuals with CP who have cognitive impairments and/or learning difficulties, which can lead to difficulty in understanding the questions, but also in identifying how they are truly feeling, which can pass on the responsibility to proxy reporting, and the results are then solely on the perception of the proxy report. In the current study, both self- and proxy-report were used to provide a more rounded insight.

Self-efficacy, as previously discussed, is an important factor in promoting health behaviours, including physical activity, and also plays an important role in the theoretical frameworks linked to behaviour change, such as Social Cognitive Theory, and the Transtheoretical Model. The results from the current study showed participants reported improving levels of both social and emotional self-efficacy post frame running suggesting the social aspects of frame running participation may have had a positive influence on the self-efficacy.

With reference to self-esteem, the participants reported frame running participation had no adverse effects on their self-esteem and can even have a positive impact on the self-esteem of participants with 80% reporting higher self-esteem post frame running. Another positive for the current study was the participants showing a trend towards improvement for their psychological wellbeing, which is important to note as children and young people with CP can be at greater risk of mental health disorders.

In reference to self-concept, there was no clear pattern with the effects of frame running participation on various aspects of self-concept, however a clear picture on 'global' self-concept can be difficult given the multidimensional nature of self-concept. On the other hand, two participants did report an improvement in some aspects of self-concept that may be related to being physically active (i.e., frame running participation), namely athletic competence and physical appearance. Overall, the results suggest frame running participation may have the potential to improve psychological aspects of children and young people with CP, such as self-esteem, but it is difficult to attribute the results solely to frame running. However, as novel research it provides an insight to the impact frame running might have on the selected psychological factors, which may contribute to an improved QoL over time.

7.1.4 Research question 4 – Does participation in a 12-week frame running training programme have an effect on the participation and enjoyment of other activities for young people with CP, & is frame running perceived as an enjoyable physical activity for children & young people with CP?

Physical activity participation is important for children with and without disabilities for their short- and long-term physical and psychological health, but participation can be restricted by various barriers (e.g., physical ability; lack of time or facilities). Individuals with CP may encounter more barriers, meaning their opportunities to participate are restricted further, thus highlighting the importance of providing suitable opportunities. The current study examined whether frame running participation impacted participation of other activities. The results did not show any clear pattern linking frame running participation to further

participation in other activities. However, what the results of the study showed were that participants had a low diversity of physical and skill-based activities compared to other activity types, but they reported high levels of enjoyment for the physical and skill-based activities.

Previous research suggests participants are more likely to report higher levels of enjoyment when participating in activities they are participating in out of choice (King et al., 2006). This is similar to the current research as the participants, who had chosen frame running, reported an average enjoyment rating of 5 (out of 5). This highlights that frame running can be an enjoyable activity for children and young people with all severities of CP, thus providing them with another opportunity to be physically active. The necessity for more suitable opportunities to be available to the CP population was also highlighted by the low diversity of physical and skill-based activities reported by participants. However, the low diversity scores are to be interpreted with caution as there may be a variety of factors leading to low diversity scores, and not just a lack of suitable activities (e.g., lack of time; fear of failure; lack of capacity for other activities).

7.1.5 Research Question 5 – What impact has frame running had on the psychosocial wellbeing of competitive frame running athletes?

Previous research has shown that participation in sports at a national level and above, which provides a competitive element, can have psychosocial benefits for athletes with disabilities (e.g., Muraki et al., 2000; Sproner et al., 2009). The current research aimed to gather qualitative data from competitive frame running athletes to explore the impact they perceive frame running to have had on their psychosocial wellbeing.

The thematic analysis showed the participants felt frame running had a positive impact on their psychosocial wellbeing, as well as bringing them new life experiences on and off the track, such as competing at a higher level, and making friends around the world. Furthermore, athletes referenced participation to positively impact psychological factors, including their self-efficacy, self-esteem, self-concept, and psychological wellbeing. Moreover, they reported that frame running had also had a positive influence on their social development. The anecdotal data is in line with previous research which has explored the impact of participation in competitive sports on athlete's psychosocial wellbeing, but the current study is novel research, providing new data for frame running.

7.2 All Roads Lead to Quality of Life

Cerebral palsy has been linked to a lower QoL due to the movement disorders, and associated impairments including cognitive, communication, learning, emotional, and

behavioural difficulties and epilepsy (Markis et al., 2019). A decreased QoL has meant that QoL is becoming an increasingly important outcome to measure for rehabilitation and therapeutic programmes, and interventions for the CP population, and this can be because QoL is not just one aspect but is viewed as a multidimensional construct. Furthermore, the multidimensional nature of 'QoL' can make it difficult to define and measure, with the World Health Organisation (WHO) defining QoL, at the very least, as '*an individual's perception of their physical state, their affect, and the interpersonal relationships and social roles in their life*' (WHOQOL Group, 1995; p.3). The definition from the WHO highlights QoL is a broad construct, and that both psychological and physical aspects can contribute to an individual's QoL, thus, a positive QoL is not always dependent on the severity of impairment (Rapp et al., 2017).

Overall, what parents and carers aim for is their child to have the best QoL possible, irrespective of their level of impairment, and they will typically do what they can to improve it. QoL can be measured as a multifaceted construct, as in the current research (see chapter 5), with instruments focussing on various aspects to provide the researcher with an insight to the perceived QoL of the participant, and this can be done with condition specific tools (e.g., PedsQL-CP module; CPQoL), or more general QoL tools (e.g., Child health questionnaire). However, QoL measures provide a snapshot of how the individual is feeling at that moment in time, and with the CP population there can also be a reliance on proxy reporting where individuals do not have the ability to self-report. Therefore, given the importance of QoL, a reliance on determining QoL based on a single set of results may not be presenting the most reliable picture.

