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Dancing with hypermobility: An exploration of the health risk and experience of generalized joint hypermobility within a classical ballet narrative

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Author Declaration

I hereby declare that:

a) I have composed this thesis
b) This is my own work
c) This work has not been submitted for any other degree or academic award

Signed

Date 22nd March 2020

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Abstract

Generalised Joint Hypermobility (GJH) is a heritable disorder of the connective tissue that can manifest as extreme range of motion in the joints and is associated with tissue fragility and injury. GJH is prevalent within classical ballet and is considered both an asset and health risk to the dancer. More recently within the general population links have also been made between GJH, anxiety disorders and poor body awareness. This research brings together three discreet yet interweaved studies that explore GJH within both vocational and professional dance environments. A mixed-methods design and a pragmatist approach was applied to this research which is situated within a biopsychosocial framework.

Study one established reliable and stable measures between the researcher and an experienced clinician for the standardised Beighton categorisation of GJH. A sample of young dancers (n=10), randomly recruited from a larger group of young dancers (n=70) during their annual dance screen, were tested for GJH. The intra-rater stability of measures was found to be moderate (k=.43) for the physiotherapist and high (k=.88) for the researcher, with a high (90-100%) inter-rater percentage of agreement (pa) for the Beighton categorisation. This provided quality assurance within the research for the researcher to confidently apply the standardised testing methods. Subsequently a larger cross sectional sample (n=90) of classical ballet dancers in Scotland were tested for GJH. They also retrospectively reported injury occurrence over the previous year of training. Within the study a high percentage of dancers were observed with GJH (56% for ≥4, 43% for ≥5 and 39% for ≥6 cut off) which was in line with the literature and suggests that people with GJH are often signposted or recruited into dance training. Interestingly, and in contrast with the literature, the injury per workload estimation provided a relatively low overall frequency of 1.85 injuries per 1000 hours of training and performance, and there was no significant relationship ($p<.001$) found between injury and GJH in study one.

Building on study one, the second study was designed to further explore the relationship between body awareness, anxiety and GJH in classically trained dancers. As the literature highlights the relationship between dance training and body awareness, the second study focused on one national level, full time vocational dance training programme. This provided assurance that the participants all received the same quality and quantity of dance training. In this study statistical analysis found no significant differences in proprioception between the dancers with and without GJH. Dancers that did not present with GJH had significantly higher scores for spatial visual memory ($p=0.001$) and not worrying ($p=.003$). Dancers with GJH scored significantly higher for body listening ($p=.012$), emotional awareness ($p=.026$) and noticing ($p=.026$). Significantly lower anxiety measures were recorded for the dancers without GJH ($p = <001$). Overall, study two demonstrated that the dancers that presented with GJH had different measures for some components of body awareness and greater anxiety than the dancers that did not present with GJH. It is argued that while GJH is reported in the literature as a signpost for entry into dance training because of the associated biological marker (distensible connective tissue) and extreme range of motion this facilitates, additional psychosocial factors, such as increased emotional awareness and body listening, may also signpost GJH to dance training. Having established GJH prevalence and associated injury rates, body awareness and anxiety levels within classical ballet dancers, this research then investigated the lived experience of GJH from both the professional

dancers' and classical dance teachers' perspective (study three) from the professional environment for classical ballet.

Study three explored the experiences of nine professional international dance artists (5 male, 4 female; mean age = 32.3yrs; SD=4.99yrs; range=25-40yrs). All were referred to the study on the basis of GJH by clinical practitioners, had a classically based training and at least ten years performance experience. GJH status was further confirmed using a validated historical self-screen questionnaire. Data was collected through retrospective recall in semi-structured interviews (35 - 45 min). Thematic analysis exposed the main themes as: openness and extent of acceptance of GJH; coping strategies towards the management of GJH including mental stillness; and the dancers agreed that GJH gave them an advantage in their careers with regards to choreographic demands. They discussed needing more time than other dancers to consolidate choreographic movement, however they did not explicitly associate this with GJH. The dancers were readily able to identify and discuss the physical aspects of GJH. Psychosocial implications and experiences of GJH were discussed, but the dancers did not make direct associations between GJH, anxiety and their struggle to find mental stillness. Semi-structured interviews (45 to 50 minutes) were conducted with four dance masters (three female & one male). The dance masters had a mean of 30.8 years (SD=6.8yrs; range 25-35) experience in their role. Thematic analysis provided emerging themes that included the hypermobile aesthetic, professional values and preconceptions towards current choreographic trends, company strategies, intellectual curiosity, pedagogy and leadership. All dance masters agreed that dancers with GJH met the direction and desired aesthetic for most of today's dance companies and choreographers. They showed a good understanding of the strengths and challenges of a physique with GJH but did not directly associate any psychosocial traits with GJH. They did however suggest that the dancers with GJH who had successfully sustained a career in dance were those who had developed intellectual and physical strategies to cope with their condition.

In conclusion, this thesis provides evidence of the multi-faceted nature of GJH within the classical ballet context. In the first instance it is evident that GJH is prevalent in young classical dancer but interestingly at this stage in their career GJH is not associated with injury, possibly due to the protective feature of their dance training. In realization of the complex nature of GJH, study two highlighted further aspects that intersect the psychosocial domain of body awareness and anxiety in dancers with GJH. Study two provides evidence to suggest that GJH signposts dancers towards dance for both physical and emotional capacities. This study also provides evidence towards altered body awareness including enhanced body – mind connections and increased anxiety within GJH. Finally, whilst the dancers and dance masters clearly recognised the physical experiences of GJH they demonstrated disconnect between the psychosocial aspects they discussed and the implications of these with GJH. This brings new insight to the experience of GJH that potentially has wider implications for dance teaching learning and performance whilst also potentially influencing the creative process for dance. This may further inform practice to provide insight in to the psychosocial ramifications of this condition and a healthier environment for dancers with GJH. This is particularly noteworthy within the education for dancers and dancer teachers and potentially other aesthetic sports where GJH is prevalent.

Publications related to this research

Invited paper & abstract accepted

Timmons, W; Sproule J; Mulholland, R
Title: Exploring the relationship between disordered connective tissue and body awareness in classical dance artists.
The Psychological and Physiological Benefits of the Arts, hosted by Dr(s) Vicky (Vassiliki) Karkou, Nisha Sajnani, Felicity Anne Baker, Jenny M Groarke, Hod Orkibi, Johanna Czamanski-Cohen, Maria Eugenia Panero, Bronwyn Tarr, Jennifer Drake, Corinne Jola in Frontiers in Psychology - section Health Psychology.

Conference presentations

Wendy Timmons, title: Interoceptive sensibility and proprioception in classical dancers with hypermobility10 min scientific presentation. International Association for Dance Medicine & Science, Helsinki, Finland 24th October 2018

Wendy Timmons, title: Exploring the relationship between interoception, anxiety and hypermobility in adolescent dance students, 20 min scientific presentation: International Association for Dance Medicine & Science, Huston Texas UAS 10th October -16th October 2017

Wendy Timmons, title: Dancing with hypermobility, the lived experience of GJH. Keynote presentation International Association for Dance Medicine & Science regional day Edinburgh 2019

Consultation

2019-2020 LTTA 5 of Erasmus+ project Shift-Dance

https://northernballet.com/shift-dance

Funded by **Erasmus+**, the project aims to:

- Develop new teaching practice for dancers with learning disabilities, underpinned by safe practice
- Enabling people to fulfil their potential
- Share good practice, initially between collaborators and subsequently to an international audience of dance artists and teachers in community, education and professional settings
- Raise the profile and skills of learning disabled dancers across Europe

Web content
Timmons, W. The interrelationship between Down syndrome, Autism and joint hypermobility within the dance teaching and learning environment
Timmons, W. A biopsychosocial ecology in talent development for dance

Keynote presentation
SHIFTing perceptions conference

https://northernballet.com/join-in/expressions/shift-conference

The interrelationship between Down syndrome, Autism and joint hypermobility within the dance teaching and learning environment.

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Abbreviations

ADHD	Attention Deficit Hyperactivity Disorder
AS	Autism Syndrome
BS	Beighton Scoring
CBTT	Corsi Block Tapping Test
EDS	Ehers Danlos syndrome
GJH	Generalised Joint Hypermobility
HDCTs	Heritable disorders of connective tissue
hEDS	Hypermobility type Ehers Danlos syndrome
HJH	Historical joint hypermobility
HSD	Hypermobility Spectrum Disorders
JH	Joint Hypermobility
LJH	Localised Joint Hypermobility
MS	Marfan Syndrome
PJH	Peripheral Joint hypermobility

Chapter 1. Introduction

1.1 Why explore generalised joint hypermobility within the context of health risk and classical ballet?

Generalised joint hypermobility (GJH), a genetic condition of the connective tissue, falls under the domain and medical expertise of rheumatology. Whilst it can affect 10-25% of the general population it is often an area of neglect in rheumatology (Hakim, A., & Grahame, 2003) because it can be asymptomatic. It remains however a heritable condition of the connective tissue and can be associated with tissue fragility, pain and injury (Armstrong, 2018; Blokland, Thijs, Backx, Goedhart, & Huisstede, 2017; Ruemper & Watkins, 2012). At a biological level in GJH enables extreme range of motion in the joints and hyperextension of the limbs. It is often characterised in lay terms as 'double-jointedness' and those who present with GJH are also described as having 'bendy bodies'. These GJH characteristics are also very conducive to the physical demands and aesthetic that is coveted within dance and some aesthetic sports and in particular within classical ballet (Armstrong & Greig, 2018a). Not surprisingly then, GJH is reported to be prevalent within classical ballet dancers (McCormack, Briggs, Hakim, Grahame, & Grahame, 2004; Mark C. Scheper et al., 2013a). At a deeper level, GJH has more recently been unexpectedly associated with anxiety and since proposed as a new "neuroconnective phenotype" (Bulbena-Cabré & Bulbena, 2018, p. 15). It has since been investigated from a psychosocial perspective and it is increasingly becoming associated with disorders that include anxiety, fear and poor body awareness (Carolina Baeza-Velasco, Bourdon, et al., 2017a; Bulbena-Cabré & Bulbena, 2018). Unlike the biological characteristics of GJH, anxiety is clearly less beneficial to a training in classical ballet. Equally, good body awareness is fundamental for the execution of classical

ballet skills and therefore poor body awareness will potentially prove to be disadvantageous to a classical ballet dancer. Whilst it is established that GJH is prevalent in dance because of its associated biological characteristics (Armstrong, 2018; C. Chan, Hopper, Zhang, Pacey, & Nicholson, 2018; Mccormack et al., 2004) it appears that there are psychosocial aspects of GJH that make its presence potentially less conducive to classical ballet training and performance. Nonetheless, with regards to GJH, the research direction within dance medicine and science currently remains completely focused on its prevalence, the implications of the associated tissue fragility, the excessive range of motion this facilitates for a dancer and injury that may occur as a result. The debate is therefore currently absorbed with the gains and health risk of GJH in classical ballet from a biological viewpoint. Reflecting on this current position, it is therefore my intention to explore yet go beyond the biological associations to discover more with regards to the psychosocial implications of GJH within the classical ballet environment for training and performance.

1.2 The researcher's attraction to study hypermobility within a classical ballet narrative

In this section I briefly delineate my own captivation with and attraction to hypermobility within dance from both a personal and professional perspective. Before entering the realms of academia, I was myself a performance artist, later I became a registered teacher of classical ballet and have been for almost 40 years. My grand entrance into the world of classical ballet commenced serendipitously at the age of three as a result of my older sister's grand exit from the ballet studio. Having been 'prescribed' ballet classes by our family doctor as a means to address her apparent lack of coordination which had manifested in a concerning and 'clumsy' gait that was pointed out to my mother by my grandfather who was himself a race walking athlete. As a timid and overly anxious child, my older sister demonstrated absolutely no

desire to engage with the ballet classes, indeed being made to partake in the ballet classes further confounded her anxiety. As a witness to the drama and scenes of my sister refusing to take part I begged my mother to allow me to take up the classes and I virtually never looked back. Dance to a great extent became my identity and my life, it was something that I was good at, and dance enabled a lifelong career that to date remains my passion and obsession. Throughout this preoccupation with dance I have worked as; a performance artist, performing as a child artist for television, later as a dance artist working internationally in musical theatre, a choreographer, and finally I qualified as a teacher of dance and became director of a dance academy. It was much later along this journey that I became fully aware of my own inherent connection with hypermobility. As a teacher of dance I was witness to many young dancers, and often sibling dancers, who seemingly had great potential to perform and execute the skills and techniques required in the dance studio, yet often struggled in other aspects of their dance learning and performance. These observations as a dance practitioner led to a thirst to know more and eventually this doctoral line of research and enquiry.

1.3 The context, scope and delineations of the thesis

Before applying empirical research within the context of current practice in classical ballet, it is perhaps noteworthy to briefly explain the practices, traditions and conducts that are historically entrenched within the art form. This may serve to demystify some of the practices and intentions within the classical ballet environment that remain today.

Classical ballet is a codified form of dance that developed in the renaissance period (14th -17th centuries) through the work of professional dance masters. This was a period in history when the discourse central to society focused on the human condition and moral behaviours that were expressed through the arts. A dance master was a teacher, choreographer and social arbitrator par excellence whose work eventually led to the

establishment of classical ballet as an art form in Western culture (Lee, 2002, p. 35). Court ballet (the origins of what we now call classical ballet) was originally practiced by males only as royal and noble education and was indeed used to establish Louis XIII's divine like status at the age of sixteen in the eyes of his nation, France (Lee, 2002, p. 48). The Renaissance royals soon also worked out the complex relationship between art and power and the first theatres since antiquity were constructed to give provenance and provide permeant residence for the practices and performances of theatrical dance. The international art form of classical ballet dance as the esteemed profession for both men and women that we recognise today sprang from these practices under the reign of Louis XIV (Lee, 2002, p. 61). Much of the classical ballet repertoire that remains today originates and is true to the very practices of these times and include specific and codified techniques that are designed to personify a dancer as an artist and athlete with supernatural powers. The objective for these supernatural powers are to transform an audience into a world of magic and spectacle, away from the daily pressures that life brings (Guest, 1962). As the art form developed, so did the physical demands on the dancer, pointe work (the ability to dance on the tips of the toes in order to appear as if airborne) for example, was once only required by female soloist dancers however by the 1870's Mr Petipa, choreographer and dance master, insisted that all female dancers were obliged to acquire this technique in order that he could push the boundaries in his own choreography, entertain and provide spectacle to his audiences (Lee, 2002). It appears then that the objective for classical ballet originate and remain in the need to defeat the norms of the human condition whilst remaining true to the traditions and cultures from which they have developed. Today ballet masters and choreographers world-wide remain the keepers of the art form and its traditions yet they are also the designers of neo classical and

modern ballet practices of the future. Inevitably then, they too use the dancers' bodies to push boundaries and enter new terrains in their work.

The current literature establishes that GJH is a disorder of the connective tissue and potentially a real health risk, it is undeniably a predominant feature within dance (C. Chan et al., 2018; Day, Koutedakis, & Wyon, 2011b; Ruemper & Watkins, 2012; Sanches, Oliveira, Osório, Crippa, & Martín-Santos, 2015). Interestingly, dance teaching and choreography remain largely as unregulated professions that are governed by the traditions and historical practices of the past. Whilst dance teaching qualifications and regulatory examining bodies do exist worldwide, experienced dance artists and performers can and often do progress to the role of teacher or dance master by default with no further professional learning once their performance career is complete. Furthermore, within the dance teaching qualifications that are delivered internationally by regulated examining bodies (for example Royal Academy of Dance, Imperial Society of Teachers of Dance), there is little to no evidence to suggest that any knowledge or understanding of GJH features or is a requirement for dance teachers. With young dancers presenting with GJH predominantly recruiting into the classical ballet environment this cautions a genuine health risk that clearly warrants further investigation.

In order to appreciate the circumstances of GJH within classical ballet, the first phase of this research explores the predominance of GJH as a genotype within the classical ballet training arena. This phase of the research extends mainly to a Scottish context and whilst this may appear to delineate the research, practices are undeniably common within the classical ballet environment world-wide. The delineation of the Scottish context is also further offset in the second phase through the establishment of experiences of GJH from dancers and ballet masters' that are situated and working internationally. In such, the context and scope of this

thesis could extend to and provide valuable insight for future practices in classical ballet worldwide.

1.4 Aims and objectives

Reflecting on the scope of this contextual backdrop for classical ballet, this thesis focused on the following overall aim and objectives:

1.4.1 Aim

To explore the health risk and experience of generalised joint hypermobility within a classical ballet narrative with a view to provide insight for dancers and practitioners who train and work in this environment.

1.4.2 Objectives

To scope the prevalence of GJH and musculoskeletal injury in a cross sectional sample of pre-vocational classical ballet dancers.

To explore GJH, body awareness and anxiety in an elite setting for pre-vocational classical ballet dancers.

To examine the embodied experience of GJH in classical ballet dance artists.

To examine dance masters' experience of classical dancers that present with GJH.

1.5 Overview of the thesis

The initial phase of this thesis (Chapters 2 to 5) defines what GJH is and quantifies its prevalence within a cross sectional sample of young trainee classical ballet dancers. This comprises of a review of the literature with regards to the past and the more recently developed understanding and definition of GJH from a biological perspective. With GJH

clearly defined, study I provides reliable assessment and categorisation methods for GJH and examines the relationship between GJH and injury. Following on from this investigation, further literature pertaining to the psychosocial aspects of GJH is reviewed in order to inform study II whereby body awareness and anxiety within in GJH are examined and quantified in one national vocational setting for classical ballet.

Building on the findings that emerge from the initial part of the research, the second phase of the thesis (Chapters 2 to 5) examines the experience of GJH within the classical ballet environment. In this phase professional dancers and ballet masters working internationally are asked to discuss their own lived experience of GJH using retrospective recall of training and performance practice.

The last phase of the thesis involves the discussion of overall conclusions, their implications and recommendations. This phase completes with future research directions and suggestions for the implications of the research findings to practice. The structure adopted across the thesis is one that builds from the first to the final phase in an attempt to provide a coherent understanding of the health risk and experience of GJH within classical ballet.

Chapter 2 A review of the literature

Understandings of the entanglement of movement and thought have led modern philosophy and neuroscience to regularly challenge former notions of the Cartesian dualist brain and body separation. With the notion of brain body unity comes the possibility that *'human minds … are at the very least in deep and critically important contact with human bodies and with the wider world'* (Clark, 2008, p. xxvi). The idea of a thinking body is indeed not new (Todd, 1937) and depicts cognition as bodily experiences that are based on our daily interactions with the world. Our bodies are then considered as *"… the primal form on which we model our thinking"* (Sheets-Johnstone, 1994, p.328). It is also proposed that linguistic arts, including expressive gesture (and therefore dance), are dependent on the body schema (Gallagher, 2005, p. 107). Indeed interaction with the physical world and bodily-kinaesthetic experience are regarded as a valid and central modes for knowledge construction (Gardner, 2011; Gilakjani, 2012; Thelen, 2000) and equally therefore it can be argued that dance (as a bodily kinaesthetic mode of interaction) is fundamental to the human condition (Lange, 1995).

Impacting on meaning-making and understanding, kinaesthetic experiences through dance are also considered essential within the development of social communication (Carr, 1997; Hanna, 1987; Warburton, 2011) and are looked upon by some even as a human biological endowment (Bond, 2008). Deep connections between dance and biology are also recognised through inquiries into kinaesthetic empathy, mirror neurons (Foster, 1997; Reason & Reynolds, 2010) and through the meanings and effects of dance for children with sensory impairments (Bond, 2008; Timmons and Ravernscroft, 2019). This work surely then also supports mind-body connections and a convincing bio-aesthetic theory of human dance

(Bond, 2009, p401). Bond's bio-aesthetic theory explains how for example a dancer uses the body as a tool for expressive posture and gesture enabling meaning to be communicated. The type of body we have and how that body develops may potentially then also influence our mind and who we become.

Historically, dance has also been a known and important part of social, spiritual and also healing rituals (Fortin, 2018). Associations between mind-body connections and the health benefits of dance have undeniably been documented since ancient times (Alpert, 2011; Arcangeli, 2008; Hanna, 2006, 2007). Current empirical thinking now also suggests that dance can improve health and wellbeing across a lifespan (Alpert, 2011; Quiroga Murcia, Kreutz, Clift, & Bongard, 2010). A recent systematic review (Yan, Cobley, Chan, Pappas, Nicholson, Ward, Murdoch, Gu, Trevor, Vassallo, and Wewege, 2018) suggests that dance of all types is equally as effective (and occasionally more effective) than other types of physical exercise for improving different health outcome measures. Similarly the implication of dance for health includes meaningful social benefits and associated positive impacts (Atkins et al., 2019) that are unique and powerful.

Dance however comes in many disciplines and technically demanding dance forms such as classical ballet (the focus of this research) are less favoured in the literature for their health benefits within 'dancer' populations. The majority of research publications that discuss classical ballet confer injury and ill health (Caine et al., 2015; J. Desmond, 1997; Ekegren, Quested, & Brodrick, 2014; McEwen & Young, 2011; Novosel, Sekulic, Peric, Kondric, & Zaletel, 2019; Russell, 2013; P. J. Smith et al., 2015; Sobrino, de la Cuadra, & Guillén, 2015; Teitz & Kilcoyne, 1998; Wainwright, Williams, & Turner, 2005). This may however be because the dancer populations investigated in this research are elite and pushing their own physical boundaries to serve their art throughout what is a relatively short career lifespan

(Moira McCormack, Bird, de Medici, Haddad, & Simmonds, 2019a). Publications that do evidence the health benefits of classical ballet are indeed therapeutic interventional studies with clinical populations such as participants who have cerebral palsy (Lakes et al., 2019), older adults , Parkinson's disease (McGill, Houston, & Lee, 2019b, 2019a) and dementia (Lapum & Bar, 2016). Within these interventions, the rational for using classical ballet as an intervention tool includes the codification, advanced technical rigour and therefore repeatability and structure of the dance form that is 'measurable' as opposed to creative or contemporary dance that is less constrained.

The reason that classical ballet technique is so far advanced is that it has been evolving and developing over hundreds of years. Indeed, it is more than 400 years since the first formal ballet school *Académie Royale de Danse* was founded in 1661 in France by Louis XIV. The forms of classical ballet practiced today have a level of technical and artistic virtuosity that surpasses these origins, many also contend that mastery of basic classical technique is also fundamental to other dance forms. The rapid development of classical ballet is attributed to international popularity and the expansion of ballet academies in every major cosmopolitan centre in the world (Ward Warren, 1996, p4). The notion of classical ballet virtuosity however, for some, proposes elitism which also points to an over-investment in specific female body types that in turn serve the image of a sylph and *"an aesthetic of beauty, grace and line"* (Albright, 1997, p306). Today, the specific body type associated with 'ballerinas' also extends beyond the stereotypical female sylphlike body to include the male dancer. Certainly, current neo-classical choreography appears as gender-neutral requiring androgynous body-types of equal athleticism, grace, beauty and elegance. Here the beauty is indeed *"an embodiment of an object of and a creator of desire"* (Pickard, 2015, p7) and is increasingly one that is potentially driven by a specific body schema. It appears therefore that

the success of a dancer today (independent of gender) is for the main part their ability to meet and embody the latest trends within the "*ballet aesthetic of beauty and perfection"* (Pickard, 2015, p7). Classical ballet has always served to afford effortless gesture and movement and an ethereal quality that historically allowed dance audiences to escape from every-day normality. Costumes and stagecraft, such as the invention of point shoes, gas lighting for the stage and the long romantic tutu seen in the 19th century (for example those worn by ballerinas such as Anna Pavlova and Marie Taglioni) were however gradually replaced with short tutus and bright lights that revealed the body and limbs in full gesture and expression. Since the birth of neo-classical ballet, and the era of Balanchine (1905-1983), (styled as *father* of 20th century American ballet and creator of New York City Ballet) aesthetic trends moved towards costumes that revealed the body in full and facilitated extensions and gestures of the limbs that went beyond normative physical bounds. Indeed, it can potentially be argued here that Sinclair's notion and 'era of excess' as a reaction to 'prohibition' also infiltrated the arts (Sinclair, 1962). Within the world of classical ballet there also became an increasing desire for 'the excess' in for example the extension of the arabesque or number of pirouettes a dancer would perform, hence it also became a pre-requisite for a dancer's body to articulate movement even further beyond the norm. This evolving spectacle of classical ballet is clearly evidenced across the visual history and evolving aesthetic of major dance companies and choreographed work internationally and also documented through empirical research (Daprati, Iosa, & Haggard, 2009; Twitchett, Angioi, Koutedakis & Wyon, 2009; Twitchett, 2011). Interestingly, this desire and phenomenon for 'the excess' also arrived at around the same time as the body types with specific *geno* (genetic make-up) and *pheno* (expression of the genetic make-up) types that had the ability to demonstrate excessive flexibility or

'hypermobility' were also being clinically observed, debated and defined in modern medicine (Kirk, Ansell and Baywaters, 1967; Grahame, 1990; Keer and Grahame, 2003).

2.1 Hypermobility

Whilst these phenotypes were becoming the body style of preference within dance at this time there is clear evidence to support the notion that hypermobility is not a 'new' condition (Jacobs, Cornelissens, Veenhuizen, & Hamel, 2018). Paleo-pathological observations suggest that hypermobility as a phenomenon has been detected since ancient times. Ceramic figures from Peru, drawings and paintings (Dequeker, 2001) and the first known clinical description of hypermobility was first recorded by Hippocrates in 400 BC (Jacobs et al., 2018, p. 1). It is however, according to Grahame (2003) *"...easy to spot if you look for it. It is easy to miss if you do not"* (page 2). So, what does this mean?

Hypermobility as a syndrome was first defined as a disabling set of musculoskeletal conditions (Kirk, Ansell and Baywaters, 1967; Grahame, 1990). In 2003, Grahame together with Keer highlight that although thirty years have passed since the syndrome was first defined, there remains a distinct lack of information available to physiotherapy practitioners within their curriculum for techniques for its recognition. They assert that until such information becomes part of routine clinical examinations *"hypermobility will continue to be overlooked"* (page 2). Indeed in 2015, a group (n=325) of physiotherapists were questioned about hypermobility and 18% had no idea of its prevalence with over 50% not knowing/understanding the further implications associated with hypermobility such as increased anxiety and poor body awareness (Rombaut et al., 2015a).

That said, often the terms hypermobility, hypermobility syndrome, joint hypermobility syndrome and benign joint hypermobility syndrome appear to be all applied

synonymously and interchangeably in the literature without definitive evidence that these classifications were one and the same thing (Keer and Grahame, 2008). Twenty years from Keer and Grahame's warning there is an apparent move to further describe hypermobility (including hypermobility, hypermobility syndrome, joint hypermobility syndrome and benign joint hypermobility syndrome) as a component within other complex syndrome(s) such as Hypermobility type Ehlers Danlers Syndrome (hEDS) and Marfan syndrome (MS). Its classification, whilst evolving due to advancements in genetics, is however still not finite (Castori et al., 2017; Malfait et al., 2017; Tinkle et al., 2017). The seemingly evolutionary nature of the terminology and its classification make reading, researching and understanding the literature challenging. To date what is confirmed is that the ability to demonstrate extreme range of motion, actively and/or passively, that goes beyond physiological axes in one or multiple joints is commonly but not always due to a genetic variance of the connective tissue (Castori et al., 2017; Juul-Kristensen, Schmedling, Rombaut, Lund, & Engelbert, 2017). This is now described in the literature as joint hypermobility (JH) (Castori et al., 2017)

2.2 Joint hypermobility (JH)

The hypermobile status of a joint can be inherent or 'acquired' and increased through intensive flexibility training (Remvig et al., 2011). Flexibility training is usually undertaken in order to facilitate an extensive range of movement that is increasingly considered as essential in some joints for dancers and other athletes to enhance their performance. The similarities to the presentation of inherent and acquired joint hypermobility mean that they can potentially be confused, there are however distinct and important differences. Acquired hypermobile joints are for example protected from injury and instability by their 'normal' tissue (so long as that tissue is not violated by the flexibility training) whereas inherent hypermobile joints result from a genetic variance that presents as 'abnormal' and fragile

connective tissue. This fragile connective tissue does not have the same protective properties as normal tissue (Castori et al., 2017; Keer & Grahame, 2008). Joint hypermobility as a genetic variance is inherent, congenital and while it can present as a symptomless trait (Castori et al., 2017) it can also be associated with complications that go beyond the physical appearance of joints that have hyperlaxity and increased range of motion (Kerr and Grahame, 2008).

Unwarranted use of the excessive range of motion within an inherently fragile hypermobile joint can lead to micro and macro trauma. Micro trauma constitutes silent recurring injury that is often not detected by the clinician or individual at the time it occurs (Castori et al., 2017). This may however lead eventually to pain, chronic pain, bone degeneration and when specific for example to the hip joint, labral tears (this is a tear the fibrocartilage attached to the rim of the acetabulum that helps keep the head of the femur in place) can occur (Devitt, Smith, Stapf, Tacey, & O'Donnell, 2017). Macro trauma would constitute for example dislocations, tendon ruptures and soft tissue injury, often occurring due to the instability and/or weakness of the joint, the fragility of the connective tissue and as a result of overuse through an excessive range of motion (Castori et al., 2017).

Poor proprioception (sense of the body in space/body awareness) is also commonly associated with inherent hypermobile joints (Chopra et al., 2017; Fatoye, Palmer, Macmillan, Rowe, & van der Linden, 2009; Ghibellini, Brancati, & Castori, 2015a; Nagai, Schilaty, Strauss, Crowley, & Hewett, 2018) and may be further diminished as a result of the micro trauma discussed above. Reduced proprioception and muscle strength are also interrelated, the one affecting the other in a vicious circle, this is turn limits the very activities that are recommended for rehabilitation, strengthening and return to healthy functioning in joints (Mark C. Scheper, De Vries, Juul-Kristensen, Nollet, & Engelbert, 2014; Mark C. Scheper et

al., 2016). Diminished proprioception may also be the cause of the apparent clumsiness that has in the past been anecdotally associated in children with joint hypermobility and more so in the literature over the past decade (Ghibellini et al., 2015a). Indeed clumsiness or delay in walking is often a silent precursory sign to the existence of joint hypermobility (Adib, Davies, Grahame, Woo, & Murray, 2005; Ghibellini et al., 2015a). Clumsiness is often a warning sign to parents and paediatric practitioners that something may not be quite right and it is also often the motive for them to prescribe cautious physical activities (such as dance) as a means to improve the apparent lack of coordination and/or delayed mature gait pattern.

2.2.1 Assessing for and describing joint hypermobility

Determining if a joint is hypermobile is achieved by comparing the passive range of motion of any joint and comparing this measurement with the normal range of motion parameters. Measurement is taken using goniometry and established protocols (Junge, Jespersen, Wedderkopp, & Juul-kristensen, 2013; Juul-Kristensen et al., 2017). The identification of JH as inherent and congenital condition has led to the development of standardised tools (Beighton, Solomon, & Soskolne, 1973). The most recognised ones being those proposed by Carter and Wilkinson (1964), Beighton, (1973) and Rotés (first described by Bulbena et al 1992) these were found to have high correlations and high predictive validity (Keer & Grahame, 2003).

The most frequently used tool for assessing JH are the Beighton criteria and scoring (Juul-kristensen, Schmedling, Rombaut, Lund, & Engelbert, 2017; Juul-kristensen et al., 2018; Juul-Kristensen et al., 2017). The Beighton criteria and scoring has also been tested as a valid and reliable tool for use with children (Smits-Engelsman, Klerks, & Kirby, 2011a). It consists of four bilateral tests (elbow and knee and 5th finger extension, and thumb

apposition) and lower back range of motion with scores ranging from 0-9. Caution is however given to the need for uniformity in applying the Beighton tests and the procedures outlined in Beighton et al 1973) are recommended for clinical use (Juul-Kristensen et al., 2017) see appendix 2.

Finally , there are now also assessment questionnaires for Generalised Joint Hypermobility (GJH) , the (Beighton Score) BS 5PQ (Bulbena et al., 2014) and Historical Joint Hypermobility (HJH) (Castori et al., 2017) that can be used to assess the presence of GJH using simple questions in combination with photographs to ascertain information about the current or historical flexibility of certain joints across the musculoskeletal system, thumbs, hips, shoulders). These are reported to have good reproducibility and accuracy.Both are implemented when appropriate as a rapid screening tools when syndromes and health issues associated with JH are being investigated in older adults (Castori et al., 2017). However Castori et al (2017) caution that although some cross-sectional research has supported the use of the Historical Joint Hypermobility (HJH) questionnaire, further research is needed (Castori, Sperduti, Celletti, Camerota, & Grammatico, 2011).

When joint hypermobility is observed in between 1 and 4 small or large joints it is defined as Localised Joint Hypermobility (LJH), this may be inherited and also acquired due to trauma, joint disease, surgery or training (Castori et al., 2017). If joint hypermobility is apparent in 5 or more joints (assessed using the Beighton criteria) the term Generalised Joint Hypermobility (GJH) is applied (Castori et al., 2017). GJH is usually congenital and inherent however in rare cases it can be caused by malnutrition, inflammatory degenerative diseases of the joints and endocrine disorders (Hasija, Khubchandani, & Shenoi, 2008). Peripheral Joint Hypermobility (PJH) is defined as a discreet form of joint hypermobility and is localised to the feet and hands. Finally Historical Joint Hypermobility (HJH) is determined using the 5

part questionnaire that retrospectively determines the flexibility of the thumbs, shoulders, and hips and therefore takes into account any decrease in flexibility and stiffening of the joints due to age or injury (Castori et al., 2017).

Within the definitions of the joint hypermobility variants a framework has now been established and is anticipated as a means to provide clearer and unified terminology for future research in this area. Castori et al (2017) strongly proposes however that the framework of JH variants are used as descriptions and not diagnoses (Castori et al., 2017). Castori et al (2017) also propose further delineation of the joint hypermobility variants into Hypermobility Spectrum Disorders (HSD). They propose that the observable characteristics of an individual with an HSD result from the interaction of their inherited genotype with the environment. It is also suggested that the observable characteristics within HSD are usually limited to the musculoskeletal system (Castori et al, 2017, p154). The descriptors within the HSD are again not proposed as clinical diagnoses, this framework however alleviates any further confusion in terminology and provides a solution for the former interchange of terms that were applied synonymously as diagnostic terms across the literature (joint hypermobility, joint laxity, double-jointedness, benign joint hypermobility, hypermobility syndrome and the generic hypermobility term etc). The variants of joint hypermobility and the delineated hypermobility spectrum disorders status (inherent or acquired) with the description criteria are summarised in Table 1[1]

[1] These are the defined descriptions and terms applied throughout this present study of hypermobility in dance.

Table 1: The Variants of Joint Hypermobility and delineated hypermobility spectrum disorder abbreviations (Castori et al., 2017)

Variant of Joint Hypermobility (JH)	Joint hypermobility Abbreviation	Hypermobility spectrum disorder delineation (HSD)	Number and situation of joints	Status	
				Inherent	Acquired
Localised Joint Hypermobility	LJH	L-HSD	≥4 single small or large joints	Possibly	Possibly
Generalised Joint Hypermobility	GJH	G-HSD	≥5 to include four limbs and axial skeleton	Yes	Not usually
Peripheral Joint hypermobility	PJH	P-HSD	hands and feet only	Yes	Not usually
Historical joint hypermobility	HJH	H-HSD	Historically ≥5 to include four limbs and axial skeleton	Yes	Not usually

The assessment for joint hypermobility can however be more complex that merely quantifying joint range of motion. This is because range of motion is also influenced by two factors

i) maturation (i.e. as joints tend to become naturally less flexible across the lifespan,
ii) Sex and endocrinal indicators (e.g. female hormones can also have an influence diminishing the tensile properties of connective tissue) (Remvig, Jensen, & Ward, 2007).

In light of this the application the scoring of the Beighton criteria has been developed since its inception (1973) and it has been modified and validated across different stages of maturation whereby different cut off points are proposed for distinct developmental ages and stages (Juul-Kristensen et al., 2017). Alongside the Beighton criteria, further tests have also been applied to determine joint hypermobility in children. These include the foot posture indicator (FPI) that is an additional and valid test to determine lower limb joint hypermobility in children as there is a strong influence of age on foot and ankle complaints in people with hypermobility (Nicholson, Pacey, Tofts, Munns, & Adams, 2014).

Finally, the retrospective BS 5PQ (Bulbena et al., 2014) and HJH questionnaires (Castori et al., 2017) founded on the Beighton criteria scoring have also been developed and refined to mitigate for individually through diminished flexibility due to age or injury (Castori et al., 2017).

2.2.2 The prevalence of Joint hypermobility

Inherent joint hypermobility can be asymptomatic which makes it problematic to estimate its prevalence because it is not usually tested for and is only registered within health reporting systems when something becomes problematic (Remvig et al., 2011). The estimation is that it occurs in a range of 5-57% of the population, due to its heritable status the frequency is also influenced across ethnic groups within different populations (Juul-Kristensen et al., 2017). The prevalence of GJH (which is the most severe within the HSDs) is reported to vary from between studies, for example in 2019 Jung et al report the prevalence of GJH as 2–57% (Junge et al., 2019) whereas Hakim and Grahame report the prevalence as 10-25% (Hakim, A., & Grahame, 2003) The range depending heavily on the assessment criteria and the population studied (Remvig, Jensen, & Ward, 2007a, 2007b). Studies reporting prevalence of JH regularly include for example study samples from specific sports settings including dance, ethnic groups, age specific groups or clinical groups (Junge et al., 2019). Whilst it is important to understand the prevalence of joint hypermobility (JH) and hypermobility spectrum disorders (HSDs) in specific settings caution must be made to avoid for example issues with reporting relating to influences on joint hypermobility from for example acquired joint hypermobility (LJH) that may occur within sports specific groups such as dance. Also bio-banding for biological maturation needs to be considered within age specific groups for HSDs such as GJH and PJH as we know that age is not a determent of physical maturation (Cumming et al., 2012; Malina, Rogol, Cumming, Coelho E Silva, &

Figueiredo, 2015). Whilst disputed by some (Castori et al., 2017) one study of 46,000 adults using the BS-5PQ (5 part retrospective questionnaire of reporting) did mitigate for influences of physical maturation (i.e. reduction in flexibility due to ageing) in a general population of adults (Bulbena et al., 2014) and the prevalence of GJH was reported as 18%. (Junge et al., 2019).

2.2.3 Joint Hypermobility with further complications

Whilst the spectrum of hypermobility disorders (HSDs) can present as asymptomatic and harmless or with symptoms that are restricted to the musculoskeletal system (Castori et al., 2017, p. 154) the literature does suggest that within the spectrum joint hypermobility (JH) can indeed also arise with further complications that are not necessarily restricted within the musculoskeletal system. These include anxiety disorders and depression (Bulbena-Cabré & Bulbena, 2018; Marco Castori & Colombi, 2015; Pasquini et al., 2014; Mark C. Scheper et al., 2016) functional gastro-intestinal disorders and manifestations (Beckers et al., 2017; Castori, Morlino, Pascolini, Blundo, & Grammatico, 2015), urinary tract infections and bladder dysfunctions (Adib et al., 2005), orthostatic tachycardia and dystonia (Kanjwal, Saeed, Karabin, Kanjwal, & Grub, 2010), and dysautonomia (Jacob & Grubb, 2012). Positive associations with JH and attention deficit hyperactivity disorder (ADHD) and the Autism spectrum have also been made (Castori & Colombi, 2015). Whilst clearly more complex, associations between this unique phenotype and other mental illnesses have also been made for example psychopathological compulsive disorders (e.g. obsessive-compulsive disorders, OCD) and some addictive behaviours (tobacco and alcohol) (Baeza-Velasco, Pailhez, Bulbena, & Baghdadli, 2015). The suggested rational for this are related to, genetic makeup (Pujana et al., 2001), dysautonomia (a dysfunction of the ANS), body awareness as in increased interoception (i.e. the sense of the internal workings of the body) (Mallorquí-

Bagué, Bulbena, Pailhez, Garfinkel, & Critchley, 2016), and decreased proprioception (i.e. the sense of the body in space) (Jacob & Grubb, 2012; N. Mallorquí-Bagué et al., 2015).

Understanding that JH can be asymptomatic within the musculoskeletal system means that these additional associations could therefore potentially manifest as primary issues before any musculoskeletal micro and macro trauma. This also implies that the links and interchangeability between the primary and secondary associations within JH can potentially go unfounded. Associations between joint hypermobility and non – musculoskeletal disorders are also further and frequently discussed as a constituent of other more complex syndrome(s) and conditions that also all present with joint hypermobility (Castori et al., 2017). These are heritable connective tissue disorders (HCTDs) of which there are two hundred and they can change the look and growth of skin, bones, joints, heart, blood vessels, lungs, eyes, and ears. Some also change how the connective tissues work. The most common are;

a. Marfan syndrome (MFS) which can affect the heart, blood vessels, lungs, eyes, bones, and ligaments and has a very characteristic tall and thin body type, with long arms and legs.
b. Osteogenesis Imperfecta (OI) which can cause bones to break easily and often for no obvious reason.
c. Ehlers-Danlos syndromes.