Throughout the thesis, and results of each of the three studies, the QoL of individuals with CP has been a consistent theme and been referenced as the 'holy grail' of outcome measures (McDougall et al., 2010). It has been referred to as the 'holy grail' because it is thought that numerous factors can be linked to, and impact an individual's QoL, whether it is physical, psychological and/or social. Participants of the Delphi study reported QoL as the fourth most important outcome to measure for physical activity interventions for children with CP, but also made reference to their view of '*all aspects can impact QoL*'. On the other hand, the results from the frame running programme (study 2) showed no clear evidence that frame running participation solely could improve QoL based on the results on the PedsQL-CP module, with no clear pattern across participants and only a trend towards improvement in some physical aspects of life (e.g., movement and balance). However, the results of the psychological factors, showed reported improvement in self-esteem, psychological wellbeing, and some aspects of self-efficacy and self-concept, which could be said to be contributing to an overall improved QoL, but may not be as clear cut as a physical factor, such as better functionality.

The psychological impact on QoL may, in some instances, play a more important role in improving QoL, for example, higher self-efficacy can provide the individual with more belief to participate in more activities whether it is day-to-day, or a new physical activity, thus enhancing their physical and social ability and independence; it can be a cyclical process, all aiming to improve the QoL. The results from this thesis would suggest frame running participation may have a positive impact on the QoL of children and young people with CP, whether it is being measured directly (e.g., PedsQL-Cp module) or indirectly via other factors (e.g., psychological wellbeing or social interaction). These thoughts are echoed by the frame running athletes in the third study, with participants referring to a variety of physical (e.g., improved strength), psychological (e.g., more self-confidence), and social (e.g., making friends around the world) factors which have improved their QoL, and have attributed to frame running. There was no direct reference of 'QoL' but the recognition that their QoL can be improved by a variety of factors suggest that frame running can be considered as an option for exercise and/or research involving rehabilitation, therapy programmes, or physical activity interventions aiming to impact children and young people's QoL.

7.3 Frame running – An Adaptive Activity for All Levels of Impairments

Individuals with disabilities can face barriers to physical activity participation all the way through childhood and adulthood, which can lead to them being inactive, and impact on their physical and psychosocial wellbeing. Children and young people with CP may encounter a variety of barriers to their participation, which can change through development, for example, risk of injury, fear of fatigue, lack of time and/or finances, unsuitable activities, not having a full understanding of their disability, mobility impairment, or lack of suitable activity (Verschuren et al., 2016). It is the lack of a suitable activity which is a theme throughout the current thesis, as the case for frame running as a safe and suitable option of adaptive activity for children and young people with CP of all severities is presented.

Previous research has shown that there are options for children and young people with CP to be physically active, such as swimming, strength training, or circuit training (Novak et al., 2013), but the activities and/or sports are not always suitable for everyone, particularly those with more severe CP (i.e., GMFCS IV and V). Participants from study one reported they wish children with CP to have access to more suitable activities, but not solely for physical benefits. Social inclusion, building relationships with like-minded people, and not being perceived as the 'odd one out', particularly when exercising and/or socialising with able bodied peers, were all suggested by participants as why it is important for individuals with CP to have access to a suitable activity.

Previous research has shown the variety of activities available to children and young people with CP can provide physical and psychosocial benefits, but there was a noticeable lack of inclusion of participants with more severe CP. This may be due to the activity/sport not being suitable because of their physical or cognitive capacity, or individuals are excluded from these types of studies because it can be difficult to assess the exercise effect on athletes with standard research equipment (e.g., in the case of high intensity circuit training; Schranz et al., 2018). On the other hand, the physical activity opportunities for those with more severe impairments, such as swimming, therapeutic horseback riding, and hippotherapy, have reported positive physical and psychosocial effects (e.g., Aidar et al., 2007; Debuse et al., 2009) , and the growing research on frame running has also made a case for it being another option for physical activity and exercise for children and young people with CP, including those with GMFCS levels IV and V (van Der Linden et al., 2018).

Furthermore, frame running research has shown participation can have physical benefits (e.g., O'Donnell et al., 2012; Bryant et al., 2015), and has been reported as an enjoyable form of exercise (e.g., van Schie et al., 2015; Bryant et al., 2015). The current research provides further evidence that frame running is a suitable activity for children and young people with CP of all severities and can potentially have a positive impact on their psychosocial wellbeing. In comparison to other activities/sports, particularly for those with more severe impairments, frame running provides the opportunity to be active at a higher level of intensity which is important as these individuals may be wheelchair bound and living an inactive lifestyle. Furthermore, one area of orthopaedic concern in relation to swimming is its non-weight bearing aspect, while frame running provides weight bearing activity and has been shown to improve the bone density of participants (Bryant et al., 2015), suggesting frame running as a good alternative or complementary activity.

Alongside potential physical and psychological benefits, frame running may also help with social development. Children and young people with CP can struggle to 'fit in' and feel socially isolated around able-bodied peers because of their disability, which can influence their activity levels and lead to a more inactive lifestyle despite the well documented benefits of being physically active. By contrast, current competitive frame running athletes reported how important frame running was for them socially, as it allowed them to meet like-minded people, and build relationships locally and worldwide. Furthermore, the current athletes highlighted that frame running provides participants of all severities the opportunity to participate and compete at competitions up to the most elite, the Paralympic Games. Competing is one aspect of disability sport that the more impaired athletes are less likely to experience, but frame running can provide them with this opportunity. Thus, the current research provides evidence that frame running should be considered as a suitable opportunity for physical activity for individuals with CP, and more importantly is accessible to

those with more severe impairments, whose opportunities to gain physical and psychosocial benefits from physical activity are even more restricted.

7.4 The Importance of Enjoyment

A key finding of the current thesis was the importance of enjoyment for physical activity and frame running participation in particular, which was highlighted by the high enjoyment ratings across the participants' frame running training programme in study two. Frame running being recognised as an enjoyable physical activity is in line with previous research (van Schie et al., 2015; Bryant et al., 2015), and this was further emphasised by the importance placed on enjoyment by the current frame running athletes, and the role it played in encouraging them to keep participating. Furthermore, enjoyment was rated by the participants in study one as the most important factor to measure when evaluating the effectiveness of physical activity interventions for children and young people with CP. This highlights the desire for the children to be involved in a physical activity they perceive as enjoyable because, apart from seeing the child happy, it can encourage them to continue participation post intervention, rehabilitation, or therapeutic programmes, which is important as there can be a drop off from physical activity when it is not part of a structured programme (Law et al., 2006).

Importantly, frame running being recognised as an enjoyable activity can facilitate participation. As previously discussed, enjoyment can be a key facilitator to physical activity participation (Verschuren et al., 2013), and can play an important role in promoting health behaviours including physical activity. For example, the Self-Determination theory suggests that enjoyment is a key component of intrinsic motivation, and high intrinsic motivation for a behaviour means it will be performed without reinforcement from an outside party (Ntoumanis, 2005). Therefore, the perception of frame running being an enjoyable activity can encourage children and young people with CP to be physically active and experience the short- and long-term benefits of being physically active such as positively influencing psychological and general health (Wankel, 1993; Jin et al., 2018).

7.5 Psychosocial Wellbeing and Frame Running

It is well documented that children and young people with CP have a lower wellbeing in comparison to their typically developing peers (Livingston et al., 2007). The research looking into the positive impact that physical activity participation can have on the physical wellbeing of children and young people with CP is also well documented, but there is less evidence of the impact on psychosocial wellbeing. Furthermore, it has been assumed that psychosocial wellbeing correlates to functional ability, thus improving an individual's functional ability

through being physically active may improve their psychosocial wellbeing, however this is not always the case (e.g., Pirpiris et al., 2006).

Throughout the current thesis the psychosocial wellbeing of children and young people with CP was an important component. The frame running training programme showed that participation can have a positive influence on some aspects of psychosocial wellbeing, including psychological wellbeing and self-esteem. Furthermore, some aspects of self-efficacy and self-concept showed improvement post frame running, however it is not possible to attribute the changes in these factors solely to frame running participation.

These are important findings because of the influence these factors can have on an individual's life. For example, self-esteem is vital for personal development, and those with lower self-esteem are thought to be at more risk of physical and mental health problems, hence the observation of a trend towards an improvement in self-esteem is a positive result in the current study (Trzensniewski et al., 2006; Brooks, 1992). Furthermore, the importance of self-efficacy in physical activity, health behaviours, and overcoming barriers has been discussed throughout the thesis, so a positive perception of self-efficacy can help the individual hold onto their belief that they can achieve their goals. The experiences and achievements of an individual can affect their self-efficacy, and in turn their level of motivation, and how they can take on their difficulties (Bandura, 1997). Therefore, the trend towards improvement in aspects of self-efficacy post frame running participation in the current research adds to the small number of studies investigating CP and self-efficacy (Dunn & Dunn, 2006).

Overall, the results show positive changes in psychological wellbeing that may be a result of participation in frame running, which is supported by the qualitative data provided by the current frame running athletes in study three. Athletes emphasised the importance of frame running for their psychosocial wellbeing, with responses referencing various aspects of psychosocial wellbeing, including their athletic identity, self-esteem, psychological wellbeing, and the all-important QoL. Although the qualitative data comes from a small sample, the responses provided insight into how the frame running athletes perceive the impact on their psychosocial wellbeing.

Furthermore, in combination with the quantitative data from study two, the research suggests frame running participation can have a positive influence on the psychosocial wellbeing of children and young people with CP of all severities. As previously mentioned, there is a gap in the knowledge of psychosocial wellbeing and physical activity for children with CP, thus the current research provides a positive insight into frame running participation, and potential benefits of frame running should be made clear to parents and healthcare professionals to encourage participation, which can help promote frame running programmes and other physical activity programmes.

7.6 The Results and the ICF-CY, SDT, SCT, and TTM

The findings from the current thesis provide new insight into the impact of frame running participation on the QoL and psychosocial wellbeing of both novice and competitive athletes. The following section will relate the findings to the International Classification of Functioning, Disability and Health for Children and Youth (ICF-CY), and the theoretical findings underpinning the thesis; the Self-Determination Theory (SDT); Social Cognitive Theory (SCT); and Transtheoretical Model (TTM). As discussed in section 2.3.1, the ICF-CY framework is useful in helping describe the current levels of health, function, activities, and participation for individuals with CP (Trabaca et al., 2012). The importance of the ICF-CY is how it presents a common language that can be used by professionals and across disciplines, which may be important for future promotion of frame running. The results from the current thesis are distributed across the body function component (e.g., mental functions through improved self-esteem), activities component (e.g., self-care through a sense of independence), and the participation component (e.g., building friendships). The results did not cover the body structures function as no physiological measures were conducted, however it is important to note that previous research has shown positive impact on body structures (e.g., van der Linden et al., 2018). The distribution of the results can help in describing the functioning and health of the individuals with CP, and the components of their health and functioning perceived to be impacted by frame running participation. Additionally, the results distributed across the ICF-CY can show the functioning of individuals from different perspectives (e.g., body functions; Trabaca et al., 2012), as well as show the complex interactions between the components due to the interactional nature of the framework suggesting the interactions frame running participation can have on an individual's health and functioning.

An important result to take from the thesis is the perceived level of enjoyment reported by participants in both study 2 and 3. This is an important finding in relation to the SDT, a prominent theory of motivation. A feeling of enjoyment can be a contributing factor to intrinsic motivation and therefore encourage participation. Intrinsically motivated activities are of benefit because the participant is not looking for a reward and participates because of personal rewards and satisfaction (Ryan et al., 2008). Furthermore, the results from study 3, suggest that frame running participation can support the three basic psychological needs of competence, relatedness, and autonomy (Teixeria et al., 2012). The Basic Psychological Needs Theory, a sub-theory of SDT, suggests each of these basic and innate psychological needs require support for motivation, and if there is a lack of support for these needs then motivation levels can fall and portray the activity as unattractive. Therefore, the perceived

presence of support for the three basic psychological needs suggests frame running can support motivation for participation.