The association between HSDs, JH, HCTDs and the further complications discussed in the literature are shown in figure 1 below

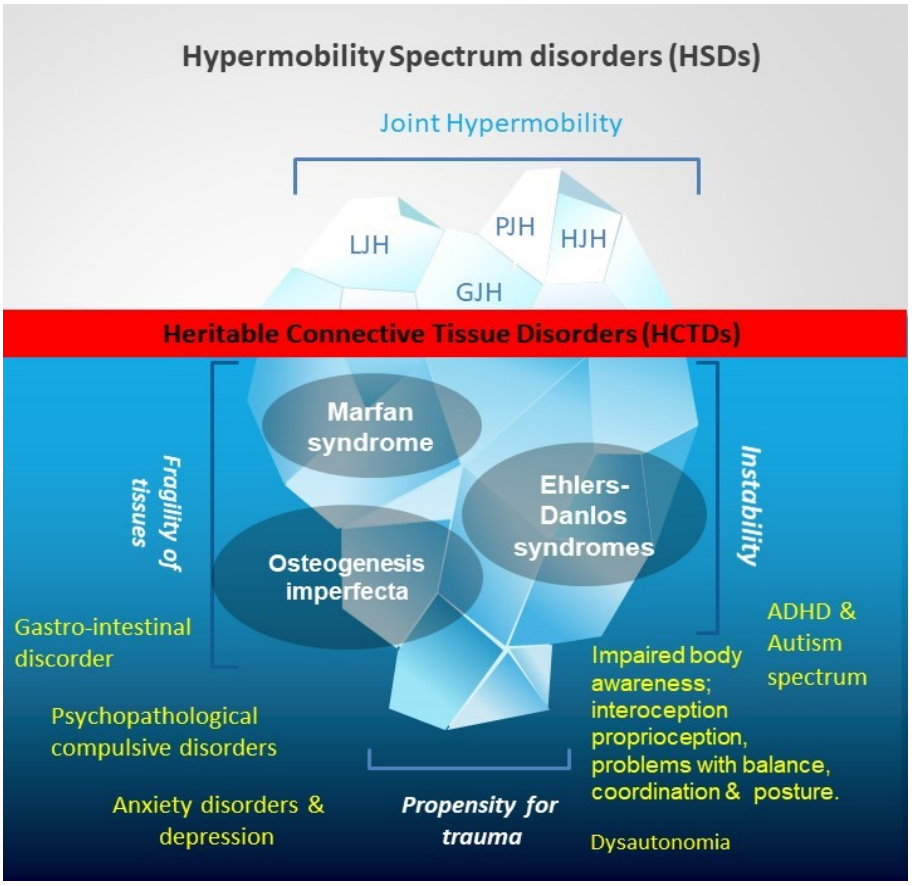

Figure 1. The association between HSDs, JH, HCTDs and the further complications

Marfan Syndrome (MFS) is currently well defined and recognisable through clinical observation of a collection of features associated with Marfanoid habitus. These are, a very tall slender body with unusually long extremities, a span: height ratio ›1.03, upper segment: lower segment ration ‹0.89, hand: height ratio › 11% and foot: height ratio ›15% (Keer & Grahame, 2003, p. 19). Marfan syndrome and Ehlers Danlos have overlapping features in that they both present with joint hypermobility (JH) and its associated clinical features and they often also both have the characteristic Marfanoid habitus. The distinguishing characteristic between these two HCTDs is that Marfan syndrome is much more serious and typically has complications that need surgery relating to aeortic root dilation and ectopia lentis (the positioning of the lens in one or both of the eyes is off-center).

Osteogenesis imperfecta (OI) also features JH however it is differentiated as it leads to severe osteoporosis and unexplained broken bones. Unlike JH and Ehlers Danlos and due to the severe nature of their characteristics, both Marfan Syndroms (MSFS and Osteogenesis Imperfecta (OI) would not normally go undetected.

Similar to the HSDs, the Ehlers-Danlos syndrome has also recently been reconsidered as a group and are now re-categorised as thirteen categories under the title Ehlers-Danlos syndromes (EDS).

2.3 Ehlers-Danlos syndromes

The notion that joint hypermobility (JH) was a compounding factor within what we now understand as the complex multi-faceted Ehlers-Danlos syndromes (EDS) only really emerged towards the end of the last century (Malfait et al., 2017). Not dissimilar to the development of thinking around joint hypermobility, and mainly as a result of progress in genetic science and sequencing, there have over the years been several changes in thinking around the grouping and categorisation of the entities within EDS.

Ehlers-Danlos was first characterised as a distinct heritable entity within the heritable connective tissue disorders known at that time and in 1908 it was named after two physicians; Edvard Lauritz Ehlers (1863–1937) and Henri-Alexandre Danlos (1844–1912) (who also observed that along with the extreme flexibility, the stretchiness and fragility of the skin were also common features). Until recently this was understood as one singular syndrome under the name Ehlers-Danlos syndrome (Parapia, Jackson, 2008). That said, within the Ehlers-Danlos syndrome, sub-types were also identified and there have again been several changes in opinion as to their number and categorisation.

In 1988 the Berlin Nosology (1988) recognised eleven subtypes within the Ehlers-Danlos syndrome. As a unifying component, the Beighton test as a known and reliable test for JH was applied within the diagnostic criteria . Ten years on and arising from developments in molecular and biochemical techniques, the Villefranch Nosology (1998) and categorisation was applied. As a result , the Ehlers-Danlos syndrome subtype classifications were reduced from eleven to six. Between the Berlin (1988) and Villefranc (1998) categorisations further clinical observations had been made and were used as additional markers to enable clearer diagnosis. These were defined as both major and minor criteria (including the Beighton 1973 criteria) and were applied in the diagnosis. In addition and where possible biochemical and molecular tests were also now introduced in the diagnostics (Castori et al., 2017).

For the past twenty years both clinical diagnoses and research have been based on the refined six subtypes of the Villefranch Nosology (1998). Following from this, advancements in medicine, genetics, the discovery of new gene sequencing and also supplementary observations and associations of the Ehlers-Danlos syndrome with other systems not related to the musculoskeletal again highlighted the need for:

a) Clearer differentiation between the six Villefranch Nosology (1998) subtypes.

b) More up to date improved diagnostic criteria.

In 2017 a comprehensive review of all the Ehlers-Danlos syndrome literature (Castori et al., 2017)by a multidisciplinary team of experts brought forth a revised classification framework that named thirteen subtypes and resulted in additional changes including the name to Ehlers-Danlos Syndromes (EDS). Ehlers-Danlos Syndromes (EDS) are the most common of a collection of heritable disorders of connective tissue (HDCTs). HDCTs are

genetic disorders that are over 200 (n>200) in number. They affect the connective tissue matrix proteins (Baeza-Velasco, Pailhez, Bulbena, Bagudadi, 2015; Castori et al., 2017). The disordered fibrous protein genes and connective tissue present as laxity and distensibility in the ligaments (Keer & Grahame, 2008, p2). This increased distensibility of ligaments in turn presents as laxity and hypermobility in the joints (JH) and this constitutes as an important unifying feature across the newly categorised thirteen subcategories of EDS (Baeza-Velasco, Pailhez, Bulbena, Bagudadi, 2015; Malfait et al., 2017). These are presented with their abbreviated terms in Table 2

Table 2: Clinical Classification of EDS adapted from Castori et al (2017)

Clinical EDS subtype	**Abbreviation**
i. Classical EDS	cEDS
ii. Classical-like EDS	clEDS
iii. Cardiac-valvular EDS	cvEDS
iv. Vascular EDS	vEDS
v. Hypermobile EDS	hEDS
vi. Arthrochalasia EDS	aEDS
vii. Dermatosparaxis EDS	dEDS
viii. Kyphoscoliotic EDS	kEDS
ix. Brittle Cornea syndrome	BCS
x. Spondylodysplastic EDS	spEDS
xi. Musculocontractual EDS	mcEDS
xii. Myopathic EDS	mEDS
xiii. Peridontal EDS	pEDS

2.3.1 Recognising and assessing EDS

The EDS have been identified for some time now through both their clinical and genetic heterogeneity (Castori et al., 2017). The genetic basis for twelve of the thirteen subtypes (1-4 and 6-13 in table 2) is currently known and provides important insight to classification and diagnosis. The genetic basis for no 5 hypermobile EDS (hEDS) however remains unknown (Castori et al., 2017, p. 10). This means that the diagnosis for hEDS remains to date through applied clinical observation criteria which were formulated from combinations of historical research findings and subsequent devised criteria. These included the 9-point Beighton criteria (1973) that is applied to define the presence of HSDs combined with further criteria, such as the Brighton criteria (1998) that originate from the Villefranch (1998) recategorization of EDS. Based on a typical history of at least seventy-seven clinical features (symptoms and associations) the Brighton criteria were drawn and complied from clinical findings and the literature (Keer & Grahame, 2003, p. 11). The Beighton & Brighton criteria were applied as combinations of major and minor criteria and were until recently the most commonly used tool for the identification of hEDS (Malfait et al., 2017).

The application of criteria to determine EDS is quite complex and again not always consistent across the literature (Malfait et al., 2017) however HSDs and joint hypermobility (JH) are common features often providing the initial insight to the presence of EDS. As previously discussed JH can be asymptomatic yet it can also potentially present with further complications (Jacobs et al., 2018; Keer, Grahame, 2003). The further complications that are beyond the musculoskeletal system and are now considered as multi-systemic disease within EDS (Colombi, Dordoni, Chiarelli, & Ritelli, 2015). This also suggests that if issues arising from EDS are to be properly addressed ideally a multidisciplinary team of clinicians is required (Jacobs et al., 2018), something that is not always readily available. Evidence

suggests that often it is only at the point of EDS becoming symptomatic as a mal-function of joint hypermobility JH within the musculoskeletal system (for example within non-specific low back pain NSLBP, arthralgia or knee pain) that it is noticed however not necessarily completely diagnosed (Allegri, Montella, Salici, Valente, Marchesini, Compagnone, & Fanelli, 2016). The literature reports that alongside the clinical features, and very similar to JH, EDS can have complications relating to quality of life and daily activities. These include neuromuscular (Donkervoort, Bonnemann, Loeys, Jungbluth, & Voermans, 2015; Rombaut, De Paepe, Malfait, Cools, 2010; Rombaut, Malfait, De Wandele, Mahieu, Thijs, Segers, De Paepe, 2011a; Rombaut, Malfait, De Wandele, Thijs, Palmans, De Paepe, 2011b; Rombaut, Malfait, DeWandele, Taes, Thijs, De Paepe, 2012; Voermans, van Alfen, Pillen, Lammens, Schalkwijk, Zwarts, van Rooij, Hamel, 2009), fatigue and chronic pain (Hakim, A; Keer, R; Grahame, 2010; Voermans, Knoop, van de Kamp, Hamel, Bleijenberg, 2010), gastrointestinal (Marco Castori et al., 2015) and psychopathological and psychiatric (Bulbena-Cabré & Bulbena, 2018; Bulbena et al., 2017a; Núria Mallorquí-Bagué et al., 2016; Sinibaldi, Ursini, & Castori, 2015).

With this in mind and with a view to further clarity around the terminology associated with hypermobility, the consortium of esteemed experts across all of the disciplines that associate with EDS met to revisit and revise the classification terms for EDS (Malfait et al., 2017). The classification was undertaken based on both molecular genetics in addition to clinical observations (wherever possible). Whilst this is still a working progress (with acknowledgement that further work is to be undertaken) Ehlers-Danlos syndromes (EDS) are now classified as a spectrum of thirteen subtype disorders to which hypermobility belongs, and they are all genetic in origin (Malfait et al., 2017). An abbreviated summary of the Malfait et al 2017 reclassification is presented below in Table 3 with indicative complications

and signs and inheritance (molecular) involvement for the thirteen subtypes. The associations between the Heritable Disorders of Connective Tissue (HDCTs) and newly categorised Ehlers Danlos Syndromes (EDS) subtypes and Hypermobile Spectrum Disorders (HSD) discussed in this chapter are also presented in figure 2.

Table 3: Abbreviated version of the 2017 reclassification with indicative complications and signs and molecular involvement adapted from (Malfait et al., 2017)

Clinical categorisation of EDS subtype	Abbreviation	HSD sub-type	Inheritance	Complications & signs		
Classical EDS	cEDS	GJH	AD[2]	Recurring dislocations, (shoulder, patella, temporomandibular joints)	Flexible flat foot Muscle hypotonia,	Pain, Fatigue Delayed gross motor development
Classical-like EDS	clEDS	GJH	AR[3]	Muscle hypotonia, Delayed gross motor development	With or without recurring dislocations, (shoulder, patella, temporomandibular joints	Pain, Fatigue
Cardiac Vulvar EDS	cvEDS	GJH or PJH	AR	Joint dislocations Easy Bruising Atrophic scarring	Hallux Valgus Pectus deformity (excavatum)	Propensity for arterial rupture at adult age Flexible flat foot
Vascular EDS	vEDS	GJH or PJH	AD	Arterial rupture at young age Uterine rupture during pregnancy Varicose veins at early age	Congenital hip dislocation Tendon and muscle rupture	Colon perforation CCSF
Hypermobile EDS	hEDS	GJH or HJH	AD	Velvety skin Atrophic scarring Mild skin hyperextensibility Unexplained stretch marks	Pelvic floor, rectal and/or uterine prolapse Arm span-height ≥1.05 Sleep disorders	Arachnodactyly Anxiety Depression Fatigue Dysautonomia
Arthrochalasia EDS	aEDS	GJH	AD	Congenital bilateral hip dislocation	Kyphosis Easily bruise Tissue fragility	Multiple dislocations Osteopenia

[2] AD: in an autosomal dominant disease, if you get the abnormal gene from only one parent, you can get the disease. Often, one of the parents may also have the disease.

[3] AR: in an autosomal recessive disease, you inherit two mutated genes, one from each parent. These disorders are usually passed on by two carriers.

Clinical categorisation of EDS subtype	Abbreviation	HSD sub-type	Inheritance	Complications & signs		
Kyphoscoliotic EDS	kEDS	GJH	AR	Hypotonia Dislocations Kyphoscoliosis	Knees, shoulders & hips in particular Hernia	Marfanoid habitus Pectus deformity
Brittle Cornea Syndrome	BCS	PJH	AR	Severe eye problems Deafness	Flat feet Halux valgus	Hip dysplasia
Spondylodysplastic EDS	sEDS	PJH	AR	Short stature Muscle hypotonia	Delayed motor development Osteopenia Delayed cognitive development	Bowing of the limbs Skin hyperextensibility Flat foot
Musculocontractural EDS	mcEDS	PJH	AR	Club foot Skin hyperextensibility Recurrent dislocations	Misshapen fingers Easily bruise	Chronic constipation Glaucoma Pectus deformity
Myopathic EDS	mEDS	PJH	AD or AR	Muscle hypotonia Problems with knees, hips and elbows	Myopathy	Motor development delay
Peridontal EDS	pEDS	GJH	AD	Peridontitis Bruising	Hernias Fragile skin	Skin hyperextensibility Infections
Dermatospraxis EDS	dEDS	GJH	AR	Tissue fragility Lax skin	Kyphosis Easily bruise	Short limbs hands and feet Tissue fragility

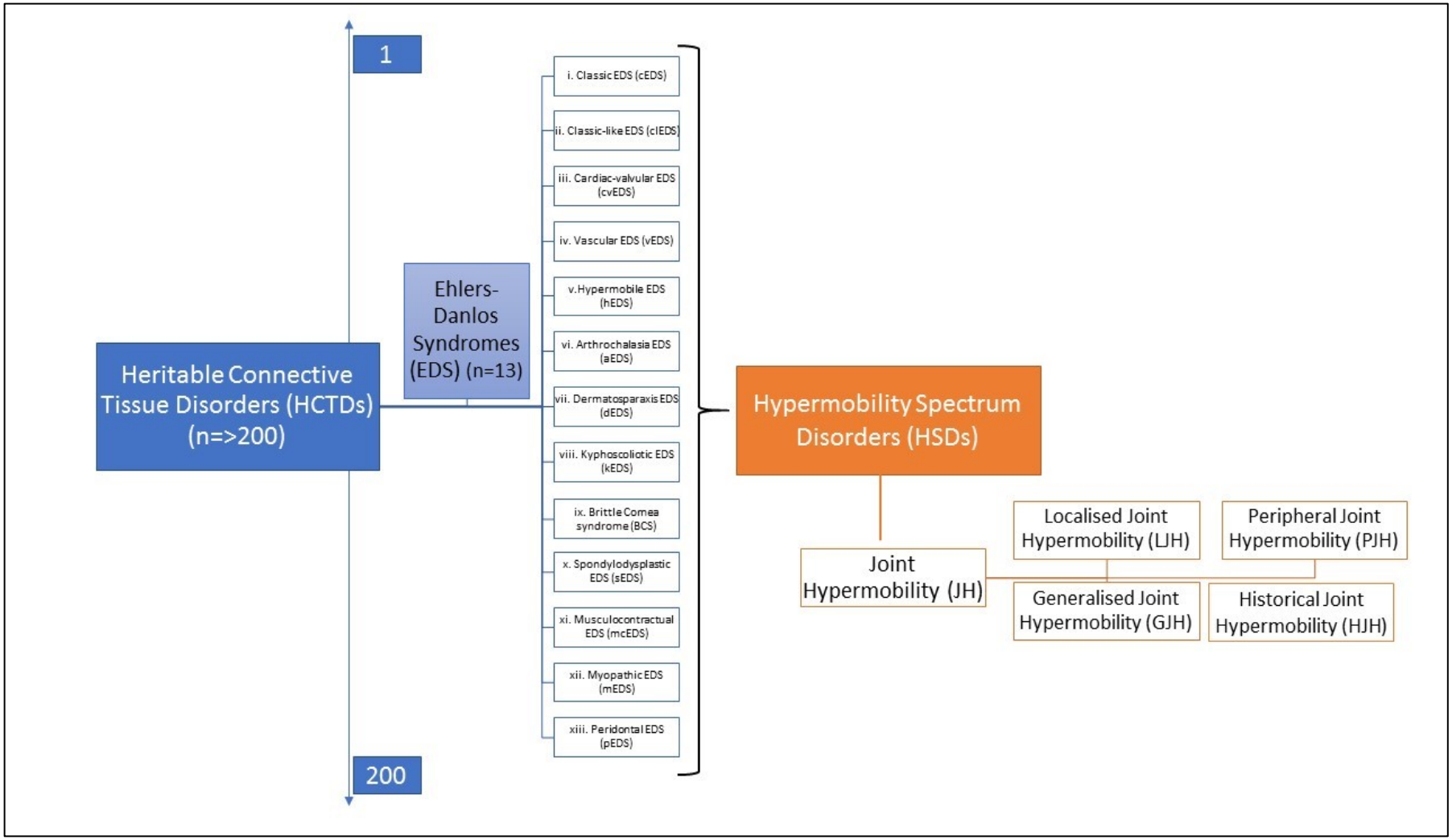

Figure 2. The association across HDCTs, EDS and HSD

Of the thirteen sub-types within the Ehlers-Danlos syndromes (EDS) the hypermobile type of EDS (hEDS) is reported as the most common hereditary connective tissue disorder (HCTD) (Keer & Grahame, 2003; B. Tinkle et al., 2017). As with the categorisation of hereditary spectrum disorders HSDs and joint hypermobility (JH), until recently there has also been a lack of clearness about the categorisation of hEDS. This is also not aided by the fact that the molecular composition for hEDS has yet to be defined and assessment for this subtype remains through clinical observations. This means that there is again a significant amount of contradiction in the terminology used in the research and literature around hEDS (Malfait et al., 2017). The confusion arises from the interchange of terms such that range from Joint Hypermobility Syndrome, Hypermobility Syndrome, Hypermobility, Ehlers-Danlos Type III and Ehlers-Danlos Syndrome Hypermobility Type. As of 2017 these have all been unified under the term Hypermobility EDS (hEDS) and the other terminology is considered as outdated (Castori et al., 2017). In line with these developments , the 2017 terminology (Bloom, Byers, Francomano, Tinkle, & Malfait, 2017; Castori et al., 2017) is also applied throughout this study.

Hypermobility EDS (hEDS) (a.k.a. Hypermobility, Joint Hypermobility Syndrome, Hypermobility Syndrome, Ehlers-Danlos Type III and Ehlers-Danlos Syndrome Hypermobility Type) was first described in 1967 by Kirk , Ansell and Bywaters (Grahame, 1990). It was (and sometimes still is) characterised the literature as a heritable connective tissue disorder, with primary identification as having generalised joint hypermobility (GJH), related musculoskeletal manifestations, and a milder involvement of the skin (B. Tinkle et al., 2017). This description however is based on former identification criteria (Brighton and Villefranche) and whilst it lacks a robust and distinguishing differentiation between hEDS and GJH it remains to be one that is applied (Castori & Colombi, 2015). This is therefore

potentially a cause of confusion as the description is not dissimilar to what we now understand as JH (within the HSD). We now know that hEDS also has considerably more features to it that negatively affect quality of life (e.g. chronic pain, chronic fatigue, anxiety and dysautonomia (Tinkle et al., 2017). In addition, when first described, many rheumatologists viewed what we now describe as the hypermobility spectrum disorders (HSDs) and JH sceptically and were therefore also sceptical of the more complex hEDS. Clinicians regarded 'bendy-bodies' more as a clinical curiosity than a rheumatic disease (Grahame, 1990, p199). They were a phenomenon regularly associated with circus acts and performance spectacles and were often (apparently) asymptomatic. In such, they were often overlooked, disregarded, discounted and on occasion regarded as an asset (Keer and Grahame, 2003). It is the hEDS subtype of EDS alongside GJH that is also commonly seen within special populations such as aesthetic sports and dance(Armstrong & Greig, 2018a; Grahame, 1990).

2.3.2 Assessing for hEDS

Assessing for hEDS is complex and requires skilled clinicians. There is currently no "gold standard" laboratory test to confirm or refute the diagnosis, it is anticipated that future research will lead to further revisions of the clinical criteria currently applied in the characterisation of hEDS (Malfait et al., 2017). In turn this may in the future necessitate further review of the relevant medical literature, however at this point in time it is the most up-to-date assessment criteria available. It is important, as this is a clinical diagnosis, that the patient's presentation does not represent one of the many other disorders of connective tissue (e.g. HSDs, Marfan Syndrome, Osteogenesis Imperfecta OI). This means that the assessment should be undertaken by a clinician who is experienced with the criteria as well the historical and clinical presentation of other HCTD and their diagnoses (Malfait et al., 2017). The

clinical diagnosis of hEDS requires the simultaneous presence of criteria 1 and 2 and 3 which are presented below as set out by Malfait et al 2017.

Table 4. Criteria for the clinical diagnosis of hEDS Malfait et al 2017

Criteria 1:

1. Generalized Joint Hypermobility
   (GJH). As GJH is a constitutional trait that is strongly influenced by acquired and inherited conditions (e.g., sex, age, past-traumas, co-morbidities, etc.), some minor adaptations to the cut-off of five should be considered for the diagnosis of hEDS. The International Consortium on the Ehlers–Danlos Syndromes (Malfait et al., 2017) proposes
   1) ≥6 for pre-pubertal children and adolescents,
   2) ≥5 for purbutal men and women >50 years of age
2. In individuals with acquired joint limitations (e.g. past surgery, wheelchair, amputations, etc.) that will affect the Beighton score calculation, the assessment of GJH can be carried out using historical information and the five-point questionnaire (BS-5PQ)
3. For scores that are lower than the Beighton score recommendations then the assessment of other joints is also often considered.

Criteria 2:

Two or more of the features (A–C) MUST be present

Feature A: systemic manifestations of a more generalized connective tissue disorder (a total of five must be present)

1. Unusually soft or velvety skin
2. Mild skin hyperextensibility
3. Unexplained striae such as striae distensaeor rubrae at the back, groins, thighs, breasts and/or abdomen in adolescents, men or prepubertal women without a history of significant gain or loss of body fat or weight.
4. Bilateral piezogenic papules of the heel
5. Recurrent or multiple abdominal hernia(s) (e.g., umbilical, inguinal, crural)
6. Atrophic scarring involving at least two sites and without the formation of truly papyraceous and/or hemosideric scars as seen in classical EDS
7. Pelvic floor, rectal, and/or uterine prolapse in children, men or nulliparous women without a history of morbid obesity or other known pre- disposing medical condition
8. Dental crowding and high or narrow palate
9. Arachnodactyly, as defined in one or more of the following: (i) positive wrist sign (Steinberg sign) on both sides; (ii) positive thumb sign (Walker sign) on both sides
10. Mitral valve prolapse (MVP) mild or 10. Arm span-to-height ≥1.05 greater based on strict echocardio- graphic criteria
11. Aortic root dilatation with Z-score >þ2

Feature B: positive family history, with one or more first degree relatives independently meeting the current diagnostic criteria for hEDS.

Feature C: musculoskeletal complications (must have at least one):

1. Musculoskeletal pain in two or more limbs, recurring daily for at least 3months
2. Chronic, widespread pain for ≥3 months
3. Recurrent joint dislocations or frank joint instability, in the absence of trauma (a or b)
   a) Three or more atraumatic dislocations in the same joint or two or more atraumatic dislocations in two different joints occurring at different times
   b) Medical confirmation of joint instability at two or more sites not related to trauma

Criteria 3:

All the Following Prerequisites MUST be met

1. Absence of unusual skin fragility, which should prompt consideration of other types of EDS
2. Exclusion of other heritable and acquired connective tissue disorders, including autoimmune rheumatologic conditions. In patients with an acquired connective tissue disorder (e.g. lupus,rheumatoid arthritis, etc.), additional diagnosis of hEDS requires meeting both Features A and B of Criterion 2. Feature C of Criterion 2 (chronic pain and/or instability) cannot be counted towards a diagnosis of hEDS in this situation.
3. Exclusion of alternative diagnoses that may also include joint hypermobility by means of hypotonia and/or connective tissue laxity. Alternative diagnoses and diagnostic categories include, but are not limited to, neuromuscular disorders (e.g., myopathic EDS, Bethlem myopathy), other HCTD (e.g., other types of EDS, Loeys–Dietz syndrome, Marfan syndrome), and skeletal dysplasias (e.g., OI). Exclusion of these considerations may be based upon history, physical examination, and/or molecular genetic testing, as indicated.

2.3.3 The elusive nature of hypermobility

Anecdotal evidence also suggests that JH and hEDS frequently find their way in to orthopaedic and emergency departments because of the failures and malfunctions they can cause within the musculoskeletal system (for example dislocations and tendon/ligament ruptures) and this raises some issues in both diagnosis and treatment. First, doctors and others encounter and are trained to examine for injury that regularly involves reduction of joint mobility rather than for an increased range (Grahame 1990), therefore hypermobility is commonly missed. Secondly, whilst orthopaedic specialists can treat musculoskeletal failures they are still not always the best informed to recognise the fuller implications of JH and /or EDS (personal communication with orthopaedic surgeon, University of Crete, 2016). Thus, the condition may be delayed or never reach the rheumatologist who is the specialist to diagnose and manage the condition. Likewise, the early connections between GJH and psychiatric disorders came as a secondary and initially anecdotal observation by psychiatrists working with their patients (Bulbena et al. 2015 and further personal communication with researcher Dr. Carolina Baeza-Velasco who works with the team of psychiatric practitioners who have been the most prolific in research in this area, 2017). This evidence and the further range of complications associated across HSDs again highlight how they may be mis or under diagnosed (Allegri, Montella, Salici, Valente, Marchesini, Compagnone and Fanelli, 2016; Rombaut et al., 2015) making Grahame's (2003) observation of it easily be *missed* very plausible.

2.4 Hypermobility as an asset

Hypermobility (hEDS) and joint hypermobility as one of its key features can present as asymptomatic within the musculoskeletal system for the whole of a lifespan

of development (Castori & Colombi, 2015; Keer & Grahame, 2003). It can also appear as symptomatic to the musculoskeletal system only at certain times during a lifespan, for example during adolescent growth spurts it is known to manifest in unexplained pain in the legs (Adib et al., 2005; Ghibellini, Brancati, & Castori, 2015b; Maillard & Pilkington, 2016). This pain is often classed as 'growing pains' with the underlying 'hypermobility' going unfounded missed or misdiagnosed.

A range of motion in some joints that goes beyond normal physiological parameters is also and often seen as an asset to those who engage in aesthetic sports and the arts such as dance and music (Castori & Colombi, 2015; Foley & Bird, 2013; Keer & Grahame, 2003; Grahame & Jenkins, 1972; Skwiot, Śliwiński, Milanese, & Śliwiński, 2019). The once viewed 'supernatural and ethereal' artists do now come under the category of 'aesthetic athletes' and this is as a direct result of developments in sports and dance science, (Day, Koutedakis, & Wyon, 2011a; Yiannis Koutedakis & Jamurtas, 2004). Progressing choreography and performance demands make the physiology of aesthetic athletes as important as their skill development. Strength and flexibility being the two most sought after features in elite ballet dancers (McCormack et al., 2019), dancers are irrefutably subject to the same unyielding physical laws and strength requirements as other athletes that do not perhaps demand comparable or extreme flexibility (Yiannis Koutedakis & Jamurtas, 2004; Russell, 2013). Hypermobility enables the demonstrations of extreme ranges of motion allowing dancers to effortlessly lift their legs higher, bend and contort their spines in all directions, hyperextend their ankles and knees and meet a desired and often choreographically required bio-aesthetic (e.g. choreographic work *Duo* by Forsythe 1996).

Industry demands do undoubtedly influence both the young and older dance student and their training regimes as they struggle to emulate seasoned professionals who have this bio-aesthetic. We also know that this aesthetic may come naturally, or it may be achieved to some extent through a considerable amount of hard work and effort. Sadly, 'naturally occurring' flexibility is often 'valued' over hard work and is considered as an extra ability or asset (Foley & Bird, 2013a)making young dancers and athletes with this capacity to be seen as 'full of potential' (McCormack, 2010, p. 5).

We now know from reviewing the literature however that genetically founded hypermobile body-types naturally occur as a result of their specific connective tissue makeup. We also know that potentially asymptomatic or misdiagnosed joint hypermobility and hEDS can manifest at vulnerable stages in physical development (e.g. adolescence) and become mechanisms for injury if the fragile tissues associated with hEDS and JH are not respected (Ekegren et al., 2014; Mitchell, Haase, Malina, & Cumming, 2016; Morris et al., 2017; Soloman, Soloman, & Micheli, 2017; Yau et al., 2017). We are also now coming to understand that the symptoms of hypermobility are not restricted to the musculoskeletal and do also manifest in what is now described as a new *"neuroconnective phenotype"* or expression of hypermobility (Bulbena-Cabré & Bulbena, 2018, p. 15)that presents with symptoms that are debilitating however the mechanisms for which are not yet clearly defined (Baeza-Velasco, Grahame, & Bravo, 2017; Bulbena et al., 2017b). This suggests that hypermobility, whilst a potential asset, may also play a more menacing and unknown role with regards to in dance and aesthetic sports. Indeed, hypermobility has already been adeptly cited by Grahame, one of the top researchers in this area, as the "Beauty and the Beast

Syndrome" (Grahame in Knight, 2015, p257). It is with this in mind that this current study has been designed and developed to further investigate the paradoxical and seemingly historical entanglement of hypermobility and dance. Where exactly is the entanglement and how might we better understand it is indeed the catalyst for this study. Associations between JH tissue fragility, risk, injury and access to ranges of joint motion that extend beyond normal parameters are unquestionably and paradoxically entangled whilst further implications of GJH now also associate with dysautonomia, emotional and psychological distress and anxiety. Whilst somehow JH is viewed as a valued sign that dancers and aesthetic artists that are full of potential (McCormack et al., 2004) JH is also known to be associated with clumsiness and poor fine motor control, reasons for which young children are often sent to the dance class in the first place. Interestingly this may suggest that '*dance*' and *'hypermobility'* are somehow predestined to coexist. Further entanglement is seated in the fact that safe and healthy practice in dance can serve as protective factors for physical health and also enable emotional expression and thus potentially maintain and increase sound mind-body functioning. Focusing on classical ballet as a very defined, technical and enduring practice where hypermobility appears to be an asset and also prevalent, this thesis will investigate the health risk and embodied experience of hypermobility with a view to further unravelling the relationship between *'beauty*' and the *'beast*'.

Chapter 3 The Principal Methodological and Philosophical Considerations and Approach to the research

The overall literature review in Chapter 2 clearly demonstrates the existence and importance of empirical research with regards to hypermobility and also within the field of dance medicine and science. In line with the development of the research and literature relating to hypermobility in general, the dance literature primarily reports around the physical manifestation and visible markers of the associated disordered connective tissue in a dancer. These are for example, very flexible or double-jointed physiques and associated shape of dancers with extreme ranges of motion in the joints. This physique has become synonymously associated with physical fragility and injury in dance (McCormack, Briggs, Hakim, & Grahame, 2004; Scheper et al., 2013)

Since the 1980's the dance medicine and science literature in general has grown. Taking a similar yet somewhat distinct path to sports medicine and science, dance medicine and science literature now also includes the psychological and social attributes and characteristics of dancers as well as the physical, this is evident for example with the development of the content of journals with impact (e.g. the Journal for Dance Medicine and Science) between the 1980's to present. The dance education literature, (e.g. the Journal for Research in Dance Education) nowadays and increasingly includes empirical research pertaining to the physical, psychological and social attributes of dancers and the associated application of dance medicine and science within the education, pedagogy and training for dancers. Seminal work from a sociological perspective by Wainwright, Turner and colleagues (2004, 2005 &2006)

likewise investigates the fractured identity and embodied vulnerability of the classical dancer, taking a retrospective viewpoint across a dancer's career in order to revisit and challenge established beliefs about dancers with up-to-date empirical evidence. Wainwright et al. (2004) focus on the epiphanies of injury, ageing and the way in which the social world of the dance environment shapes and may even threaten a dancer's personal identity (p328). More recently, Pickard (2013) also reconsidered Ballet as "*a social practice"* (p16) one that operates as a bi-directional influence between the dancing and the dancer. Pickard (2013) argues, "…*there is a strong connection between the size, shape and aesthetic of the ballet body and identity"* (p16).

Whilst there is clearly some debate in the dance literature around the complexities associated with hypermobility as a unique yet very common feature within the dance environment (Foley & Bird, 2013a; McCormack, Briggs, Hakim, Grahame, & Grahame, 2004; Rietveld, 2013), this work remains primarily associated with the physical injury reporting and prevention context. The explicit presentation and full contribution of hypermobility as an in-built and encapsulated feature within the aesthetic and culture for dance and in particular classical ballet is to date, not fully conferred, presenting as a gap within the dance medicine, education, and science literature. Demonstrating a need for further investigation not only around the physical characteristics, but also the psycho-social characteristics and connotations of hypermobility within dance. With this a deeper understanding of the experience of hypermobility in dance, and in particular the classical ballet environment, there may also come a clearer knowledge about the complexities of hypermobility in general and how it may be managed.

From the perspective of a dance practitioner, the drive and the catalyst for this thesis was indeed to find out how the heritable disorder of hypermobility not only manifests but is understood in the dance environment. Of particular interest were the manifestations of hypermobility from the perspective of the embodied experience and development of the dancer with hypermobility. Within this, the attitudes towards hypermobility of those who operate in this environment, to include the dancers and dance teachers were also of interest.

3.1 Epistemological and ontological perspective of the research

Situated within the social sciences the focus of this study was to appreciate and make reasonable claims about human understanding and behaviour regardless of paradigmatic positioning (Johnson & Onwuegbuzie, 2004). Within the research paradigm for social sciences there are four elements to a methodological approach that interlink to inform ways of knowing and understanding. These are; the underlying assumptions (epistemological and ontological), theoretical perspective, methodology and the methods (Crotty, 1998, pp3-4). In this next section the framework, theoretical underpinning, methodological approach and overall design of the study are discussed.

Assumptions that underpin all research are epistemological and ontological (Gray, 2014; Robson, 2011). The epistemological deals with the nature and forms of knowledge and how we can gain access to these (Cohen, Manion, & Morrison, 2007, p7; Maxwell, 2013, p42). The epistemological assumptions therefore provided the philosophical background to the research for deciding what kind of knowledge would be legitimate and adequate to answer the underpinning research question (Gray, 2014, p19). The epistemology, concerned with truth , beliefs

and the justification pertaining to how knowledge can be created, acquired and communicated and is "*what it means to know"*(Scotland, 2012, p9), in this research the knowing is with reference to hypermobility within the dance environment. The ontological deals with the study of reality and being (Crotty, 1998, p. 10, Maxwell, 2013, p42). Ontological assumptions are concerned with what constitutes reality, or the *"what is"* (Scotland, 2012, p9), in this research the reference is again to hypermobility within the dance environment. The overall research and thesis is therefore established as a case study of hypermobility within the classical ballet narrative.

The primary aims that underpin this case study are to discover if, how and is therefore ***to what extent is hypermobility known and understood in the classical ballet environment?***

Whilst asking 'how hypermobility is known and understood in the dance environment?' it was important to appreciate what we mean by 'knowing and understanding'. The word understanding indicates a bodily connection to being upright (Münker, 2019) and derives from the old English language meaning to be 'between' or 'among'. Münker, (2019) uses somatic practice beliefs (Feldenkrais) and enactive relationality to provide a convincing rational for an embodied explanation of understanding as

> *"...how we are and how we bring ourselves in relation to our environment creates our understanding and forms our experiences and conceptions of the world."*
>
> (Münker, 2019, p1)

Opposing views (reductionist and non-reductionist) of knowing and understanding are also eminently presented by Paulina Silwa in 2015 (at the meeting

of the Aristotelian Society and published in proceedings from this meeting). Based on Silwa's reductionist/non-reductionist explanation; a reductionist view of hypermobility for example would rely on quantitative data and testimony such as scientific facts that are presented for example in research publications. However, a non-reductionist would argue that "…*testimony is a source of understanding, but not always of very deep understanding"* (Sliwa, 2015, p16). Therefore, to 'really understand' hypermobility, testimony and scientific fact alone may present as potentially insufficient evidence. A non-reductionist view of hypermobility on the other hand (again according to Silwa's theory) would provide a description of an epistemic phenomenon (such as that of the experience of hypermobility within the dance environment for example). Experience indeed plays a pervasive and important role in ordinary epistemic practice (including scientific practice) and one that traditional epistemology with its single-minded focus on knowledge may hitherto neglect (Sliwa, 2015, p16).

Complex ways of knowing and understanding are indeed pertinent to this research as it was equally important for the researcher to understand to what extent hypermobility is understood by those in the dance environment and also how hypermobility is experienced by the dancers who present with it. It was also very important to gain insight as to how the exchange and processing of information in the dance environment relative to the body such as; spatial patterning and technical details relating to where the body is space might be affected by hypermobility. Meanwhile, solely applying scientific rigour to social and cultural contexts such as learning could potentially also be problematic as this may lead to the oversimplification of individual experience by looking for rules and patterns based on

numbers and statistics alone (Peim, 2018). Therefore including the dancers' (with hypermobility) experience of learning and performing dance, and consequently also the teachers' experience of working with dancers with hypermobility was important, however it was significant to initially to gain a deeper understanding of how hypermobility (as a genotype) is phenotypically expressed in a dancer in the dance environment.

Empirical knowing is on the other hand reductionist and evidenced based and enables the testing of a hypothesis. It would for example permit testing for conditions such as hypermobility through the application of standardised and validated tests such as the Beighton and Brighton criteria. Sliwa (2015) also presents a convincing debate for the reductionist viewpoint whilst also providing a thrifty account of 'understanding'. In doing so she pertains that the non-reductionists approach affects that 'knowing' is not sufficient for 'understanding' and that a

> *"…lack of understanding need not generally imply lack of knowledge: rather it may just be that the agent fails to 'cognitively grasp' the known proposition in the special way that's required for understanding."* (p15)

This suggests that a non-reductionist approach relies on the somewhat subjective notion that cognition can be 'grasped' whereas the reductionist viewpoint acknowledges that knowledge is seeded in facts and comes in varying degrees and amounts. The notion that knowledge is measurable in degrees and amounts also supports the recognition and role of expertise in providing a deeper understanding and that the level of understanding can and will impact on practice. This meant adopting both reductionist and non-dualistic, non-reductionist approaches in this research in order to really understand the potential impact of hypermobility within the teaching

and learning environment for dance. It meant considering the facts surrounding (knowing and understanding) and (understanding) the experience of hypermobility as an exchange of merged phenomenon. This within the dance environment included knowing about the learner (with hypermobility), the entity to be learnt (dance) and the instruction or exchange of information (Pring, 2015).

3.2 The philosophical positioning of this thesis

Whilst researching the facts about hypermobility and also the experience of hypermobility, it very was important to apply a worldview and paradigm that would provide methods of research that were appropriate for exploring the phenomenon of the genetic condition (GJH) and how it is experienced. The philosophical positioning for the case study was therefore pragmatist whilst components within the investigation shift between deductive and inductive paradigms using positivist quasi-experimental and phenomenological designs. The pragmatists method allowed for practical and pluralistic approaches that enabled a combination of methods that together could facilitate a better understanding of the focus of the research (the health risk and experience of hypermobility in dance) (Johnson, Onwuegbuxie, & Turner, 2007; Kivunja & Kuyini, 2017). In such it provided practical and pluralistic approaches to discover; if and how dancers with hypermobility are affected by their genotype and how it is expressed in, and affects, dance as an art form; how hypermobility is experienced by those who operate closely (dancers and dance masters/choreographers) in the dance environment; and also it provided a means to discover more about the actual behaviour of the participants, the beliefs that stand behind those behaviours and the consequences that are likely to follow from different behaviours

Within the case study three discreet yet interwoven studies are situated. Each is designed to build on the understanding of hypermobility within the dance environment. Study I is deductive and quantitative, it has a quasi-experimental design to objectively survey the prevalence of hypermobility and its relationship to injury within the vocational dance environment. Study II also adopts a deductive quantitative approach and has a quasi-experimental design of empirical enquiry that is based upon scientific observation. Both studies I and II use quantitative methods for the data analysis. Study III is on the other hand inductive and qualitative, it is a hermeneutic phenomenological study. The qualitative data is collected through retrospective interviews that are analysed using thematic analysis. It examines how hypermobility is experienced within the dance context and environment. Overall, the pragmatist approach to the case study provides an epistemological rational for the biopsychosocial evidence presented in the literature and justification for the mixed methods applied overall (Onwuegbuzie et al., 2009). The pragmatist approach also opens opportunities for unpicking and facing the problems associated with hypermobility in relation to pedagogical methods and the practical consequences of hypermobility within the dance environment (Gray, 2014, p28). The methodological approach employed overall therefore extended beyond empirical knowledge and seeking a means to 'know better' and to 'understand' within the context of the relationship between 'knowing and *understanding* understanding' (Sliwa, 2015, p1; Munker, 2019, p1). The use of reductionist and non-reductionist methodologies overall also potentially provided a better 'understanding' (within Sliwa's sense) as to how, in a neurobiological prenoetic sense, interaction with the environment in which we choose to operate actually shapes our 'doing' and 'being' (Gallagher, 2005, p149).

Within Studies I and II investigating, yet not assigning priority to either the epistemological or the ontological metaphysical knowledge was important. This enabled a truly systematic and deductive methodology to provide an account of hypermobility within the dance environment. This approach allowed the researcher to work with "*what is real and what is known"* about hypermobility and *"account for the data"* (Lehrer, 2008, p3). The rational for the three-study design was further seeded in the desire to generate practical consequences and actions for hypermobility within the dance culture and environment. This was realised using the mixed methodologies design overall for the case study. Quantitative evidence provided explicit knowledge about hypermobility within dance in in studies I & II. Study III was developed using these findings to provide qualitative evidence of the lived experience and impressions of hypermobility within the dance environment. The *pragmatist* positioning within the case study also provided and epistemological justification for the rational and reasoning for mixing approaches and methods across the three discreet studies (studies I, II & III) (Onwuegbuzie et al, 2009 in Gray, 2014, p29). The design of these mixed methods was both developmental and interactive (Gray, 2014, p197; Maxwell, 2013) whereby the quantitative data from study I informed study II and both of these studies informed the development, implementation and analysis of studies III.