In relation to the social cognitive theory, a pivotal construct of this framework is self-efficacy, and though no clear results were found in study 2 regarding self-efficacy, themes identified in study 3 suggest that frame running participation may have a positive influence on self-efficacy, thus having an impact on health behaviours (e.g., physical activity), and other factors of the SCT, outcome expectations and socio-cultural factors. Through a perceived influence on self-efficacy the results suggest frame running participation can positively impact the social outcome expectations (e.g., building and developing friendships; and a sense of inclusion) and self-evaluative outcome expectations (e.g., life changing experiences; and a growing sense of independence). The outcome expectations relate to the individual's judgement of the possible outcomes as a result of performing, thus the outcome expectations of frame running may encourage participation. Furthermore, results from the research suggest frame running participation can help with the socio-cultural factors (i.e., barriers and facilitators to a behaviour) to enhance self-efficacy of frame running whether it is psychological (e.g., boosting of self-esteem), social (e.g., the joy of meeting new people), or environmental (Involvement in a competitive environment).

In relation to the TTM, the identification of facilitators to participation in frame running suggested through the identified themes and sub-themes in study 3, as well as the high enjoyment scores from study 2, suggest frame running participation can be achieved through the stages of change. It is dependent on the stage the individual perceives to be at, but the facilitators identified can help to promote physical activity and ideally change behaviour. Furthermore, self-efficacy is also an important component to the TTM, with it thought to increase in the individual across each stage of change (Marshall & Biddle, 2001). The perceived influence on self-efficacy from frame running is important due to the array of barriers individuals with CP wanting to be physically active may encounter to be confident in their ability to be physically active. Overall, the results suggest there are aspects of frame running participation that may impact behaviour change, whether it is through motivational factors, increasing self-efficacy, or improving knowledge of facilitators and overcoming barriers.

7.7 Personal and Project Reflections

During the PhD there were many highs and lows intertwined with great experiences and valuable lessons along the way, as may be expected on the rollercoaster that is a PhD journey. The following sections will reflect on decisions made throughout the PhD with a focus on the methodological decisions, the sustainability of feasibility research moving forward with frame running, and why frame running works.

7.7.1 Methodological Reflections

From the beginning of a PhD journey many decisions must be made around the directions the research is to go to answer the research questions, and these can include questions around participants, design, outcome measures, relevance to previous research, and how the current research can fit into the literature. However, with the unpredictability of research and the possibility of logistical and time constraints, ideas and designs need to be altered with further decisions having to be made, and throughout the current PhD decisions had to be made.

In study 1, the aim was to identify which factors participants viewed as most important when evaluating the effectiveness of physical activity interventions for children with CP using the Delphi technique. The design of this study was based on previous research by Vargus-Adams and Martin (2009; 2010) which aimed to identify the domains of importance in therapeutic interventions for children with CP and therefore the initial methodology of the study was suitable. Furthermore, as this was a new area of research, the methodology was thought to be appropriate to provide some guidance on what areas participants viewed as important for physical activity interventions for children with CP. One of the main decisions to be made was the populations to include as 'experts/panelists' to be able to collect data with diverse backgrounds. In previous research, Vargus-Adams and Martin (2009) included youth with CP, parents of children with CP, and health care professionals as their groups of participants to identify a range of areas they view as important for therapeutic interventions. In the current study, the decision was made to include parents of children with CP and health care professionals as in Vargus-Adams and Martin (2009), but to include frame running coaches in the place of children with CP to provide a different voice, and one related to the focus of the PhD, frame running. Furthermore, the exclusion of children with CP as participants was to try and avoid overlap of children and parent participants responses that may occur with children with CP requiring assistance to participate given the possible cognitive and communication deficits associated with CP.

If the research was conducted again, one amendment that would be made is the inclusion of children with CP as participants as I feel it would be a valuable addition to the data collection. Furthermore, as discussed in section 3.3, it is important for children to be included in research, and even more so for those with disabilities as their voice can be underrepresented due to ableist and adultist views present in research. They are the only ones who can provide a data from first-hand experiences and give a true representation of how the population feel. Additionally, the inclusion of children with CP may have also

increased the number of participants in the study, therefore providing additional information in the exploration of the new area of research.

The second study explored the impact of a frame running intervention on the psychosocial wellbeing of youths with CP. The design of the study was single case design and collected data from participants over four time points. The decision to go with a single case design was implemented due to the small frame running population, as well as the limited access to space and equipment. The design of the study was deemed suitable for the population given the diversity of participants (i.e., age, gross motor function level, and frame running experience) thus allowing for more in-depth analysis across multiple points to identify if there was influence from frame running on their psychosocial wellbeing and QoL and provides initial insight in the relationship between free running, QoL, and psychosocial wellbeing. Furthermore, as novel research for frame running and with limited research in psychosocial wellbeing in youths with CP, there were seven constructs measured, which were informed by the previous study, and the limited literature looking at physical activity and participation and psychosocial wellbeing in this population. The decision to include all seven constructs may have been casting too wide a net, which may have had a negative methodological impact. Firstly, the use of the six measures meant that the load for the participants was high, thus limiting the number of data collection points. If the study was to be conducted again, the single case design would be maintained because it provides the researcher the ability to individually track a diverse set of participants, however, there would be a narrower focus for the outcomes, for example, self-concept only as it is a complex and multifaceted construct, thus allowing for more in-depth data collection and a smaller load on participants. Furthermore, the smaller load means a stable baseline can be established, and the lack of a stable baseline was a methodological issue present in this study. Additionally, the use of condition specific measures, such as myTREEHOUSE Self-Concept Assessment (Cheong et al., 2017), would be another methodological amendment. Some of the outcome measures used may not have been able to detect subtle changes that would be present in individuals with CP compared to able-bodied peers. The sensitivity of condition-specific measures may provide greater insight to the influence of frame running participation.