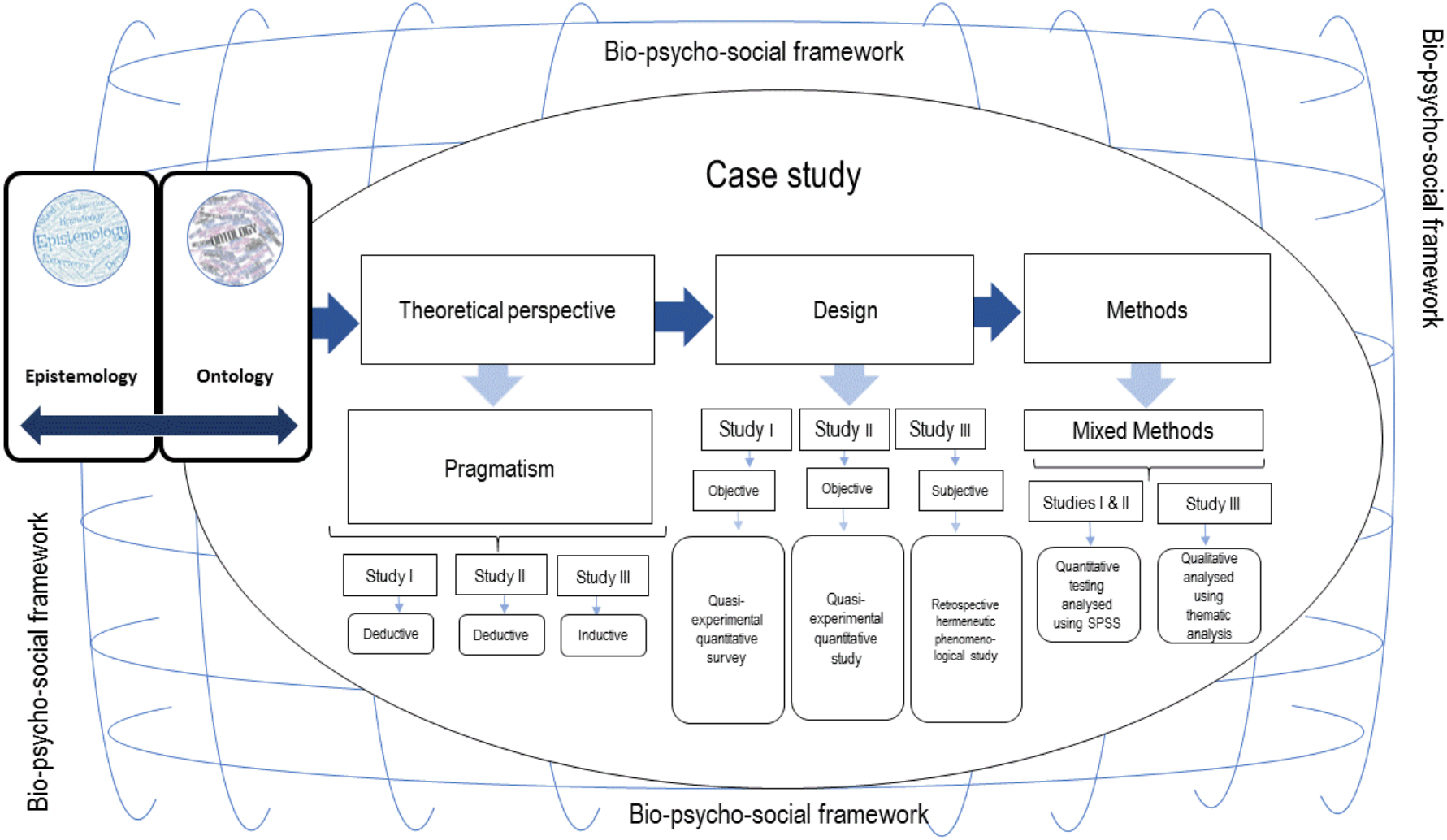

Figure 3 The underpinning framework, positioning, theoretical perspectives, methods and design of the research

The four-study design was considered necessary not only for the required intellectual development of knowledge about hypermobility but also its application into dance practice in the future. This would require a good level of 'understanding' of the facts surrounding hypermobility in dance and also the application of different forms of knowledge, both tacit and explicit, that aid to make sense of experience (Nutley, Walter, & Davies, 2003). The three studies consequently investigated both the critical formative years of a dancer and also scoped the professional career of a dancer. Studies I and II were situated within a typical classical ballet environment for talented pre-adolescent/adolescent dancers in a vocational school at a national level. Alternatively, study III was situated in the performance environment for professional dancers and the participants (dancers and dance masters) were able to draw and reflect on their experience of hypermobility within this environment.

3.3 The framework for the overall thesis

Despite the assumption by some that hypermobility is an asset to dance (Castori & Colombi, 2015; Foley & Bird, 2013b; Rietveld, 2013), hypermobility remains a genetic disorder of the connective tissue that is considered by the NHS as a disability (NHS, n.d.). Therefore, a framework used within the healthcare perspective is considered as important within the overall approach and design of this research. Gomm and Davies (2000) suggest that within healthcare settings there are three overarching and overlapping concerns, the biological, psychological and social, and it is these concerns that drive the desire to know more in order to understand better (Gomm & Davies, 2000, p5). These concerns are therefore thought as lenses through which health issues may be viewed. The lenses are used as a rational to, improve the capacity for prediction and control, develop understanding and explore meanings and address ethical issues and promote desired values within health (Gomm & Davies, 2000). In this context the approach to health research also takes on one or a combination of all of these three lenses to act as the view finder and may also be

interchangeable. It is challenging to present a succinct and simple map of research methods and over simplification, in an attempt to do so, can at times prove detrimental to the research itself. The methodological approaches to defining 'understanding and knowing' hypermobility within the classical ballet culture and environment therefore also adopted a three-lens approach (bio, psycho and social). Likewise determining how the knowledge presented was understood was also complex. The approach taken to this research therefore at times used both Silwa's thrifty reductionist and her non-reductionist perspectives to investigate how hypermobility influences the young dancer during the formative years of their craft. In such this approach to the research aspired to act as the mirror and lens through which not only the external but also the internal state of the dancer was reflected in order to discover more distinct ways of knowing and understanding hypermobility within the classical ballet culture and environment (see Figure 5).

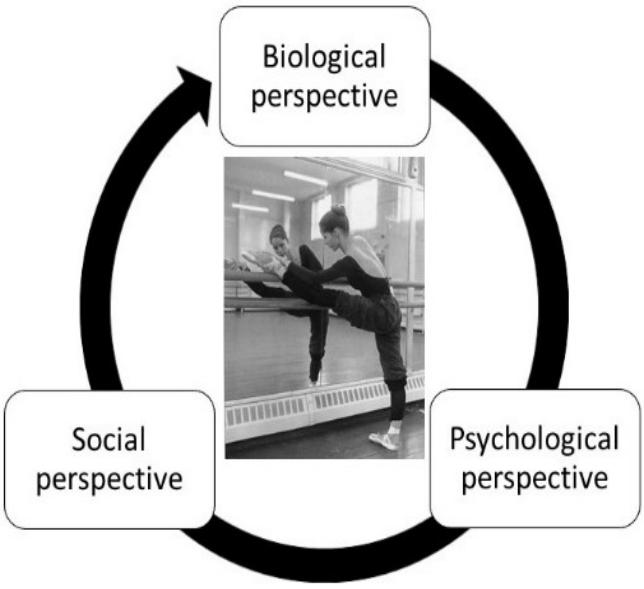

Figure 5. The intricacies of hypermobility that are at play within the classical culture and ballet environment

Within dance, practitioners (dance artist and teachers) regularly operate, observing and intervening in every-day practice, this type of work facilitates assessing, reflecting and predicting outcomes and options in an environment where the results are often subjective and

therefore uncertain. Knowing, within the dance environment, is therefore a complex accumulation of different types of knowledge that are not always explicit and clear cut.

From a purely epistemological viewpoint, we have knowledge of the external world through the evidencing that it exists (Lehrer, 2008, p3). Empirical knowing about dancers with hypermobility involves knowledge and knowing based on evidence that has been generated through empirical research and presented in the literature and is in such an explicit form of knowledge. The approach presented in studies I and II for example provides quantitative evidence for empirical knowing about hypermobility within a context of elite adolescent dancers. This within the context of this overall study implies that knowledge of hypermobility within a dance context can be evidenced from the external starting point of how it presents.

Alternatively, theoretical knowing in the dance context involves 'weighing up' and reflecting upon different theories around the training of skills and techniques and their application to choreography and the art form in order to shed light on appropriate means to approach a desired aesthetic with a very specific hypermobile physique. This type of knowing is often intuitive and informal and anecdotally known as the 'intuition of a practitioner'. It is tacit knowledge that is often accumulated through many years of practice. Similarly, experiential knowing also builds up over time and years of experiencing and 'doing', it involves a tacit understanding that is often hard to explain or pin down. This subjective and often complex type of knowing associates with the metaphysical thinking of Hume (1711-1776) and the internal focus as the starting point of ideas (Lehrer, 2008). Here Hume proposes that thoughts and knowledge arise internally through the impressions of external things (Lehrer, 2008, p3). Within a dance context an example of this type of knowing associates with a practitioner's personal impression and experience of a dancer with hypermobility. Alternatively, this type of knowing also identifies as the way in which a

dancer with hypermobility gets to know and understand the strengths and limitations of their own body. In both of these examples the knowing has been formed internally through the impressions and experience of working a hypermobile physique and relates to the type of information and knowing that is explored in study three.

3.3.1 Dance within a health context

Since the presence of ancient societies and throughout pre-modern European cultures, dance has been regarded as having a bearing on health. At times dance was believed to be one of the symptoms (e.g. St Vitus dance or *Sydenhams chorear* a form of rheumatic fever that affected children) and yet it was also considered a remedy for some illnesses and a means to wellbeing (Arcangeli, 2008). The effect of dance on the general well-being of its practitioners and also in some cases of its spectators was an area that found its place both in the literature of dance and of medicine (Arcangeli, 2008, p3). Indeed both *Plato* and *Lucian* both debate in their ancient dialogues the virtue of dance with respect to physical and mental wellbeing in their ancient scripts (Lucian, n.d.; Plato, 1980). The supposition then that disordered health does not exclusively occur within cells tissues, and organs but rather the state of the whole organism and with equal significance for the biological, psychological and social factors, is indeed not new (Havelka, Lucanin, & Lucanin, 2009).

Purely scientific approaches have however influenced Western thinking for many years offering a 'way of knowing' that is explicit and rigorous. This thinking also stems from the a Descartes' dualistic approach and reductionist mechanistic separation of the body and mind (and soul) into separate entities (Arcangeli, 2008; Havelka et al., 2009). This was the foundation for the biomedical model within medicine that had successfully prevailed in the 19th and early 20th century when disease and infection prevailed however the practice of this biomedical approach became highly questionable at the occurrence of non-infectious chronic diseases. From this point the additional psychological and social risk factors within ill health

and ill-being were also considered (Havelka et al., 2009; Rogers, 2018). In 1948 the World Health Organisation (WHO) defined health as "a state of complete physical, mental, and social well-being and not merely the absence of disease or infirmity"(WHO, 2006a and 2006b) and it is also currently recognised that chronic disease has become the primary concern for ill health in the 21st century (Rogers, 2018)

In 1980 Engel identified and presented challenges that the educators for health professionals were facing when he proposed the need for a new medical model (Engel, 1980). His proposed model went beyond the scientifically rigorous biomedical model that was at times restrictive as it was applied to patients using a factor-analysis method that only gave one dimension of an illness, pathology or condition when it was applied within health education and care settings. As a result, practitioners in health contexts now draw on empirical, experiential and theoretical knowledge and research in this context often assumes the biopsychosocial model proposed by Engel (1980) and focus on the interplay between genes, environment, and physical and psychosocial factors. Within the biopsychosocial model, the mind-body interaction approach to chronic ill health is indeed an integration of the holistic approach to early medicine with modern reductionist understanding of disease processes (Rogers, 2018, p6)

The biopsychosocial model was adopted as the framework to underpin the theoretical perspective, methodology and methods used in the three studies within this research (studies I, II & III). The focus and lens within the model for each of the discreet studies was varied in an attempt to organise the data and also to collectively inform the practice of dance with hypermobility. Building on Engel's work, Gomm and Davies (2000) also pertain that the biopsychosocial model can be taken one step further to include the cultural aspects that influence the research context (2000, pp 7-8). This approach was of particular value within study no III as the experience of teaching and learning environment for dance incorporates a

unique history and culture that is very specific to the art-form and in particular within classical ballet (Thomas, 2003, p98).

3.4 Methodology and methods

A positivist approach to research enables the testing of a hypothesis using quantifiable objective measures and this counterbalances the variable and potentially lesser reliable personal and subjective accounts (Kivunja & Kuyini, 2017). The positivist approach is achieved through an empirical methodology that facilitates the accumulation of facts discovered through controlled, replicable and objective observation or experiment (Gomm & Davies, 2000, p11). Within this methodology laws/rules and theories can be derived from facts and these may have the power to make accurate predictionsin other settings. Such laws nevertheless are difficult to achieve and define in 'real-life' health and wellbeing settings (Gomm & Davies, 2000). This is because the interaction between lifestyle, illness, stresses and environment are detrimental to any law or objective prediction and will make it almost impossible to prove beyond doubt. Scientific quantitative methods in a real life setting are appropriate yet challenging and are limited in the variables that can be assessed. Such quantitative methods were however appropriate for studies I and II of this research as both of the studies were designed to accumulate facts through standardised testing and observation. An interpretivist approach to research falls in the broad category of qualitative research. The interpretivist approach provides researchers with a means to make sense of the research by shifting between pre-assumptions and what is being revealed in the research. Grounded theory is for example an interpretivist approach that focuses on unravelling experiences and studying the interrelationships within these experiences (Kivunja & Kuyini, 2017). In doing so the researcher is able to understand the meaning of a particular experience for a particular group in a particular setting. This approach also builds on Kant's (1724-1804) realisation of

the phenomenological world which can be explained as the world that is lived in, how it appears and is experienced from an individual perspective. Each of these exists through our individual ability to filter and understand which ultimately comes from experience (Kafle, 2013). How and if we understand other people's experiences at all is questionable and relevant to our own experiences, these are also subjective, transient (e.g. emotion and pain) and often related to context and social structure. Hermeneutic analysis focuses on consciousness and experiences whereby an interview protocol gives an important account of conscious experience(Kafle, 2013). The reflection and interpretation of the protocol allows for a fuller and more meaningful understanding (Moustakas, p10). Further to this Dilthey (1976) believes that in order to understand human experience it was also necessary to consider the historical groundings and context of the description. Empirical phenomenology is also grounded in experiences, this approach involves a return to experience in order to determine what an experience means to the individual. From individual descriptions universal meanings are derived as the essence of the experience. This approach was adopted for studies III as the focus of the studies was to investigate the experiences of hypermobility within the classical ballet environment dance using retrospective recall.

Gomma & Davies (2000, p 18) identify tensions and dimensions within both positivist and interpretivist approaches to research. Rigour and relevance are one of these tensions. On the one hand rigour is achievable if a small number of variables are constant however this may limit the depth and richness of the research. Increased variables on the other hand will mitigate for this limitation however there will be less control over the procedures. This necessitates a level of detail within recorded data. Generalisability and differentiation are also questions of concern in any research. It is important that knowledge gleamed from research can be generalised and inform other contexts however the specifics and context to the

research questions are also very important and the premise for pragmatism (Kivunja & Kuyini, 2017). Finally the reality and relativism of research come in to play, the truth and accurate predictions may be required however no one truth may ever be assumed and research also seeks to explore explanations and meanings relevant to situations and contexts (Kivunja & Kuyini, 2017).

The rational of proposed methodological design and approach to the specific research project therefore draws on theories that surround 'knowing' and are embedded in theoretical perspectives that inform proposed plans of action and underpin the choice and use of methods for the desired outcomes. The focus of this research required a pooling together of physical, experiential, social and cultural aspects of hypermobility and dance practice in order to determine the health risk and embodied experience of the hypermobile dancers. It was however hard to know where the biological, psychological, social/cultural boundaries lay in this context and to which extent each influences the embodied experience of the dancers. Hypermobility is a complex genetic condition that is often understood through the rigour of empirical quantification of biological and physiological parameters that warrant a positivist approach. The context for which this condition was researched however was specific to the teaching, learning and performance environment and therefore also encompassed the psychological, cultural and sociological complexities that require an interpretivist approach for which qualitative measures were also essential.

The necessary design for the research therefore included epistemological and ontological assumptions that were framed within a pragmatic perspective and lens that focused on the biopsychosocial aspects of the experience of hypermobility within dance. An amalgam of methodologies that would yield both quantitative and qualitative data were used for consideration and analysis across the three very discreet yet interrelated studies. The design for the research was therefore mixed methods (Johnson et al., 2007) and the paradigm

or approach pragmatist as two very different types of data collection procedures were applied across the three studies. Within studies I and II a range of standardised testing procedures enabled quantitative yielding and analysis of the data whereas study III yielded qualitative data for interpretation and analysis. This approach enabled a focus and triangulation of the quantitative and qualitative data ensuring that any potential bias of measures across the studies were counterbalanced (Gray, 2014, p196). The mixed methodological approach providing further insight in to the embodied experience of young hypermobile dancers and represented the interplay between genes, environment, biological, psychosocial and cultural factors that influence dance in a highly specialist environment.

3.5 Considerations for the data analysis and sampling across the thesis

3.5.1 Data analysis

Within the mixed methods design of this thesis, both quantitative and qualitative data sets were analysed

3.5.1.1 Quantitative data

The only definite means to know the exact parameters of a population (of dancers for example) is to measure them all, however measuring the entire population of classical dancers in Scotland for this research was unrealistic. Statistical techniques were applied to the data that represented the characteristics of a sample of the dancer population in question (young dancers with GJH). These were used to estimate the values of interest within classical ballet with a view to explore the health risk and experience of the dancers in relation to GJH (Vincent, 1999, p. 13). Statistics were also used in this research to summarise larger sets of quantitative data into smaller numbers of meaningful figures. This was important as it helped the researcher to understand the distribution of the quantitative data with regards to the central tendency and this in turn determined the appropriate presentation of descriptive

statistics and the choice of statistical tests to be applied (parametric or non-parametric) (Chan, 2003a). Distribution of data can be normal, skewed (to the right or left) or not normal, testing for distribution across studies I and II was carried out using a range of methods such as graphs (Histograms and Q-Q plots), descriptive statistics (skewness and kurtosis) and formal statistical tests (Komolgorov Smirnov one Sample test and Shapiro Wilk test) (Chan, 2003a, 2003b).

a. Descriptive statistics.

Descriptive statistics are useful for defining groups of participants, in such the dancer populations that participated in the four studies discreet studies were all defined using descriptive statistics. These included the gender of the participants and mean and median, range and standard deviations of the measures collected (for example age and experience in dance) and which simply showed the central tendency of a data set, the range within the data and the standard deviation from that tendency (Chan, 2003). A sample mean is simply the average of the data collected and is calculated and reported if the data set follows a normal distribution. Alternatively, the sample median is applied when data is not normally distributed as it is a ranked value that lies in the middle of the data and the point that divides the distribution of scores into two equal halves.

The statistical data analysis throughout the thesis was carried out using IBM* SPSS* version 24 software.

b. *Parametric v non- parametric statistics*

Both parametric and non-parametric tests were applied in this research. Parametric tests assume normal distribution and uniform variance of data between groups whereas non-parametric tests are used for data that does not follow normal distribution. The term non-parametric applies to the statistical method used to analyse data, and is not a property of the

data itself (Chan, 2003b). The parametric statistics used in this research were the frequently applied t tests, analysis of variance that are applied for comparing groups, and least squares correlations for studying the relation between variables (Altman & Bland, 2009). The alternative non-parametric Mann-Whitney test, Spearman's Rank order Correlation (rho) were used for the data that was not normally distributed. Non parametric tests work by using the rank order of observations rather than the measurements themselves (Altman & Bland, 2009).

3.5.1.2 Qualitative data

Qualitative data was collected in study III, this took the form of interview transcripts. Initially visual representations of data were constructed to help the organisation of the research data as pictorial representations can enhance the clarity and support for research findings (Dickinson, 2010). Pictorial presentations of data and findings can also allow researchers to more effectively observe patterns and trends that might not be apparent through tabular or other means of presentation (Dickinson, 2010; Wainer, Word Cloud 2005). These can then be used as an entry point if further analysis, such as thematic analysis (Braun & Clarke, 2006, 2019) is to be applied. Visual displays of data also provide a different way for researchers to effectively tell the story of their data (Dickinson, 2010). Both pictorial representations and thematic analysis methods were applied in this research using the bio-psycho-social framework to guide the analysis.

3.5.2 Sampling

It is not possible to study a whole population of interest because of cost and time constraints. Therefore, studies such as those contained in this thesis, that are specific to a population such as classical ballet dance can use a sample which is sufficiently representative and includes participants with a cross-section of varied backgrounds from within the specific

population of interest (Valsiner & Sato, 2015). Sample size estimation is important and greatly assists planning and evaluation of research situations (Naing, Winn, & Rusli, 2006). For example being aware of an appropriate sample size allows a researcher to evaluate the time and resources required to complete a study and this in turn informs the feasibility and viability of the project. Sample size can be calculated for both quantitative and qualitative study designs however the methods differ and also depend on the calculations to be used in the data analysis (Malterud, Siersma, & Guassora, 2016; Naing et al., 2006). The estimation of a parameter of a population that is based on sample statistics does (as an estimation) for example contain some error and the size of that error depends on the variability and size of the sample used in the research (Vincent, 1999). Whilst being transparent about sample sizes, in reality sometimes researchers have to use the pragmatic sample that is available to them within the timeframe that is allocated for the research (Vasileiou, Barnett, Thorpe, & Young, 2018). Sampling considerations for the quantitative (studies I and II) and qualitative elements (studies III and IV) within this thesis are now discussed.

Sampling calculations made for quantitative studies depend on the nature of the study. For example the aim of the calculation in prevalence studies is to determine an acceptable sample size to estimate the population prevalence with a good precision (Naing et al., 2006). This form of sampling was necessary for study I as the purpose of the study was to estimate the prevalence of GJH within a population of vocational dancers in a cross sectional sample with a good precision. The minimum sample required for prevalence studies can be calculated using GJH prevalence reported in the literature for a similar context and the simple formula shown below and recommended for use within prevalence studies (Daniel, 1999; Naing et al., 2006),

$$n = \frac{Z^2 P(1-P)}{d^2}$$

Where;
n = sample size
Z = Z statistic for a level of confidence
P = expected prevalence or proportion (in proportion of one; if 20%, P = 0.2)
d = precision (in proportion of one; if 5%, d = 0.05).

Using a 95% level of confidence (Z=1.96) and the expected prevalence for adolescent dancers of 62% (Schmidt, Pedersen, Junge, Engelbert, & Juul-Kristensen, 2017) the required sample size calculated for study I was 36 participants.

Similarly, sampling can also be made to calculate adequate sample sizes in order to explore relationships between variables with good precision, as with study II. The focus of the study II is to determine the relationship between body awareness, anxiety and GJH. The aim of this study is therefore to get significant result ($p < 0.05$) with sufficient power (80%) to detect at least correlation coefficient of 0.4. Ideally this requires a minimum sample size of 46 for which the formula for calculation is based on two-tailed test (Bujang & Baharum, 2016).

Unlike quantitative studies, for qualitative interview studies, there are no statistical means for estimation of sample size. It is often therefore highlighted that qualitative research demonstrates a low level of transparency regarding sample sizes and poor data adequacy (Malterud et al., 2016; Vasileiou et al., 2018). Sample size in qualitative research methods is also often smaller than that used in quantitative research methods as the emphasis is often to garner an in-depth understanding of a phenomenon and often focuses on the 'how and why' of a particular environment and the interactions within it (Dworkin, 2012). Suggested sample size for phenomenological studies vary between ≤5 (Creswell & Poth, 2016) to ≥6 interviews (Morse, 1994). The objective of in-depth interviews for a phenomenological study is to create

a rich data set from which themes and categories can be derived to investigate relationships and commonalities while attending to and understanding the lived experience of the participants.

Saturation of information is regularly used by researchers as an indicator that the sample for this type of data collection was adequate, sometimes this method can fail to take in to account the suitability or power of the information gathered in the interviews (Malterud et al., 2016; Vasileiou et al., 2018). Information power means that the more detail the interview data holds that is relevant for the actual study, the lower number of participants is needed (Malterud et al., 2016). Malterud et al. provide the model presented below in figure 3 that clearly depicts how less participants are needed when the study aim is focused and the participants are also highly specific to focus of study. Information power is further increased if the research is supported by established theory and when the interview dialogue is strong. The power is also increased when the analysis is carried out systematically and includes in depth explorations of longitudinal in-depth exploration of narratives or discourse details.

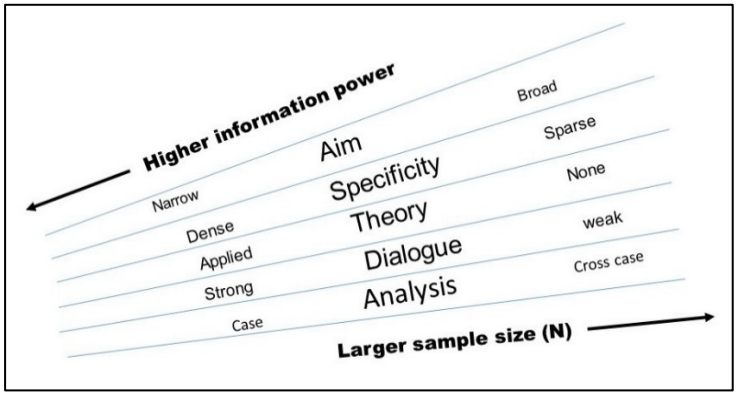

Figure 4 Information power model adapted from Malterud et al. page 1756

Within the construct of information power, methods used for sourcing participants for interview are therefore also important. Criteria for recruitment are usually set based on the focus, scope and aims of the research and sampling is purposefully specific to these

(Maxwell, 2013). Information power and strict criteria that determined the specificity, and experience of the participants were used in studies III.

The common sampling method used in qualitative studies of snowballing, whereby at least two individuals who fully meet the criteria and are valid for the topic of research are purposefully selected and are then asked to suggest others (Malterud et al., 2016) was adopted in studies III and IV. The advantage of this method as opposed to putting out a call for participants was that the participants referred others to the researcher from their network and provided strong communication links for the next interview. A disadvantage of this method is that the distinction within the participants may be limited or preconceived because the sample comprises of participants from the same network. For this reason it was important to have at least two different additional entrances in the community of focus (Sandelowski, 1995; Shankar, 2007) who in this case were four experts and clinicians in the field.

3.5.3 Validity, trustworthiness and declaration of bias

Validity is the "*degree to which data in a research study are accurate and credible*" (Gray, 2014, p692). The validity of the methods chosen to address the research questions within each of the discreet studies (I, II, III) are presented in the reporting of each of the studies. In all research however, both positivist and interpretivist, operates within a framework and moral concern for any researcher's drive to test out a hypothesis. This potentially means that there is more often than not an invested interest in the research (Gomm & Davies, 2000, p18). Given my own background, ≥40 years of experience in the dance environment with practice in both performance and teaching. This was further complicated with the potential that I myself have historically experienced (directly and indirectly) some degree of joint hypermobility. There was indeed then moral concern that my own invested interest and personal goals towards the research topic might also manifest in personal bias.

That being said, my extensive experience of the dance environment and hypermobility could, if appropriately articulated, also prove to be a valuable asset in the research in terms of motivation and insight (Maxwell, 2013, p24).

In order to mitigate personal bias within this research there was a conscious attempt to render these influences transparent. This was achieved through considered declaration and reflection of goals, experiences, assumptions, feelings and values that related to the research. This was undertaken with a view to discover personal resources yet also potential personally biased views the researcher's experience and identity might generate (Maxwell, 2013, p34-35). In doing so the researcher was able to objectively reflect on valuable experience of hypermobility while addressing and putting aside invested viewpoints. This process left the researcher open to new knowledge and insight within the topic of research. In order to raise this experience into conscientiousness whilst mitigating potential bias, a *researcher's identity memo* (Maxwell, 2005, p47) examining the assumptions and values associated with practical experiential knowledge was created for the overall study that is presented in this thesis. The identity memo took the form of a *blogs* and reflective writing that examined the researcher's own experience of hypermobility within the teaching and learning environment for classical ballet.

3.5.4 Ethical considerations

Any research that involves data collection and contact with others (human or animal) needs ethical consideration (Gray, 2014, p73). The purpose of this consideration in the overall research (Studies I to III) was to ensure that harm, ranging from physical to mental and emotional harm, was avoided. This included avoiding causing anxiety or stress to participants or producing negative reactions (Gray, 2014, p74) which may for example potentially have occurred during the retrospective interviews (Study III). For this

reason, a range of ethical guidelines were followed (BASES, BERA and BSP) across the three studies and ethical approval was sought from the ethics committee at Moray House School of Education, University of Edinburgh, for each of the stages and studies in the research. Informed consent was ensured for all participants, permissions to withdraw from the research at any time in the research process were clearly communicated and where appropriate (participants under 18 yr.) parental or *in loco parentis* consent was also ensured. The privacy of all participants was also respected, and in order for GDPR compliance additional safeguards were integrated. These safeguards were technical and organisational and included for example the use of encrypted *onedrive* storage for electronic data and locked safe storage for hard copies e.g. consent forms. The use of only the absolute minimum of personal data required for the purpose of the study was applied and personal data was anonymised using pseudonyms within the thesis. All participant informed consent forms are presented in Appendix 1.

Chapter 4 Study I: Surveying the prevalence of hypermobility and musculoskeletal injury in young pre-vocational dancers within a Scottish context

It is established from the literature that a dancer's ability to demonstrate the extreme range of motion and joint extension that goes beyond normal parameters in the limbs and spine is associated with Joint Hypermobility (JH) and some also perceive this as an advantageous asset within dance (Day, Koutedakis & Wyon, 2011; Grahame & Jenkins, 1972; McCormack, Briggs, Hakim & Grahame, 2004; McCormack et al., 2019). The balance of strength and flexibility for control and virtuosity of the limbs within aesthetic performance is also cited as an instrumental component of dance expertise and are the most important factors within classical ballet training (Bläsing, Calvo-Merino, Cross, Jola, Honisch, and Stevens, 2012; McCormack et al., 2019). This balance is also imperative when preventing musculoskeletal injury in young growing dancers (Storm, Wolman, Bakker, & Wyon, 2018).

The prevalence of injury in elite pre-vocational dancers is high and when measured per 1000 hours of dancing is reported to be between 0.62 and 5.6 (Luke, Kinney, D'Hemecourt, Baum, Owen, and Micheli, 2002; Gamboa, Roberts, Maring, & Fergus, 2008). Clinical incidence of injury (i.e. medically attended injuries) are recorded as 1.42 injuries per dancer and with a risk of injury as 76% over a year within pre-professional ballet training (Ekegren et al., 2014). This study (Ekegren et al., 2014) also suggests that, compared to young adolescent sports-persons, elite pre-professional dancers have a higher risk of injuries and the only other athletes that have similar injury to work-load ration are aesthetic gymnasts. Evidence suggests that professional classical dancers with joint hypermobility do not reach their full potential within the ranking of dancers (from corps de ballet to etoile) as they are generally excluded due to significant injury (McCormack et al., 2004). So, is the high risk for

injury related to the aesthetic demands for extensive range of motion and the prevalence of hypermobility in dance?

A review of the literature (Storm et al., 2018) relating to flexibility (which includes to range of motion and extension) and injury occurrence in dancers suggests that during the often challenging period of development of adolescence, changes do occur in the range of motion in the joints. This can lead to a decrease in neuromuscular control and dynamic stability and control of the knee for example. Storm et al's review (2018) also concludes that growth spurts, an overall decrease in joint stability and an increase in joint laxity in females due to hormonal changes, (all of which occur during adolescence) may potentially be linked to musculoskeletal injury. Furthermore, Gamboa et al (2008) previously investigated the association between measures of strength and flexibility in upper and lower aspects of the body in healthy and injured adolescent dancers with a view to discover potential deficits as a reason for injury patterns. No significant relationships between the groups are reported. The rational for these findings is discussed around the lack of robust quantifiable testing methods and the use of manual (and therefore subjective) strength testing undertaken as part of the study potentially and hence not being a sensitive and accurate measure (Gamboa et al., 2008). This research (Gamboa et al., 2008) however omitted to investigate and report the association between the strength and flexibility in the assessed joints for each dancer and proportionate levels of strength and flexibility in a dancer are an indicator of control within the joints (Koutedakis, Stavropoulos-Kalinoglou, & Metsios, 2005; Koutedakis & Jamurtas, 2004; Toledo, Akuthota, Drake, Nadler, & Chou, 2004). Control within the joints acts as a protective factor and may have given further insight into injury patterns (Koutedakis et al., 2005). Furthermore, it is unclear how Gamboa et al (2008) recorded these measures for the injured dancers and if they had mitigated for potential deficit in neuromuscular control, range of motion and strength because of the injury sustained.

4.1 Joint Hypermobility (JH) and dance

The relationship between joint hypermobility, health risk and injury across a dancer's career is discussed as a potential liability for classical dancers in particular (McCormack et al., 2004). In particular evidence suggests that within the Hypermobility Spectrum Disorders, (HDS) Generalised Joint Hypermobility (GJH) may be related to increased risk of injury in dance and aesthetic sports that require increased flexibility such as gymnastics (Bojan Bukva, Goran Vrgoč, Dejan Madić, Goran Sporiš, 2018; Schmidt et al., 2017). Such a relationship however has not been found in other sports such as women's football where increased range of motion is secondary to strength and speed and also knee and ankle braces are acceptable and do not distract from the aesthetic of the performance (Blokland, Thijs, Backx, Goedhart, & Huisstede, 2017).

Joint laxity and related potential deficits in neuromuscular control and proprioception are characteristic within GJH (Bronner & Bauer, 2018) and are also considered as intrinsic risk factors for knee injury within adolescents with GJH (Junge, Wedderkopp, Thorlund, Søgaard, & Juul-Kristensen, 2015) . Neuromuscular control and proprioception are however fundamental to all dance training and performance (Barlow, 2018; Bläsing et al., 2012) and GJH within the dancer population is reported as high (Moira McCormack et al., 2004). Dance is also *"undoubtedly associated with numerous injuries"* (Russell, 2013, p2) and musculoskeletal injury risk in elite pre-professional dancers is high (Ekegren et al., 2014; Gamboa et al., 2008; Russell, 2013; J. Smith, 2009; Storm et al., 2018).

Beyond the asset-liability debate that centres around JH (Foley & Bird, 2013b; Keer & Grahame, 2003; Klemp, Stevens, & Isaacs, 1984) a paradox is unfolding. Hypermobility spectrum disorders HSDs and JH manifest not only in tissue fragility but also extensibility and laxity of the joints (Castori et al., 2017) yet these same joints also articulate and control the aesthetic movement required by classical dancers. This aesthetic sensibility is

fundamental to both classical ballet and gymnastics whereby many hours of the training are indeed also dedicated to the fine neuromuscular control and articulation of the body so that it can perform to an elite level (Bläsing et al., 2012). The fragility of the connective tissue and joint laxity that present within JH however also provide a practical rational for the occurrence and known risk of injury in dance (Gamboa et al., 2008; Russell, 2013; Storm et al., 2018). It is therefore rational to propose a potential relationship between JH and injury in classical ballet yet also perhaps to debate that there are protective factors within the dance training that have historically allowed many dancers with JH to achieve mastery and elite performance. Examples of which are historically selected and portrayed in art, Valentin le désossé (figure 1 in outline) otherwise known as Valentin the *Boneless* (1843 – 1907); seen a paintings by Toulouse-Lautrec (1864-1901); the media as well as dance performance. Sylvie Guilliem who was a *Danseuse Étoile* or dancer of the highest ranking at the Paris opera for example was also famously used her extreme flexibility for a Rolex advertisement (figure 2); and in film, Sergi Polunin. Known as the "*Graceful Beast"* (Official *Dancer* Trailer, 2016), the youngest ever principle dancer at the Royal Ballet and also an actor and model.

Figure 5 Valentin le Désossé at the Moulin Rouge, painting by Toulouse-Lautrec

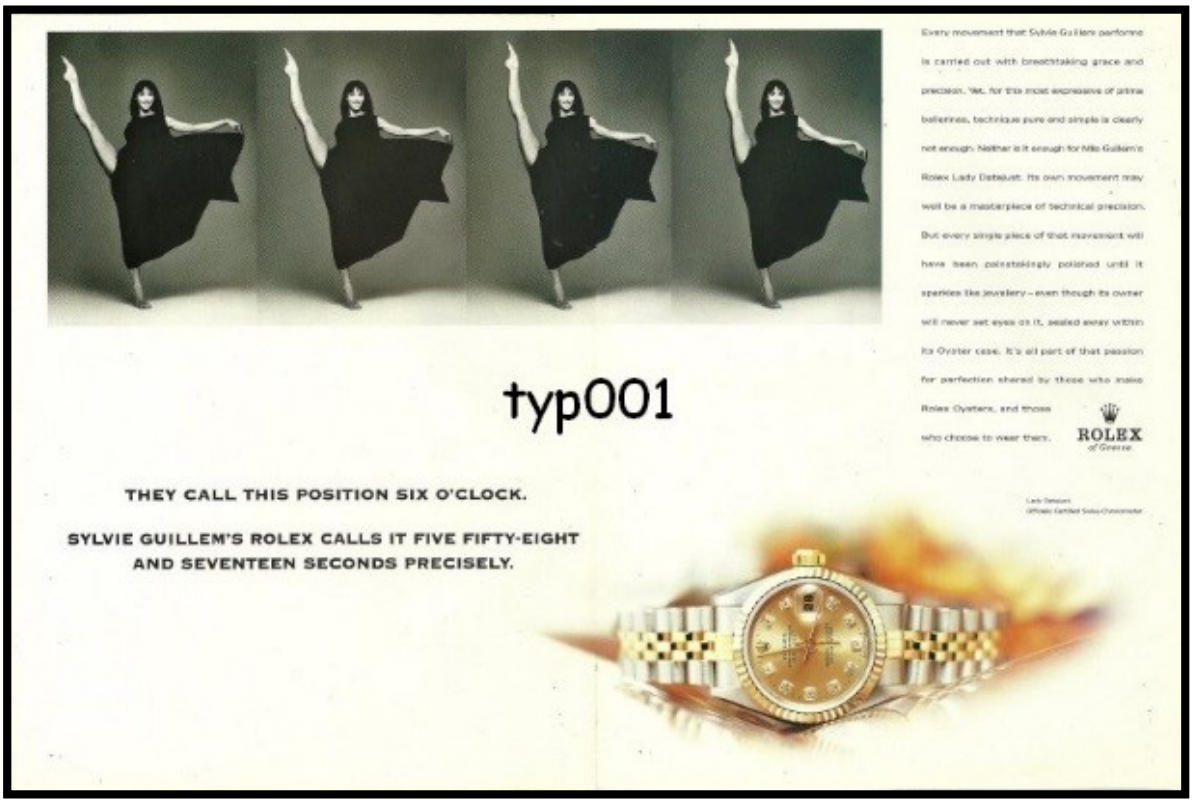

Figure 6 Advertisement for Rolex© watch 1997

4.2 Assessing for Joint Hypermobility

Joint Hypermobility (JH) in adults is classified using the Beighton criteria where an agreed score of ≥4 is generally used as the cut-off point and indicator for JH (Smits-engelsman & Kirby, 2010). As discussed in the literature review however we now understand JH to be a Hypermobility Spectrum Disorder (HSD) of four different types. These are; localised joint hypermobility (LJH) that is either acquired or inherent and uses the score of ≥4 for identification, generalised joint hypermobility (GJH) which is usually inherrant and is determined by a Beighton score of ≥5, peripheral joint hypermobility (which affects only hands and feet), and historical joint hypermobility (HJH) which is only measurable in older adults (Castori et al., 2017).

The Beighton score is commonly used to determine JH in children however the cut off criteria are not as clear or well documented and range in the current literature from 4/9 to 7/9 (Schmidt, Lykke Pedersen, Junge, Engelbert & Juul-Kristensen, 2017). This ambiguity within the cut off criteria for JH classification in children means that the literature currently reports a prevalence of JH in children anything from two percent to fifty seven percent depending on age, sex, ethnicity, and the tests and criteria applied (Schmidt et al., 2017).

Within a general population of schoolchildren aged 14 years, Clinch et al. (2011) applied a cut off marker of 4/9 on the Beighton scoring and reported a prevalence range of 11-28%. The authors propose that these findings provide a rational for using a cut–off score that is greater than ≥4 as they indicate a higher than usual prevalence of JH in the UK (2011). Schmidt et al. (2017) also recommend a need to consider further the cut-off point when determining the prevalence of JH in children and young adults. They report an overall GJH prevalence in adolescent athletes using a Beighton score of ≥4 as 27.3% and 15.9% using a cut-off Beighton score of ≥5. This study also reported GJH prevalence between sports activities with the highest prevalence for dance and specifically classical ballet. The authors

suggest that this is potentially due to the overall requirement for flexibility in dance activities in order to meet the necessary aesthetic demands of the activity (Schmidt et al., 2017). Schmidt et al's study reports GJH across a range of cut off points, a Beighton score of ≥4 determines the highest prevalence of GJH in ballet dancers (68.2%) with gymnasts second (24.6%) and handball players third (13.2%). This prevalence ranking remained the same when cut-offs of ≥5 and ≥6 were also applied. This suggests that adolescent ballet dancers, compared with other adolescent athletes (gymnasts and handball players) and the general adolescent population demonstrate the highest prevalence of GJH (Schmidt et al., 2017).

4.3 Overall aims of Study 1

The aims of this study were;

a) To explore the prevalence of GJH in adolescent dancers in pre-vocational training within a Scottish context.
b) To determine if there was a relationship between GJH and injury prevalence in the same group of dancers.

The focus in this study was the inherent form of GJH as opposed to LJH, which is often acquired, or PJH, which only affects the hands and feet and therefore does not present as such a threat to injury in dance.

4.4 Design

Study I was designed in two parts. Part 1 was a pilot to examine intra and inter-rater reliability of Beighton scoring. Part 2 investigated the prevalence and relationship between GJH and injury prevalence within pre-vocational classical dance students in Scotland. In such study I focused only on biological markers of the dancer's physique (GJH positive or negative), prevalence for injury and potential relationships between GJH and injury. All of the measures for this study were taken over a short period within one context (Scotland), this

was therefore a fixed design cross sectional study that applied quantitative empirical investigation (Robson, 2016, p124).

4.4.1 Research tools

a. Injury recording Inventory

Injury prevalence within this study was recorded via self –reported questionnaire and was retrospective over the year of training prior to the testing. The questionnaire used was a short version of the standardized questionnaire that is administered within the annual screening by the National Institute for Dance Medicine & Science in the UK (see appendix 3). Each dancer was asked to recall and report any single or reoccurring injuries that had stopped them training for one day or longer throughout the current academic year. The participants were also asked to report separately the medically attended injuries and non-attended injuries. These were clearly defined to the participants as it is known that injury definition is important for injury inventories (Kenny, Palacios-Derflingher, Whittaker, & Emery, 2018; Kenny, 2017). These definitions were also given to the dancers along with an outline of the body in order to help them determine and clearly report the location, severity and type of injury that had been sustained. Definitions included a medical-attention injury definition, i.e. an injury that had been seen by a clinical practitioner, and an all-complaints injury i.e. any physical complaint leading to difficulties participating in normal dance class, rehearsal, or performance, irrespective of the need for medical attention or time lost from dance activities (Kenny et al., 2018).

b. Testing for Generalized Joint Hypermobility (GJH)

The Beighton criteria were first developed in 1973 as an adaption of the Carter Wilkinson scale (Simmonds, 2017), the 9-point Beighton scale was designed as an epidemiological tool for identifying generalized hypermobility (Beighton 1973) and was

initially intended for adults. The Beighton criteria are applied to provide a scoring out of nine and are commonly used as a relatively simple and effective tool to identify Generalized Joint Hypermobility (GJH). A cut of ranging from ≥3 to ≥4 is generally applied in adults (Armstrong & Greig, 2018, Beighton 1973). There are however debates as to which cut off points should be used for children and young adults, cut-off points ranging from ≥5 to ≥8 have been used in younger populations as a means to mitigate for natural flexibility in youth. Authors of a recent study undertaken in Australia (Singh et al., 2017) critically justify the case for using Beighton score cut-offs closest to the uppermost 5%, which correlates to 2 S.D. above the mean. This rational is in line with the 1965 recommendation of the American Academy of Orthopaedic Surgeons also seeking to prevent over-diagnosis of joint hypermobility (Simmonds, 2017). This rational may however not apply to very specific populations such as dancers where GJH is over-represented as opposed to over-diagnosed (Schmidt et al., 2017). It was therefore decided that in this research scores of ≥5/9 to ≥6/9 would be examined and considered for the young dancers across study I and that once these methods were standardised they would also be carried into study II (Smits-Engelsman, Klerks, & Kirby, 2011b).

The Beighton criteria are time-efficient and easy to administer and are therefore the most commonly used (Simmonds, 2017). Simmonds does however highlight limitations of the Beighton criteria one of which is the fact that there is an *"upper limb focus"* (2017, p1832), this may be a strength when assessing dancers if we consider that the focus for acquired flexibility and hypermobility is on the lower limb and hips. The Beighton criteria and scale are currently considered as trustworthy and valid in paediatric populations and also demonstrate high inter-examiner reproducibility at all ages (Junge et al., 2013; Juul-kristensen et al., 2017; Smits-engelsman & Kirby, 2010). The Beighton criteria and scale have been applied and validated in several studies for the assessment of hypermobility in

children and adolescent athletes (Smits-Englesman, Klerks & Kirby, 2010, p. 4; Boyle, Witt & Riegger-Krugh, 2003, p. 283). The Beighton criteria (Beighton, 1973), as a method for determining the presence of JH are customarily applied by trained clinicians (General Practitioners, Rheumatologists or physiotherapist) within a diagnostic clinical environment and comprise of a 9 point scoring whereby joints are categorised as positive or negative as shown in table 5 below.

Table 5 Nine points Beighton criteria and scoring for the detection of GJH

The points measured in the Beighton score of hypermobility	**Bilateral Testing**	**Scoring (Max. points)**
With the palm of the hand and forearm resting on a flat surface with the elbow flexed at 90°, if the metacarpal-phalangeal joint of the fifth finger can be hyperextended more than 90° with respect to the dorsum of the hand, it is considered positive, scoring 1 point.	Yes	2
With arms outstretched forward but hand pronated, if the thumb can be passively moved to touch the ipsilateral forearm it is considered positive scoring 1 point.	Yes	2
With the arms outstretched to the side and hand supine, if the elbow extends more than 10°, it is considered positive scoring 1 point.	Yes	2
While standing, with knees locked in genu recurvatum, if the knee extends more than 10°, it is considered positive scoring 1 point.	Yes	2
With knees locked straight and feet together, if the patient can bend forward to place the total palm of both hands flat on the floor just in front of the feet, it is considered positive scoring 1 point	No	1 **Total =9**

The scope of this whole study (and study II that follows) did not permit the presence of a clinical practitioner for all data collections and so a smaller pilot sample of testing was

carried out to determine the feasibility and reliability of any further data collections by the dance-teacher researcher alone. The pilot testing was administered by two researchers, a clinical practitioner (physiotherapist) and myself the dance teacher-researcher.

4.5 Study I, part 1:Pilot study to identify the agreement of inter-rater and intra-rater reliability of meeting the Beighton criteria and scoring.

Statistical methods based on a repeated-measures study design are recommended when the reliability of measures are being assessed, this design and statistical method also closely reflects the way in clinician researchers (e.g. physiotherapists) collect data (Eliasziw, Young, Woodbury, & Fryday-Field, 1994). Consistent and reliable measures give assurance that a method of measurement and observation is in general accurate (Mchugh, 2012). Whilst Beighton scoring and methods for categorisation of GJH have for example been widely assessed for reliability (Aslan, Çelik, Cavlak, & Akdağ, 2006; Boyle, Witt, & Riegger-krugh, 2003; Erkula, Kiter, Kilic, Er, & Demirkan, 2005; Junge et al., 2013; Juul-kristensen et al., 2018) these studies were all undertaken by trained clinicians. From the perspective of the measurement, scoring and categorisation for GJH for the participants in study I, the reliability of measures for GJH was very important as it was to be explored in relationship to injury in dance.

4.5.1 Pilot study aims

The purpose of this pilot study was to identify if there was an acceptable level for:

a) Test- retest, intra-rater stability in repeated measures for each individual rater within a small sample of young dancers.

b) Inter-rater reliability for the Beighton scoring between Rater A, the clinical practitioner (certified physiotherapist) and Rater B, the dance teacher-researcher.

Stability within the measures for each independent rater (intra-rater reliability) would support the reliability of the measures applied to the Beighton scoring and criteria for GJH categorisation. Good inter-rater reliability would also support the rational that the dance teacher- researcher's assessment for GHJ in young dancers was reliable from this point forward for the purpose of studies I and II of this thesis. Piloting data collection methods and good intra and inter-rater reliability would also increase the validity of the data collection in studies I and II (Robinson, 2016)

a. Intra-rater reliability

Consistency for one rater is known as intra-rater reliability and provides evidence for a rater to produce reliable and stable measures repeatedly (Eliasziw et al., 1994). Intra-rater reliability can also report the statistical agreement between repeated applications of criteria for a diagnostic test (such as the application of the Beighton criteria to test for GJH) by a single rater (Oremus et al., 2012).

b. Inter-rater reliability

Repeated measures recorded by more than one rater gives reliability across the measures for both raters and is known as inter-rater reliability and represents the extent to which data are accurate representations of the variables measured (Mchugh, 2012). Inter-rater reliability is important when for example one rater is more experienced than the other (as with study I). When two raters are deciding between two conditions that are sharply defined, e.g. if a person has survived or not from a life debilitating illness, reliability is

usually high. When however two (or more) raters are required to score using measurements (as with the measures required for the Beighton scoring criteria), reliability is much more difficult and requires careful instruction and agreement or a standardised protocol as to how the measures are acquired and recorded (Mchugh, 2012). Inter-rater reliability is therefore a useful statistic to establish as it determines the quality as well as the reliability of measures (Oremus et al., 2012).

4.5.2 Statistical methods to measure agreement of measures for categorisation

Percentage of agreement (PA) is a simple statistic that is often used in research and is both easily calculated and interpreted (Mchugh, 2012). An example of its application would be to obtain PA for the ordinal categorisation of GJH (e.g. positive=1 or negative=2) between two raters, R1 and R2. PA is calculated by subtracting R1's categorisation from R2's categorisation and the number of zeros that result are divided by the number of participants in the study group and multiplied by 100 to calculate the result as a percentage. This provides the PA statistic that can be applied to directly interpreted as the percentage of times that raters agreed on the GJH categorisation (Mchugh, 2012). A known limitation for PA is however that this measure does not take in to account chance or the probability of one of the raters guessing and may overestimate true agreement among raters (Mchugh, 2012) and this may be the case in this study for example as the specific disorder (GJH) being tested for is known to be prevalent in the population to be tested which is again the case for dance (Kee and Grahame, 2003; McCormack, Briggs, Hakim, Grahame, & Grahame, 2004).

The intra and inter-rater reliability of ordinal measures (positive or negative categorisation) can also be calculated statistically using Cohen's kappa which is proposed as a more a robust correlation statistic as it mitigates for random agreement (Cohen, 1960). This statistical measure was therefore also used for the calculations of the Beighton scoring

criteria (categorisation of each of the 9 joints assessed) and the overall GJH categorisation (score out of 9). Cohens kappa is represented by the lower case Greek letter, (κ). It can range from -1 to +1, where 0 represents the amount of agreement that can be expected from random chance, and 1 represents perfect agreement between the raters. Values below 0 are possible however improbable in practice and indicate a worse level of agreement than expected (Marston, 2010). Interpretation of κ values is reported as; >0.80 = very good, 0.61-0.80 = good, 0.41-0.60 = moderate, 0.21-0.40 = fair and <0.21 = poor and as with other correlation statistics, κ is a standardized value and can therefore be interpreted uniformly across multiple studies (Cohen, 1960; Fleiss, Levin, & Paik, 2003; Pallant, 2016).

Both PA and κ statistics are reported in the literature to have strengths and limitations for measuring levels of agreement (McHugh, 2012). Whilst the percent agreement (PA) statistic is easily calculated and directly interpretable, it does not take account of the likelihood that raters guessed on the scores that they allocated (McHugh, 2012). Choen's κ on the other hand mitigates for the 'guessing' however it is reported to make assumptions that lower the estimate of agreement excessively (e.g. rater independence); the *k* statistic is also harder to interpret which in turn means that lower κ scores are often accepted in studies for interrater reliability (McHugh, 2012). McHugh (2012) therefore proposes that both Cohen's κ and PA are calculated for clinical and health settings and that κ levels for measures of agreement are also in line with the literature (McHugh, 2012).

4.5.3 Participants

Ten young dancers aged between 12 and 17yr. (mean 14.2yr. SD 1.83) and two rater/observers took part in the pilot study. The ten young dancers were selected during the annual informative screening of dancers at a private Dance Academy that took place over two days in a physiotherapy clinic and included over fifty young dancers, and multiple

measurements and assessments. Criteria for inclusion in the pilot study were that the dancers were not injured at the point of testing and that they themselves and their parents had read the information about the pilot study and signed consent to take part as an additional part to their annual screening. Thirty nine dancers met this criteria from across the two days of screening and the ten dancer participants were allocated from across the two days of screening for inclusion by the screening administrator/secretary. The allocation of the 10 participants from within the intense screening and overall volume of measures taken during the intense two day screening ensured the unlikelihood of raters remembering scores between measures. The two raters (physiotherapist and dance teacher-researcher) were both part of the regular screening team at the Dance Academy that the participants attended which made the process less intrusive for the dancers as they were familiar with both the testers and the test.

4.5.4 Procedures

Signed informed consent was obtained from the parents/guardians and young dancers. The nine-point Beighton scale shown in table 1 was the measure that was tested for reliability. The Beighton testing protocol and criteria applied in this pilot study were ones that the two raters were used to applying in the annual screening of dancers which were standardised methods (Junge et al., 2013) taken from the National Institute for Dance Medicine & Science (NIDMS) screening protocol.

Beighton scoring and GJH categorization cut off scores of ≥4, ≥5 and ≥6 were tested for reliability in this pilot study. Two *66fit* goniometers widely used in clinical physiotherapy were used to measure the range of motion bilaterally in the metacarpal-phalangeal joint of the fifth finger, elbows and knees. A standard protocol for the goniometry was also agreed and followed by both raters who applied standard goniometry guidelines (Smits-Engelsman et al., 2011b).

Each of the ten dancers were scored against the Beighton criteria independently by each rater-observer, these evaluations took place in two separate rooms so that the raters were blinded against each other's evaluations. The measures were repeated by each rater two times for each dancer during the annual screening. During the screening session; no warm up was undertaken and confirmation was recorded that the dancers had not taken any dance or exercise classes on the day of testing. At least 30 min lapsed between each of the Beighton scorings so that the range of motion in the joints evaluated was not affected by repetition. An independent assistant recorded the measures in order to keep the raters unfamiliar with the previously recorded measures.

4.5.5 Results

The two independent raters scored the ten dancers on the 9 point Beighton scale twice. In total, 360 Beighton scores (0-9) were recorded and the Beighton categorisation was made 40 times overall.

The overall mean, SD and range of Beighton scoring was 6.05 (2.08, 2-9), the mean, SD and range of Beighton scoring for Rater A was 6.05 (2.06, 2-9) and 6.05 (2.06, 3-9) for Rater B. Cohen's κ and percentage of agreement (PA) were calculated to determine intra and inter-rater reliability and are presented in tables 6 and 7.

Table 6 Intra-rater reliability of Beighton scoring and categorisation

Rater	Percentage of Agreement	Cohen's κ	Level of agreement
Rater A (Physio) intra- rater agreement for all individual Beighton scores	50%	.430	Moderate
Rater A (Physio) intra- rater agreement GJH categorization (≥4/9 Beighton score)	90%	.737	Good
Rater A (Physio) intra- rater agreement GJH categorization (≥5/9 Beighton score)	80%	.380	Moderate
Rater A (Physio) intra- rater agreement GJH categorization (≥6/9 Beighton score)	100%	1.00	Very Good
Rater B (DT) intra- rater agreement for all individual Beighton scores	90%	.880	Very Good
Rater B (DT) intra- rater agreement GJH categorization (≥4/9 Beighton score)	100%	1.00	Very Good
Rater B (DT) intra- rater agreement GJH categorization (≥5/9 Beighton score)	90%	.740	Good
Rater B (DT) intra- rater agreement GJH categorization (≥6/9 Beighton score)	80%	.583	Moderate

Table 7 Inter-rater reliability of Beighton scoring and categorisation

Rater	Percentage of Agreement	Cohen's κ	Level of agreement
Raters A & B inter- rater agreement for individual Beighton scores	70%	.650	Good
Raters A & B inter- rater agreement GHJ categorization (≥4/9 Beighton score)	90%	.800	Very Good
Raters A & B inter- rater agreement GHJ categorization (≥5/9 Beighton score)	95%	.860	Very Good
Raters A & B inter- rater agreement GHJ categorization (≥6/9 Beighton score)	100%	1.00	Very Good

4.5.6 Discussion

The prevalence for GJH across the sample of participants was high (62.5%) compared to the literature where GJH is prevalent in 4-40% in children (Junge et al., 2013; Schmidt et al., 2017). This result however was within the expected range for dance populations (Foley & Bird, 2013a; Moira McCormack et al., 2004). The intra-rater percentage agreement calculated for the actual categorisation of the 9 point Beighton scoring indicated that the dance teacher-researcher's repeated scoring were more accurate (90% compared with 50%) across trials. These results were also indicated in the *k* scores with a moderate level of agreement across the two trials for the physiotherapist (k=.43) and a much

stronger and very good indicator for the dance teacher-researcher (k=.88). Reasons for this may be due to a certain amount of bias due to the teacher's familiarity with the participants' physique as she also taught the participant dancers regularly in the dance classes that they attended. However, the dance teacher also had more years of experience working with, manoeuvring and assessing young dancer physiques. It may therefore be the case that the participants were more relaxed with the dance teacher's assessment facilitating less tension in the body generally; this would allow the rater to assess the true 'passive' range of motion in the knee and elbow joints.

The intra-rater categorization for the three cut-off points for GJH tested (≥4, ≥5 and ≥6) had a very high percentage of 80-100% of agreement (PA) across trials for both raters. Cohen's κ and level of agreement of intra-rater agreement across cut off points was moderate to very good (.380 to 1.00) for the Physio (Rater A) and also moderate to very good (.583 to 1.00) for the dance teacher (Rater B). These results indicate that the repeated scoring for GJH categorization are moderately to very reliable for both the physiotherapist and dance teacher-researcher.

The inter–rater PA for the overall actual Beighton scores (score out of 9) was also high (70%). The inter-rater k value (.86) also indicated a very good level of agreement between the raters for the overall Beighton scoring again indicating valid and reliable measures between raters. The inter-rater reliability for the categorization of GJH across the three Beighton score cut off points was also very high ranging from 90-100% and k values across these cut off categorization scores were also reached a very good level of agreement ranging from .80-1.00.

The results from this study for GJH categorization between raters using the Beighton scoring and cut off criteria were also compared to findings in the literature. Schlager et al

(2018) report inter-rater reliability with a PA of 98% and *k* value of 0.96 where a cut off criteria ≥5 was applied, the participants were however adults. Jung *et al*. (2013) also applied a cut of ≥5 and the *k* value was reported as 0.64 when a similar assessment protocol to this one was applied. Overall, the pilot study reported here demonstrated a very good levels of agreement and *k* values, the agreement was also in line with Schlager's study (2018) and slightly higher than Jung et al (2013) .

Interestingly, whilst credible within typical populations (Simmonds, 2017) Singh et al's (2017) justification were also applied to this population and specific group (whereby a Beighton score cut-off closest to the uppermost 5%, that correlates to 2 S.D. above the mean), the cut off was calculate as 10.21 which is in fact beyond the 9 pint score maximum confirming that this group is specific and does not have a typical representation of GJH prevalence.

4.5.7 Conclusion

In relation to the reliable administration, scoring and categorisation of the Beighton scale and category, the applied measures of GJH for both the clinical practitioner and the dance teacher-researcher were stable and showed a good level of agreement across the categorisation cut offs for GJH. This meant that dance teacher, who was less experienced in clinical measures, could take forward the procedures for GJH categorisation independently as measures and categorisation were reliable with a good level of agreement, in line with the literature and therefore valid.

4.6 Study I, part 2: Exploring the relationship between GJH and injury prevalence within pre-vocational classical dance students in Scotland

For Study I, the piloted Beighton protocol, criteria, scoring and categorization (Beighton, 1973; Juul-kristensen, Schmedling, Rombaut, Lund, & Engelbert, 2017) were

taken forward and administered by the dance teacher researcher only, these were used I combination with the standardized injury inventory questionnaire (NIDMS). Participants were recruited from five pre-vocational dance schools across Scotland. The data collection took place over a period of three weeks towards the end of one academic year, this minimised differences across the schools that may have occurred due to workload during the year of testing. The University of Edinburgh's ethics committee granted ethical approval for the study and informed consent forms were sent by the schools to all the potential participants' parent/guardians for signatory. Informed assent forms were also given to each of the participants and opportunities for the potential participants to speak to the researcher prior to the data collects were made, this also allowed time for the participants to discuss with the researcher and clarify queries about the testing. Once this procedure was complete the data collection was undertaken.

4.6.1 Participants and measures

A total of 94 young dancers (n=94) participated in the study, this number fulfilled the sampling criteria (n=36) discussed in chapter 3 for a prevalence study relating to GJH and adolescent dancers. The Beighton testing protocol (appendix 2) and a questionnaire inventory (adapted from the NIDMS screening protocol, (appendix 3) were administered to all participants who were all attending intensive dance training at the time of the study. The foundations for the participants training included technical classical ballet training at a vocational level.

4.6.2 Methods

The Beighton scoring and categorisation criteria are validated for assessment of hypermobility in children and athletes (Smits-Englesman, Klerks & Kirby, 2010, p. 4; Boyle, Witt & Riegger-Krugh, 2003, p. 283). The Beighton 9-point criteria (1973) was applied in

this part of the study with the same protocol and methods used and validated in the pilot study previously reported. This allowed the participants to be categorized as either GJH positive or negative. As with the pilot study cut off points ranging from ≥4 to ≥6 were applied, the relationship of each of the GJH categories (≥4, ≥5, and ≥6) with injury prevalence was statistically examined.

Injuries were surveyed retrospectively (reporting for the 12 month period prior to testing) using the injury questionnaire inventory that was given to the participants to complete after the Beighton testing. The participants completed the questionnaire individually and privately while in the presence of the researcher who remained available for questions and clarifications throughout the time taken for completion, while taking steps to refrain from influencing any responses. It was made clear to the participants that their individual answers would remain anonymous and that it was important they answered each question as best and honestly as they could.

4.6.3 Results

Ninety-four (n=94) participants took part in the study however based on complete data sets, ninety (n=90) comprised the final sample. Sixty-five participants identified as female (n=65) and twenty five (n=25) identified as male. The participant's age ranged between eleven and seventeen with a mean of 13.65 years (SD=1.66 years).

The characteristics of the recorded injury prevalence and Beighton scores were investigated for normal distribution. A Shapiro Wilk's test (p ›.05) (Shapiro & Wilk, 1975; Razali & Wha, 2011) and a visual inspection of their histograms, normal Q-Q plots and box plots showed that the data were normally distributed. When data is approximately normally distributed parametric methods are recommended in preference to non-parametric methods as they provide greater statistical power (Altman & Bland, 2009).The parametric testing of the

data assumed that the underlying distribution of the surveyed injury frequency and Beighton scores was normal.

4.6.3.1 The prevalence of Generalized Joint Hypermobility (GJH)

Table 8 shows the Beighton scores (mean, SD and range) and prevalence of GJH within the group (n=90) according to the cut off scores of ≥4, ≥5 and ≥6.

Table 8 Beighton scoring, GJH categorisation and % of GJH prevalence for all dancers

Beighton scores (mean, SD and range)	Beighton Criteria ≥4, total GJH positive dancers (%)	Beighton Criteria ≥5 total GJH positive dancers (%)	Beighton Criteria ≥6 total GJH positive dancers(%)
3.97 (2.03, 0-8)	50 (56%)	39 (43%)	39(43%)

4.6.3.2 Injury prevalence

The injury inventory data showed that across all the participants with a full data set (n=90) one hundred and seventy five (175) injuries were sustained over the year. Twenty-seven (30%) of the participants did not report any injuries at all and the remaining sixty-three (70%) dancers reported being injured during the period of recall, injury incidence for the 63 dancers was therefore 2.8. Of the injuries sustained, these were also categorized as non-medically attended and medically attended, one hundred and five (60%) injuries were non-medically attended and seventy (40%) were medically attended.

4.6.3.3 Injury frequency

Calculations for injury frequency are based on the number of injuries that occurr in relation to the exposures to dance during the period of the survey (i.e. participation in a class, rehearsal, or performance in which the dancer was exposed to the possibility of a dance injury) (Allen, Ribbans, Nevill, & Wyon, 2014). The hours of dance practice per student each year was calculated in this study as 1000 (calculated at 25 hours of practice per

week over the 40 weeks of term in one year), this was also confirmed by the school administrator. The workload estimation provided us with a frequency of 1.85 injuries per 1000 hours in this study.

4.6.3.4 Relationship between Beighton scores and injury prevalence

The relationship between the Beighton scores and the injury prevalence was investigated using the Pearson product moment correlation coefficient. Preliminary analysis were performed to ensure no violation of the assumptions of normality, linearity and homoscedasticity. The Pearson product moment correlation was selected as the distribution of the data and scatterplots showed no indication of curvilinear relationships between the two variables (Pallant, 2016). No correlation was found between the number of injuries sustained and the Beighton scores and cut off criteria.

Table 9 Correlations between Beighton scores and the injury prevalence

		Beighton total	Beighton ≥4	Beighton ≥5	Beighton ≥6
Number of injuries	Pearson's correlation	.051	-.019	.063	.011
	Sig.(two-tailed)	.634	.862	.557	.917
	N	90	90	90	90

4.6.4 Discussion

As predicted within this group of young dancers the percentage of GJH positive dancers was high was for the cut off points of ≥4 and ≥5 , 56% and 43% respectively. This is much higher than the representation of GJH in non-dancer populations of young adolescents which is reported as between 11 and 28% with a cut off of ≥4 (Clinch et al., 2011) and 15.9% as reported by Schmidt et al (2017) within adolescent athletes with a cut off of ≥5. The representation of GJH positive dancers in the group (56%) also mirrors the results for classical ballet dancers that were GJH positive in Schmidt's study (2017) which were reported at 62.2% with a cut off of ≥4. This study therefore confirms an overrepresentation

of GJH in this specific group of ballet dancers and confers with Schmidt's assumptions (2017) that this is due to the overall requirement for flexibility in dance activities in order to meet necessary aesthetic demands.

Injury prevalence was high within the group 175 injuries in total which calculates to 1.94 injuries per dancer over the year, although not all medically attended this number (1.42) is representative to that reported by Ekegeren et al (2014). The frequency of injury reported in the literature per 1000 hours of exposure is reported between 0.62 to5.6. The workload estimation for this study provides us with a frequency of 1.85 injuries per 1000 hours of exposure which is at the lower range of what is reported in the literature for pre-professional dancers (Chan et al., 2018; Gamboa et al., 2008).

Finally, this study finds no significant relationship between the presence of GJH and injury in pre-vocational ballet dancers. This non association suggests that these ballet dancers may be protected through their training from the musculoskeletal injuries that are often related to GJH such as dislocation, sprains, and tendinopathies (Fatoye et al., 2009; Keer & Grahame, 2003; Knight, 2015; Sanches, Oliveira, Osório, et al., 2015). This protection may be provided by the corrective neuromuscular and motor control training and strengthening that are fundamental within the training of classical ballet.

4.6.5 Conclusion

Study I has provided a valid and reliable method for the researcher to test for GJH in young adolescent dancers, this GJH testing protocol can therefore be taken forward and applied in the methodology and procedures for study II by the researcher. Within the broader and overall biopsychosocial considerations for the case study, study I is an investigation only of the bio. The psycho-social implications of JH within the classical ballet environment are therefore developed and given further consideration in studies II, and III.

Chapter 5 Study II: A case study exploring body awareness indicators of young dancers within a vocational setting in Scotland

Hypermobile Spectrum Disorders (HDS) that commonly present with joint hypermobility (JH) are prevalent in dance and aesthetic sports (Armstrong, 2018; Armstrong & Greig, 2018a). This was further documented within classical ballet in study I where the prevalence of JH within the cross-sectional sample of dancers in Scotland was reported as 56% when applying a Beighton cut off scores of ≥4, and 43% when applying a Beighton cut off scores of ≥5. The reported percentages were much higher than those found in general and specific populations of children where JH is between 11 and 28% with a cut off of ≥4 (Clinch et al., 2011) and 15.9% in young athletes with a cut off of ≥5 (Schmidt et al., 2017). However study I findings did not validate any relationship between injury prevalence and GJH, suggesting that nature and rigour of vocational dance training may potentially act as a protective factor against injury in the presence of joint hypermobility.

Usually what a researcher brings to research from their own background and identity has been considered as 'bias' (Maxwell, 2013, p. 45). Nonetheless and unlike study I which only used known methodological procedures and protocols, the research design of study II also draws on evidence from the researcher's forty plus years of experience teaching, observing and developing dancers across their lifespan within the research design. With a view to make of use whilst attempting to maintain objective rigour within the research, this anecdotal evidence has been carefully considered, articulated and documented as a researcher memo (presented in appendix 4). The memo was then used cautiously in study II as supplementary evidence and insight to enable both the research design and validity checks (Maxwell, 2013, p. 45) .

5.1 Psychosocial markers in hypermobility

The overall literature review towards the biopsychosocial approach to this thesis (Chapter 2) suggests that JH is also often entangled with further complications that go beyond the physiological musculoskeletal manifestations reported in study I. The literature also suggests that complications within JH may also be potential indicators of other, more serious, heritable connective tissue disorders (HCTDs) such as Ehlers Danlos syndromes (EDS), Marfan Syndrome (MFS) and Osteogenesis imperfecta (OI). The major complications often associated with both MFS (serious heart condition) and OI (very fragile bones) do however almost certainly exclude young adolescents from reaching the point of entry into an elite vocational dance training environment that has high physical demands. This does not mean however that MFS and OI may not present in recreational dance settings that do not have the same entry requirements. Whilst remarkable, this is not within the scope and focus of this research and will therefore not be part of this investigation.

The musculoskeletal health complications associated with HSDs such as JH and some types of EDS do not necessarily present as a barrier for entry into vocational training for dance. Indeed, it has already been established from the literature that musculoskeletal associations with JH and EDS may very well present as asymptomatic, mis or undiagnosed (Foley & Bird, 2013; Grahame & Jenkins, 1972; Castori et al., 2013; Rietveld, 2013). Indeed at times JH and hypermobility type EDS (hEDS) are evaluated as an asset to the training, signposting pupils in audition circumstances as being *"full of potential"* (Grahame & Jenkins, 1972; McCormack et al., 2019). Whilst psychosocial complications relating to JH are now becoming better established in the literature (Baeza-Velasco, Bulbena, Polanco-Carrasco, & Jaussaud, 2018; Bulbena et al., 2017b, 2017a), to date, there is no conclusive evidence pertaining to these complications in relation to the classical ballet environment and elite vocational training. Of specific interest in the study reported here was the representation

and awareness of the body which are fundamental within dance yet also known to be potentially disordered in the presence of JH (Baeza-Velasco, Bulbena, Polanco-Carrasco, & Jaussaud, 2018; Bulbena et al., 2017b, 2017a). To add to this the researcher also has evidence of apparent disordered body awareness in young dancers with JH drawn from personal longstanding anecdotal observations within the teaching and learning environment for dance (see appendix 4). The contradiction presented here then is that sound body representation and awareness are integral to classical ballet training, performance, and choreographic processes (Bailey & Pickard, 2010; Waterhouse, Watts, & Blassing, 2014) and in such should be requirements for entry into elite training. However, JH appears to favour individuals towards selection for dance training despite the fact that JH is also associated with poor body representation and awareness (Carolina Baeza-Velasco, Bourdon, et al., 2017a; Bulbena et al., 2017b).

Of particular interest in the study reported in this chapter (study II) were adolescent dancers who were following a programme of elite vocational training with a view to enter the profession upon completion. The rational for this choice of participant group was that this stage and level of training is critical to a dancer's career and professional success (Mitchell et al., 2016). Furthermore, from a biopsychosocial perspective, adolescence is a period of developmental turbulence (Granz, Schnell, Mayer, & Thiel, 2019) and within dance adolescence also has intense physical, cognitive, social and emotional demands that can increase a young dancer's vulnerability (Grove, Main, & Sharp, 2013; Pickard, 2013). This is mainly due to the fact that for classical ballet early specialisation is key as a dancer's career starts young and is relatively short (Pickard, 2007; Wanke, Groneberg, & Mill, 2012). Added then to these vulnerabilities and pressures for the adolescent dancer is also the potential prevalence of JH.

Whilst the end goal for professional ballet dancers is common, the nature and demands of vocational dance training can potentially vary between institutions, training systems and establishments. The present study was therefore designed as a discreet case study within one homogenous national level elite training environment. It was an environment for adolescent classical ballet dancers between the ages of twelve and seventeen. In this chosen environment the researcher was confident that the selection process, training methods, programme and rigour were assured and common across all dancer participants.

Whilst study I examined solely the physical presentation and prevalence of JH and injury in dancers, the literature reviewed thus far also demonstrates that the fragility and vulnerability within JH is not restricted to the physical (Carolina Baeza-Velasco, Bourdon, et al., 2018; Carolina Baeza-Velasco, Grahame, et al., 2017; Sanches, Oliveira, Osorio, Crippa, & Martín-Santos, 2015). Entangled with JH are also many psychosocial elements and factors. Anxiety for example can amplify and individual's risk of encountering an injury and is also potentially career threatening to a young dancer (Ford, Ildefonso, Jones, & Arvinen-Barrow, 2017; Mark C. Scheper et al., 2013b). It is also known that anxiety can exacerbate pain perception during training or after an injury, and high levels of fear of re-injury (kinesophobia) and pain-related fear can lead to poor rehabilitation outcomes (e.g. avoidance behaviours) (Ford et al., 2017; Walker & Nordin-Bates, 2010). The association between pain sensation and JH are reported in the literature, but JH is commonly overlooked as a cause for chronic pain (Kumar & Lenert, 2017). As a result, inappropriate interventions are often prescribed, and JH can even be totally disregarded because it appears as an advantage in dancers and gymnasts (Kumar & Lenert, 2017). Baeza-Valesco et al. (2011) made clear recommendations for psychological problems associated with JH, directly referring to and describing a potential genetic commonality between JH and panic disorder, agoraphobia,

simple phobia and social phobia. This connection however remains to be scientifically substantiated (Baeza-Velasco, Gély-Nargeot, Vilarrasa, & Bravo, 2011).

Thus far, it is inferred that dancers with JH within the rigour and stresses of vocational elite training may potentially also be exposed to risks of psychological issues such as altered body-awareness and worrying thoughts because of the complications associated with hypermobility. Whilst these have been scoped in the body of the literature review in chapter two further literature pertaining specifically to body-awareness is reviewed and critiqued here. In particular body-awareness with reference to dance training and education, and joint hypermobility are discussed in order to substantiate and inform the methodological approach to study II.

5.2 Body-awareness and joint hypermobility in dance.

More recently research has indicated that individuals with JH may also have disturbances in their perception of body-awareness (Bulbena et al., 2017a). Body-awareness manifests twofold in the literature with regards to JH. First as sense of self, also known as *interoception*, and second as a *sense of motion,* that is the sense of the body moving in space (Mallorqui--Bague et al., 2014; Mallorquí-Bagué et al., 2016). Unfortunately, *sense of motion* is historically described across the literature using a range of alternating and interchangeable terminology such as; kinesthesis, kinaesthetic awareness and/or proprioception (Bulbena et al., 2017b; Fatoye et al., 2009; Jacobs et al., 2018) making clarity within and across the research difficult.

Body-awareness as a concept was first defined in the literature in by Sherrington (1907) who built on Scottish physiologist Charles Bell's (1826) notion and anatomical basis of the connection between the brain and limbs and English pathologist/anatomist Henry Bastian's notion of "kinaethesia" (Han, Waddington, Adams, Anson, & Liu, 2016a).

Kinaesthesia, is derived from two Greek words "*κινώ*" (to move) and "*αίσθηση*" (sensation) [*Κιναισθησία*] and refers to the *"...sensation which results from or is directly occasioned by movements ... kinaesthesis"* (Bastian in Han et al., 2016a, p80). Sherrington further differentiated within the concept of body-awareness, describing proprioception as *"the perception of joint and body movement as well as position of the body, or body segments, in space"* (Sherrington, 1906), and interoception as the awareness and sensitivity to stimuli originating from within the body. He further distinguished this from exteroception which he described as the awareness and sensing of stimulation from outside the body (Han, Waddington, Adams, Anson, & Liu, 2016b). Current thinking now proposes that these aspects of body-awareness (proprioception, exteroception and interoception) and the sense of the body from within and outside integrate to provide a sensory feedback-forward loop between the brain and the body to produce a sense of body ownership and "*the experience of this body as mine"* (Tsakiris, Tajadura-Jiménez, & Costantini, 2011, p. 2470).

Current thinking proposes that interoception plays a key active role in this feedback-forward process as the brain integrates body-related multi-sensory perceptions (Tsakiris et al., 2011). Recent interest in interoception has developed alongside theories of embodied cognition and the notion that cognition and emotion influence and are influenced by both mind and bodily processes (Garfinkel, Seth, Barrett, Suzuki, & Critchley, 2015). Interestingly, whilst interoception is heightened in JH, mind bodily processes such as visuo-spatial problem-solving, attention, memory, and awareness of performance are reported as much lower in JH than control subjects (Baeza-Velasco, Bourdon, et al., 2017).

Interoception, a component of body-awareness, has been defined as the sensing of the physiological condition of the body and its internal state and is a complex process with several facets (Murphy, Brewer, Catmur, & Bird, 2017; Vaitl, 1996). Interoception regulates for example the detection of subtle changes in bodily systems, including muscles, skin, joints,

and viscera (Craig, 2009). It also regulates all major biological systems involved in maintaining bodily homeostasis, including the cardiovascular, pulmonary, gastrointestinal, genitourinary, nociceptive, chemosensory, osmotic, thermoregulatory, visceral, immune, and autonomic systems. Most of these interoceptive processes however occur outside the realm of conscious awareness (Khalsa et al., 2018). Nevertheless, interoception also involves conscious viscero-motor sensations, such as the fullness of bladder or bowel that motivates controlled voiding, or the abdominal sensations that accompany nausea or hunger. Sensations that the researcher has unequivocally yet anecdotally witnessed as being over-represented in young dancers with hypermobility and ones that less experienced teachers find disruptive to teaching practices (researcher's memo page X lineXX).

As science progresses, theories are continually developing and interoception is now also described using three different dimensions which can at times make the literature hard to follow (Garfinkel et al., 2015). The first dimension, interoceptive accuracy, describes the ability to accurately attend to bodily signals. It is determined through testing for the detection of bodily functions such as for example heart beat discrimination tests (Garfinkel et al., 2015; Khalsa et al., 2018). Research in dance has established that interoceptive accuracy is higher in professional dancers compared to control participants, and this work also theorises the idea that interoceptive accuracy and timing are related (Christensen, Gaigg, & Calvo-Merino, 2018). This suggests that those with traits for higher interoceptive accuracy may again be susceptible to signposting for dance training (Christensen et al., 2018) and further supports the notion that not only the musculoskeletal advantages of JH but also associated heightened interoceptive accuracy may be perceived as an asset within dance. On the other hand it may be that interoceptive accuracy can be enhanced through engagement with any training involving body awareness and attention to bodily signals (such as heartbeats, sweat response,

muscle contraction) in addition to the "*use of these signals for the expression of states and emotions"*(Christensen et al., 2018, p. 9).

The second dimension of interoception is known as interoceptive sensibility and is described as the tendency to focus on internal bodily signals, this can measured through self-reporting questionnaire/surveys (Farb et al., 2015; Mehling et al., 2012; Porges, 1993). The use of self-reporting questionnaire/surveys does come with some limitation as they can reflect participant threshold bias (Garfinkel et al., 2015). Despite this they do give insight and explanations as to how internal sensations are experienced (Garfinkel et al., 2015; Weineck, Messner, Hauke, & Pollatos, 2019). Tskiris et al (2011) described interoceptive sensitivity as the ability to notice and represent internal signal from the body compared to the magnitude of the change in body image as measured with a multi-sensory task such as the rubber hand experiment. This research demonstrated that low interoceptive sensitivity results in greater adaptability to body representations. The rational proposed for this is that in a low interoceptive situation more attentional resources can be allocated to external multi-sensory processing (exceteroception and proprioception) due to the abridged experience of the internal states (interoception) (Tsakiris et al., 2011, p. 2475). In such, does this then not also support a theory that heightened interoceptive sensitivity could have the opposing effect and less attentional resource allocation for the external sense of the body? This also may potentially be interpreted as an internal-external focal imbalance within body awareness (i.e. heightened interoception and diminished proprioception) that is also associated with JH (Mallorqui-Bague et al., 2014; Ghibellini et al., 2015b). Therefore it may also play a critical role in the experience of a body with hypermobility.

The third and final dimension of interoception is known as interoceptive awareness which is described as the

> *"conscious perception of sensations from inside the body that create the sense of the physiological condition of the body, such as heart beat, respiration, satiety, and the autonomic nervous system sensations related to emotions* (Mehling et al., 2012, p. 2).

Interoceptive awareness also provides a marker for metacognitive awareness of interoceptive ability and can be quantified using combined measures of interoceptive accuracy (e.g. heartbeat discrimination task performance) with a measure of subjective confidence in performing the task (Garfinkel et al., 2015). It can also be measured using a validated and widely applied self-reporting questionnaire that provides a *"differentiated assessment of psychological aspects of the perception and evaluation of body sensations"* (Mehling et al., 2012, p. 20). Whilst it is widely used to assess internal body awareness, this assessment tool is self-reporting and therefore may be viewed as subjective and not an instrument that can be used to diagnose determine clinical conditions such as anxiety. It does however provide an understanding of the aspects of body awareness that are related to outcomes such as anxiety and can also inform the design of mind-body therapies aimed at treating those conditions (Mehling et al., 2012).

While heightened interoception is linked with anxiety (Mallorquí-Bagué et al., 2016), the literature also evidences direct links between balanced interoception and mental wellbeing (Weineck et al., 2019). Interoceptive processes are associated with intuitive judgment and decision-making, emotional experience, emotional processing , behavioural self-regulation and body image are also implicated (Weineck et al., 2019). Weineck et al. (2019) applied a bottom up approach (i.e. from body to brain) to their research using a physical embodiment of '*positive power'* postures. These were found to significantly related to improved interoceptive awareness (Weineck et al., 2019, p. 13). In agreement with Tsakiris et al (2011), it was also found that influences between sensory external and internal signals were the reason for an increase in interoceptive awareness (Weineck et al., 2019). This work

concluded that the adoption of a positive physical pose meant focusing on the external bodily aspects of the self which in turn provided feedback resulting in a balanced accuracy and awareness of internal bodily states (Weineck et al., 2019). That said, could we then theoretically propose that increased focus on proprioception such as awareness of the body's posture in space could potentially re-balance body-awareness and decrease the focus and attention to internal interoceptive processes that are associated with anxious states?

With clear indications arising thus far from the literature to suggest that a balanced bodily-awareness and arousal are important for mental wellbeing, it is proposed here that a window of tolerance within bodily awareness processes and states could also be important for emotional state regulation. The notion that a window of tolerance within body-awareness is further elaborated here and framed using the hierarchical model of autonomic activity, also known as the polyvagal theory (Porges, 2011). Unlike pure physiology (that has its focus on mechanism, structure and clinical status) the polyvagal theory is rooted in psychophysiology and is historically driven by paradigms derived from psychology, often treating physiological parameters as if they were observable behaviours (Porgess, 2007). The Polyvagal theory (Porges, 1995) introduced a new perspective relating to the autonomic nervous system's (ANS) function. It encourages a level of inquiry that challenges scientists to incorporate an integrative understanding of the role that neural mechanisms play in regulating bio-behavioural processes. It emphasizes that physiological states support different classes of behaviour, and that it is the two branches of the vagus that also support different adaptive behavioural strategies. The polyvagal theory articulates three phylogenic stages of development within the autonomic nervous system (ANS). These three stages can be conceptualised as dynamic circuits providing adaptive responses to safe, dangerous or life-threatening situations and contexts. Porges (1995) applied the poly-vagal theory to provide a convincing theory towards the neurobiology of 'feeling safe' (Porges, 2017, p33). He

suggested that protective physiological adaptations seen in perceived dangerous or life threatening situations can either manifest in arousal (e.g. increase in blood flow to support the fight or flight mechanism) or a physiological shut down (e.g. passing out) and/or psychological disassociation. The theory also proposes that this evolution of the autonomic nervous system (ANS) provides neurophysiological substrates for emotional experiences and affective processes that are critical in social behaviour such as attachment (Heilman et al., 2008; Porges, 2003) and autism, whereby a threat or diminished feeling of safeness can result in a triggering of disassociation as a defence mechanism (Porges, 2017, p50). This also supports the bio-directional brain body connection in that a person's physiological states can also limit the range of behaviour and psychological experience (Porges, 1995). The evolution of this nervous system determines the range of emotional expression, facial gestures, vocal communication and social behaviour and provides a plausible explanation for social, emotional and communication behaviours and disorders (Heilman et al., 2008; Porges, 1995, 2003).