Within the third study, there were numerous methodological decisions that had to be made around the participants, how to present the questions, and how to collect the data. The constructs forming the basis of the questions for the frame running athletes were informed by the previous study to provide consistency throughout the thesis, and I feel that this is important for the study as it provides an additional viewpoint around the same constructs. The use of an online web-based survey was a methodological decision for the study. Web-based surveys were selected over interviews as they were informed by potential verbal communication deficits associated with CP, and in an attempt to gather richer and more

diverse data, web-based surveys were selected. This methodology allows participants to communicate in their own way, and their own time. Furthermore, the decision to use web-based surveys was influenced by COVID, as data collection began around May 2020. If the study was to be conducted again, I feel the use of in person, face-to-face interviews, with suitable communication abilities, may allow for richer data collection. Finally, the participants in the study were all athletes that competed in frame running at some level. The inclusion of a larger sample size, possibly novice frame runners, provides both a larger sampling pool and a comparison between athlete's perception through qualitative data.

Overall, on reflection of the methodological decisions made during the PhD, I feel the basis of them were well informed, and provide initial insight into a new area of research. However, with the methodological amendments suggested, it would provide a more focused and robust research project.

7.7.2 Is Feasibility Research Suitable?

With the limited evidence base for frame running, it is important that moving forward, the development, evaluation, and implementation process of future research is done correctly, and one aspect that can help in the development of complex interventions is feasibility research. Feasibility research appears as the next logical step for frame running research as it will provide important information which can impact the design of interventions, such as testing how acceptable procedures are, providing information on the recruitment, and informing the sample size (Craig et al., 2008; Skivington et al., 2021). Furthermore, feasibility research is often used to determine if the intervention is appropriate for further testing (Bowen et al., 2009). Therefore, the use of suitability research involving frame running would help with the design and implementation of future interventions.

As discussed in previous sections, some of the methodological issues included recruitment and sample size (i.e., acceptability), practicality (i.e., high load placed on participants), and implementation, and these are areas of focus that can be addressed through feasibility research. Furthermore, with the diversity of participants involved in frame running, feasibility research can help to inform the design of suitable interventions, whether it is an individualized randomised trial, or stepped wedge design. The feasibility research can improve confidence in moving forward with the development of interventions and help to ensure the intervention is delivered in the desired manner. As the popularity of frame running is growing and the limited knowledge base around frame running interventions, I feel feasibility research is the most suitable next step.

7.7.3 Why does Frame Running Work?

The results discussed in the thesis suggest that frame running participation may have a positive influence on the psychosocial wellbeing and QoL of individuals with CP, but why does frame running work? Previous research has shown the short- and long-term benefits of participation for individuals with and without disabilities, whether impacting on physical, psychological, and/or social aspects, I think that frame running can work in that way too. But why? In my opinion, frame running works because of the diverse areas that it can appeal to (e.g., social, psychological, and physical), and the barriers it can potentially overcome. For example, frame running offers a unique and safe opportunity for individuals may have had limited opportunities to be physically active and therefore providing them the opportunity to benefit from the short- and long-term benefits associated with being physically active, and the cyclical effect it may have on mental and physical health.

Frame running works as it can be used as a complementary activity with other options, such as swimming or cycling. It does not have to be frame running, and only frame running, it adds to the possibilities for individuals with CP. Frame running can provide opportunities for participants that they may not have had before, whether it is experiencing independence for the first time, running for the first time, or the opportunity to meet like-minded individuals from the four corners of the world. The social aspect of frame running is an important reason behind why it works. Individuals with CP can suffer from social isolation, and frame running may appear as an attractive alternative to meet people in a similar situation. Again, this may instigate a domino effect with the potential to improve physical and psychological wellbeing as they feel motivated to continue participating.

Finally, the uniqueness and adaptability of how frame running can lend itself to a recreational activity all the way up to an athletic event being showcased at the World Para Athletics championships, as it can appeal to everyone. Overall, I think that frame running works because of its ability to address the complex interactional nature of life experiences of individuals with CP, whether it is psychological, social, physical, emotional, or environmental, frame running works.

7.8 Limitations of research

There are a number of limitations in the current research, therefore the findings should be interpreted with caution. The recruitment of participants for study two was one of the biggest limitations to the research, and although the recruitment process was done over a 12-month period, and frame running clubs were contacted around Scotland and England, as well as “come and try” days organised to introduce the sport, there was still only a small uptake by participants. Frame running is still a relatively new and growing sport, so parents

may have been wary to sign their child up to an activity that they did not have experience in. Furthermore, the frame running training sessions were provided within the community, and so there may have been transport, time and/or financial barriers to participation, as these are barriers identified by previous research (e.g., Shimmell et al., 2013). Moreover, the participants were keen on frame running as they had prior knowledge about the sport and were already signed up to frame running, thus as a convenience sample the results of the frame running training programme may not be generalisable to the CP population. Future research is needed to confirm the trends observed in this thesis.

Furthermore, the recruitment and sample size for study three was also a limitation, with the recruitment process spanning an 8-month period with contact being made with the frame running athletes as potential participants at the CP World Games, a sample size of 10 was viewed as small. The reliance on gatekeepers, whether it was parents or coaches of the athletes, may have had an impact on the participant uptake. Additionally, a gap between the initial contact and follow-up contact with further information regarding the research may have caused participants to lose interest, or not be able to participate for unknown reasons. On the other hand, across studies two and three, only one participant dropped out, highlighting the participants' motivation to be involved in frame running and share their experiences, which is admirable.

There were some design related limitations throughout the research, and for a more in-depth discussion on the limitations of each study see sections 4.7, 5.5, and 6.5. The main design related limitation was a lack of control data for the frame running training programme, and therefore it is not possible to attribute changes in psychosocial wellbeing reported by participants solely to frame running. Furthermore, some participants participated in physical activity outside of frame running, again not making it possible to attribute any changes solely to frame running. The case study design provides only a small insight into the effects of frame running, but with participants ranging in age from 5 to 17 years, and in GMFCS levels from II to V, there was a substantial difference between participants in physical, psychological, and social capacity and development, making it difficult to pinpoint the impact of frame running on the different aspects of psychosocial wellbeing.