Emotions are produced primarily in the amygdala and hypothalamus of the brain's limbic system and the amygdala is central to this process. The amygdala is a group of nuclei in the cerebral hemispheres of the brain and can send direct signals to the frontal cortex which is part of the brain that controls important cognitive skills in humans, such as emotional expression, problem solving, memory, language, judgment, and sexual behaviours. The amygdala is a crucial region of the brain that supports motivation and emotional memory and it is also implicated in the autonomic nervous system's (ANS) threat processing, bodily arousal reactions and the expression of moods (Eccles, Owens, Mathias, Umeda, & Critchley, 2015). The amygdala can also send signals indirectly to the hypothalamus, which releases hormones such as endorphins. These are associated with both positive emotions and cortisol which is critical for homeostasis and is also related to hyper-arousal, stress and anxiety

triggering the body and brain in to a state of emergency (Minton & Faber, 2016, pp. 64 & 65). This state of emergency is also otherwise proposed as 'bodily crisis' in the literature pertaining to skill development (Toner, Jones and Moran, 2016). Hormonal changes that induce hyper-arousal, stress and anxiety are triggered via an indirect signal that is relayed to the somatosensory cortex and on to the frontal cortex of the brain where they result in emotion (Carter, 2010). Interestingly, structural differences in amygdala volume comparable to, or greater than, observations from volumetric studies of clinical psychiatric populations have also been observed in JH (Eccles et al., 2012). Sinibaldi et al (2015) proposed that musculoskeletal complications alongside interoceptive and proprioceptive aspects of body awareness represent visceral, cognitive and emotional counterparts of JH (Sinibaldi et al., 2015). Interestingly people more sensitive to visceral signals, as experienced in heightened interoception, do experience more intense emotions (Critchley & Garfinkel, 2017) and the neural structures that are responsive for the regulation of interoception are also intertwined in emotional processes (Damasio, Grabowski, Bechara , Damasio, Ponto & Parvizi, 2000).

Evidence of links between JH and Ehlers-Danlos syndrome (hEDS) and altered brain development and activity in children (e.g. attention deficit disorders ADD, attention deficit hyperactivity disorders ADHD , Autism spectrum disorders ASD, clumsiness, poor coordination proprioception and visio-spatial skills) are also presented as potential mechanisms through which JH and psychopathological symptoms interrelate (Sinibaldi et al., 2015). Likewise, it is proposed that a somatic condition such as JH could act as a stressor that could induce maladaptive psychological responses that are at times diagnosed as an adjustment disorder (Sinibaldi et al., 2015). In addition, the importance of considering hereditary disorders of connective tissue (HDCT) as underlying Early Symptomatic Syndromes Eliciting Neurodevelopmental Clinical Examinations in children, also known as ESSENCE (Gillberg, 2010), is also highlighted in the literature and in particular when there

is a family history of pain or joint hypermobility (Baeza-Velasco et al., 2017). Baeza-Velasco, Grahame and Bravo (2017) recently highlighted the need for research in this emerging area in order to combine and make better understanding the somatic and developmental/psychiatric aspects of JH and EDS.

So what does this all mean within JH and dance? The proposed links between the bi-psycho-social aspects relating to disordered connective tissue and amygdala volume, emotional processing and body-awareness, i.e. interoception and proprioception could potentially signpost that 'bodily-crisis' occurs when these processes are disordered or imbalanced and this would alter how the brain-body functions are experienced. For example, heightened visceral interoceptive sensitivity may be experienced as stress/anxiety whereas a diminished sense of where the musculoskeletal apparatus of the body is in space i.e. proprioception may be experienced as fear and a feeling of not being safe. This suggestion is based on the understanding that the processing and memory of reactions such as fear and stress are modulated by interoceptive awareness and that these also affect how emotions are experienced and expressed (Pollatos & Schandry, 2008). Further supporting evidence is found in somatic experience therapy used for trauma and chronic stress. Here bottom-up processing within the brain-body function is used to direct a client's attention to regulating bodily awareness (Payne, Levine, & Crane-Godreau, 2015). This includes attending to both visceral (interoceptive) and musculo-skeletal (proprioceptive and kinesthetic) experiences as opposed to cognitive or emotional experiences (Payne et al., 2015). The experience of JH can then be further explained through the intention that there is a 'window of tolerance' within body awareness within which the focus of interoceptive and proprioceptive functions are balanced. The regulation of the internal and external focus is challenging and therefore easily upset by additional stressors, such as having to adapt to new situations or learn new skills or choreographic materials. Again this is something that the researcher has observed and

documented in the researcher's memo (Appendix 4). A window of tolerance is typically described as the zone of arousal in which a person is able to function and adapt most effectively (Slegel, 2010; Ogden, Minton & Pain, 2006). It is also proposed here that the 'window of tolerance' within the regulation of body awareness associated with JH may be further implied using the hierarchical model of autonomic activity and vagal tone, otherwise known as the polyvagal theory (Porges, 2001).

A diagrammatic representation of the proposed polyvagal theory and bodily crisis within JH is presented in figure 5 below.

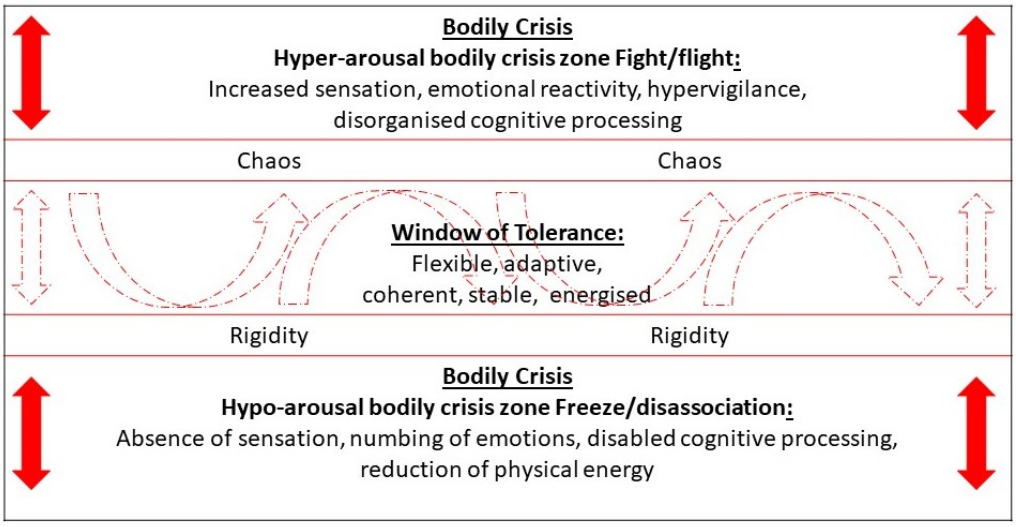

Figure 5 A model of Bodily Crisis within JH applied to Polyvagal and Window of Tolerance theories. Adapted from Siegel and Ogden, Minton & Pain (Siegel, 2010; Ogden, Minton, & Pain, 2006)

Within the model presented in Figure 5 it is proposed that 'normal' functioning and balanced body awareness could occur within the 'window of tolerance'. However, trauma or turbulence potentially caused for example by poor or imbalanced body awareness and/or additional stressors within the model could potentially result in 'flipping ' between and or getting temporarily stuck within either the hyper and hypo arousal states (Ogden et al., 2006; Siegel, 2010). This is further explained using the polyvagal theory and the notion that three defence mechanisms (fight, flight and dissociation) are available when danger or turbulence

is sensed (Porges, 2017, p. 101). The polyvagal theory (Heilman et al., 2008; Kemp, 2017; Porges, 2017) also proposes that when the body is in any one of these defence mechanisms it is using metabolic resources and cannot be for example creative or heal. The implications for this within dance are significant as creativity is fundamental to the art form and injury is prevalent within elite sports. Porges (2017) further proposed that the *"neural pathway for healing overlaps with the neural pathway for social engagement"* (p 101) and is the vagal pathway that sends information between the brain and the body signalling safety and providing calm. This pathway, in addition to providing feelings of safety and calm, also enables social engagement and expression in the face and body (Porges, 2017, p.101-102). This is significant in dance as expression through the face and body is critical. It is further proposed here that the neural pathway for social engagement that Porges discussed, including the interchange of brain-body signalling, could also include behaviours that are pertinent to effective teaching and learning environments. This suggests that 'feeling safe' is a condition that is conducive to learning as a form of social engagement. Indeed a recent paradigm shift within learning theory and practice now recognises the important role of emotional well-being for learning alongside the more stablished theories of motivation (Rowe, Fitness, & Wood, 2015). Rowe et al (2015) further built on previous research (Fredrickson, 2004) to provide additional evidence to suggest that positive emotions enhance cognitive and social functioning. In particular within this functioning Rowe et al (2015) pertain to memory, concentration and communication (Rowe et al., 2015, p. 7). They also discussed how negative emotions, with particular reference to fear (i.e. not feeling safe) were obstructions to this process. This further corroborated Porges' theory that 'feeling safe' enables social engagement such as learning that requires concentration and memory and expression in the face and body (i.e. communication). However memory is neither conscious nor a function of learning unless it can be recalled and the most powerful cue known for recall is the emotional

context of the memory (Zull, 2002). Nonetheless negative memories are known to inhibit recall and learning (Minton & Faber, 2016, p. 92).

Intense emotions are associated with hypermobility (Eccles et al., 2015) and intense emotions can also invigorate the homeostatic fight/flight mechanism, influencing the vagal states within the proposed window of tolerance. Intense emotions are also a catalyst for art and in such "*fuel the dance-making process and ignite audiences"* (Minton & Faber, 2016, p. 63). Indeed dance training involves the cultivation, manipulation and communication of emotions from a relatively young age. Could this training then act as a protective factor for dancers with JH as it will inevitably train them to a deeper understanding of feelings and emotions and unavoidably also to self-regulation?

Emotion processing, the key affective brain processing areas, and higher brain responses are also associated with the anxiety that includes somatic symptoms (e.g. panic disorders) that present in people with heritable collagen variance and Joint Hypermobility (Mallorquí-Baguéa, Bulbena, Roé-Vellvéc, Hoekzemac, Carmonac, Barba-Müllera, Fauquetc, Pailheza, & Vilarroya, 2015). Whilst advances in affective neuroscience provide a subsequent renewed empirical understanding of the relationship between bodily sensation and emotional experience. This new understanding also suggests that body awareness is an appropriate, if at times controversial (Garfinkel & Critchley, 2013), concept that emerges from both clinical experience and theoretical models of emotion (Garfinkel & Critchley, 2013; Mallorquí-Bagué et al., 2016). For example, theoretical models of emotion define complex states of feeling which can manifest in physical and psychological changes, and these in turn can influence thought actions and motivation(Meyers, 2004). The main theories of motivation that are directly associated with emotional arousal are principally arranged into three categories; physiological, neurological, and cognitive. Whilst this is understood there is still a lot of debate and ambiguity as to why we have emotions (Griffiths, 2017; Meyers,

2004). Meanwhile, whilst people who can judge their bodily signals to a high level of accuracy are more likely to experience higher levels of anxiety, they are also able to experience emotions more intensely (Bulbena et al., 2014). What is clear is that emotional states (such as for example, fear, anxiety or love) are associated with physiological changes in the body (e.g. tremor). However it is contested as to whether the physiological changes arise from the efferent emotional state or the emotional state is a result of the afferent physiological change (Schachter & Singer, 1962). Similarly, the neurologically based homeostasis and poly-vagal mechanisms of fight, flight or disassociation are linked with the cognitive appraisal of situations which in turn manifest in physical responses (Myers, 2004).

These theories clearly corroborate the notion of a bio-directional efferent/afferent flow of information between the brain and the body. They also substantiate the brain-body connections proposed within philosophically underpinned embodiment theory and the notion that, yes the body does shape the mind (Gallagher, 2005, p. 149).

Furthermore, whilst a relatively new concept within neuroscientific terms, interoception, when broadly defined as the sensing and awareness of signals originating within the body (Farb et al., 2015), actually supplements our sense and understanding of embodiment, motivation, and well- being (Farb et al., 2015). Interoception regulates a persons' sensitivity to changes in the internal physiological state of the body which are also known to be linked to anxiety, physiological arousal and the resultant physical sensations (Mallorquí-Bagué et al., 2016). As a component of body awareness, interoception is also strongly influenced by mental processes, including attention, interpretation, appraisal, beliefs, memories, conditioning, attitudes, affect and potentially connects internal physiology to socio-emotional competence (Murphy et al., 2017) and social behaviours (Garfinkel & Critchley, 2013). Accelerated interoceptive sensitivity and autonomic abnormalities such as accelerated pain sensitivity, IBS, fibromyalgia, chronic fatigue, postural tachycardia

syndrome are indeed known to overlap with both JH and anxiety disorders. Additionally these abnormalities are also associated with an accelerated (hyper) body-awareness (Mallorquí-Bagué et al., 2015; Mallorquí-Bagué et al., 2016). Indeed, research proposes that the brain structure of people with JH presents an overrepresentation (up to 16 times) of people with anxiety and altered autonomic reactivity (Eccles, Owens, Mathias, Umeda, & Critchley, 2015).

Let us further consider then bodily metaphor as proposed by the embodied neural theory of language and expression whereby understanding in one domain is transferred to another and the mind and body are one (Feldman, 2008, p195). This surely also confirms that thinking is functional and ideas are things that are relative to the body's functioning (Lakoff and Jonson, 1999; Feldman, 2008, p 194). In turn this also explains how bodily sensations such as pain are closely associated with emotions (that are also common and fundamental across expressive choreographic communication) for example passion, anger, sadness and love. Could then the experience of intense emotions, if managed and regulated, also provide increased awareness and artistic ability in a dancer as a means towards the expressive communication of emotion? May then the notion of increased body-awareness in JH also support, to some degree, a view that JH in dance is indeed an asset that goes beyond the musculoskeletal ability to extend the limbs and spine beyond normal parameters?

Whilst this is an interesting proposition, what does remain is that people with JH have an increased ability to perceive actual changes in the physiological state of their body and they therefore have a tendency to focus on physical symptoms with magnification which can also have negative out-comes (Eccles et al., 2012). These findings also improve our understanding of the mechanisms through which vulnerability to anxiety and compulsive disorders arise in people with hypermobility (Baeza-Velasco et al., 2011; Eccles et al., 2015).

There is clear evidence to support the prevalence of JH in dance, yoga, aesthetic sports (Beighton, Grahame, & Bird, 2011; Remvig et al., 2007a) and some further sports that require and overexpressed range of motion in some joints (e.g. swimming, netball, gymnastics) (C. Chan et al., 2018; Liaghat et al., 2018; Soper, Simmonds, Kaz Kaz, & Ninis, 2015). However what is perhaps more unusual in sport yet prominent in dance and yoga practices is the intense somatic training that occurs to heighten the internal sense of the body (interoception) (Sze, Gyurak, Yuan, & Levenson, 2010) and the sense of the body in motion (proprioception) (McCormack et al., 2019). Research has indicated a linear relationship among groups comparing yoga and dance practitioners with controls (Sze et al., 2010) where the relationship between emotional and physiological states was explored in relation to the role of visceral and somatic body-awareness in the specific practices. Specifically it showed that those with training in yoga and meditation focusing on visceral body-awareness (i.e. attention to breathing and heartbeat) had the greatest coherence between emotional and physiological states, next were those with dance training and somatic followed by the controls who did not have any body-awareness training (Sze et al., 2010). Following the most recent summit on interoception (2016) it was proposed that sensing, understanding, and integrating information about the bodily systems is understood and therefore measured as interoception awareness. This includes interoceptive attention, detection, discrimination, accuracy, insight, sensibility, and self-report (Khalsa et al., 2018).

Given the biological importance of interoception for regulating bodily functions there is without doubt a clear rational for the association between interoception and both physical and mental health. Furthermore, current research now also connects interoception with less apparent functions such as higher-order cognition, emotional regulation (Pollatos & Schandry, 2008), learning and decision making (Werner, Jung, Duschek, & Schandry, 2009), further confirming that emotional state is also important for cognitive processing, motivation

and learning (Blankson et al., 2013; Immordino-Yang & Sylvan, 2010). This further supports Porges' theory of 'feeling safe' as being conducive to social engagement activities such as learning (Porges, 2017). Also the researcher has anecdotal observations of issues with emotional states (anxiety and panic) retention and recall of learning (remembering choreography and spatial patterns) in the dance environment with dancers with JH (see memo appendix 4). Of further significance here are the links proposed between the ability to describe emotions and interoceptive awareness which were presented by the 2016 interoception summit committee (Khalsa et al., 2018). Finally the summit noted that interoceptive awareness can also change over a lifespan and what is of interest to the present study is that it may decrease in adolescence (Khalsa et al., 2018).

Psychiatric studies also explore links between anxiety, interoceptive awareness and hypermobility in sub-clinical populations (Mallorquí-Bagué, Garfinkel, Engels, Eccles, Pailhez, Bulbena, & Critchley, 2014). This work suggested that there was a significant relationship between state anxiety and joint hypermobility (JH) with interoceptive awareness being heightened in JH and assumed to play the facilitatory role between state anxiety and hypermobility.

Eccles et al. (2015) proposed that JH has specific yet common variants in physiological reactivity that are related to the autonomic nervous system (ANS) and also have major influences on emotional state (Eccles et al., 2015). Meanwhile, Seibert and Ellis (1991) also believed that both positive and negative emotional states can lead to potential cognitive impairment and more irrelevant thoughts are produced during emotional mood states (Seibert & Ellis, 1991). Their research showed that emotional mood states are related to alterations in social and personal judgments, alterations in spatial judgments, mood congruence effects, and recall impairment (Seibert & Ellis, 1991). Thus, emotional states may be considered as a function of a state of physiological arousal and/or of a cognition that is

appropriate to this state of arousal. Further, emotional states influence cognitive processes and in most emotion inducing situations the two may indeed be interrelated (Schachter & Singer, 1962).

It is argued then that physiological reactivity is proposed as bodily crisis that could potentially have major influences on emotional states and may also present as a rational for the researcher's anecdotal yet compelling observations of altered cognitive processing and recall when learning new skills in young hypermobile dancers (appendix 4). In order to examine this further it is important to discuss the interrelation between physiological and emotional arousal and cognitive processes.

Emotion processing, the key affective brain processing areas, and higher brain responses are also associated with the anxiety and somatic symptoms that present in people with heritable collagen variance and Joint Hypermobility (Mallorquí-Baguéa, Bulbena, Roé-Vellvéc, Hoekzemac, Carmonac, Barba-Müllera, Fauquetc, Pailheza, & Vilarroya, 2015). Whilst advances in affective neuroscience provide a subsequent renewed empirical understanding of the relationship between bodily sensation and emotional experience. This new understanding also suggests that somatic body-awareness and interoceptive awareness are appropriate, if at times controversial (Garfinkel & Critchley, 2013), concepts that emerge from both clinical experience and theoretical models of emotion (Garfinkel & Critchley, 2013; Mallorquí-Bagué et al., 2016). Theoretical models of emotion define complex states of feeling which manifest in physical and psychological changes that in turn can influence thought actions and motivation (Meyers, 2004). The main theories of motivation that are directly associated with emotional arousal are principally arranged into three categories; physiological, neurological, and cognitive. Whilst this is understood there is still a lot of debate and ambiguity as to why actually we have emotions (Griffiths, 2017; Meyers, 2004).

What is clear is that emotional states (such as for example, fear, anxiety or love) are associated with physiological changes in the body (e.g. tremor), it is contested however as to whether the physiological changes arise from the efferent emotional state or the emotional state is a result of the afferent physiological change (Schachter & Singer, 1962). Similarly, the neurologically based homeostasis and fight or flight mechanism associates with the cognitive appraisal of a situation, which in turn manifests in physical responses (Myers, 2004).

Together with the uncertainty around terms used within the interoception literature, proprioception, a facet of body-awareness that is controlled by the sensory cortex of the brain is also discussed in the literature from both scientific and philosophical viewpoints (Barlow, 2018; Batson, 2009). Lack of application using common definitions and terminology across research are also highlighted as an issue and potential barrier to a clearer understanding of proprioception (Barlow, 2018). Indeed, within dance research both the scientific and philosophical viewpoints for proprioception are pertinent. The latter being the most common in the dance literature as until recently there really was very little scientific evidence to support what exactly the mechanism for proprioception involved other than a means to a specific dance aesthetic or when in deficit a potential mechanism towards injury (Batson, 2009). Latterly, balance and postural awareness have been recognised as an important means to define and determine bodily awareness within dance and their enhancement are cited as a means to prevent injury and enhance performance (Batson, 2009).

From a strictly biological viewpoint proprioception is understood as *"typically absent from conscious perception"* (Tuthill & Azim, 2018, p. 187). A 'sixth sense' that is critical to human experience and without which there is a total inability to coordinate movement (Tuthill & Azim, 2018),without which a body must also remain unreal and unpossessed (Sacks, 1987, p. 43). Knight further corroborated this notion in her autobiographic description

of a deficit in proprioception as "*frightening"* (Knight, 2015a, p. 124). She also described how only correct management of her hypermobility allowed her to eventually develop self-awareness and be in control as opposed to being at the "*mercy of her body"* (Knight, 2013, p. 141). Whatever the focus and debate, biological, philosophical or motor control, our understanding of proprioception remains as a feedback mechanism for the sensation of body position, posture and movement (Barlow, 2018; Batson, 2009).

Body posture, control and balance are a form of dynamic equilibrium that require body-awareness as detected by proprioceptors known as Golgi tendon organs (Tuthill & Azim, 2018). Biologically these lie at the interface between muscles and tendons, each Golgi tendon organ is attached to individual muscle fibres and contains mechanosensory endings that are wrapped around strands of collagen (Tuthill & Azim, 2018). These afferent (as opposed to efferent) innervate tendon organs carry nerve impulses from sensory stimuli towards the central nervous system and brain encoding muscle force. At rest Golgi tendons are silent only increasing their firing frequency in the muscle resulting in increased tension during for example resisted movements (Tuthill & Azim, 2018). If we pause for a moment here then to reconsider joint hypermobility and its status as a HCTD then it is plain to see that the involvement of the associated biological tissues (including collagen, Golgi organ mechanoceptors, muscles and joint tissue) that provide the mechanisms for movement control and proprioception will, in the presence of JH, be potentially altered and in deficit. Furthermore, from a motor control perspective within elite training regimes, deficient or altered sense of the body in space may also present as a 'bodily crisis' in skilled performance. Bodily crisis arising when the adaptive and flexible capacity of the performing body is challenged (Toner, Jones, & Moran, 2016).

Indeed, it now seems that an intriguing paradox is emerging whereby JH is prevalent in dance and is additionally associated with poor body-awareness that manifests as

heightened interoception and diminished proprioception. Despite this, it is accepted that dance practice enhances a sense of motion (proprioception) and the somatic practices that are included in dance training serve to develop and regulate body-awareness. Therefore, could it be that dance potentially not only has a protective factor against injury in JH (as proposed in study I) but also has protective factors against altered or imbalanced body-awareness?

The theories presented above provide an interesting, albeit conceptual, explanation to the researcher's career long observations of dancers in the teaching and learning environment who were suspected to fall within the JH category. These dancers whilst physically very competent in the execution of dance skills and techniques required of them in class have been described as being challenged with remembering, recalling and memorising spatial patterns and dance sequences or choreography. Likewise they were also often void of any emotional or expressive communication in their performance. That said, it is then also proposed here that these dancers had surpassed their window of tolerance and were in some form of 'bodily crisis'. This also suggests that they were possibly void of safe feelings and positive emotions thus inhibiting their abilities to remember, recall and communicate the dance sequences they were learning. An interesting proposition perhaps, yet unsubstantiated and to the authors knowledge never before tested in dance.

Further literature relating to JH, classical ballet training, body awareness and emotional expression as an important component of that training has now been reviewed. It is suggested that within the biopsychosocial focus of this research, some physical attributes desired within the art of classical ballet and JH also signpost towards classical ballet training. For example, extreme range of motion and extension of the limbs. Meanwhile, it appears that psycho-social elements of body awareness are paradoxically entangled within JH and classical ballet. For example, proprioception is enhanced with classical ballet training but in deficit within JH. Similarly the capacity to express emotions through the body is fundamental

to the art-form and again essential within classical ballet training yet paradoxically, feeling pain and emotional sensitivity are flagged as impairments within JH. Equally, visio-spatial skills are fundamental to dance training yet the research suggests that these are in deficit in JH (Baeza-Velasco, Bourdon, et al., 2017). The researcher has anecdotal evidence to suggest that dancers with joint hypermobility have difficulty with visuo-spatial memory, and in particular short term working memory, when under the stressors of learning new movement sequencing (documented in the researcher's memo appendix 4). The challenge in the study reported here then was to design research that could objectively measure and quantify these aspects of bodily awareness in dancers and compare those in the group with JH to the dancers that did not present with JH.

5.3 Research aims

Additional evidence towards the entanglement of biopsychosocial components within joint hypermobility is reviewed in this chapter and it is clear that there are implications within JH that go beyond the musculoskeletal elements that were investigated in study I. The literature and evidence presented in this chapter suggest that there are implications of vulnerability yet signposting towards dance training within JH that relate to interoceptive and proprioceptive mechanisms of body awareness. Additionally there is evidence in both the literature and the researcher's anecdotal observations to suggest that visio-spatial memory (a component of proprioception) may also be altered in JH. Finally anxiety, which is interrelated with interoception and therefore body awareness, is also evidenced (in the literature presented in this chapter and documented in the researcher's memo) as an additional consequence of JH. Therefore, the study reported here was designed to further explore these biopsychosocial implications of body awareness within JH. Situated within the classical ballet environment for elite vocational dancers, the main research focus was to answer the following question:

Is there a relationship between generalised joint hypermobility and body awareness in elite vocational classical ballet dancers?

Following on from this main question were sub-questions.

i. Do dancers with GJH have lower proprioception than dancers without GJH?
    a. Do dancers with GJH have poorer balance than dancers without GJH?
    b. Do dancers with GJH have poorer spatio-visual awareness than dancers without GJH?

ii. Do dancers with GJH have more intense interoception than dancers without GJH?

iii. Do dancers with GJH have higher anxiety than dancers without GJH when engaging with motor and cognitive tasks simultaneously?

Finally and in order to mitigate for the potential effects of classical ballet training towards improved body awareness a further question was explored in connection with the above sub-questions, as follows:

iv. Do more experienced dancers have improved body awareness?

5.4 Ethical considerations

Ethical approval for the study was confirmed through the ethics committee at the researcher's institution (University of Edinburgh). Further permission was obtained from the secondary school head and the dance director to approach the parents and pupils in order to recruit participants to this study undertake the data collection. Information about the research and parental/legal guardian informed consent forms were sent to the homes of all pupils for approval. Information about the research and pupil informed consent forms were also disseminated to all pupils in the school. The researcher was also available for further clarification and questions about the research procedures and intent via e-mail or upon

appointment at the school, at the convenience of the school director, parent/legal guardians and/pupils.

5.5 Conceptual context and study design

The literature reviewed in this chapter support that body awareness, and in particular proprioception and interoception are important for the execution and communication of dance (Barlow, 2018; Batson, 2009; Christensen et al., 2018; Potter, 2008). The literature and also the findings from study I also provide evidence for the prevalence of GJH in young adolescent dancers. Evidence connecting GJH and body-awareness within the training environment for classical ballet remains however largely theoretical, anecdotal and experiential. It was therefore important to design methods to this investigation that would provide objectivity and rigour and it was for this reason that a quantitative design was applied to survey the dancers for GJH and body awareness. The conceptual context for the study drew on four major sources, prior theory, pilot studies, experience, and thought experiments (Maxwell, 1996, p. 46). The prior theory provided the standardised protocols and tests that were used in the data collection whilst the researchers experience and thought experiments provided the framework for the application of the protocols to young adolescent dancers in a classical ballet context. In such measures for GJH and body awareness were compared across groups of dancers with and without GJH and body awareness measures were also explored in relation to GJH and the level of dance experience within the group.

The research was carried out in a full time elite training environment for young vocational classical ballet dancers in Scotland. This specialist dance school is situated in a state secondary level school in Glasgow for dancers aged between 11 and 18 years that is fully funded by the Scottish government. The dance pupils are integrated in to the secondary school for their general education and undertake their dance training daily in the afternoons and evenings Monday to Friday. They board at the school, their training is free, and entry to

the school is for children of Scottish residents only. Selection for the classical dance training is determined by a rigorous process of application and audition. The school director, tutors, independent dance experts and members of the classical ballet profession in Scotland (Scottish Ballet) make up the selection panel. The applicants are also assessed for general health upon entry, but screening for hypermobility is not part of this assessment. The school has representation of talented young dancers from all socio-economic areas and diversities within Scotland (Glasgow Community Planning Partnership, 2019) .

5.6 Overall procedures

The surveillance for GJH and body awareness was undertaken over a period of two consecutive weeks towards the end of the school year and the total data collection took eight days. Both parental or *in loco parentis* and participant informed consent was required before the screening. Only participants that were available during the allocated two week data collection period were included in the study to ensure that they were all at the same stage in their training during that year. The testing time for each dancer varied between 45 and 60 min. Each dancer was screened individually in a room in the school's health suite where equipment could be set up and left without disturbance and participants had privacy and quiet away from the dance studio and peer pressures.

5.7 Participants

Thirty two dancers participated in this study, seven were male and 25 female. The mean age of all participants was 14.2 years, (SD = 1.7, range 11-17yr.) mean male age was 13.4 years (SD = 1.4, range 13-17) mean female age was 13.5 years, (SD = 1.7, range 11-17yr). The average of overall dancer training was 9.5 years (SD = 2.8, range 4-15yrs) within the group and this ranged from 4 to 16 years. Whilst their training experience varied, all participants had the required skills and experience levels and had been successfully selected

to train at the dance school at the time of the data collection. This design, as opposed to a comparative experimental design using a control group of non-dancers, was chosen because of the potential effects of the intense dance training on both hypermobility and body awareness. All participants were undergoing a common training regime and who were surveyed by the researcher using the same data collection methods each time. This ensured that there was no variance in the application of the testing protocols and the dance training received was consistent across participants during the data collection. The participants were treated as a homogenous group from one vocational dance training setting and those that were classified as GJH positive were compared to those that were not.

Finally, it is well known that both maturation and growth are by no means linear between the ages of 11 and 18 years (e.g. Cumming et al., 2012) which was the target age group. It is also acknowledged that age at peak height velocity (PHV) for both sexes within aesthetic sports comparable to elite dance training (such as artistic gymnastics) can often occur later than other groups (Malina et al., 2015). Biological maturation is a process that occurs in all bodily tissues, organs and systems (Cumming et al., 2012; Malina et al., 2015) whereby the outcomes of underlying processes associated with maturation can be measured to provide an indication of progress towards a biologically mature state. Maturity assessment has specific application in the classification of children for sport during the adolescent period (Mirwald et al., 2002) as the range of variability of biological maturation status between individuals of the same chronological age in somatic and biological growth is large and can be accentuated around the adolescent growth spurt (Mirwald et al., 2002). Therefore it was important to determine each participant's maturational status. This was important because some stages, for example the adolescent growth spurt, can greatly influence flexibility (Malina et al., 2015; Storm et al., 2018). Importantly, hip flexion, hamstring extensibility and the ability for a dancer to reach the floor with their hands from a standing position with

extended knees (Mitchell et al., 2016) is also one of the nine points on the Beighton criteria and scoring (C. Chan et al., 2018; Singh et al., 2017) used in the present study.

It was therefore essential that the effects of maturation were controlled for within this research investigation (Mirwald et al., 2002) . This would allow the researcher to determine at which stage the dancers were within their maturation and if they had indeed surpassed the adolescent growth stage and reached their predicted peak height velocity (Mirwald et al., 2002). Whilst radiograph-based methods are reported as the most suited in predicting actual PHV they do involve exposing young children to ionizing radiation and are also costly (Mills, Baker, Pacey, Wollin, & Drew, 2017). Anthropological measures such as foot length increase is also an accurate surrogate to radiographic measures however changes in foot length need to be monitored over time using standardised measures for this method to be accurate. Equation based methods such as the one proposed by Mirwald et al (2002) are reported as frequently applied and reliable methods for predicting maturation status, however it must be noted that there are criticisms that report the data used to inform PHV equations is potentially now out-dated and does not reflect current generation adolescent (Mills et al., 2017). There is now a consensus that the prediction of maturity can be improved using anthropometric variables and a maturity ratio (Fransen et al., 2018). However, at the time of this study and data collection the accepted equation based method was advised by the supervisory team for application and use in this research as it was the most ecologically viable for this study as it is an accepted measure for maturity offset or the number of years pre or post maturity (Mirwald et al., 2002). In such this method provided an indication of a homogenous group of participants that were not affected by adolescent growth spurts.

5.8 Measures and research tools

The variables measured for in this study were; maturation, generalised joint hypermobility (GJH) and body awareness (BA). Body awareness was characterised by applying measures for; proprioception (balance and spatio-visual memory), interoception (multidimensional assessment of interoceptive awareness MAIA), and anxiety (galvanic skin response GSR and heart rate variability HRV). Generalised joint hypermobility (GJH) was measured using the Beighton criteria and the standardised methods applied in study 1 were carried in to this study. The choice of research tools for the investigation of body awareness were carefully considered within the research context. Whilst drawing on current understandings of GJH, it was also important to take in to account and draw on the researchers own experience (Maxwell, 2013) of GJH within the teaching and learning environment for dance. The tools selected for this study are reviewed, outlined and discussed below. The main criteria for inclusion were that they should be ecologically viable for dance yet valid, simple to apply in the field and relatively non-invasive yet appropriate to the elite classical ballet environment.

a) Testing for maturation status

On day one of the data collection, the 32 participants were gathered together and PHV was explained within the context of the study. Students were made aware that if they had not yet reached PHV they would not be used further in the data collection and there was time for the students to ask further questions about the study or opt out. Following this information and question and answer session all dancers agreed to proceed. Measures of height, sitting height, date of birth and weight were taken and applied to Mirwald et al's (2002) methodology and calculation of PHV. In this process a portable stadiometer (Seca 213) was used to record the anthropometric measures of height and sitting height and these were

measured to the nearest mm. A recently calibrated digital floor scale (Marsden M-420 Digital Portable Scale) was used to record the dancers' body mass to the nearest 0.1 kg. Measurements were repeated twice and an additional third measurement was taken if the first two differed by more than 4 mm for height and sitting height and 0.4 g for weight (Mirwald et al., 2002). The two measurements for each anthropometric measure were averaged. If three measures were taken, the median value was used. The anthropometric techniques for measuring the dancers are presented in appendix 5 (Ross & Marfell-Jones, 1991, pp 223-308.).

b) Testing for Generalised Joint Hypermobility

The dancers were surveyed for joint hypermobility using the Beighton criteria the application of these methods was also tested as reliable in study I. Joint goniometry protocols using a *66fit* goniometer and guidelines widely used in clinical physiotherapy were used to measure the range of motion bilaterally in the metacarpal-phalangeal joint of the fifth finger, elbows and knees (Smits-Engelsman et al., 2011b). The Beighton criteria and a cut-off point of ≥5 for JH+ was applied (Schmidt et al., 2017).

c) Testing for Body-awareness

a. Proprioception

Proprioception was investigated using measures for balance and spatial visual memory.

i. Balance:

Applying Sherrington's original concept of proprioception whereby proprioception is seen as the *"perception of the position of the body, or body segments, in space"* it was considered appropriate to assess how the brain perceives joint position within a multi-segment posture. Whilst balance tests with a more dynamic component to them may be more suitable for testing some athletes (Hrysomallis, 2007), a balance test involving several joint

angles (hip, knee, ankle) was considered more appropriate for the adolescent dancers. The rational for this was that balance and the sense of where the body is in space in terms of the specific posture that is maintained, is important in dance. An example of this would be applied to balance postures such as a *retiré devant* also commonly known in ballet as the 'pirouette position', is a fundamental and integral part of all ballet training (Batson, 2009), and therefore something that the dancers would be comfortable performing. The proprioception of the young dancers was therefore assessed using methods as described by Emery et al. (2002) using a unipedal balance test with eyes open and closed. The balance test was carried out barefoot to eliminate potential differences in shoe support. Each dancer asked to balance on one leg with eyes open on a flat surface and this was then repeated using the other leg and again with eyes closed on both sides. Time was recorded with digital stopwatch when the subject's balance was lost, eyes opened, or the maximum allowable time (180 seconds) was reached (Emery, Cassidy, Klassen, Rosychuk, & Rowe, 2005). In order to make this test more functional for the classical ballet setting a *retiré devant* posture with arms in 1st position was used as the unipedal balance posture and 'balance lost' was defined when the balance posture was no longer technically held or the eyes opened. The complete test was repeated three times bilaterally in both eyes open and eyes closed conditions. The order in which the test was commenced, right or left leg was alternated for each participant (i.e. participant 1 right leg first, participant 2 left leg first). The mean of each of the three repetitions for each leg and condition was calculated and recorded.

ii. Spatio-visual memory (SVM)

Spatio- visual memory is an important component associated with proprioception, testing for spatio-visual memory was undertaken using an electronic version of the standardised corsi-block tapping test (CBTT). The Corsi block tapping task (Corsi, 1972) has been described as the single most important nonverbal task in neuropsychological research

(Berch, Krikorian, & Huha, 1998). It is a nonverbal task that is applied in neuropsychological research and is applicable to children and adults (Brunetti, Del Gatto, & Delogu, 2014; Lefevre et al., 2010). It is commonly used in diagnostics for the assessment of visuo-spatial working memory (VSWM) and spatial attention. Compared to the traditional physical board, e versions of the CBTT have been cited as having several advantages, including: simple installation, set-up, and use; considerably increased accuracy in presentation timing, automatic measures of span and reaction times, in both the forward and backward response modalities (Brunetti et al., 2014). An electronic version of this test was used in this research and the setup consisted of a nine square rubric on the computer screen (Lefevre et al., 2010). The squares flashed on a computer screen in a random sequence, the sequence commenced with two squares and as the test progressed the sequence was lengthened by one further square each time. Participants reproduced the sequences by using the computer mouse to click on the blocks (Lefevre et al., 2010). The test terminated when the participant could no longer accurately reproduce the sequence. The awarded score therefore represented the number of blocks within the sequencing that each participant could reproduce successfully (e.g. up to four blocks, score=4).

b. Interoception

The Multidimensional Assessment of Interoceptive Awareness MAIA (Mehling et al., 2012) was used to assess interoception in this study. MAIA is a validated self-reporting questionnaire that is a 32-item multidimensional instrument with eight separately scored scales (see appendix 6 for full questionnaire and scoring).The scales report scores for;

a) attention regulation
b) not worrying
c) body listening
d) noticing
e) emotional regulation
f) self-regulation
g) not distracting
h) trusting

Originally developed to assess Interoceptive Awareness (IA) in individuals performing mind–body practices the Multidimensional Assessment of Interoceptive Awareness (MAIA) has more recently been used to assess the relationship between IA and eating disorders in both adolescent and adult populations (Brown et al., 2017) and also to assess emotional dysregulation (Price et al., 2019). It was therefore evaluated as appropriate for this study was chosen over testing using standardised heart beat tracking and discrimination protocols. The rational for this being that as it was considered that the questionnaire would be less complex and intrusive and more appropriate for the research setting and age of specific participant group. The questionnaire was administered at the end of the physical testing and the researcher was available to clarify any of the questions if required.

d) Anxiety

Anxiety was measured during some of the tests using two parameters, heart rate variability (HRV) and Galvanic skin response measures (GSR).

i. *Heart rate variability*

The Equivital monitoring system® includes a Heart Rate Monitor, which records inter-beat interval (IBI) data. The data was imported from the sensor through the equivital software for analysis using the USB 2.0 adapter provided. The imported data was automatically presented in an electronic spreadsheet for further analysis. semi-automated identification and mean replacement of outliers (defined here as a high-IBI value of “1200 and above” or low-IBI value of“200 and below”) (Allen et al., 2007). As the heart rate data were collected during the completion of the balance, corsi block and tapping tests the data collection time varied. In order to calculate the mean for a near-standard length for the

collected data, levels of cardiac responding during the first four 1400 IBI segments and the mean MSD of these four segments was to be utilized in this analysis.

ii. *Galvanic Skin Response (GSR)*

GSR can also be referred to as Electro Dermal Response (EDA) and is used as an indicator of stress. It is a proven psycho-physiological measurement and has for example been used to evaluate and quantify levels of anxiety in clinical settings as well as within social interaction settings for children and adults (Najafpour, Asl-Aminabadi, Nuroloyuni, Jamali, & Shirazi, 2017). In this setting GSR was measured whilst the participants performed a simple motor speed tests called the tapping test (TT). Galvanic Skin Response, (GSR), is a measure of conductivity of the skin, it provides an indication of changes in human sympathetic nervous system (SNS) (Shi, Ruiz, Taib, Choi, & Chen, 2007). It is well established that the magnitude of GSR is affected by changes in general mood, emotional reactions and is applied in psychophysiology experiments to infer emotional state and anxiety in response to stressful situations (Appukuttan, 2016; Najafpour et al., 2017). As a person's stress levels increase so does the GSR and likewise a decrease in stress will decrease the GSR (Shi, et al 2007). GSR readings have also been linked to cognitive activity and function and provides a theoretical basis for the use of GSR to measure cognitive load and its variations (Shi et al., 2007). GSR and anxiety can be measured because sweat on the skin is released in anxious states and provides a low-resistance pathway for electrical current that can be recorded and measured as an indicator for anxiety. Using this method a skin galvanometer attachment was applied to the Equvital system and was used to measure and record the galvanic skin response through the passage of a small harmless electric current. The GSR Sample rate was set to 16Hz and was measured in Siemen units. The GSR sensor connected to the expansion port on the Equivital SEM that enabled the recording of the GSR signal. The expansion port was worn on the wrist of the non-dominant hand and the short leads

terminated in standard 'Snap Lead' connectors that were used with the disposable electrodes that were placed on the surfaces of the first and third medial phalanges of the non-dominant hand. This set up allowed the participants to freely use their dominant hand to complete the tapping test. The tapping test is a test for fine motor control that is free to download and is delivered through an app for iPad or iPhone. This digital finger tapping test is used as an analytical tool in a basic research environments to measure a person's motor speed. The TT was applied in this testing for anxiety as the app provided a relatively easy-to-use researching tool in the environment as all of the participants were very familiar with the use of iPad technology and the test required no prior or specific skills. The testing was set up and undertaken as described on the digital finger tapping test app for iPhone (see https://apps.apple.com/tr/app/digital-finger-tapping-test/id439751108). This test was chosen because it was a very simple non-dance specific task and in such did not present technical challenges to the dancer. The test was administered and measured under two conditions, a) only tapping (TT) and b) tapping with additional mental stressors (TT+). The mental stressor took the form of simple mental arithmetic challenges that were delivered by the researcher and the participants were instructed to respond as quickly as possible. For the condition where a mental stressor was applied, a set of twenty five simple mental arithmetic challenges were devised and administered across the three trials in the TT+ condition for each participant. Participants were instructed to respond to as many of the mental arithmetic challenges as they could in the given time across the three trials (3 x30s). The scores for the mental arithmetic challenges were not recorded as the purpose of the additional challenge was to allow the researcher to measure how the dancers' GSR levels potentially fluctuated when presented with synchronous motor control and mental tasks. A familiarisation trial was undertaken for each of the participants prior to the data collection, this was necessary as non-of the dancers had encountered this app before. The tapping test challenge was repeated three

times using the dominant hand for each participant in both conditions (TT and TT+). The average number of taps was automatically recorded in the app across the three trials and a spreadsheet of the results was sent electronically to the researcher through the app. These procedures were accurately repeated across all participants. The galvanic skin response measures were recorded using the Equivital monitoring system® and the tapping the tests (TT and TT+) were commenced once the data stream had been assured. The data was imported from the sensor through the Equivital software for analysis using the USB 2.0 adapter provided and the imported data was automatically presented in an electronic spreadsheet for further analysis. Before any analysis took place the GSR data was cleaned for outliers otherwise characterised as artifacts. These were defined using methods published by Hubbard (2002) where GSR measures of less than .12 Siemen units and greater than 50 Siemen units were classed as artifacts (Hubbard et al., 2002). Once artifacts were removed, the GSR data for each tapping test (TT and TTMS) was used in subsequent data analyses. The tapping test is timed in the app to 30 seconds per trial, there were three trials for each of the test conditions (TT and TTMS). Whilst the app times each test to 30 sec. the setting for each trial is done manually in the app, this meant that the data collection time varied slightly for each participant. The mean GSR values were therefore calculated for the participants during each of the test conditions (TT) and (TTMS).

5.9 Data analysis

The considerations for the statistical analysis (parametric and non-parametric) of the data collected in relation to research aims are tabulated in table 10 below

Table 10 Considerations for the statistical analysis

PURPOSE	QUESTION	STATISTICAL ANALYSIS		VARIABLES		FEATURES THAT ARE ESSENTIAL
		PARAMETRIC	NON-PARAMETRIC	INDEPENDANT	DEPENDANT	
Comparing groups A & B	Is there a difference between the Body Awareness (BA) indicators for dancers with (JH+) and without (JH-) joint hypermobility? Do dancers with JH have lower proprioception that dancers without JH? Do dancers with JH have poorer balance than dancers without JH? Do dancers with JH have poorer spatio-visual awareness than dancers without JH? Do dancers with JH have higher interoception than dancers without JH? Do dancers with JH have higher anxiety during a TT and TT with mental stressors than dancers without JH?	Paired t test	Wilcox Signed rank test	(category) Group A (JH-)& and Group B (JH-)	**BA indicators** *Proprioception* 1) Balance time 2) CBTT score 3) Interoception 4) MAIA (9 scores) *Anxiety* 5) TT & TTMS 6) GSR-TT & GSR-TTMS	
Comparing levels 1 & 2	Is there a difference between the intermediate and advanced dancers?	Paired t test	Wilcox Signed rank test	Int. level 1 and Adv. level 2	**BA indicators** *Proprioception* 7) Balance time 8) CBTT score 9) Interoception 10) MAIA (9 scores) *Anxiety* 11) TT & TTMS GSR-TT & GSR-TTMS	
Group A	Is there a relationship between proprioception (CBTT) and Interoception (MAIA scores)? Do dancers (JH-) with good spatio-visual memory have good interocetive awareness?	Pearson product-moment correlation coefficient (r)	Spearman's Rank order Correlation (rho)	CBTT and MAIA scores		One group 2 scores
Group B	Is there a relationship between proprioception (CBTT) and Interoception (MAIA scores)? Do dancers (GJH+) with good spatio-visual memory have good interocetive awareness?	Pearson product-moment correlation coefficient (r)	Spearman's Rank order Correlation (rho)	CBTT and MAIA scores		One group 2 scores
Group A	Is there a relationship between the anxiety levels (GSR) for the TT and TTMS for JH- dancers	Paired t test Two-way between groups	Wilcox Signed rank test	Two tests- TT and TTMS	GSR scores	Same group two different occasions
Group B	Is there a relationship between the anxiety levels (GSR) for the TT and TTMS for GJH+ dancers	Paired t test Two way between groups	Wilcox Signed rank test	Two tests- TT and TTMS	GSR scores	Same group two different occasions

5.10 Results

5.10.1 Peak Height Velocity (PHV)

Within the 32 participants in this study, the maturity offset (number of years to PHV) ranged from -2 to +3.1, five participants had not reached PHV, all of which were male participants. A diagrammatic representation of the maturation status for all participants is shown in figure 5 below. The 27 participants who had a maturity offset above 0 (indicated by the red cut off line) were used in the further data collection for the study reported here, the mean age was 14.6 yrs (SD=1.5 yrs; range 12-17 yrs), two were male and 25 were female.

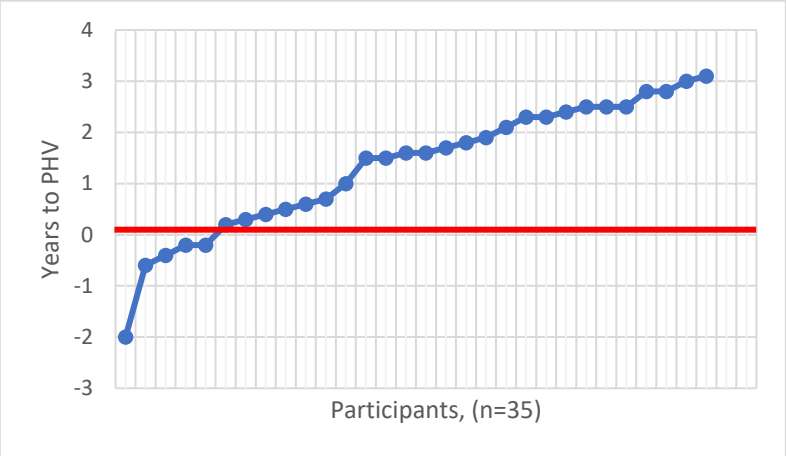

Figure 5 Maturation status of participants

5.10.2 Generalised Joint Hypermobility (GJH)

Classification of dancers, without (GJH-) and with hypermobility (GJH+) were made using the Beighton score cut off of ≥5. Out of the participant group of 27 dancers, there were nine dancers who were GJH- and eighteen who were GJH+. The GJH- dancers were allocated into group A and the GJH+ dancers into group B.

5.10.3 Balance

The balance time for the eyes open condition exceeded the cut off of 180 seconds for all the dancers and this test was therefore discarded.

For the eyes closed condition, the balance times for all tests (right and left leg) for the whole group were tested for normal distribution and were normally distributed. The mean balance time was 17.24 sec (SD=10.44 sec; Range=2.8-39 sec).

Following this an independent samples *t* test was carried out to compare the balance times between the right and left legs in the whole group. There was no significant difference in balance times for the right leg (M= 17.74 sec; SD=11.22 sec) and the left leg (M= 16.74 sec; SD= 9.76 sec); t (54) = .379, p=.706, two tailed. The magnitude of differences in the means (mean difference = 95% CI -4.26 - 6.25). The effect size was small (Eta squared=.005). Therefore the data from the balance tests for right and left legs were combined as an average score for each participant in the eyes closed conditions.

The balance times for group A (GJH-) were then compared to group B (GJH+).

An independent samples *t* test was carried out to compare the balance times between group A and group B. There was no significant difference in balance times for group A (JH-) (M= 18.44 sec; SD=7.28 sec) and group B (M= 14.36 sec; SD=7.42 sec); t (27) = 1.35, p= 1.88, two tailed. The magnitude of differences in the means (mean difference = 95% CI:-2.12 - 10.27). The effect size was small (Eta squared= .004).

In order to test if the level of dance training was related to the balance times, balance times for the whole group (group A and group B combined) were then analysed according to their level of training (Intermediate and Advanced).

An independent samples *t* test was carried out to compare the balance times between level 1 and level 2 dancers. There was no significant difference in balance times for level 1 (M= 8.12 sec; SD=2.25 sec) and level 2 (M= 7.09 sec; SD=1.89 sec) dancers; t (13) = -.515, p= .611 two tailed. The magnitude of differences in the means (mean difference =-1.51, 95% CI: -7.54 - 4.52). The effect size was small (Eta squared= .01).

Each group was then analysed to see if there was a relationship between balance times and levels. Group A (GJH-); there were nine dancers in total in group A, 4 were at level 1 and 5 at level 2. An independent samples *t* test was carried out to compare the balance times between level 1 and level 2 dancers in group A. There was no significant difference in balance times for level 1 (M= 16.35 sec; SD=9.07 sec) and level 2 (M=20.11 sec; SD=6.05 sec) dancers; t (9) = -.746, p=.48, two tailed. The magnitude of differences in the means (mean difference = -3.75, 95% CI: -15.64 to 8.14). The effect size was large (Eta squared=.142)

Group B (GJH+); there were 18 dancers in total in group A, 13 were at level 1 and 5 at level 2. An independent samples *t* test was carried out to compare the balance times between level 1 and level 2 dancers in group B. There was no significant difference in balance times for level 1 (M=14.54 sec; SD=7.12 sec) and level 2 (M=14.54 sec; SD=7.12 sec) dancers; t (18) = .154, p= .879 two tailed. The magnitude of differences in the means (mean difference = .62, 95% CI: -7.90 to 9.14). The effect size with a moderate effect (Eta squared=.06).

Overall, no significant differences between balance times and levels (intermediate and advanced) or balance times and group A (GJH) and group B (GJH+) were found.

5.10.4 Spatial visual memory: Corsi block tapping test (CBTT)

The results from the Corsi block tapping tests were scored by the maximum number achieved in the series of blocks. The results for the Corsi Block Tapping Test (CBTT) entered into the data analysis software and tested for normality across all dancers (n=27). The mean score for all participants was 8.00 with a range of 5 to 11 and SD=1.71. The data was normally distributed.

An independent t test was conducted to compare the CBTT scores for group A (GJH-) and group B (GJH+). There was a significant difference between the CBTT scores for Group A (GJH-) (M=11.19, SD=1.80) and Group B (GJH+) (M=8.32, SD=1.80, t (27) =3.90, p=0.001, two tailed. The magnitude of the difference in the means (mean difference= 2.87, 95% CI: 1.35-4.38) was very large (eta squared = 3.6).

The CBTT scores were also analysed to see if there were any differences in the scores according to the level of training (level 1 and level 20 in the whole group (group A and group B). An independent t test was conducted to compare the CBTT scores for the level 1 dancers and level 2. There was no significant difference between the CBTT scores for level 1 (M= 9.13, SD=1.88) and level 2 dancers (M= 9.41, SD=2.60, t (27) =-.315, p=.756, two tailed. The magnitude of the difference in the means (mean difference= -.28, 95% CI: -2.09 -1.5) was very small (eta squared =.003).

Each group's CBTT scores were then compared across training levels. Group A; an independent t test was conducted to compare the CBTT scores for levels 1 and 2 dancers in group A (GJH-). There was no significant difference between the CBTT scores for level 1 (Group A, GJH-) (M= 10.50, SD=1.29) and level 2 (Group A, GJH+) (M=11.73, SD=2.10, t (9) =-1.02, p=.341, two tailed. The magnitude of the difference in the means (mean difference= -1.23, 95% CI: -4.08-1.62) was small (eta squared = .01).

Group B; an independent t test was conducted to compare the CBTT scores for levels 1 and 2 dancers in group B (GJH+). There was no significant difference between the CBTT scores for level 1 (group B, GJH-) (M= 10.50, SD=1.29) and level 2 (group B, GJH+) (M= 8.38, SD=1.80, t (18) =.258, p=.800, two tailed. The magnitude of the difference in the means (mean difference= .251, 95% CI: -1.82 - 2.31) was very small (eta squared = .004).

These results suggest that there was a significant difference in the CBTT scores between group A (GJH-) and group B (GJH+) where the scores for group B (GJH-) were significantly lower than group A. This suggests that the Spatio-visual memory in dancers with joint hypermobility (GJH+) is significantly worse than the dancers who did not screen positively for joint hypermobility (GJH-). There were no significant differences for the overall CBTT scores between levels of expertise.

5.10.5 Interoception (MAIA)

The data from the MAIA questionnaire provided a single score out of five for each of the eight components (noticing, not distracting, not worrying, attention regulation, emotional regulation, self-regulation, body listening, trusting), the Mean, SD and Range for these scores for the whole group are presented in table 11 below.

Table 11. MAIA scores for groups A and B

MAIA components	All (n=27) Mean (SD)	Group A (n=9) Mean (SD)		Group B (n=18) Mean (SD)
ATTENTION REGULATION	3.42(.511)	3.65 (.460)		3.31 (.508)
BODY LISTENING	3.04 (1.053)	2.18 (.973)		3.45 (.836)
EMOTIONAL AWARENESS	3.41(1.160)	2.64 (1.232)		3.77 (.974)
NOT DISTRACTING	2.14 (1.016)	2.92 (1.053)		1.77 (.778)
NOT WORRYING	2.28 (.763)	3.67 (.612)		4.25 (.527)
NOTICING	4.06 (.611)	2.92 (.641)		1.97 (.623)
SELF REGULATION	3.13 (.780)	3.53 (.631)		2.93 (.786)
TRUST	3.14 (1.325)	4.45 (.408)		2.51 (1.133)

The data were not normally distributed and therefore non-parametric tests were used in the analysis.

i) MAIA scores between groups, (group A and group B)

a) Attention Regulation

Attention regulation scores were analysed between groups A and B. The Mann-Whitney-U test revealed a significant difference in the attention regulation score between group A (*MD* =3.5, *N*=9) and group B (*MD =4.5, N=18), U*=124, *Z=2.24, p=.027, r*=.43 with a medium to large effect using Cohen (1988).

The attention regulation scores were also analysed for differences across levels. Whole group; The Mann-Whitney-U test revealed no significant difference in the attention regulation score between the level 1 dancers (*MD* =3.57, *N*=13) and level 2 dancers (*MD* =3.42*, N*=14*), U*=92, *Z*=.49*, p*=.961*, r*=.09 with a small effect using Cohen (1988). The separate group (A and B) scores were then analysed between levels. Group A (GJH-); The Mann-Whitney-U test revealed a significant difference in the attention regulation score between the level 1 dancers (*MD* =1.33, *N*=4) and level 2 dancers (*MD* =2.33*, N=5), U=* 16.5, *Z=1.62, p=.111, r*= .54 with a large effect using Cohen (1988). Group B (GJH+); The Mann-Whitney-U test revealed a significant difference in the attention regulation score between the level 1 dancers (*MD* =3.75, *N*=13) and level 2 dancers (*MD* =4.00*, N*=6*),* *U*=50.5, *Z*=1.09*, p*=.275*, r*= .257 with a small to medium effect using Cohen (1988).

These results suggest that there is a difference in attention regulation between group A (JH-) and group B (JH+), group B had poorer attention regulation than group A (JH+). There were also significant differences in the attention regulation across levels of training suggesting that dance training improves attention regulation.

b) Body Listening

Body listening scores were analysed between groups A and B. The Mann-Whitney-U test revealed a significant difference in the body listening score between group A (*MD =3.5*, *N*=9) and group B (*MD =4.5, N=18), U=129*, *Z=2.52, p=.012, r*= .48 with a large effect using Cohen (1988).

The body listening scores were also analysed for differences across levels. Whole group; The Mann-Whitney-U test revealed no significant difference in the body listening score between the level 1 dancers (*MD* = 3.75, *N*=13) and level 2 dancers overall (*MD =2.49, N=14), U=76*, *Z=-.74, p=.457, r*= .142 with a small effect using Cohen (1988).

Group A (GJH-); The Mann-Whitney-U test revealed a significant difference in the body listening score between the level 1 dancers (*MD* =1.33, *N*=4) and level 2 dancers (*MD* =2.33*, N=5), U=* 16.5, *Z=1.62, p=.111, r*= .54 with a large effect using Cohen (1988). Group B (JH+); The Mann-Whitney-U test revealed a significant difference in the body listening score between the level 1 dancers (*MD* =3.75, *N*=13) and level 2 dancers (*MD* =4.00*, N=6), U=*50.5, *Z=*1.09*, p=*.275*, r*= .257 with a small to medium effect using Cohen (1988). The analysis suggests that there was a significant difference for the Body listening scores between groups A and B with group B (GJH+) having significantly higher scores than group B (GJH). There were no significant differences between levels of training for the whole group or groups A and B.

c) Emotional Awareness

The emotional awareness data was tested for differences between groups. The Mann-Whitney-U test revealed a significant difference in the emotional awareness score between group A (*MD =2.2*, *N*=9) and group B (*MD =3.9, N=18), U=124*, *Z=2.22, p=.026, r*= .42 with a medium to large effect using Cohen (1988).

The emotional awareness scores were also analysed for differences across levels. Whole group; The Mann-Whitney-U test revealed no significant difference in the emotional awareness score between the level 1 dancers (*MD* =3.6, *N*=13) and level 2 dancers (*MD* =3.8*, N=14), U*=90, *Z*=.049*, p*=.961*, r*= .009 with a very small effect using Cohen (1988). As no significant differences were found between levels for the whole group, no further analysis was carried out for separate groups between levels. The analysis suggests that there was a significant difference in emotional awareness between group A (JH-) and group B (JH+). The dancers with joint hypermobility had a greater emotional awareness. There was no difference in emotional awareness across the levels of training.

d) Not distracting

The data for not distracting was analysed for differences between group A (GJH-) and group B (GJH+). The Mann-Whitney-U test revealed a significant difference in the not distracting score between group A (*MD =3.0*, *N*=9) and group B (*MD =1.66, N=18), U=30.50*, *Z=-2.62, p=.009, r*=.5 with a large effect using Cohen (1988).

The not distracting data were also analysed for differences across levels. Whole group; The Mann-Whitney-U test revealed no significant difference in the not distracting score between the level 1 dancers (*MD* =2.33, *N*=13) and level 2 dancers (*MD* =1.66*, N=14), U*=84.5, *Z*=.319*, p*=.750*, r*=.006 with a very small effect using Cohen (1988). The analysis suggests that there is a significant difference in the not distracting scores for group A (GJH-) and group B (GJH+) with group B scoring significantly lower than group A. There were no differences in the data across levels of training suggesting that there was no relationship between the training level of the dancers and how much they were distracted.

e) Not Worrying

The data for not worrying was analysed for differences between group A (GJH-) and group B (GJH+). The Mann-Whitney-U test revealed a significant difference in the not-worrying score between group A (*MD =3.0*, *N*=9) and group B (*MD =2.0, N=18), U*=23.00, *Z=-3.02, p*=.003*, r*= .58 with a large effect using Cohen (1988)

The not worrying scores were also analysed for differences across levels. Whole group; The Mann-Whitney-U test revealed no significant difference in the not worrying score between the level 1 dancers (*MD* =2.33, *N*=13) and level 2 dancers (*MD* =2.16*, N=14), U*=90, *Z*=-.049*, p*=.961*, r*=.009 with a very small effect using Cohen (1988). There was a significant difference in the not worrying scores between group A (GJH-) and group B (GJH+), group B scored lower, meaning that they worried more than group A. There were no differences in the scores between training levels suggesting that the training level was not related to how much the dancers worried.

f) Noticing

The data for noticing was analysed for differences between group A (GJH-) and group B (GJH+). A Mann-Whitney U Test was used to test for differences between groups for the noticing score. The Mann-Whitney-U test revealed a significant difference in the noticing score between group A (*MD =3.5*, *N*=9) and group B (*MD =4.5, N=18), U*=124, *Z*=2.24*, p*=.025*, r*= .43 with a medium to large effect using Cohen (1988)

The noticing scores were also analysed for differences across levels.
Whole group; The Mann-Whitney-U test revealed no significant difference in the noticing score between the level 1 dancers (*MD* =4.25 *N*=13) and level 2 dancers (*MD* =3.75*, N=14), U*=76, *Z*=-.738*, p*=.460*, r*= .13 with a small effect using Cohen (1988). There was a significant difference in the noticing scores between group A (GJH-) and group B (GJH+),

group B scored significantly higher, meaning that they noticed more than group A. There were no differences in the scores between training levels suggesting that the training level was not related to how much the dancers noticed.

g) Self-Regulation

The self-regulation data was analysed for differences between group A (GJH-) and group B (GJH+). *MD* =3.25, *N*=9) and group B (*MD =2.27, N=18), U=50*, *Z=-1.606, p*=.108, *r*= .30. Whole group; The Mann-Whitney-U test revealed no significant difference in the self-regulation score between the level 1 dancers (*MD* =3.25, *N*=13) and level 2 dancers (*MD* =3.12*, N=14), U*=101.5, *Z*=.513*, p*=.608*, r*= .09 with a small effect using Cohen (1988). The analysis did not find any significant differences in self-regulation between groups or levels of training.

h) Trust

The trust data was analysed for differences between group A (GJH-) and group B (GJH+). A Mann-Whitney U Test was used to test for differences between groups for the trust score. The Mann-Whitney-U test revealed a significant difference in the trust score between group A (*MD* =4.3 *N*=9) and group B (*MD* =2.0*, N=18), U=13.5*, *Z=-3.50,* p=<001*, r*= .67 with a large effect using Cohen (1988). *MD* =4.0, *N*=13) and level 2 dancers (*MD* =3.33*, N=14), U*=76.5, *Z*=-.710*, p*=.478*, r*= .14 with a small effect using Cohen (1988). There was a significant difference in the trusting scores between group A (JH-) and group B (JH+), group B scored significantly lower, meaning that they trusted less than group A. There were no differences in the scores between training levels suggesting that the training level was not related to how much the dancers trusted.

5.10.6 Anxiety:

i) Tapping Test (TT)

First the TT and TTMS results were analysed for the whole group of participants (n=27). Then they were analysed to see if there were any differences between conditions (TT & TTMS) and between groups (group A, JH- & group B, JH+). The results for the tapping test (TT) from all participants (Group A and B combined, n=27) were tested for normality. The mean score for the TT was 87.16 (range of 49 to 155 taps, SD=25.5) and the data was normally distributed.

For the tapping test with mental stimulation (TTMS) the mean score was 85.18 (range of 43 to 141 taps, SD=23.8) and the data was also normally distributed. A paired sample t test was conducted to see if there was a difference between the scores for the TT and the TTMS tests for the whole group, there was no significant difference between the TT scores and the TTMS scores.

Group A and B's tapping scores were then analysed separately using a paired sample t test to see if there was a difference between the mean scores for the TT and the TTMS conditions. p= .347 (>.001). The mean decrease in taps between the TT and TTMS conditions for group A was 4.33 with a 95% confidence interval ranging from -5.67 to 14.34. .820 (>.001). The mean decrease in taps for between the TT and TTMS conditions group B was .808 with a 95% confidence interval ranging from -6.54 to 8.15. There were no significant differences between the scores for the TT and the TTMS conditions for either group A (JH-) or group B (JH+)

The tapping test scores (TT and TTMS) for groups A and B were then analysed using independent samples t tests to determine differences between the two groups Group A (JH-) and Group B (JH+). t (25) =-.433, p= .669 (two-tailed). There were also no significant

differences for the TTMS scores between groups A, (M=79.78, SD=21.04) and Group B (M= 87.89, SD=25.25); t (25) = -.828, p=.415 (two-tailed).

ii) Galvanic skin response (GSR)

The mean GSR values for the whole group (group A and B) were tested for normal distribution in both conditions. A paired sample t test for non-parametric data (Wilcoxon Signed Rank Test) was conducted to see if there was a difference in GSR values between the TT and the TTMS tests for the whole group. The Wilcoxon Signed Rank Tests revealed a statistically significant increase in the GSR values for the Tapping test with Mental stressor (TTMS), *z*=-4,493, p=<001, with a large effect size (*r*=.865) using Cohen (1988) criteria. The median value of the GSR increased from the TT (*Md*=8.01) to TTMS (*Md*=13.20). Group A; the Wilcoxon Signed Rank Tests revealed a statistically significant increase in the GSR values during the TTMS, z=-2,43, p=.015, with a large effect (*r*=.58) using Cohen (1988) criteria. The median GSR value increased from the TT (*Md*=4.97) to TTMS (*Md*=5.89). Group B; the Wilcoxon Signed Rank Tests revealed a statistically significant increase in the GSR values during the TTMS, z=-3.72, p=<001, with a large effect (*r*=.62) using Cohen (1988) criteria. The median GSR value increased from the TT (*Md*=10.87) to TTMS (*Md*=17.68).

Between groups; A one way between groups analysis of variance (Mann-Whitney U Test) was conducted to see if there was a difference for the GSR values between group A (GJH-) and group B (GJH+) during the TT test. The Mann-Whitney U Test revealed a significantly lower GSR values for group A (GJH-), (*Md* =4.98, n=9) than group B (GJH+), (*Md* = 10.87, n=18), U=157, *z*=3.91, p = <001, with a large effect size *r*=.75 using Cohen (1988) criteria. A one way between groups analysis of variance (Mann-Whitney U Test) was also conducted to see if there was a difference for the GSR values between group A and

group B during the TTMS test. The Mann-Whitney U Test revealed a significantly lower GSR values for group A (GJH-), (*Md* =5.89, n=9) than group B (GJH+), (*Md* = 117.68, n=18), U=162, *z*=4.17, p = <001, with a large effect size *r*=.80 using Cohen (1988) criteria.

5.10.7 Between group comparisons of body awareness (BA)

The data was analysed in order to determine if there was a relationship between groups and the body awareness indicators (proprioception and interoception)

Relationship between variables were investigated using the Spearman's rho correlation coefficient as preliminary analysis performed on the MAIA scores did not ensure violation of the assumption of normality, linearity and homoscedasticity.

Relationships between

i) the spatio-visual memory (as measured by Corsi Bloct tapping test, CBTT) and interoceptive awareness (as measured by the MAIA questionnaire) and joint hypermobility (as categorised by the Beighton scale)

Group A (GJH-); the results demonstrated no correlation between the CBTT and interoceptive awareness (MAIA scores).

Group B (GJH+); the results demonstrated a very strong correlation between two variables, Spatio-visual memory and body listening rho=.505, n=18, p=.032, with good spatio-visual memory associated with body listening. No other variables were correlated.

ii) Balance (as measures in time in seconds) and interoceptive awareness (as measured by the MAIA questionnaire) and joint hypermobility (as categorised by the Beighton scale).

Group A (GJH-); the results demonstrated no correlation between balance and interoceptive awareness (MAIA scores). Group B (GJH+); the results demonstrated no correlation between balance and interoceptive awareness (MAIA scores).

iii) Anxiety (as measured by GSR values) and interoceptive awareness (as measured by the MAIA questionnaire) and joint hypermobility (as categorised by the Beighton scale).

Group A (GJH-); the results demonstrated no correlation between anxiety (GSR measures) and interoceptive awareness (MAIA scores).

Group B (GJH+); the results demonstrated a very strong negative correlation between two variables, GSR (tapping test with mental stressor TTMS) and emotional awareness rho= -.514, n=18, p=.029, with anxiety negatively associating emotional awareness, meaning that increased emotional awareness related to decreased anxiety. No other MAIA variables correlated with the GSR values.

5.11 Discussion

In the present study the prevalence of joint hypermobility within the participant group was 66%, and this is typical of an elite dancer group (C. Chan et al., 2018; Ruemper & Watkins, 2012; Sanches, Oliveira, Osorio, et al., 2015). There were seven students (5%) of the participant group who had yet to reach PHV which again is representative if not slightly lower than that found in groups of aesthetic athletes (Cumming et al., 2012; Malina et al., 2015). These figures therefore suggest that the participant group within this study typically represented dancers within elite training to become professional dancers.

The main research question for the study reported here investigated if there was a relationship between joint hypermobility and body awareness in elite vocational classical ballet dancers? In order to gain an understanding of body awareness within the participant

group as a whole and also between the dancers with and without joint hypermobility, the results pertaining to each sub-question are firstly addressed and discussed.

The first sub-question proposed to discover if dancers with JH have lower proprioception than dancers without JH and arose as a result of scoping the literature on GJH whereby it is suggested poor proprioception features in children and adults as a component of GJH (Ghibellini et al., 2015a; Scheper et al., 2013). Tests were devised to determine if dancers with GJH have poorer balance than dancers without GJH however the results were inconclusive and no significant differences in balance times were found across the GJH- and GJH+ groupings. Interestingly the eyes open condition of maintaining balance for 180 seconds, which is a standard test used in research (Springer, Marin, Cyhan, Roberts, & Gill, 2007), demonstrated that the participants all had enhanced balance skills. Not a surprising result when one considers how much time dancers spend learning to balance as this is part of many of the fundamental skills required within the examination syllabi for dance from an early age (5 yr. +) (e.g. Royal academy of Dance, Imperial Society of Teachers of Dance, British Ballet Organisation). This assumption was further established when the data was further analysed in the eyes closed condition and across levels of expertise (intermediate and advanced levels). Again no significant differences were found suggesting that the test was not sensitive enough to differentiate balance across the levels and body types (GJH- and GJH+) in the group. Finding a test for future research purposes that would be sensitive enough for dancers may therefore be a challenge and something to consider.

The second question asked if dancers with JH had poorer spatio-visual awareness than dancers without GJH? This question again arose from the literature (Carolina Baeza-Velasco, Bourdon, et al., 2017b) and also anecdotal observations that are recorded in the investigator's researchers memo (appendix 4). The Corsi Block test was applied as it was non-dance specific and undertaken at a desk using a computer app. The results provided evidence to

suggest that there were significant differences between the GJH- and GJH+ groups (p=0.001, two tailed). The magnitude of the difference in the means was very large (eta squared = 3.6) suggesting that spatio-visual memory in dancers with GJH is significantly lower than dancers who do not have GJH. As a key skill within learning, retaining and performing movement sequences, diminished spati-visual memory could then have a detrimental effect on a dancer's development and career.

As a major component of body awareness it was also important to discover if dancers with JH have more intense interoception than dancers without GJH? Whilst never before explored in dancers, the literature suggests that within GJH in general an enhanced sensitivity to how the body feels (interoceptive awareness) can present as an issue for those who have GJH (Eccles et al., 2012, 2015; Mallorqui--Bague et al., 2014). Interoception was measured using eight derivatives of interceptive awareness, attention regulation, body listening, emotional regulation, distracting, worrying, noticing, self-regulation and trust. Attention regulation was found to be significantly greater in dancers with GJH with a medium to large effect (Cohen 1988). These results suggest that attention regulation could potentially be a further asset to dancers with GJH. When analysed across the skills levels across the whole group (intermediate and advanced) there were no significant differences however within each group (GJH- and GJH+) attention regulation was significantly different across the skill levels with attention regulation increasing as the skill levels increased. There was a larger effect across the skill levels in the GJH-group than the GJH+ group which also suggests that the GJH- group have poorer attention regulation at a lower level of skill however this improves with a greater effect than the GJH+ group. This provides evidence to suggest that dance training can improve attention regulation in general.

Body listening scores also showed a significant difference between groups A and B with the GJH+ dancers showing larger scores (p=.012) with a with a large effect (r=.48)

using Cohen (1988). Interestingly, in the GJH literature it is suggested that people with joint hypermobility report a greater awareness of what is happening inside their bodies which causes anxiety (Kemp, 2017; Mallorquí-Bagué, Bulbena, Pailhez, Garfinkel, & Critchley, 2016; Mallorqui--Bague et al., 2014; Sinibaldi, Ursini, & Castori, 2015). This type of body awareness may well also relate the level at which they listen to their body. As with the previous body awareness indicators, the level of body awareness also increased significantly across levels of training within the GJH- and GJH+ groups with a larger effect for the GJH- dancers. These results suggest that body listening increases with dance training. However it increased more for the GJH- group. The question that arises here is do we want dancers with GJH+ to increase their body listening as this may potentially be a source of anxiety?

When the MAIA scores for emotional awareness were analysed a significant difference was found ($p=.026$), the GJH- dancers having a much greater emotional awareness with a medium to large effect using Cohen (1988). Whilst emotional awareness is an asset to a classical dancer as it allows greater expression and communication of emotions, it is also a potential cause of anxiety. This gives potential evidence for signposting for dancers with GJH and supports the discussion in the literature reviewed in study II. Also, it could be a cause for concern as it could potentially negatively associated with anxiety for dancers with GJH.

Interestingly, the scores for not distracting were also significantly different between group A and B, ($p=.009$) with a large effect using Cohen (1988) ($r=.5$). The scores revealed that GJH+ dancers were distracted significantly more than those without GJH. This appears to contradict the attention regulation scores which were greater for the dancers with GJH, but would explain the links discussed in the literature with disorders such as ADHD (Bulbena et al., 2017; Ghibellini, Brancati, & Castori, 2015).

The indicator for not worrying was also significantly different between the groups (A and B). The GJH+ dancers scored significantly lower on this part of the MAIA questionnaire and therefore worried significantly more than those without JH. This again is supported in the general literature for GJH (Carolina Baeza-Velasco, Pailhez, Bulbena, & Baghdadli, 2015; Sinibaldi et al., 2015). However, it has never before been evidenced in young dancers and may be of potential interest to those who teach and support young dancers in the learning environment. There were no apparent differences in worrying across the two levels of dance experience suggesting that dance training does not enhance a dancer's capacity to not worry.

The scores for noticing were again significantly higher for dancers with GJH. This also potentially supports the findings for body listening, emotional awareness and not distracting as noticing would presumably relate to listening to the body, being emotionally aware and not distracted. Again these associations for dancers are paradoxical as increased noticing could be seen as important in dancer practice whilst it may also be a source of anxiety. Again this is something that would need to be kept in check within the dancers training environment as it could lead to too much attention to detail, perfectionism and even potentially obsessive behaviour. Attention to detail is again seen as an asset in dancers as are adaptive perfectionist traits ((McEwen & Young, 2011). However these can easily become maladaptive and obsessive behaviours which again are known to be linked to GJH (C. Baeza-Velasco, Gély-Nargeot, Vilarrasa, & Bravo, 2011; Bulbena et al., 2017).

Trusting was also a significant issue for the dancers with GJH. Their scores were significantly lower ($p=<001$) than the dancers without JH with a large effect using Cohen (1988) ($r= .67$). Again trusting could also potentially be related to not worrying and the scores across both of these components of interoceptive awareness do appear to be related.

What is clear from these results is that indicators for interoception and one of the indicators for proprioception (spatio-visual memory) are clearly significantly different for the dancers with GJH. Interoception being greater for the dancers with GJH whereas proprioception is diminished. These results are again supported by the general GJH literature (Keer & Grahame, 2003; Mallorquí-Bagué et al., 2016; Voermans NC, van Alfen N, Pillen S, Lammens M, Schalkwijk J, Zwarts MJ, van Rooij IA, Hamel BC, 2009). The second indicator for proprioception (balance) appears to have been influenced by the dance training that all participants received, irrelevant of training levels and this would have to be taken into consideration for further research into the relationship between interoception and proprioception in dancers.

Relationships were further investigated by exploring potential correlations between the variables for body awareness (proprioception and interoception) within both groups. Correlations were found for group B but not for group A. For the dancers with GJH it was the spatio-visual memory (CBTT) and body listening that positively correlated. This finding suggests that improvements in spatio-visual memory could potentially also be associated with improved body listening. Again, this finding is potentially important for those who teach dancers with GJH. No other variables correlated.

Whilst the mental stressor applied to the tapping test (TT) did not appear to significantly affect the motor speed of any of the dancers the GSR values did indicate that all the dancers were significantly more anxious during the tapping test with mental stressors (TTMS) with a large effect size (r=.865) using Cohen (1988) criteria. This gives evidence to suggest that the mental arithmetic tasks did actually stress the dancers. When analysed as two separate groups, the dancers with JH also had a significantly larger increase in their GSR values than those without GJH during the TTMS. This suggests that the dancers with GJH were less able to cope with the mental overload applied to the task. This again potentially

provides significant insight for those who work with dancers with JH in a teaching, learning and performance capacity. Of particular interest would be to further investigate this finding in order to understand how dancers with GJH cope in situations where new materials are being delivered over a short period of time. This is typical during preparation periods for examination, competition and performance situations.

The measures for anxiety (GSR) were also correlated with the variables for body awareness independently across both groups A and B. A negative association between anxiety and emotional awareness (p=.029) was found for the dancers in group B (GJH+) with a large effect (rho= -.514). This means that decreased emotional awareness was related to increased anxiety. Again this has implications for dancers with GJH in the learning environment for dance where increased anxiety could lead to a decrease in emotional awareness and the ability to express and communicate emotions within a performance.

5.12 Conclusion

Study II has brought further some insight in to the entanglement of body awareness and joint hypermobility. Differences in the applied objective measures were found between dancers with and without hypermobility. The results also suggest connections between interoception and proprioception, but the relationship between these within GJH is still not clear. This may be because their quantification is just too complex due to the entangled nature of body awareness and dance. For example, whilst the balance test was designed to be dance specific and appropriate for the participants' environment it was potentially not sensitive enough to detect changes nor mitigate for the training effect of dance to balance. The tests that were more successful in terms of measuring differences between participants were in fact the ones that were not dance specific, (CBTT, TT, TTMSS, MAIA). This needs further consideration for research in dance in general.

It was also unfortunate that the HRV data could not be analysed to be included in the overall analysis as this may have given further insight into interoceptive awareness and anxiety. Equipment choices for research with dancers who have a very specific body type that is potentially different to sporting populations also needs further consideration. Whilst research in dance science is advancing the wide majority of equipment is made for sport and potentially not suitable in a dance environment. This makes quantifying the components that relate to a dancers performance and development challenging.

Despite these limitations the significant and therefore conclusive findings clearly have implications that can influence a dancer's biopsychosocial development and interaction within the teaching and learning environment. In order to further investigate the experience of joint hypermobility within this environment, one further studies was designed. This explored both the dancers' and teachers' perspective and experience of joint hypermobility.

Chapter 6
Study III: *Psyche* (*Ψυχή*)[4], a retrospective view of the lived experience of GJH in the professional dance environment.

6.1 Introduction

Previous research of GJH in dance is characterised by an emphasis on the biological markers associated with attributes and detriments concerning injury that in turn influence career progressions and physical well-being (Day et al., 2011b; Foley & Bird, 2013a; Klemp et al., 1984; Mccormack et al., 2004; Moira McCormack, Bird, de Medici, Haddad, & Simmonds, 2019b; Sanches, Oliveira, Osório, et al., 2015). The research perspectives in the current literature are also largely derived from dancers' response to injury surveys, questionnaires and/or scales rather than from their first person description of their experience. In such, apart from one autobiographical text (Knight, 2013), there is a real lack of understanding and a need for a grounded theoretical context that analyses and explains the lived experience of hypermobility.

Study I investigated the pre-existing theories relating to the dance environment and physical risk of GJH. Study II explored the connections between body awareness (interoception, proprioception and anxiety). Theories pertaining to the lived experience of GJH in the dance environment from a bio-psycho-social perspective do however not exist, the study presented in this chapter therefore attempts to provide further insight in to this experience.

[4] *Psyche* (*Ψυχή*) - The most beautiful girl on earth, always admired but never really loved

1

GJH is one of the hypermobility spectrum disorders (HSD) that are genetic disorders of the connective tissue (Castori et al., 2017) and are therefore a constant part of a dancer's biological functioning as opposed to being an injury or condition that can be medically treated and cured. From an bio-ecological perspective (Bronfenbrenner & Morris, 2007) the degree to which the symptoms and health risks of GJH (as a biological factor) therefore develop, fluctuate and change over the career and lifespan development of a dancer depend to a large extent on the work and training environment that the dancers are situated in at any given time. The development of risk reduction strategies within health was first advocated by the World Health Organization's (WHO) through a bio-psycho-social model of functioning (WHO, 2001). This classification and model of functioning recognizes for example that the health risk of injury in dance is due to the interaction of causative factors over time (Liederbach, Hagins, Gamboa & Welsh, 2012; Meeuwisse, 1994). In addition and from a bio-ecological perspective, (Bronfenbrenner & Morris, 2007) therefore the degree to which the causative factors and health risks of GJH may develop, fluctuate and change over the career and lifespan of a dancer depend to a large extent on the environmental, contextual and personal factors of the dancer at any given time. These two perspectives and are brought together in the model of functioning and health risk shown in figure 6 which diagrammatically represents the entanglement of GJH (as a potential health risk) in the dance environment.

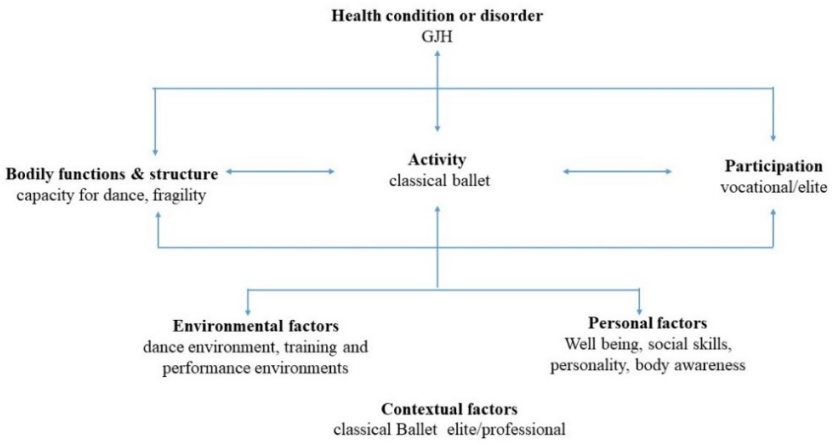

Figure 6 Model of functioning and health risk of GJH in professional dance, adapted from the ICF model (WHO, 2001)

In the study reported in this chapter, GJH is investigated (as a potential health risk) within the context of bio-directional influences through the body's function and structure (disordered connective tissue, capacity) and dance (activity) which interact at an intense elite level of functioning. Within the classification of functioning model in figure 6, the factors that are also influenced by context relative to this study are the elite dance environment (environmental) and the dancers' capacity to flourish (personal factors).

This study therefore provides a theoretically suggestive, integrated depiction of GJH in the 'dance environment'. The '*dance environment*' refers to a physical and metaphorical space and the interactions that happen in this space such as training, teaching, choreographic, rehearsal and performance and will be referred to as such herein. The depiction of GJH is based on the descriptions and experiences of both dancers with GJH and those who teach and coach them in the dance environment, this refers to teachers, choreographers, directors and coaches, all of whom are described as the 'masters' of the dance environment irrelevant of sex or gender orientation and for the purpose of this study will be referred to as *'dance masters'* herein.