7.9 The Directions for Future Frame running Research

Based on the current research, future studies should look to incorporate control data for children and young people with CP, and a larger sample size to carry out a randomised controlled trial, if the participant numbers will allow it. This will allow for comparison of data and provide greater insight to the impact that frame running can have on the psychosocial wellbeing. Furthermore, the addition of qualitative data from novice frame running athletes

may be able to provide further insight to how the participants feel regarding the impact on their psychosocial wellbeing, and then there is not sole reliance on the quantitative data. Additionally, it may provide comparison data for study three, and show how novice athletes perceive the impact of frame running in comparison to more elite level athletes.

There were a number of physical factors identified in study one by parents and health care professionals as important to measure for physical activity interventions, thus, future research could explore the impact frame running has on physical factors, such as limb strength, or motor function. Previous research has shown physical activity participation can improve the gross motor function of children and young people with CP (e.g., Akinola et al., 2019), and research exploring the impact of frame running participation on gross motor function could add to the literature presented by Bryant and colleagues (2015).

The inclusion of participants with a more homogenous set of impairments, such as matching mobility and/or cognitive impairments, may allow for a better comparison of data, however, given the nature of CP, it may be difficult to match the participants and recruit the desired sample size. The same could be said for the age range of participants; a smaller age range (e.g., only young children, or only adolescents) would increase the homogeneity of results, but it may not be feasible given the need to maximise the number of participants for the study. Overall, frame running is a growing sport, and the current evidence base is small leaving a lot of scope for future research.

7.10 Final Conclusions

As one of the most common motor disabilities, children and young people with CP encounter complicated issues which can impact on their physical, psychological, and social development and wellbeing, and the lifelong associated motor impairments can limit their activity participation (Blair, 2010). Furthermore, the restriction in their activity participation can increase their levels of inactivity which can cause a cycle of deconditioning and barriers to participation become greater, thus providing activity opportunities to overcome barriers and break the cycle of deconditioning is important for development. Previous research has shown the physical benefits of being physically active for children and young people with CP (e.g., Reedman et al., 2017), but there is a more limited evidence base regarding psychosocial impact of physical activity, and no evidence on the impact of frame running on psychosocial wellbeing. Furthermore, the importance of psychosocial wellbeing being measured was highlighted by the Delphi study by participants, and given a gap in the knowledge, a study to examine the effects of frame running training on psychosocial wellbeing was carried out.

This is the first study to examine the effects of frame running participation on the psychosocial wellbeing of children and young people with CP. Implementing a multiple case design, five participants completed a 12-week frame running training programme and completed outcome assessments measuring aspects of their psychosocial wellbeing and QoL. Furthermore, it was important the enjoyment of frame running sessions was measured as enjoyment is a key factor for motivation and maintaining a physically healthy lifestyle.

The results suggest that frame running participation can affect aspects of psychosocial wellbeing such as self-esteem and psychological wellbeing, which is in line with previous research involving children with CP and physical activity interventions (e.g., Novak et al., 2013; Groff et al., 2009). However, it is not possible to attribute the changes reported by participants solely to frame running. Importantly, participants reported high levels of enjoyment and showed high adherence to the frame running training sessions, indicating enjoyment can be key and should be measured in future studies. Furthermore, the perception of an activity as enjoyable can increase participation and in turn the benefits associated with participation.

Additionally, the qualitative data from current frame running athletes on how they perceive frame running to have affected their psychosocial wellbeing suggests that frame running participation can positively influence psychosocial wellbeing, but future research is necessary to provide more robust evidence. Therefore, as novel research, the current thesis provides an insight into the effects frame running can have on children and young people with CP. Finally, it must be noted that frame running provides an enjoyable, safe and alternative opportunity to be physically active, exercise, and even compete, which the CP population have restricted opportunities to do, particularly for those with more severe impairments, thus helping to break a cycle of deconditioning, and improving the all-important QoL.

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Appendices

Appendix A – Information and Consent for Study 1, Round 1

Invitation

Hello,

I would like to invite you to take part in a survey, which is part of my PhD study at Edinburgh University. The survey is split into 2 rounds. This survey is round 1, and a further link will be sent out for the second round once the first has been completed. Each round should take no longer than 5 to 10 minutes of your time. The results have the potential to help with the development of disabled/adapted sport.

My PhD involves evaluating the benefits of a physical activity intervention for young people with cerebral palsy (CP), and it is important to measure what matters. Interventions may have physical, psychological, and social-emotional effects on individuals. The aim of this survey is to establish what you view as the most important factors to measure when evaluating a physical activity intervention for children with CP.

The Research Team

The research is being carried out by Hamish Johnson, a PhD candidate at Edinburgh University, and supervised by Dr. Martine Verheul, a lecturer in motor control, and Dr. Amanda Martindale, a lecturer in sports and performance psychology, both at the University of Edinburgh.

What will happen?

The following survey will ask you as a parent/guardian, frame running coach or health professional to name the factors you see as most important to measure when evaluating a physical activity intervention.

Your Rights

Your participation is voluntary and anonymous, and all responses will be stored confidentially. Whether you choose to take part or not is entirely your personal choice and you can withdraw at any time whilst participating in the survey, with no questions asked. By completing and submitting the survey you are providing consent for the information you have provided to be used by myself and research supervisors to help construct the second round of the survey.

At the end of this survey you will be asked to leave a contact email address, so we are able to contact you with the second round of the survey.

If you have any questions, or you wish to withdraw following the completion of the survey, please email the researcher, Hamish Johnson or supervisors, Dr. Martine Verheul and Dr. Amanda Martindale.

Thank you for taking the time to complete the survey.

Hamish Johnson

Appendix B – Information and Consent for Study 1, Round 2

Invitation

Hello,

You are being contacted as you have recently completed a survey related to physical activity interventions and the effect of participation for children with cerebral palsy (CP). I would like to invite you to take part in the second round as part of my PhD.

Here is a quick reminder of what my research is about: I am evaluating the benefits of a physical activity intervention for young people with CP and aim to identify the most important factors to measure. Interventions may have physical, psychological, social-emotional effects on individuals. The aim of this survey is to establish what you view as the most important factors to measure when evaluating a physical activity intervention for children with CP.

By collecting views on which factors are most important this survey aims to improve the evaluation of interventions in the area of adapted sports.

What will happen?

This is a very quick survey and will take you around 5 minutes to complete. The survey will ask you to rate each factor on a 5-point scale from **1=not at all important** to **5=very important**.

Your Rights

Your participation is voluntary and anonymous, and all responses will be stored confidentially. Whether you choose to take part or not is entirely your choice and you can withdraw at any time.

By completing and submitting the survey you are providing consent for the information you provide to be used as part of the thesis, future presentations, and papers.

If you have any questions, or wish to withdraw following completion of the survey, please email Hamish Johnson or supervisors, Dr. Martine Verheul and Dr. Amanda Martindale.

Thank you for taking time to complete the survey.

Hamish Johnson

Appendix C – Information Sheet for Study 2



THE UNIVERSITY
of EDINBURGH

Study Title: Effects of a Frame running intervention on the psychological and social wellbeing of children with cerebral palsy.

We are looking for volunteers to participate in a frame running study.

What is the purpose of the study?

Frame running is a relatively new sport, which is aiming to be included in the Paralympic games. It is different from other disability sports as it allows individuals who can't run independently, to use their legs in a running like motion. There is a steady increase in participation, and this highlights the potential of RR within disability sport. Results from past physical activity studies makes us believe that taking part in Frame running can improve children's psychological and social wellbeing. However, at this moment in time there is no scientific proof of this. Therefore, the aim of this study is to explore the effects of a 3-month Frame running intervention on the psychological and social wellbeing of children with cerebral palsy.

Who will take part?

Your child has been invited to take part because they have recently started taking part in Frame running training sessions.

Do they have to take part?

NO! It is up to you and your child. If you do, you (and your child) will have to sign a form giving consent to take part.

What will happen if my child takes part?

They will be asked to complete some questionnaires, with or without the help of a parent or carer. If a parent or carer is used for physical assistance to complete the questionnaires, please try to ensure that the responses are the child's own. The questionnaires will be made available online, so that you are able to take your time when completing them and should take around 1 hour to complete. Apart from that, you just have to keep going along to the at RR sessions once a week and be asked to use smiley faces after each session to express how you are feeling.

What if I change my mind?

That is not a problem. Taking part is your choice, and completely anonymous, with all responses being stored confidentially. You may withdraw at any point with no questions asked.

I have a question/Where can I find out more information?

If you have any further questions, please do not hesitate to contact the PhD researcher (Hamish Johnson) or one of the research supervisors, Dr. Martine Verheul and Dr. Amanda Martindale.

Thank you for your time!

Appendix D – Example Information Sheet for Participants



Information sheets for participants

We are looking for volunteers to take part in our frame running research.

We are asking you if you would agree to take part in this study. Before you decide on whether to join or not, it is important to know why this research is being done and what would be your role as part of it. Please read this information sheet carefully. Talk about it with your family and friends if you need to, and if you have any questions or worries, you can talk to us via the contact details at the end of this sheet.

Why are we doing this research?

Frame running is a relatively new sport, which at some point may be included in the Paralympic games. We want to find out if taking part in frame running makes you feel healthier and happier.

Why have I been asked to take part?

You have been invited to take part in this study because you are about to join a frame running club or are already attend frame running training sessions.

Do I have to take part?

No. It is entirely up to you to decide whether or not to take part in this study. If you decide not to, you do not need to give a reason. If you decide to take part, you are free to stop taking part at any time during the research without giving a reason, and you can just carry on with your frame running training.






If you decide you would like to take part, we will ask you to sign a form that shows you agree to take part.

What does taking part involve?

You will be asked to complete a pack of questionnaires about yourself and what activities you like to do. If necessary, you can ask your parent or carer for help, but please try to make sure the answers are your own. As the questionnaires will be handed or posted to you, you are able to take your time when completing them. The questionnaires should take around 1 hour to complete but these can be done at your own pace.

Where does the research take place and how long will it last?

As the questionnaires are using a pen/pencil and paper, you can complete them in the comfort of your home. We would like you to do the first set of questionnaires as soon as possible, and three more times over the course of the study. We also would like you to keep going along to your frame running training sessions, and after each session, using smiley faces, show how you are feeling. For example:

DATE OF SESSION	HOW MUCH DID YOU ENJOY YOUR FRAME RUNNING SESSION?				
					
<i>Example: 1/06/2018</i>				X	

Why would it be good for you to take part, and is there anything to be worried about?

We hope that our research will prove that taking part in frame running can make people feel healthier, stronger, and happier, but we cannot guarantee this. We are hoping that this research will increase our understanding of the benefits of frame running. This can help open the door to inclusion in the Paralympic Games and can lead to more funding and support for CP sport and exercise programmes at a local, national, and international level. We believe that the risks of taking part are very low, but if you are worried about anything and would like to speak to someone, you or your parents/carers can contact one of us involved with the research.

Thank you for taking your time to read this information sheet!

Where can I find out more information about the study? You can contact any of us if you have any questions:

PhD Researcher:

Hamish Johnson
University of Edinburgh

Frame running Contact:

Peter Drysdale
CPIIRA President

Supervisors:

Dr Martine Verheul
Institute of Sport, PE, and Health Sciences
University of Edinburgh

Dr Amanda Martindale
Senior Lecturer in Sport & Performance
Psychology
University of Edinburgh



Appendix E – Parent Consent Form



PARENT CONSENT FORM

Participant Number:

Please mark box



1. I confirm that I have read and understand the information sheet for the mentioned study. I have had the opportunity to consider the information, ask questions, and have had these answered satisfactorily.
2. I understand that participation for my child is voluntary, and that I am free to withdraw my child at any time, without giving any reason, and without any consequences.
3. I agree to a summary of the anonymized data collected being shared with research supervisors.
4. I understand and agree that the data collected will be used for a research thesis, will be published in scientific literature, and presented at scientific conferences.
5. I agree with information provided and give consent for my child to take part in the above study.