6.2 The research focus

The experience of GJH within the dance environment with particular reference to classical ballet was the focus of study III. Here, the associations between the biopsychosocial experience within classical ballet are further articulated.

Training to be an elite ballet dancer is often paralleled to becoming an elite athlete and many of the physical and mental attributes are similar (Koutedakis & Jamurtas, 2004b; Moira McCormack et al., 2019b). Ballet is however at the same time an art-form that is process driven, the processes within this art-form are indeed literally inscribed on the dancers' body in both technique and artistic interpretation (Wainwright, Williams, & Turner, 2006). This is where the demands of elite performance in dance greatly differ from the majority of sport, in dance the aesthetic experience is the intentional outcome whereas this is secondary to the achievement of an objective score or result in sport (Carr, 1997)

The acquisition and refinement of technique is key to becoming an expert in both dance and sport, however within dance the ability to communicate feelings and emotions through a refined technique is paramount to being a successful performer (Carr, 1997; Maxine Sheets-Johnstone, 1999). This is largely achieved through a lengthy process of repetitive practice, analysis and critical reflection of both the physical and emotional aspects of performance over an extended period often exceeding ten years(Chua, 2014; MacNamara, Á., & Collins, 2017). In such, a dancer's development continues across the lifespan of their training and career, and whilst a dancer's physicality may diminish with age their artistry and ability to communicate emotionally continues to develop and mature. A dancer's body is highly trained to have specific skills however a dancer is are also the embodiment of particular aesthetic ideas and ideals (Aalten, 2004; Alexias & Dimitropoulou, 2011; Pickard, 2013). A dancer can expect to repeatedly practice the physical and emotional capacities required from the very beginning stages of training (often at the age of seven years) and this

practice and training remains in place throughout a professional career. Foster also believes that

> "…*the repetitive exercise and instruction that is practiced in dance alongside the images used in this process become integral to the body itself and over time this training is part of creating and reconfiguring a dancer's body"*
>
> (Foster in Desmond, 2014, p239) .

Establishing control of the body gaining and maintaining stamina, flexibility, strength and technique through physical repetition represent the principal dimensions of 'learning' in dance (Adair 1992, p. 35). Becoming an accomplished artist undoubtedly requires enhanced emotional integrity, in dance, this is developed through personal critical reflection and also working closely with teachers and masters of the art-form (Karin, 2016). The close relationship between the dancer and 'dance master' often (but not always) also allows the 'dance master' to have greater insight in to how a dancer thinks and learns. Training normally begins at a young age for dancers and the practical nature of the dance class is the foundation for all learning in dance performance. Klob (2015) advocates that gaining an understanding through experiencing is the best way to learn and dancers are undeniably very articulate in describing how their bodies feel (Jackson, 2005). The art-form indeed shapes both the physical and emotional aspects of a dancer and the brain-body connection is paramount to success (Karin, 2016).

Here we come to understand that the learning in dance is physical, emotional and cognitive and is without doubt, shaped through the body. This idea draws on the work of Gallagher (2005), his approach to *neo*-Aristotelian neurobiology and the work of Sheets-Johnstone (1990). Gallagher (2005) in his interdisciplinary work addresses the contribution of embodiment to cognition proposing a challenging and *neo*-Aristotelian approach to the notion of mind-body connections. He suggests an entanglement between the human soul (*psyche)* and the human shape (*schema*)proposing a *neo*-Aristotelian understanding of the body's form

(*morphe)* that encapsulates the ability to rationalise (associated with *psyche*) and the interdependence of this on shape. This approach suggests that the biological and neurological development of the human form (*morphe*) are indeed connected and influence the way in which we perceive the environment, how we act in it and how we experience it. Biological evidence to support this notion can be found in an elite dance environment where, young developing dancers (11-14yr.), who train for more than six hours a week, biologically develop a very specific skeletal geometry. The torsion of the femoral head for example matures in association with intense training and the use of external rotation from the hips, this development is in turn a means to increased functional range of motion and a desired means to express the (more latterly) required *dance aesthetic*. This bio-environmental development enables the dancer to achieve ideal range of motion for the external rotation from the hips (known as *en dehors* or *turnout*) using fewer compensatory strategies and consequently also reduces the risk of injury (Hamilton, Aronsen, Løken, Berg, Skotheim, Hopper, Clarke, Briffa, 2006).

Similarly, Clark (1997) through his work on perception and action coins a term '*soft assembly'* whereby he proposes that the neurological system learns to modulate physical parameters (such as stiffness or laxity of a joint) and work with both the intrinsic bodily and external environmental constraints in order to yield the desired movements or outcomes (Clark, 1997 p45). Johnston also proposes that we make sense of the spatial-temporal world that surrounds us through our physical being, suggesting that human experience and meaning depend very much on the body (Johnston, xxi, 1987). Equally, the biological bodily systems associated with homeostasis are *prenoetic* (operating below the threshold of consciousness) and are relevant to perception and the ability to perceive and cognate without distraction. For example, the body physiologically reacts to cold by shivering; this is a pre-conscious *prenoetic* reaction however, the meaning or cognition of this discomfort is perceived through

and affects the mind. Sheets-Johnston (1999) provides both empirical and phenomenological evidence to confirm the integrity between muscular activity and mental activity and the relationship between emotion and movement. Finally, this concept is also upheld within the theory of *Dynamic Systems* (Kelso, 1995) that proposes that pairs such as body and mind or nature and nurture are not mutually exclusive, but complementary. These complementary aspects and their dynamics are necessary in order to understand the complex phenomena and systems in life, mind, society and nature (Kelso, 2008).

Evidently, the reviewed theories of embodiment suggest that the body does indeed shape the mind. From this the question then arises as to how might an atypical body, one of a hypermobile dancer for example, shape the mind? In order to examine this concept further it was necessary to capture the experience of hypermobility in the dance environment. A phenomenological approach was applied as the study sought to generate theory that related to the experience of hypermobility and the recall of actions, interactions and processes of those affected by hypermobility (Moustakas, 1994, p. 4)

6.3 Methodology

GJH within the dance environment is experienced by both the dancer with GJH and those who work closely with and manage them. Phenomenology, a compound word (synthesizing the Greek words *Φαινόμενο* and *λόγος*) literally means, a rational account (*λόγος*) of the ways in which things appear (*Φαινόμενο)* (Authors own translation). Sheets-Johnston (1990, p358) discusses the importance of phenomenology within sensory –kinetic experience, in particular with reference to the experience of one's own body. She also discusses the subjective nature of experience as proposed by Nagel (1974) and the critical methodological difference between *objective phenomenology* and *hermeneutical phenomenology*. Whilst both the objective and hermeneutic propose to reveal the body in its primal physiognomy, hermeneutic phenomenology focuses on subjective experience of

individuals and groups and exposes the world as experienced by the subject through their life-world stories (Kafle, 2011, p25). Alternatively, objective phenomenology is not solely reliant on the subjective experience of individuals and groups, its goal is to describe the character of experience in a way that can be objectively understood by those not capable of such experience (Nagel, 1974, 538). An objective phenomenological perspective for example, would attempt to explain to a dancer who does not present with GJH; what it is like to be a dancer with GJH? The intention of this study was however to understand the lived experience of GJH within the dance environment with a view to inform the practice and procedures that take place in that environment and determine potential harm or risk to dancers with GJH in that environment. A hermeneutic phenomenological approach was therefore adopted (Sheets-Johnstone, 1990, p347) in study III.

6.4 Research design

The design for this research did not follow a logical or linear strategy as the experience under investigation was captured from a range of view-points. These were tracked back and forth in order to account for the different opinions and explanations. Each set of experiences was indeed unique to each situation, and the research design selected was a qualitative interactive research design (Maxwell 2011)

This provided a design for the study that was flexible, interactive and interconnected, the components of which were; goals, conceptual framework, research questions, methods and validity (Maxwell, 2013, p3-4). These are represented in figure 6 below which is adapted from Maxwell's model for qualitative research designs (2011, p5)

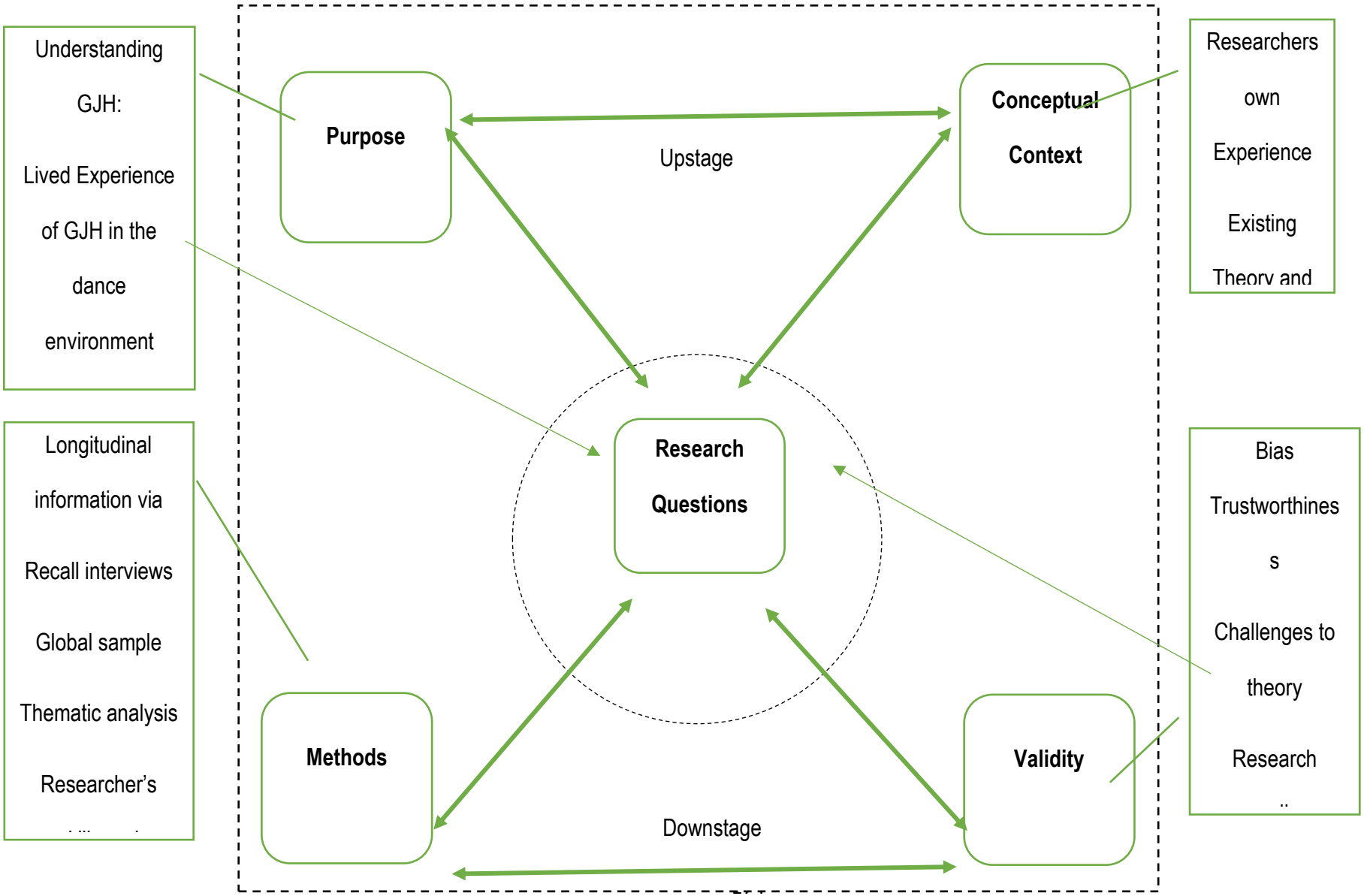

Figure 7. Mapping the contextual factors that influence the interactive research design for study III

A longitudinal method was ideally the best way to provide an in depth understanding of how GJH affects the dance environment across the lifespan development and career of a professional dancer, however longitudinal studies as the name suggests, conventionally follow particular individuals over prolonged periods, often years or decades (Caruana, Roman, Hernández-Sánchez, & Solli, 2015, p538). A longitudinal approach and design is therefore out-with the timescale of this study however this may be considered for future follow up research beyond this thesis. Retrospective studies are designed after participants have already experienced events that are of relevance, with data for potential exposures in the identified cohort being collected and examined retrospectively(Robson, 2011, p. 128). Retrospective interviews have for example been effectively used to gain insight in to the talent development of elite athletes across their careers (Côté, Ericsson, & Law, 2005) and interviews are used to generate the recall of longitudinal knowledge. This approach allows experiences to be recalled, this may be considered as a subjective and less valid approach however, when views are substantiated by asking participants to exemplify experience using the recall of specific incidents or key moments in their careers this can clearly evidence and validate the experience further (Côté et al., 2005; Wylleman, Reints, Côté, Ericsson, & Law, 2010). Similarly, the focus in this study was the experience of GJH throughout the lifespan and development of dancers' careers both from a dancer's and manager's perspective. Côte et al.'s retrospective methodology (2005) to gather the required longitudinal evidence was adopted and facilitated. The participants were questioned about their understanding and experience of hypermobility in the dance environment across their careers and they were also asked to recall and make further references to specific retrospective experiences and events during the interviews. In addition to the retrospective recall, for those participants who were still practicing, the interviews also probed current experiences of hypermobility. This approach effectively gave a cross sectional design that endeavoured to simulate a longitudinal

approach (Robson, 2011, p128). The overall aim of the study reported here was to investigate individual experiences and events relating to GJH over a career lifespan in the dance environment. This environment is itself highly interactive, dancers are required to interact with their own and others' physical being and their art-form, non-of which are static. They also interact with those who manage the environment, which includes the dance masters, in order to achieve common and desired outcomes. This is however a bi-directional process as the dance masters equally interact and negotiate the physical dialogue with the dancers, the art-form and the environment. This is undeniably and by no means a linear process, it is more an entanglement of the shifting nature of the biological, psychological and social components within the dance environment at any given time. The validity of the overall approach to the study was of course not only reliant on participants' ability to accurately recall and describe experiences but also the interviewer's ability to expose and interpret information.

Within the qualitative paradigm, the researcher is viewed as the primary instrument for data collection and analysis (Maxwell, J, 2013; Robson, 2011). Therefore the researchers own long career and background in the dance environment (as both a dancer and dance master) were indeed key to the inception and development of the overall study. In addition the researcher has across this career come to understand that she has direct experience of hypermobility. For this reason it was vital for the researcher to disclose potential sources of bias within personal histories or beliefs surrounding hypermobility. To enable this a researcher identity memo (Maxwell, J, 2013) was created and this allowed the researcher to challenge her own personal assumptions and practice around hypermobility in the dance environment. This document was developed throughout this thesis and was completed before the interviews took place. Detailed personal recollections about the researchers own past experiences, current work and intellectual knowledge that had been accumulated over a career in the dance environment and also whilst undertaking studies I and II**.** The

development of the researcher's identity memo allowed previous assumptions and practice to be grouped into personal, practical and intellectual categories. From this document (recorded in researcher's memo appendix 4) evidence regarding the importance of understanding how the body shapes the brain with regards to hypermobility could be corroborated. Personal thoughts and experiences that were not substantiated in this process were put aside for 'future deliberation' outside of the research process in this study. The memo was initially used to assist the formulation of the interactive research design for the study. It was consulted further during the development of the interview questions. This enabled the researcher to objectively draw on over thirty five years of practice in the dance environment and introduced another voice to the discussion and debate. This grounded yet interactive approach provided a framework for the research design and interviews. As the elements of the experience of GJH unravelled, their interrelationships provided a clearer understanding of the nature and meaning of the experience of hypermobility for all who operated in the dance environment (Moustakas, 1994, p. 4). This method also provided procedures for the research that were flexible yet systematic. In such, the research was not led by initial decisions and opinions but the interactive design provided a systematic and valid approach to the data collection process. Alongside the qualitative data that were generated from the retrospective recall of experiences and events in the interviews, important demographic information about each participant was also collated. This process was particularly useful for evaluating the relationship between the bio-psycho-social experience of GJH, potential risk factors and the development and manifestation of signs associated with GJH that were specific to the dance environment (Caruana, *et al.,* 2015, p538).

6.5 Interview focus and research questions

The interview focus for both groups of participants was designed to explore and gain insight in to the lived experience and understanding of GJH within the professional dance environment. The interview questions for the participants in group 1 (dancers) were designed to gain insight in to a professional dancers' own understanding and perception of GJH, the research questions pertinent to this group were:

1. Do dancers understand hypermobility?
    i. Do dancers recognise hypermobility in themselves (and how)?
    ii. Does hypermobility affected them (and how)?
    iii. How do dancers manage their hypermobility?
    iv. Do dancers perceive hypermobility as an asset or a liability?

The interview questions for the participants in group 2 (dance masters) were developed to explore and gain insight in to a dance masters' perceptions of hypermobility in the dancers that they managed, the research questions for this group were:

1. How do dance masters understand hypermobility?
    i. And what level is that understanding?
    ii. What are their beliefs about hypermobility?

6.5.1 The development of the interview questions

Study III builds on the findings for studies I and II however as the participants in these studies were not yet professional dancers the development of the semi-structured questions were primarily grounded in information assembled from a scoping of the academic and *grey* literature which is reviewed in chapter 2. The overall literature on hypermobility is at this time increasing, however it is clearly evident that the main body of literature still

relates to the biological markers, psychological traits and potential consequences associated with hypermobility in general. To a much lesser extent some of the social implications of hypermobility are discussed indirectly and documented in narrative or autobiographical literature (Clark & Knight, 2017; Knight, 2013, 2015b). Overall, the phenomenon and 'lived experience' of hypermobility is not widely documented and is limited to reports of patients with hypermobility in a clinical setting and not at all in a dance environment (Murray, Yashar, Uhlmann, Clauw, & Petty, 2013). Some experiences of hypermobility are documented in '*grey literature*' such as personal or informative health blogs and web sites about hypermobility that do not necessarily refer to dance performance. The autobiographic text by Knight (2011) provides a detailed personal account of living with hypermobility whilst also referring to dance, this text brings together some of the grey literature from blogs and the more reliable internet sources. Knight's hypermobile experience is evidently insightful as she is herself a Bowen practitioner it is however filtered inevitably through her own personal experience of dance and this is not at the professional level under scrutiny in this study. Seeking to emphasise the lived experience of hypermobility and dance, the Knight (2011) text was however included as a source of reference for the development of the interview questions. Additionally, the researcher's own experience of hypermobility contributed to the development of the interview questions as this inevitable and distinct 'component of practice' played a role in the overall conceptualisation of this research. The researcher's own practice had indeed drawn her to the focus of this study and revealed evidence, if only anecdotal, that was collated through longitudinal observation (›40 yr.) and scrutiny of movement through performing, teaching, analysing, managing and working with hypermobile dancers in both vocational and professional dance environments. Personally and frequently encountered in others, hypermobility was indeed part of the researcher's daily practice, rather than being dismissed this experiential knowledge is capitalised on in this

study (Grey, 2017, p49). As a researcher, drawing on and reflecting on one's own practice and experience within the setting or field of investigation can bring credibility and substantially inform the research (Maxwell, 2005, p46). However, a "*quality of awareness*" is required when reflecting on personal experience for research purposes (Reason, 1988. P.12). The *researcher's identity memo* was therefore conscientiously considered alongside the relative literature (peer reviewed and *grey*) throughout the development of the interview questions for the participants in both groups 1 and 2. The questions were developed so that they were clear and unambiguous, did not contain prejudicial language, jargon, not leading or hypothetical, and did not require the participants to recall knowledge that did not have (Grey, 2014, p395). Possible prompts or sub questions were also anticipated. The interview questions for all participants included both exploratory (open ended) and confirmatory (closed) questions. The open ended questions were designed to gather information about the participants professional experience, whereas the confirmatory questions related to the descriptive information and facts gathered, for example, personal details e.g. how many years practice and what type of practice did the participants have including information about their current practice. For group 1 it was also important to gather information about symptoms and features that the dancers experienced that provided additional information alongside the criteria for assessing hypermobility (Malfait et al., 2017).

The interview questions for groups 1 and 2 were developed in parallel as the overall aim of these interviews was to discover more about the experience and understanding of hypermobility from the inside (dancers' perspective) and also as externally observed (dance master perspective). It was also important to determine potential common themes that might triangulate information shared with the researcher by both groups. The interviews commenced with questions that would enable the researcher to gather confirmatory data from the participants, this included descriptive data such as the number of years that the

participants had practiced as a dancer and/or dance master and in which environments they had trained and performed. Exploratory interview questions were also used to gather both ontological and epistemological information (Saldana, 2013). For example, some questions were designed to capture the opinions and the reality of hypermobility in the teaching and learning environment for dance (ontological) and others were designed to capture the participants' knowledge and understanding of the phenomenon of hypermobility in dance environment (epistemological).

Understanding is described as an abstract process of the mind and brain, understanding exists in varying degrees and also in different modes (Kelp, 2015). Kelp (2015) outlines levels of understanding and articulates these in relation to knowledge. He proposes an account of 'maximal understanding' of a given phenomenon in terms of fully inclusive and maximally well-connected knowledge of the phenomena. He pertains that with 'maximal understanding' comes an ability to explain the phenomena while he also presents 'degrees of understanding' in terms of approximations to knowledge, i.e. the lesser the degree of understanding the lesser the aptitude to explain the phenomena. In addition to these explanations Kelp further proposes 'outright understanding' whereby the knowledge can also be conceptualised and applied within other contexts (2015, p3813). Whether knowledge and what type of knowledge is required in order to understand is undecided and debated by Polanyi (1966), however what is clear is that knowledge comes in many shapes and ways. It can be factual or explicit, also described as declarative or the "know- that" of facts and theories (Boshoff, 2014). In terms of the dancers' and dance masters' knowledge this would relate to the musculoskeletal and psychological traits associated with hypermobility and also the knowledge of the dance style, steps and choreography that are applied to the dancer's body. Practical or procedural knowledge on the other hand is implicit, experiential knowledge and includes tacit abilities such as skills, and understanding ; this is the "know-

how" that is applied to the factual knowledge (Boshoff, 2014) . For the dancers and dance masters the "know-how" refers to the knowledge and understanding of both the art form (dance) and the dancers' body and the dance environment that facilitates the transformation of factual explicit knowledge about dance and dancers to create dance-artists and art. The "know-that" and "know-how" therefore relate to associated ways (explicit, implicit and tacit) and degrees of understanding which are involved in understanding (Boshoff, 2014). In order to reveal 'how the participants understood hypermobility and their beliefs about this condition' it was therefore important to gather information through interview that related to ways of understanding hypermobility within dance.

Determining the participants' explicit knowledge and understanding of hypermobility was relatively straight forward and direct questions were used, for example

Group 1 "…*have you been diagnosed as hypermobile and if so, do you know what this means?"*

Group 2 *"…in your role as a Ballet Master, Director or Choreographer, have you worked with hypermobile dancers?"*

This question was then further prompted *by "…and how do you recognise hypermobility?"* in order to get a clearer understanding of what the dancers and dance masters understood to be hypermobility.

Further questions relating to the explicit knowledge and understanding of hypermobility were also developed, for example

Group 2 *"…have you ever had any concerns about the physical health of a hypermobile dancer?"*

Determining implicit 'tacit knowledge and understanding' was more complex, indeed tacit knowledge is described as "to *know more than we can tell"* (Polanyi, 1966, p18), it is less tangible than knowledge that is explicit and easily recalled and therefore more difficult to elucidate. Indeed, tacit knowledge exists deep in memory and consciousness and stems from the interactions between an individual their environment and day to day practice (Kumagia, 2014). Kumagia discusses tacit knowledge and understanding within professional practice and describes this from a doctor-patient perspective and this is not dissimilar to the dance master-dancer relationship within the professional dance environment. Both he believes are based on common interests, Kumagia explains, "*knowledge is not generated in a value free vacuum; rather, it is created in response to specific interests or goals*" (Kumagai, 2014, p979). The interests within the dance environment for both the dance master and dancer are common and ultimately the art-form and how successfully this is presented. Frequently within the dance environment, common interests are somewhat intense (Wainwright et al., 2005) and may further intensified not only by the fact that most (if not all) dance masters have a) themselves been elite performers , but also b) the dance masters may also be hypermobile. The connection between the practice of dancers and the art-form habitually goes beyond desire, often becoming obsessive passion. This is not only represented in the art-form itself whereby protagonists 'dance themselves to death' (e.g. The Red Shoes, Les Sylphides, Swan Lake) but obsessive passion is also discussed in the literature as a mechanism and aetiology leading of injury (Markula, 2015; Angela Pickard, 2013; Wainwright et al., 2005, 2006)

Polanyi (1966) proposes that tacit knowledge and understanding consists of two interrelated parts: the "proximal" and the "distal". He suggests that we are aware of the proximal through its effect on the distal (Polanyi, 1966 p 11). Thus, our immediate attention drawn to the physiognomy, "the distal", and what this means in terms of its features. This is

the manifestation or the appearance of what we are attending to or in this instance 'the hypermobile aesthetic' or hyperextension of the limbs and bendy bodies. What this signifies and 'means' from an ontological, semantic and phenomenological perspective however is transposed through our tacit understanding of the "proximal" (Kumagai, 2014, p978; Polanyi, 1966, pp9-16). The proximal in the context of the dance master-dancer relationship in terms of the bio-psycho and social features of hypermobility would be information available to the dance master, gained through experience that is either embodied or through professional experience or both. The proximal for the dancers however would be their experience of hypermobility in their bodies. The "distal" in this context is the overall manifestation or physiognomy of hypermobility. Polanyi proposes that the proximal and distal features of tacit knowledge are distinct, he explains that *"...we may know a physiognomy without being able to specify its particulars"* (1966, p12). For this reason, when describing a dancer or event in response to the interview questions, the participants were also asked wherever possible to retrospectively recall and describe specific occurrences, people or events and to put these in to the context. This way both their proximal and distal understanding could be investigated. For example, '*Can you briefly describe in what way(s) (if any) are these dancers different from a non- hypermobile dancer and are there any specific occasions or events that you could recall and describe for me to illustrate this?'*

6.5.1.1 Piloting the interview questions

Once the interview guides had been formulated a pilot interview was undertaken with one ballet master and one dancer. This provided; not only a gauge as to how long the interviews process would be and to also pilot the use of the recording device and app for the interviews, but also insight in to the effectiveness of the questions including the consideration of leading, over-complex and judgemental questions. The pilot interviewees were asked to feedback on any of these points and also add any additional comment they felt appropriate.

As is transpired the dancer who was asked to undertake the pilot interview was in fact herself not hypermobile, she did however have some knowledge of hypermobility as she had danced as a soloist for many years in a large ballet company where hypermobility featured greatly. She had also, fairly recently transitioned to a role where she was required to teach the company dancers. Likewise the ballet master that was interviewed for the pilot study was able to give further insight into the complexity of the questions in the interview and also the language and vocabulary that would be appropriate and well received by the ballet masters. In particular the questions were adapted to exclude clinical terms where at all possible and where this was not possible, simple explanations or examples were provided. The pilot process was also effective in determining the best way to open the interviews and the overall tone, that is by being friendly but not overly so, as this distracted the focus of the interview and also the capacity of the researcher to really pay attention and listen to the information being shared and steer the interview process.

6.6 Participants

The participants were two distinct groups as the perspectives of interest in this study were those of both dancers and dance masters from within the professional environment.

a) Group 1: dancers who presented with GJH themselves and for simplicity from herein this study are named dancers and
b) Group 2: dance masters, who were individuals who managed dancers in dance environments within a teaching and learning context.

Purposive sampling methods (Robson, p149) following the inclusion criteria and using snowballing techniques with two entrance points were adopted for all participants (Malterud et al., 2016; Vasileiou et al., 2018), the criteria for selection are shown in table 12.

The two entrance points for the sample of dancers in Group 1 were; clinical practitioners who worked with dance companies and company directors or dance masters. These individuals either confirmed (clinicians) or suspected (teachers) that the dancers they had approached to be part of the study presented with GJH. Once the dancers had agreed they were introduced to the researcher electronically and they were consequently invited for interview via e mail. Similarly, group 2 the dance masters, were approached using snowballing methods for which the initial entry points were recommendations and e introductions from two professionals within the researcher's network, the sampling was again criterion based and purposeful. The criteria applied in this selection also provided assurance that the information gained from experienced dance masters would be powerful and indepth

Table 12 Inclusion criteria for participants

Group 1 Dancers with GJH	**Group 2 Dance masters**
Ten years or more experience within the professional dance environment, five years performing and currently still practicing professionally.	Ten years or more working with and managing professional dancers in a teaching and learning context.
Classical ballet as one of their fundamental techniques of training.	The capacity for this work was as teacher, choreographer, director and /or dance master
Validation of hypermobility status using the self-screening hypermobility test (Bulbena; Mallorqui-Bague; Pailhez; Rosado; Gonzalez; Blanch-Rubio & Carbonell, 2014)	

The sampling procedures and inclusion criteria provided two groups of homogenous participants who were working in dance environments globally. This was demonstrative of the characteristics of the professional dance environment today where dancers and dance masters from all over the world work together in companies that are not necessarily in their native land. This provided an international scope for the study and allowed the researcher to capture the experiences of a sample of nine seasoned elite dancers and a smaller sample of

four dance masters. The interviews that followed were semi-structured in their primary approach; retrospective recall of events was also applied in order to put the information in to context and help the participants recall as much information as possible from a lived 'dance environment' perspective.

Nine dancer interviews were undertaken that lasted between 35 and 45 min whilst four dance master interviews were carried out that were longer, 45-60 min. All interviews were conducted via skype and recorded as MP4 audio files using an *audio memo* app for Apple© *iphone* and *ipad*. On completion of the recording the audio file was automatically sent to the researcher's e mail account and downloaded and stored on her encrypted onedrive. This allowed the researcher to immediately delete the file from the device it was recorded on for security purposes. The files were all anonymised during the transcription into word documents. In order to ensure the trustworthiness and validity of this process, a copy of each transcribed interview was sent to the interviewee. Validity in this instance refers to the correctness and credibility of the transcript (Maxwell, 1996, p 87) allowing the participants to reflect on the interview and verify that their comments and the information that they had shared in the interview was correct and had not been taken out of context. This procedure was important as it helped the researcher to avoid the influence of further unconscious bias in the interpretation and transcription of the audio file.

Demographic data for the participants including Beighton scores for group 1 are reported in table 13 below.

Table 13 Demographic data for groups 1 and 2

Group 1, Dancers	**All (9)**	**Male (5)**	**Female (4)**
Age in Yr.	Mean=32.33, SD=4.99 Range=25-40	Mean=32.8 SD=5.19 Range=27-40	Mean=31.75 SD=4.66 Range=25-38
Age started dance in Yr.	Mean=7.56, SD= 3.50 Range=4-14	Mean=10 SD=3.50 Range=6-14	Mean=4.5 SD=1.19 Range=3-6
Performance experience (Yr.)	Mean=11.89, SD=3.92 Range=6-18	Mean=11.9 SD=3.91 Range=8-16	Mean=13 SD=4.84 Range=6-18
JHS SS score /10 (≥3=GJH+)	Mean=7.78, SD=1.90 Range=4-10	Mean=6.6 SD=2.04 Range=4-9	Mean=9.25 SD=0.23 Range=9-10
Group 2, Dance masters	**All (4)**	**Male (1)**	**Female (3)**
Experience as dance-master (Yr.)	Mean= 31 SD=6.48 Range=25-38	Mean=15 SD=N/A Range=N/A	Mean=17 SD=2.66 Range=15-20
Experience as a performer (Yr.)	Mean=16.5 SD=2.38 Range=15-20	Mean=20 SD=N/A Range=N/A	Mean=32.67 SD=6.80 Range=25-38

6.6.1 Group 1 (Dancers)

There were nine dancers in group 1 and the mean age was 32.3yr. (SD=4.99, Range=25-40) five were male, mean age 32.8yr. (SD=5.19, Range=27-40) and four female, mean age 31.75yr. (SD=4.66, Range=25-38). The mean age for starting dance classes for the group was 7.56yr. (SD=3.5, Range 4-14) with the female dancers starting on average 5.5 years younger than the male dancers which is consistent with common practice in dance and in particular classical ballet (Weiss, Shah, & Burchette, 2008). The dancers had a mean of 11.89 yr. (SD 3.92) professional performance experience which ranged from 6-18 years confirming that all dancers were eligible to meet the inclusion criteria of more than 5 years of professional performance experience. All dancers had a classical ballet training and eight out of the nine also had a training in contemporary dance. Seven out of the group worked and performed in the classical ballet and neo-classical dance genre, two were currently contemporary dancers and one worked in the commercial field. Interestingly four out of the group had initially started training in gymnastics at a very early age (between 4-7 yr.) these

four dancers were encouraged/selected into gymnastics because of their flexibility. One of the dancers had also been selected as a talented tennis player at an early age. All the dancers in the group were generalised joint hypermobility positive (GHS+) according to the self-screening test that has a ≥3 positive score criteria (Bulbena et al., 2014), this was also the inclusion criteria to participate in this study. The mean scores on the self-reported screen (Bulbena et al., 2014) for the whole group were 7.79 (SD=1.9, range 4-10). The female dancers however scored considerably higher, mean score= 9.25 (SD=0.23, Range 9-10) than the male dancers, mean=6.6 (SD=2.04, Range=4-9).

With regards to other characteristic symptoms associated with GJH and discussed in the literature (Castori & Colombi, 2015; Keer & Grahame, 2003; Knight, 2013), only one dancer reported to have asthma and respiratory issues however all dancers reported the characteristic head rush when returning swiftly from a horizontal to vertical plane or recovering from a forward or backward extension of the spine that is associated with hypermobility (Keer and Grahame, 2003; Knight, 2013). Eight of the dancers reported difficulties and discomfort in standing still for any length of time. Bowl pathologies (e.g. IBS, constipation etc.) were only reported by one of the dancers. Difficulties with sleep were reported by eight of the group, fatigue and chronic pain were reported by four of the group and mood swings were also an issue for four of the group. When asked about psychological issues associated with GJH (Sanches, Oliveira, Osorio, Crippa, & Martín-Santos, 2018) anxiety was prevalent for eight of the dancers in the group, panic was reported by four dancers however depression was less common and only reported as an issue by one of the dancers. Injury was a common feature reported by all of the dancers and common injuries reported by the dancers were tendon pathologies, hernias and stress fractures. The areas where injury most commonly occurred in the group were ankles, feet, knees, hips, spine, shoulders and the upper body in general.

6.6.2 Group 2 (Dance Masters)

There were four dance masters in the study, three female and one male. They had a mean performance experience of 31Yr. (SD=6.48, range 25-38) and had all reached and exceeded soloist status during their careers. They all performed nationally/internationally in acclaimed ballet companies which had 30+ dancers within them and performed masterworks regularly in their repertoire. Their mean experience as a dance master was 19.25Yr. (SD=2.89, Range=15-22) and they were all practicing as dance masters at the time of the interview.

6.7 Data analysis

Initially the transcript texts for each separate group of participants were combined and imported into a spreadsheet. The transcript verbatim was organised in to three categories which were the categories that underpinned the research framework (Bio-Psycho & Social). This process was done manually by the researcher who having undertaken the transcription was familiar with the content and context of each of the interviews. Word frequencies calculations were made allowing the data to be presented objectively according to the bio-psycho-social framework. These are presented below in figures 7 and 8 the parameters set for the generation of these word clouds were that the most frequent words were to appear also as the largest.

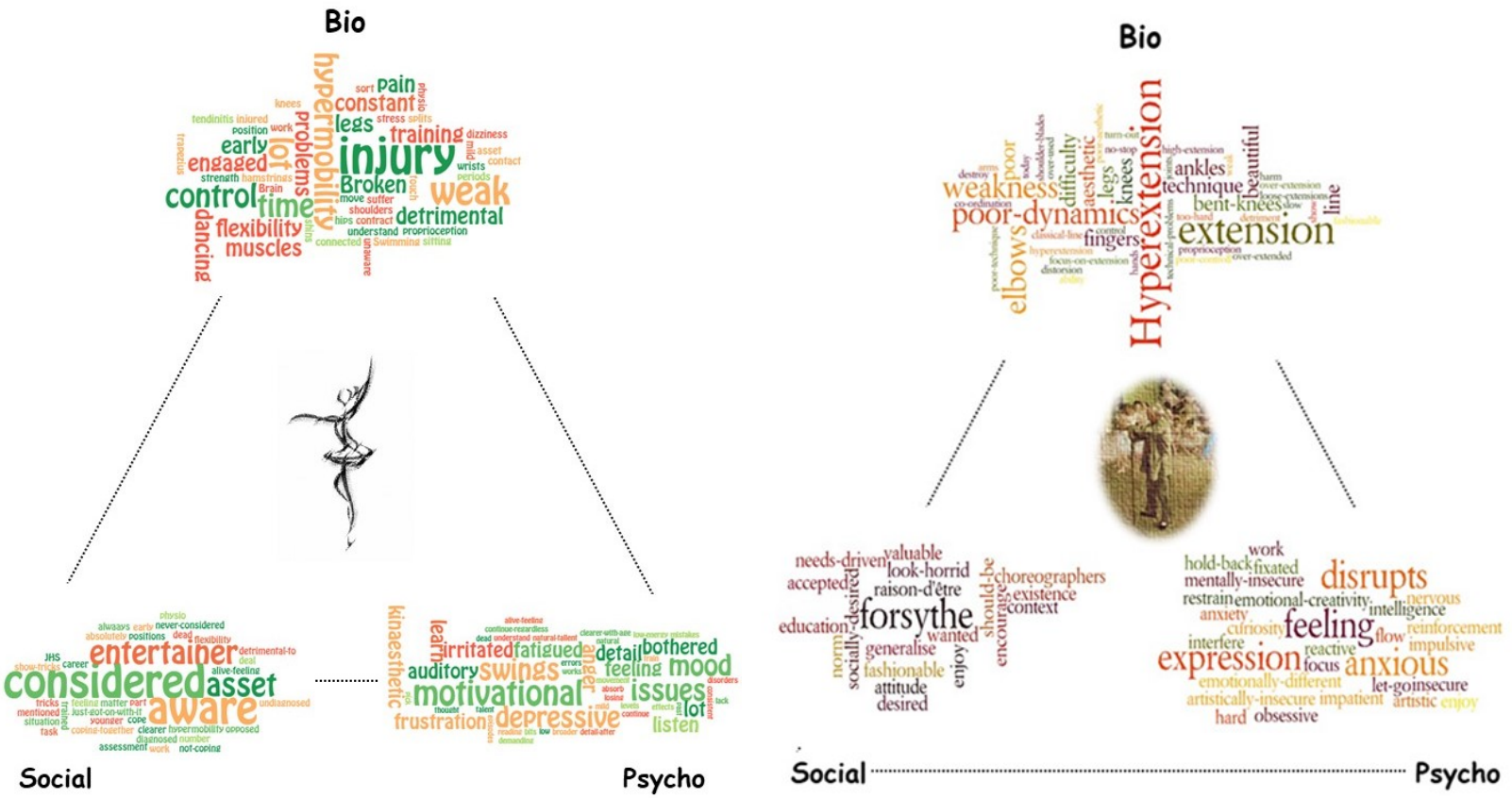

Figure 8 Diagrammatic representations of group 1 (dancer) and group 2 (dance master) transcripts

The verbatim data collected from the direct responses to questions and also more complex descriptions including those explained through the use of retrospective recall of events and times in the participants' careers was then further probed and analysed using thematic analysis. This was considered necessary in order to objectively examine, decode and understand the comments and opinions provided by the participants. The longer responses were rich in detail in both words and feelings as the participants expressed unique thoughts about the physical, emotional, and cognitive wellbeing and the experience in the dance environment across their careers and sometimes also their training. The participants were also asked wherever possible and relevant to retrospectively reflect and describe events that would exemplify the experience that they were describing. This helped the participants put things in to context and also assisted the thematic analysis making sure that the comments were also analysed within the right context and not misinterpreted. In this process the interviews (nine dance and four dance master) were read and re-read several times in order for the contents to be familiarised this process was strengthened further by the fact that the researcher had personally undertaken the initial transcription of the interviews to text. The features of the interview data were considered alongside the research questions for the both groups and the dataset for each group was primarily re-checked for reoccurring features and second for themes (and sub themes) to determine potential common stories and opinions that addressed and clarified the research questions.

The reoccurring features that emerged from the dancer and dance master's interviews are presented below in tables 14 and 15.

Table 14 Group 1 (the dancers)

Reoccurring features
Time management – needing more time
Achieving stillness, physical and mental
Overthinking, ruminating and obsessive compulsions
Awareness of JHS and lack of awareness
Learning through injury
Insecurity
Not being understood
Need for routine
Being overwhelmed
Joint hypermobility as talent , an aesthetic to be admired by others

Table 15 Group 2 (the dance masters)

Reoccurring features
Types of repertoire (classical v contemporary/neo-classical)
The dancers reason for being in the profession
Current choreographic trends
Pressures put on the dance companies to please the audiences (including funding implications)
Dance training
Technical differences (dancers from West and East)
The influence of social media (you tube, face book) and the millennium influence - push button society
Changes and current trends in a dancer's training
Company demands
The dancer's experience
Dance injury
The dancer's ability to cope and mechanisms for this
Thinking dancers, intelligence and the role of experience
The age of the company, capacity for mentorship in the company

Further analysis of each of the groups' data was then undertaken, here a thematic analysis approach was also applied. Many qualitative researchers now agree that this approach should be considered as a discreet methodology for qualitative data analysis that creates sensitive, insightful, rich, and trustworthy research findings (Braun & Clarke, 2006; King, 2004; Leininger, 1992; Nowell, Norris, White & Moules, 2017 & Thorne, 2000). It is a method that

can be applied to identify, analyse, organise, describe, and report themes found within a data set (Braun & Clarke, 2006). A thematic analysis process is considered robust and trustworthy when it follows a known and tried process with criteria that are set out to demonstrate trustworthiness at every stage of the process. Lincoln and Guba (1985) relate the trustworthiness criteria to the pragmatic choices that researchers make concerning the acceptability and usefulness of their research for a variety of stakeholders. Lincoln & Guba's trustworthiness criteria as;

i. Credibility
ii. transferability,
iii. dependability
iv. confirmability
v. audit trails and reflection

'Credibility' associates with how the readers of the research relate to and recognise it, this was operationalized in this study through the process of reading-through and confirming the interpretations of each interview with the participant (Lincoln & Guba, 1985). 'Transferability' on the other hand allows for generalisability and application of the inquiry to other contexts. The exploratory nature of this study however, meant that the researcher was unaware of the contexts to which other researchers may want to generalise the findings. The researcher tried, nevertheless and wherever possible to provide opulent descriptions so that they may be used as a means of rich information that may assist future transferability judgements to be made by others (Lincoln & Guba, 1985). To achieve dependability, the researcher tried to ensure the data analysis process was clearly documented, logical, and traceable (Tobin & Begley, 2004) and for which Braun & Clarke's process and methods (2006) were brought in to play. Confirmability requires the researcher to demonstrate how conclusions and interpretations have been reached (Tobin & Begley, 2004), this again was established through the clear documentation of the process towards credibility, transferability, and dependability (Lincoln & Guba, 1985). Finally the 'reflexivity' and 'audit trail' across

the formation and implementation of the interview questions and data collection were documented providing readers with evidence of the rational for decisions and choices made by the researcher regarding theoretical and methodological issues throughout this study .