Name of participant: _____

Name of person providing consent: _____

Signature of person providing consent: _____

Date: _____

Appendix F: Participant Consent Form



PARTICIPANT CONSENT FORM

Participant Number:

Please mark box



6. I confirm that I have read and understand the information sheet for the mentioned study. I have had the opportunity to consider the information, ask questions, and have had these answered satisfactorily.
7. I understand that participation is voluntary and that I am free to withdraw at any time, without giving any reason, and without any consequences.
8. I agree to a summary of the anonymized data collected being shared with research supervisors.
9. I understand and agree that the data collected will be used for a research thesis, will be published in scientific literature, and presented at scientific conferences.
10. I agree with information provided and give consent to take part in the above study.

Name of participant: _____

Name of parent providing consent: _____

Signature of parent providing consent: _____

Date: _____

Appendix G: Parent Instruction Sheet








Instruction Sheet for parents

Thank you very much for agreeing to take part in our Frame running research. Below are some steps to make sure that everything is clear for you to complete the pack. Here is what to do:

1. Open the pack
2. Complete the **participant ID sheet**
3. Read the **information sheets**
4. Complete and sign the **consent forms**

Now onto the questionnaires:

5. Open the questionnaire pack for **parent/carer**. Complete the two questionnaires; there are instructions for each.
6. Open the questionnaire pack for **children**. Your child should work their way through the five questionnaires at their own pace. There are instructions for each. If your child needs any help in marking the answers that is fine. If they need help answering the questions, then that is also fine, but try to ensure as many of the responses are their own. We will ask you at the end to estimate what percentage are their own answers.
7. Open the **green/blue record form** booklet and help your child work through the booklet (and assist them when and where is necessary).

DATE OF SESSION	HOW MUCH DID YOU ENJOY YOUR FRAME RUNNING SESSION?				
					
<i>Example: 1/06/2018</i>				X	

8. The **session enjoyment sheet** is for you to keep for the duration of the research. All you have to do is have your child mark how much they enjoyed the session after each frame running session they attend, for example:
9. Please **return** the **participant ID sheet, consent forms, questionnaires, and final checklist**, in the stamped envelope, and **hang onto** the **session enjoyment sheet** for the duration

Thank you again for all of your help, & if you have any further questions please do not hesitate to get in touch (contact details can be found on the information sheets).

Appendix H: Social Validation Questions

Perceptions of Frame running Intervention

Name:

Instructions: Please indicate, by circling on the scales, to what extent you agree or disagree with each of the following statements. Please use the box below the scale to explain your answer, and fill out the second box with your child (if possible) to give their point of view:

1. The Frame running intervention has been important to me/my child.

1 2 3 4 5 6 7
Not at all Neutral Extremely

Parent Comments: Why has it been too important me or not?

Child Comments: Why has it been too important me or not?

2. The procedures used throughout the frame running intervention were appropriate.

1 2 3 4 5 6 7
Not at all Neutral Extremely

Parent Comments: What did you think of the frame running sessions and the questionnaires used throughout the intervention?

Child Comments: What did you think of the frame running sessions and the questionnaires used throughout the intervention?

Appendix I: Information and Consent sheet for Study 3

Invitation

Hello,

We would like to invite you to complete a survey as part of my research on the impact of participation in frame running on various aspects of life for children, adolescents, and adults with cerebral palsy (CP).

My name is Hamish Johnson, and I am a PhD student at the University of Edinburgh, and I am involved in racerunning research at the university.

The research is investigating the impact that Frame Running has had on different aspects of your life.

We are looking for participants aged 14 and over, who are currently involved and competing in frame running (current winter breaks are included as being currently involved). The questionnaire is in English; therefore, a good level of English would be beneficial but not a requirement, and participants should not have a cognitive impairment.

I understand that the frame running community is a small place, so if you have been sent this questionnaire already, apologies, and a big thank you if you have already completed it.

The Research Team

The research is being carried out by Hamish Johnson, a PhD candidate at Edinburgh University, and supervised by Dr. Martine Verheul, a lecturer in motor control, and Dr. Amanda Martindale, a lecturer in sports and performance psychology, both at the University of Edinburgh.

What will happen?

If you choose to participate, you will be asked to complete this questionnaire. We ask you to take time to consider each question carefully and answer as fully as possible. The questionnaire will begin with basic demographic questions (e.g., gender, age, schooling, sporting history), and then will move to ask questions about your involvement in Frame Running and how it has impacted you and your life.

What are the risks and benefits of taking part?

There are no direct benefits for you by taking part in this research. However, we are looking to gather information about the impact of frame running, as a relatively new sport, on the lives of current racerunning athletes.

The information gathered can be used to build a case for frame running as a para-sport at all levels and help to influence and improve future research.

There are no known risks to participating in the research. However, if you do become stressed, anxious, or upset by any questions asked, please stop the questionnaire. If you wish for further support, there will be contact details provided for helplines at charities (e.g., Scope - www.scope.org.uk, 0808 800 3333; Mind - www.mind.org.uk, 020 8519 2122).

Your Rights

Your participation in this study is voluntary, anonymous, and all responses will be stored confidentially (please note that we only ask for your name at the beginning for consent, and this will **NOT** be used in the analysis). You may decide to stop taking part at any time during the research without any questions asked, and you have the right to refuse to answer any questions asked.

Most of the questions will ask you to provide longer answers. We may use direct quotes from your answers, but these will remain anonymous, and we will simply refer to you by your participant number.

By completing and submitting the survey, you are providing consent for the information you have provided to be used in a PhD thesis, and possibly in future journal articles, conference presentations.

If you would like any further information, have any questions, or wish to withdraw from the research after the completion of the survey, then please contact one of the following:

Hamish Johnson (researcher)

Dr. Martine Verheul (supervisor)

Dr. Amanda Martindale (supervisor)

Thank you for taking the time to take part in the research, it is greatly appreciated!