The different features of thematic analysis as set out by Braun & Clarke, (20006) are six. These features are implemented in a recursive process, meaning that this is far from a linear process and there is often an element of back-and-forth between the different features This same process was followed for the analysis of data for each of the groups 1 & 2 of participants and alongside the reoccurring features, codes were developed for each of the groups, the transcripts were re-read and coded accordingly. The coding enabled themes and sub-themes to be identified, this process is documented below in table 16 for group 1 and table 17 for group 2.

Table 16 Group 1 (dancers) emerging themes, sub-themes and codes

Thematic analysis process		
Codes	**Sub themes**	**Themes**
○ Insecurity ○ Overthinking ○ JHS, degrees of ○ Injury ○ Aesthetic to be admired ○ Learning styles	Understanding of hypermobility and awareness of the positive and negative associations	**Openness to JHS**
○ Time ○ Stillness ○ Routine ○ Chaos/Confusion ○ Rhythm ○ Determination	Coping strategies	**Managing JHS**

Table 17 Group 2 (dance masters) emerging themes, sub-themes and codes

Thematic analysis process		
Codes	**Sub themes**	**Themes**
○ Trends ○ Choreographic ○ Beauty	Taste	**Aesthetic**
○ Social media ○ Funding ○ Technical training	Driven by current cultural strategy *Mise-en-scène*	
○ Technique ○ Emotion ○ Resilience ○ Passion ○ Intelligence ○ Injury ○ Coping	Intellectual curiosity/thinking dancers	**Professional values & preconceptions**
○ Teaching & learning ○ Technical training ○ Trust ○ Cultural shift	Pedagogical beliefs & leadership	

The interviews provided opportunity for the participants to also discuss others, for example the dancers and dance masters referred to other dancers that they had trained with in the past or ones that they had worked with who ranged from *corps* and *soloist/principle* level dancers. As the participants were asked to recall specific events and happenings from across their career in order to 'bring to life' and exemplify explicitly and implicitly the information that they were sharing, they were also requested, wherever possible, to not disclose the identity of the dancers discussed in the interview process, if names were mentioned these were also anonyimised in the transcription process. At times , and in response some of the questions, participants also found it useful at times to refer to and recall their experiences with younger less experienced dancers with whom they had trained, worked or observed at vocational ballet schools. Whilst not directly about the dancers themselves, this information was considered in the analysis as it added to their experience of hypermobility and also allowed the participants to discuss the professional dancers in comparison with less experienced dancers.

6.8 Discussion

6.8.1 Group 1 (the dancers)

Only one dancer had a clinical diagnosis of what they named as 'hypermobility', it was not clear if this was GJH or hEDS because at the time of the diagnosis the most recent clarification of HSD's and EDS was not available (Malfait et al., 2017). The remaining eight dancers had all been referred to the study for their hypermobility and this was further confirmed prior to each interview using the self-screen questionnaire. The outcomes of the screening were however not discussed as part of the interview and interestingly none of the dancers asked questions about the screening questionnaire.

From the discussions it was made clear that all of the dancers had an awareness of hypermobility and double-jointedness and they all suspected that they had this to *'some degree'* however only one of the group had been clinically diagnosed with GJH. Seven of the group believed absolutely that being hypermobile (or GJH+) was an asset for a dancer, two considered it to be a liability, and one participant considered it to be both an asset and a liability for a dancer to be GJH+.

Two main themes emerged from across the nine dancers' transcripts, *openness to hypermobility* and *managing hypermobility*. These are now discussed under two separate headings and then together in relation to the research questions for this group.

a. *Was there an openness towards hypermobility?*

The dancers' degree of awareness and associated 'openness' to hypermobility differed across the group. None of the dancers had been clinically diagnosed as hypermobile however responses to the question 'are you hypermobile' ranged from a straight "*yes*" (Ron line 29 p 1, Elsi line 23, p1) and *"yes I suppose I am"* (Bella, line 18, p1), this response was almost as if Bella had never considered this before, to "*not diagnosed but I was a very flexible dancer"* (Liz line13, p1) who appears to only understand hypermobility through the presentation of flexibility. One dancer (Ed) totally contradicted himself saying "*personally I don't believe that I was a flexible dancer…I can do four of the positions on the self-screen test"* (Line 29, p1). A score of three or more on this scale indicates Joint Hypermobility Syndrome (JHS), (Bulbena et al., 2014) Ed also disclosed that his physiotherapist believed that he had hypermobility in his upper body (shoulders and thoracic spine however it appears that the lack of flexibility in the legs and hips has led Ed to assume that he was not hypermobile - or at least not in the right way for a dancer perhaps? Ed was indeed referred to me by a physiotherapist for inclusion in this study of hypermobile dancers. Another dancer, Tom, suggested that he *"had a slim degree of hypermobility"* (line18, p1) and *"…had a lot of*

injuries that could be associated with hypermobility" (Line 19, page 1). Tom also discussed how his hypermobility had been *"detrimental to the measurements of control he had over his body"* and in particular because of the very early age he started intense training (Tom, lines 28-32, p1). The overall responses suggested that the dancers considered hypermobility to be significant if it was visible in the knees and elbows or it facilitated extreme extensions of the legs and hips. They also tended to compare their own 'degree' of hypermobility with other dancers that they recalled who were visibly more hypermobile than them and who could achieve extreme extensions of the legs and hips. One dancer described how her mother *"knew that her body was very different from others"* (Gill, line24, p 1) which is why she was sent to gymnastics, the difference she was describing (and also showing to me in the interview) was purely physical , for example, the way her fingers, elbows and knees hyperextended beyond a normal range of motion. The interview data suggests that only after many years of professional training did the dancers come to understand hypermobility better, for example Tom describes

> *"I knew in my training that I had sway-backs"* referring to his knees *" I was never told that this was anything other than a term used to describe some dancers' legs…it was only post professional education that I realised what hypermobility was and the effects of it"* (lines 18-22, p1).

Elsi also discussed how she discovered the implications of being hypermobile through academic study towards the end of her performance career,

> *"I knew I had hypermobility but I was unaware of how it could potentially influence my training and cause injury"* (Lines 27-28, page 1)

Despite the fact that some dancers clearly knew or suspected that they were hypermobile and this may also potentially lead to injury, none of them had considered it necessary to seek a proper diagnosis or advice. Most of the group undeniably considered being hypermobile an asset for dancers, for example when asked Ron replied

> *"my hypermobility enabled me to get lots of dance jobs that I might not have otherwise and I could afford to have a great life in Europe, … on another practical level, I can still bend down and pick something off the floor with not much effort – I can even pick things up with my feet…"* (Ron, lines 41-45, p2)

One dancer discussed how she believed that hypermobility was understood to be an asset in the professional performance domain, she believed however that *"…it was a blessing that I did not stay with one particular company or choreographer for a long period of time…"* (Elsi line 36-38 p2) inferring that that the extra capacity for range of motion was not exploited over time and in the same way by the same choreographer. She went on to discuss however that "*near to the end of every contract that she had undertaken I had been injured"* (Elsi, lines 39-40, p2), this, she retrospectively put down to her own lack of strength and fatigue.

Gill discussed in her interview that although she believed that being hypermobile was *"mostly an asset…"* she also disclosed that at times "*it really hindered my development"* she had difficulty as parts of her body *"… did not fire up quickly"* explaining how she *"over-indulged in the movement and went further",* (Gill lines 35-39 p2), demonstrating an extreme position to me in the interview. Gill described how she assumed that she had come to the assumption that she had "*more collagen"* in her body and this stopped her getting strong and technically fast, also giving her *"a slow start in her career"* (Gill, lines 43-45, p2). Gill further clarified how at the beginning of her career she clearly looked 'the part', referring to her 'slim fragile ballerina appearance' but when it came to delivering the roles with the required strength she *"did not know how to handle the abilities she had".* Bella also agreed with this suggesting that hypermobility was an 'aesthetic' asset however she believed that it was *"much harder to perform classical ballet with hypermobility"* (Bella, line30 p1). Despite these declarations, none of the dancers actively sought out help. The dancers who did not have the type of hypermobile knee and hip joints that facilitate high extensions really did not consider themselves as hypermobile and overall the group's understanding and openness

to hypermobility was purely in relation to the associated positive visible attributes and negative physical weaknesses.

In general there was a clear lack of openness or understanding towards the known psychological and social issues that are associated with hypermobility (Carolina Baeza-Velasco, Bulbena, et al., 2018; Bulbena et al., 2017b; Gurer, Sendur, Gultekin, & Ozcan, 2010). Between the members of the group the dancers discussed feelings of being overwhelmed, obsessive in their thoughts at times, over-anxious, not coping with stressful situations and two of the group had suffered from depression for which they has sought help. None of the dancers had associated these feelings and conditions with hypermobility. In the cases of anxiety and depression where clinical help had been required, there were no suggestions that there may be links to hypermobility.

b. *How did the dancers managing hypermobility?*

Only two of the dancers acknowledged that they had received additional support and advice about hypermobility during their initial training, this support was however solely in relation to coping with the physical aspects of hypermobility. None of the dancers discussed getting support or advice from dance company members related to coping and managing hypermobility during their performance careers. Three dancers did receive advice and information about the physical aspects of hypermobility whilst they were being treated by physical practitioners (physiotherapists, osteopaths, giro-tonic practitioners) they were seeing for injury and weakness related physical issues. The practitioners addresses the dancers' issues, explained the physical weaknesses but did not refer directly to hypermobility. The reluctance of the practitioners to name or label the dancers as hypermobile is interesting and one that the researcher encountered when initially contacting clinicians to ask for potential referrals to the study. It remains unclear whether this was done deliberately or

inadvertently, however what this might suggest is an attitude in the dance environment whereby if something is not given a name then could it be considered to be non-existent or at best insignificant?

Despite the lack of clear and direct information about hypermobility, the dancers had all managed to work with and around the physicality of their hypermobile bodies. Using the extra range of motion for aesthetic and choreographic gains, they had also learned to work with the weaknesses in their physicality that resulted from the hypermobility. This had come to some of the dancers at a cost of pain and injury, something that they considered natural in dance,

> *"…when we did Forsythe and Balanchine, we were pushing out of our point shoes, I had a lot of bunion pain…"* (Gill, line 88-89, p3)

By 'pushing out' Gill was referring to the over-arching or hyperextension of the foot and ankle in order to achieve the aesthetic that these choreographers often demanded. She also described having to over use or over-extend her arabesque for these choreographers which also caused her pain in her feet and back.

All of dancers were asked about working as a group as opposed to dancing solo, they all proposed that they preferred to dance solo. Of course this may be because the attention of the audience is focused on a solo performer however some gave reasons for this saying that it was difficult to co-ordinate exact movements with other dancers that did not have the same range of motion as them. Elsi describes working in a group configuration as hard because she was aware of her lack of sense of where she and her body parts were in space and having less control for the centre. Aiden also discussed working in a group formation describing it as *"challenging"* and *"something that needed persistence and extra focus"* (Aiden, line 144-146, p5). Partner work and the challenges of working with another dancer in duets was also

discussed most of the dancers agreed that this is something that comes with experience, Liz for example discussed how when she initially started partner work she was always very conscious of being criticised for being too floppy,

> *"…like you are too heavy in his arms… it can be very personal getting these comments and corrections…to the point that you begin to feel that you are too heavy and need to lose weight"* (Liz lines 111-117, p4)

Bella also discussed how it took her longer that other dancers when she started *pas de deux* work while in training, *"…I really was not aware of my body in space"* (Bella, 77 p3) she also described how she had to work with experienced partners who understood that the line of gravity and forces through her ankle, knee and hip were very different to a dancer who did not have GJH I particular when they lowered her to the ground. This is because of the need to increase the torque in the muscles and ligaments of the joints in the leg in order to remain balanced on pointe which inevitably results in hyperextension in the knee (Huwyler, 2002, p. 77). The male dancers also discussed the challenges of hypermobile shoulders when lifting other dancers and the constant need to strengthen in this area in order to avoid dislocations. The dancers had come to these modification in technique through experience, trial and effort and pure determination to succeed, in some instances this also involved learning the hard way through injury.

The biggest challenge to all of the dancers was managing stillness. Physical stillness is an important and necessary part of a dance performance and contrasts with the complex dynamics of the dance movement. Classical repertoire in particular often requires the dancers to hold a position or pose for several minutes whilst present on stage.

> *"In a performance it can be very difficult especially when you have just been dancing and then you have to stand on stage for a long time … your feet and legs cramp up … I am not a fan of standing still"* (Aiden, lines 67-69, p2).

The dancers referred to this in a range of contexts, for example, the more experienced dancers discussed how they had learned to deal with what they termed as the restlessness (both physical and mental) over time. Some had help from experienced teachers or had been directed towards alternative practices such as yoga. Gill explains,

> *"I have learned to get to the still point quicker, but it took me a long time...in earlier years I was constantly adjusting or finding my centre" (*Gill, lines53-56, p2)

Bella also described the physical discomfort of not being able to stay still *"...I cannot stay still at all, I need to shake them out regularly"* (Bella lines 38-41, p2) by this she was referring to her legs and how when she 'sat' into the natural hyperextension of her knee she was initially comfortable however this became painful after some time. Likewise Elsi discussed her inability to sit for any time because of the aches and pains she experienced in her body. Liz also suggested that she felt physically ill when she was made to stand still for any length of time and she described the extra effort and concentration that was required when having to hold a pose. Sometimes for her this extra effort was required at points in the choreography that were actually designed to give the dancer a 'breathing pause' or a moment of stillness on stage that allows the dancer to catch breath to continue. Mark also declared that in general *"...oh I hate standing still"* (Mark, line 49, p2) and Ron discussed how he had latterly changed to a standing desk that allowed him to move around because he found sitting for any amount of time painful.

Mental stillness was also discussed in terms of agitation, mood swings and difficulty in switching off. Bella discussed how she needed to focus and concentrate hard in order to get things right to the point where she considered that she ruminated becoming 'obsessive and compulsive in her thoughts' at times, in particular when she was tired. Tom discussed how in times of stress and anxiety that *"...my brain goes faster than my mouth"* (Tom, line 66, page 20). Mark spoke of the peaks and troughs in states of minds when he was under pressure, for

example when on tour and in unknown environments. Gill described an obsessiveness with doing her best and coping with this through self-punishment for not working 110% all of the time, this she knew resulted in swings of mood that she had learned to cope with over the years. Ron likewise disclosed that his depression was linked to the fatigue that he felt daily at times and this also caused anxiety. Elsi found that she needed routine when coming out of a very busy contract of performance period *"like going to the gym…I have that to hang on to because if I don't then I don't feel very well…I need the structure"* (Elsi, lines 89-91, p3). Elsi also described how she needed to be in control in order not to feel stressed however this obsession with control did at times also make her feel exhausted.

When asked about managing to learn new works or movement styles, all of the dancers referred to preferred ways of learning that was described as needing to understand the 'bigger picture' of a piece of choreography before working on the detail. By this they meant that they needed to be aware of the orientation and pattern of the work before they could add any technical detail. Most of the dancers disclosed that they knew that in order to grasp complex and/or lengthy choreographic pieces they would need more time than other dancers. This extra time would be used working alone at their own speed until they were confident of the piece. Elsi interestingly used the rhythm of a movement in order to remember it and did not *"follow verbal instruction well… if people give me instructions and directions at the same time, I get stressed"* (Elsi, line 95-103, p 3). Elsi disclosed that even though she was now an experienced dancer in order to not get anxious and stressed she still had to go away and practice repetitively more than anyone else in the group. Aiden also disclosed that he knew he had to really concentrate hard to retain new choreography, harder than other dancers he felt. Bella considered that she needed to focus on patterns before she could cope with the details. This for Bella would depend on the choreographer's or ballet master's style of delivery, when given all of the choreographic information at once, working this way for her

this sometimes meant spending extra time and energy correcting and filling in the details *"...like correcting and changing basically whatever your body remembers"* (Bella, lines 98-99, p4), this for Bella would depend on the choreographer's or ballet master's style of delivery. Likewise, Ron also described how he liked to *"get the wash of a sequence of movement"* and then he would *"find the details afterwards"* (Ron, lines 164-165, p6).

The dancers also all preferred learning repertoire from actual, 'bodies in the room' despite the fact that learning from a screen could potentially enable them to have more time and working at their own individual pace. The dancers discussed how that this was for them often disorientating, time-consuming and therefore much more stressful. Once the dancers had spent time and were in command of the choreography they were learning they did not however have problems remembering it over time.

c. Do hypermobile dancers understand hypermobility?

With regards to the research question and sub-questions, the interview data suggests that the dancers all had an awareness of hypermobility which they described this as having hypermobility 'to some degree'. Their understanding, including the dancer that had been diagnosed at an early age, was predominantly experiential, tacit and not explicit. This is not surprising as within specialist and clinical practitioners it is only recently that an understanding of what is now clinically described as the HSD. The further links between HSD and conditions within EDS (Castori et al., 2017), and more specifically hEDS which is potentially a greater risk for dancers (Foley & Bird, 2013a; Mccormack et al., 2004), are also only now becoming clear. The intricacies and differentiation between GJH, LJH, HJH and PJH and their connections with hEDS are then undeniably still developing in the clinical world (Malfait et al., 2017).

For the dancers, recognition of hypermobility was essentially made through the identification of for example hyperextended legs, hips, arms and ankles. For them the aesthetic gain that this was perceived to achieve was of course an asset. Several of the dancers did also recognised that hypermobility also came with weaknesses that had to be compensated for and managed through additional training. Managing and coping with hypermobility in this way had led them to a tacit understanding of how to deal with with, for example, the associated pain (when standing for any length of time and having to shift the position and line of gravity so that it did not fall through the hyperextended joint), weakness (through injury, dislocations, sprains and fractures), or the difficulties they had with strengthening and their core in order to allow them to successfully undertake partner work (Bella). Whilst the psychosocial implications of GJH are clear in the literature, no one had ever discussed these with the dancers in relation to some of the anxiety and mental wellbeing issues they had experienced across their careers. Again they had learnt to manage and cope with these by compensating and using strategies that they had devised over time. Sadly for most of the dancers this had come through time, and for some perhaps too late as it was towards the end of their career.

6.8.2 Discussion group 2 (the dance masters)

For the purpose of the discussion that follows and maintaining the anonymity of the participants assigned to group 2, all ballet master participants were given pseudonyms; Michael (M), Carolyn (C), Jannis (JA), Jean (JE), and they are referred to by these names and codes throughout the thesis. The findings are organised and driven by the research questions and are initially discussed in terms of the emerging themes and sub themes.

First, the interview responses were considered with regards to the two themes and sub themes, these were interrelated and of equal significance therefore they are presented and discussed below in no specific hierarchical order.

a) Theme: The 'hypermobile aesthetic'

The Subthemes within this theme were*; taste, driven by strategy and Mise-en-scène'*

All four dance masters were readily able to descry the biological markers, features and attributes associated with a hypermobility and referred to hypermobility not so much in terms of a 'condition' but as a 'hypermobile aesthetic'. In this context the features that they all identified were; '*overextension, joints, fingers, knees, ankles, hands, elbows, and the significant range of motion that facilitated high extensions'.* Michael stated that in the company where he worked, the directorate actively sought out and had developed a 'taste' for the hypermobile aesthetic and dancers who mainly came from the Eastern-bloc (referring to the former Communist states of Eastern and Central Europe). Michael believed that extreme flexibility (although not explicitly in the entrance criteria) is almost a prerequisite for entrance in to the conservatoires and state schools in Eastern and Central Europe. Michael remarked that he now sees that in general for today's dancers the aesthetic line of extension has shifted to over-extension "*the splits are no longer 180 degrees but easily 200 degrees"* (M line 19, p1). He confirmed that for the company where he worked, *"90% of our dancers are hypermobile, the others, are the minority, yes"* (M, line 64-65, p2). Carolyn also discussed how there was a trend for the 'cultivation' of this type of dancer in companies and that she could not quite understand it because "…*they are so*" in her words *"much harder to train"* (C, line 115-116, p3). In this Carolyn was referring to the known weaknesses and imbalances of strength and flexibility, lack of proprioception, and other physical vulnerabilities associated with hypermobility that the dance masters referred to in their interviews. Carolyn also went on to point out that this aesthetic was currently "the norm" in

St Petersburg and a now a trend in some European companies such as the Paris Opera. Commenting also on the reality that today in several ballet companies *"the dancers are just all basically hypermobile"* (C, line 10, p1) Carolyn also established that some dance companies were now actively identifying and only seeking out dancers with a body type and physiognomy that is able to produce this 'hypermobile aesthetic' for their audiences. She strongly believed however that there was a difference between the range of dance styles that were needed in the Eastern bloc and in Europe. The '*Russian'* style she described as typically *"pose orientated and not dynamically moving"* (C, line 118, p3) whereas the demands from choreographers and repertoire currently popular in Europe were much more dynamic and moving.

On several occasions in her interview Jean also discussed the shift away from classical aesthetic and repertoire where, to her mind, the hypermobile aesthetic was detrimental to the *"alignment of the classical positions"* and the expressive capacity that was within the classical vocabulary. Jean is referring to the aesthetic shift within classical ballet that has come from evolving traditions and expanding repertory. Ballet progressed from its origins as the codified dance form of the 17th Century French Court to become a dramatic art form and spectacle in the 19th century, associated with the 'Romantic classical style' and narrative works such as *La Sylphide*, and Swan Lake. It was in the 19th century that the evolution and professionalism of ballet companies also led to a new generation of ballet masters and dancers (Scholl, 1994). From this point onwards the aesthetic within choreography shifted further to develop into the neo-classical style that we associate with *Balanchine,* founder of the School of American Ballet. Interestingly this neo-classical style is also associated with the typical '*Balanchine body'* that in many respects facilitated *Balanchine's* fascination with the body and its movement in three dimensional space (Ritenburg, 2010). The '*Balanchine body'* also moves towards the 'hypermobile aesthetic',

the typical *Balanchine* ballerina was very lean with long legs and arms, small hips, short torso, small ribcage, and long neck that we associate with today. Jean confirmed that the trend for choreographers to seek out this specific body type and abilities was also still a current one,

> *"I personally know choreographers demand it"* she continues*...with many choreographers they encourage and want to choreograph in a way that extends mobility to its nth degree* (JE Lines 28-29, p1) *…you know who I am talking about … and they require that, to my mind it is brutal and I fear that sometimes the dancers will pay the price of exactly what this choreography is asking of them…"* (JE, lines 66-68, p2)

For Michael the true classical aesthetic and current trends were also a feature, he too believed that there was a lack of understanding in some hypermobile dancers who now struggle to maintain the classical line and aesthetic awareness of the classical postures and gestures. They can be seen *"kicking the legs"* and (when wearing the classical tutu as opposed to a more contemporary costume) exposing an angle of the body that *"you don't want to show and see in classical ballet"* (M, lines 118-119, p4).

There was also common agreement between participants that, from a physical perspective, hypermobility was associated with and could also be recognised through a weakness in the legs, in particular the hamstrings and shoulder blades and that a 'poor aesthetic' and 'winging' for the back was characteristic of this. The dance masters all made comment on the fact that the winging and associated weaknesses often caused and technical problems and poor aesthetic in the arms, often (but not always) a lack of dynamics , and poor jumping capacity. The dancers were often as Carolyn described it *"not as fast, and lacked dynamics"* (C, lines 34-35, p1). Jannis discussed how many of the hypermobile dancers have, weak spines, often even scoliosis and a tendency to appear very thin and lacking in muscle tone, this in turn she believed meant that they were often incorrectly 'branded' as anorexic (JA, lines 60-73, p2).

The dance masters all recognised that this trend was driven by current taste and cultures and is associated with the popular *mise-en-scène*, a concept borrowed from cinematography that currently is increasingly associated and applied with dance. Originating from the world of cinematography *mise-en-scène*, refers to the arrangement, design and composition of a production. In a dance context the *mise-en-scène* relates to the positioning or arrangement and design of body positions and movement that create a visual theme as opposed to the more traditional mimetic expression that depicted the 'narrative or story' associated with classical repertoires of the past (e.g. Giselle, Swan Lake). It is the design of body positions that Michael, Jean and Jannis refer to when they discuss the choreographic practices of today in, for example, works by Forsythe, Maliphante and McGregor who regularly seek to use hypermobile dancers to make and perform their work (e.g. Guilliem http://bit.ly/GuillemDocumentary). Jannis, Michael and Jean also discoursed how some dancers do not have what they refer to as intelligence (bodily) and are (they believe) unable to differentiate between the styles of movement needed for classical and current contemporary works. They recognise that dancers today are often fixated on the visual theme or *mise-en-scène* of extreme ranges of motion associated with the 'hypermobile aesthetic' and apply this across all performance.

Interestingly all dance masters agreed and recognised that the development of the current hypermobile aesthetic within the realms of classical ballet is related to choices that are largely driven by current strategy, the need to please audiences, fill theatres and ultimately satisfy the funders. These are typically states or governments who fund the art-form via arts bodies (such as Arts Council England and Creative Scotland in the UK) and who are increasingly influencing what is programmed in theatres (Burns and Harrison, 2009). Their interviews suggested that this strategy was ultimately influencing current aesthetic trends and demands in the dance environment. There was also a strong consensus that freely

available digital platforms on social media (You tube, Vimeo, Facebook etc.) had a significant role to play in this phenomenon. Less experienced audiences and dancers are, as Jannis describes, influenced by a *mise-en-scène* design whereby they are fixated on still images and video clips of dancers achieving the impossible.

> "*fixated on aiming at the best possible photograph, or what they think is the best possible photograph to put on Face book, and they forget that dance is about movement"* (JA, lines 29-30 , p1) ,

There was a strong overall consensus from the dance masters that this type of fixation means that the young hypermobile dancers are no longer considering the meaning, artistry, artistic communication and expression within the performance, their concerns are only with the physical aspects of the work. Jannis further discusses,

> "*It is an attitude of today, because of the likes of Sylvie Guilliem*[i]*, everybody thinks that they have to lift their legs as high as they possibly can. It looks as if they are aiming at the best possible photograph (or what they think is the best possible photograph) to put on Facebook and they forget that dance is about movement".* (JA, lines 27-30, p1)

The interviews manifested both individual and common views and beliefs from the dance masters, and these are presented under the second theme

a. Theme: Values and preconception

The subthemes within this theme were; *Intellectual curiosity and thinking dancers, pedagogical values and leadership*

All four dance masters agreed that hypermobility was more than just a specific yet very current 'aesthetic or look'. They valued hypermobility in different ways and to different degrees and expressed individual views and preconceptions about it. They also discussed associated opposing views of professionals such as directors, teachers, coaches and the dancers themselves in relationship to the value of this specific body-type within dance performance. For example, Jannis initially described hyperextension as "*beautiful"* (JA, line 13, p1) however she also iterated the important value of correct training for the dancers and a

mutual understanding of hypermobility (for both the dancers and dance masters) and using the hypermobile capacity fittingly and for the appropriate style of choreography. In contrast, Jean had a very clear view believing that *At a certain point there is no control – the limbs will continue and that of course distorts the classical line* (JE, line 24-25, p1) , she did however agree that it was a necessity and therefore professionally valued within certain choreographic designs and styles, undeniably all dance masters noted and attributed this style to the American choreographer Forsythe (1949-present) and works by the Forsythe Company (2005-2015). Michael discussed how there was currently a lot of demand for hypermobile dancers as it was now a valued and an assumed given "*part of the show"* (M, line 24, p1). Carolyn discussed the dance company funders' pre-conceptions (Ministry of Culture in this specific case) which were that theatres and ballet companies could put up programmes with very little rehearsal time which she believes *"...ideally needs dancers who are very fast at creating and absorbing new work...and who are less prone to injury"* (C, lines 122-125, p3). This conflicts the participants' general consensus that a hypermobile aesthetic can come with weaknesses (both physical and technical). Carolyn also questions the preconceptions and pedagogical role of a dance teacher and master when influencing a dancer's career choice, commenting on how further research and consequently a clearer understanding of hypermobility may influence a company directorate's choices and professional values in the future. Indeed she discussed a recent shift within New York City Ballet towards dancers who have a more athletic aesthetic and *"less curved and bendy legs"* (C, lines 53-54, p2 making reference to and demonstrating in gesture the hyperextended knee that is characteristic of hypermobility). Jannis also discussed how values, pre-conceptions, pedagogy and intellectual curiosity play a role in the 'hypermobile aesthetic' and current trends. She discussed the lack of pedagogy (teacher-led direction) within current dance training to guide young dancers to discover *"what the different styles of dance mean even within classical ballet"* (JA, lines 34-

35, p1). She believed that classical ballets from the 19th and 20th century repertoire are increasingly now all perceived the same way.

Carolyn also discussed the dancers' training and pedagogy and described dancers who *'in spite of being'* hypermobile were incredible dancers because they had the *"intelligence and exposure to different training"* (C, lines 111-112, p2). Interestingly, Carolyn infers a preconception that not all dancers who are hypermobile have this capacity. Jean likewise confirmed that dancers needed the correct teaching and knowledge in order to have the right information to support and work properly with their hypermobile state. Jean referred repeatedly to the 'intelligence' of dancers with a hypermobile aesthetic, she believed that some of these dancers

> *"...would genuinely have you believe that they are working hard, and they probably are, but I would like them to let go of the working hard (with the hypermobility) and work more with their intelligence, curiosity for artistry, and creativity"*(JE, lines 114-116, p3)

When asked to describe if and how these dancers differed from non-hypermobile dancers straight away Jean described the weaknesses that they show,

> *"...because the hypermobility lets them extend as far as they possibly can, with that comes a certain weakness, because there is no stop, they have no control…the feeling of having that looseness and mobility is a lovely feeling and they enjoy that feeling…mentally you have to bring them back to a study period to understand how much is valuable and how much will destroy the line".* (JE, lines 25-29, p1)

Jean was very clear in her view that a dancer with hypermobility needed to have the knowledge and information to support the extreme ranges of motion, hyperextension and other associated issues such as poor control. She believed that it was a matter of education and intelligence for the dancers but more so for the teachers. She described hypermobility as being valuable as part of the dancer's technique which she believed is however only the starting point or base line,

> *"…a dancer can develop a fixation…Dancers needed to learn to either 'disguise it'* [i.e. the hypermobility]*or let go of the fixation' at that level of performance if they were ever to tread where angels fear to go"* (JE, lines 79-80, p2)

Jean believed that if hypermobility was professionally valued as *"the thing or IT"* that was going to propel the dancer forward in their careers in order to a great dancer then intellectually and emotionally the dancer would be weakened. She also believed that some hypermobile dancers put too much value on their ability to hyperextend and they exploit it as an asset that the dancers believe is wanted by today's choreographers. Jean goes on to explain that if dancers really want to translate that 'language' (referring to the aesthetic communication) perfectly and beautifully and correctly then they should not distort their bodies in that way. Indeed Jean believed that emotionally today's dancers are "*in a quandary"*(JE, line 120, p3) Jannis confirmed this view discussing how often a hypermobile dancer is chosen for his/her hypermobile physique and not the burning desire, passion and intrinsic need to become a dancer.

The dance masters shifted their discussions away from the physical manifestation of extreme body postures, and gestures and the 'hypermobile aesthetic', towards some of the psychological behaviours that they associated with the hypermobile dancers. Between them the ballet masters noted fixations, passion, excessiveness, coping (or not), emotions and resilience. Jean discussed

> *"…if a dancer is working too hard with the hypermobility and is concentrating on the height of the limb, it can distract them allowing themselves to emotionally explore the work because they become fixated on technique and that you know disrupts the flow of the artistic output" (*JE, line 45-47, p1))

Jean and Jannis repeatedly referred to the capacity of the dancers to understand what they were really striving to achieve, Jean regularly used the term *"intelligent or thinking dancers"* , both had a clear understanding of the role of experience and coping in relation to their own and the dancers' understanding of hypermobility . Jean referred to the difficulties the

younger hypermobile dancers had in understanding how to control their bodies because in her words *"...they are far too impatient"* (JE, line 40, p1). She discussed how certain skills (associated with the extreme range of motion they had) had come *"easy"* and they were not able to cope with *"putting in the work"* in order to achieve the required strength and control. Carolyn also discussed the dancers in terms of age and experience, she mainly works with younger and less experiences dancers and noted how often, "*they would have a tendency to be emotionally insecure and hold back"* (C, line 46, p. 2) even for things that should be quite routine given their level of expertise and training. She gave an example of her surprise she worked with them on proprioceptive skills placing them on an instable surface, *"I mean these are normally very skilled dancers and should be able to cope with that – after all this is what they need"* (C, line 45, p2) . Carolyn also attributed the dancers' insecurity and lack of resilience to the fact that dance companies are increasing now made up of younger and less mature dancers. In the company where she currently worked out of eighty dancers there were only five over the age of twenty five. She explained how the young dancers were *"socially and emotionally lost"* line 89-90, p3) she believed that it was vital to have older and more mature dancers in the company as they were the ones in her day who helped them cope *"...who gave us the hold*" (C, line 86, p3) meaning that the younger inexperienced dancer could to go to and learn from the senior dancers. Carolyn (again reflecting on her own experience as a dancer) discussed how companies 30 years ago did not have the same exposure to hypermobile dancers. She believed that *"now companies want to be on a par for example with the Paris Opera*" (C, line 116-117, p3) where she considered that the leadership and directorate no longer value the way dancers are trained in the West. She sensed that the strategic shift towards the 'hypermobile aesthetic' has led to the development of dancers who, in spite of an 'assumed resilience' that she believed was also associated with the hard, highly competitive and often brutal training received in the Eastern bloc, find it hard to cope

emotionally and who demonstrate *"a tense fear factor"* (C, line 79, p2) in their posture. In this Carolyn was referring to the breath holding and apical breathing (as opposed to lateral-costal breathing) that is commonly associated with the restraints of the classical ballet posture and technique (Hamilton in Ryan, 1987). This type of breathing is also commonly observed in hypermobility where altered and ineffective motor patterns are recruited in an attempt to stabilise the trunk (Simmonds & Keer, 2008). The hypermobile dancer will focus on the muscle recruitment in the gesture limb and compensate for postural instability by using muscular force and tension in the intra-abdominal-pressure (IAP) or core (Karin, 2016). The dancers' fixation on the height and hyper-extension of the gesture limbs and deficit in postural stability is discussed by Jean. Likewise Carolyn refers to postural instability in the hypermobile dancers she works with, this is also supported in the literature (Fatoye et al., 2009). Both would seemingly contribute therefore to the commonly observed tense posture of the hypermobile dancer (McCormack, Briggs, Hakim and Grahame, 2004).

When addressing the research question, *do dance masters understand hypermobility?* It was clear from the thematic analysis of the interview verbatim that the dance masters clearly understood features of hypermobility and were all able to describe hypermobility within a dance context. How they expressed their understanding of the condition and how they articulated this however varied. For example, Carolyn and Michael readily accepted and used the term 'hypermobile' and they discussed the dancers. Jean and Jannis on the other hand chose to dispute the term 'hypermobile' and preferred not to use it in the dancer context. The varying degree of acceptance suggest that there were possibly also differences in the dance masters' degrees or ways of understanding of hypermobility. The mixed acknowledgement also suggests that their understandings of hypermobility were entangled in their personal perception, experience, tacit and explicit understandings of the condition. Their experience of hypermobility and therefore personal knowledge of it was a) either resulting from experience

of teaching hypermobile dancers (which is why they were selected for interview) or b) conceivably also through personal first-hand lived (or embodied) experience of hypermobility and this lived experience was integral to their tacit understanding. Being hypermobile was regrettably not one of the dance master selection criteria and in hindsight, hypermobile status of the dance masters should have been tested. However, knowing that all dance masters had themselves been elite performers prior to taking the role of dance master and also knowing the prevalence of hypermobility in professional ballet dance is generally high compared to normal populations (Day et al., 2011b) it was hypothetically plausible that some of the dance masters were indeed hypermobile themselves. The potential impact of this was therefore also considered and further confirmation of all the dance masters' hypermobile status was investigated. This was achieved through observation of readily available archival footage and photographs of the dance masters in their prime as dancers and using this method it was confirmed that Jean and Jannis did indeed appear to display the tell-tale hypermobile characteristics of hyperextended knees and elbows.

Whilst considering what level is that understanding? Understanding requires the application of explicit, implicit and tacit knowledge. Explicit knowledge is factual and principally objective, in dance for example it relates to contextual aspects of dance (history, style genre) and the dance techniques and skills themselves. In this context, the dance masters' professional experience (Mean 31yr) working with professional dance companies and also personal performance experience (Mean 19yr.) provided a basis to assume that their expertise gave them excellent explicit knowledge of dance. Implicit knowledge is understood and contained however, it may not necessarily be articulated within an applied context. Furthermore, tacit knowledge arrives from the articulation, experience and application of implicit and explicit knowledge within a context, for example, through the act of teaching dance. Implicit knowledge, although a component of understanding, does not necessarily

have a tacit dimension, likewise tacit knowledge is also not necessarily articulable (Polanyi, 2005 p 17). All acts of knowing rely on a subsidiary levels and meaning (as a degree of understanding, see Kelp, 2015) and are best understood as a combination or interaction between degrees of implicit and tacit knowing (Polanyi, 2005, p18). When considering how dance masters' understand hypermobility it was therefore important to discuss the 'ways' of understanding this condition. Polanyi proposes this in his theory of tacit knowing,

> *"…suppose that tacit thought forms an indispensable part of all knowledge, then the ideal of eliminating all personal elements of knowledge, would, in effect, aim at the destruction of all knowledge."* (1966, p20)

Peck provides an interpretation of tacit knowledge as "inarticulate" he describes the act of tacit knowing as more than being able to express it or *"…to indwell the particulars, to attend to them from the clues and focus on the meaningful whole"* (Peck, 2005, p16). This interpretation indeed provides us with a rational for Jean and Jannis' reluctance to apply the term hypermobile to dancers.

The dance masters' expertise in dance performance and teaching dance implies that they all had explicit, implicit and tacit knowledge of dancers as required in the act of teaching, however they potential had different experience of hypermobility. Their different experiences of hypermobility were therefore potentially instrumental to their understanding. For example, the two who were hypothetically hypermobile themselves (Jean and Jannis) had personal and potentially a 'different understanding' to Carolyn and Michael.

all dance masters demonstrated a common capacity to identify hypermobility through specific physical attributes. For example all dance masters described and understood particular characteristic of hypermobility. This common understanding can be further discussed in relation to Polanyi's proximal –distal theory of tacit knowing (further proposed as focal-subsidiary by Peck, 2005). That is, within hypermobility, particular characteristics (such as

the hyperextension) associate with the biological manifestations of hypermobility and were for the dance masters the 'proximal' or focal aspects whereas, the aptitude to contort the body into shapes and postures and express the art form (dance) through the proximal or focal aspects pertains to a 'distal' or subsidiary understanding of hypermobility. For Peck "…*to know something is to indwell the particulars"* (2005, p16) which were in this case the proximal or focal particulars and "*to attend from these clues and to focus on a meaningful whole"* (ibid), referring to the 'distal' or subsidiary of tacit understanding. Peck also pertains to a notion that experience also acts upon different levels of focal and subsidiary tacit awareness (2005, p16).

The diagrammatic word-cloud representations of the data further confirm that the biological (bio) particulars of hypermobility were foremost in the dance masters' understanding of this condition. These (muscular strength, flexibility, condition) together with the associated behavioural manifestations (psycho) of the anatomy and physiology (e.g. fragility, dynamic capacity of movement) and associated social aspects (e.g. anxiety, insecurity)were, for all dance masters, significant and identifiable characteristics of hypermobility. For example when Jannis was asked to comment on what is 'characteristic about hypermobile dancers' she responded

> "*sometimes they have difficulty holding their turn out or holding their positions as opposed to being able to get in to very high extensions"* (JA, lines 13 & 14).

The dance masters clearly agreed and understood that the proximal (focal) attributes or features of hypermobility were characteristic. These proximal attributes are couched under the theme 'hypermobile aesthetic'. The dance masters all referred the hypermobile aesthetic using the legs, arms, ankles, fingers and the spine as point s of reference. These observations also reflect the origins of hypermobility within the literature, the term

'hypermobility syndrome', first coined by Kirk et al in 1967, was initially defined solely by the presentation and identification of generalised laxity within joints (Grahame, 1971). More specifically the dance master observations also corresponded to the later nine point Beighton criteria (Beighton, 1973). This is despite the fact that some of the points recognised in the criteria (and also by the dance masters) represent extreme mobility in joints, such as the fingers, that are not normally exercised and trained for extreme flexibility in classical ballet. This again confers with Grahame's initial (1971) observations and clarification between acquired and heritable hypermobility (Grahame in Kerr and Grahame, 2003, p2) however the dance masters did not refer to these clinical terms.

Both Jean and Jannis readily discussed and showed a strong consensus that there were also intellectual and pedagogical implications connected to hypermobility however they were still hesitant in applying the term hypermobile directly to dancers when they recalled them in their interviews. Both also recognised that hypermobility would influence a dancer's performance pathway. For example Jean discussed the implications of a dancer's 'fixation' referring to this as a fixation with 'technique' whilst implying that hypermobility was the technique that they were fixating on. She discussed how this technique was detrimental and obstructed the dancer to 'move on' and explore the intellectual, expressive and creative side of their performance. In Polanyi's terms therefore Jean is referring to a dancer's fixation on the proximal or focal internal processes and features of hypermobility and is unable to move on to the distal application of these processes to create meaning and art (see Polanyi , p19).

Jean also articulated her views of the 'intellectual' in a similar dance context to Forster (1997) who coins the term 'bodily Intelligence' (Forster in Desmond, 1997). In a similar way , the seminal text ' *The Thinking Body'* observes the dynamic body (Todd, 1937)and confirms the reciprocal affection between mental and bodily attitudes. This reciprocal affection has latterly been re-articulated through the interest of cognitive scientists who

ascribe positive or negative intensification of a sensation or feeling to a learned response in the brain (Fensham, 2014). It is also argued that the "discovery" of mirror neurons and the plasticity of the brain can be applied to the human capacity through 'Kinaesthetic empathy' whereby the observer of dance can feel and respond the others moving (Reason & Reynolds, 2010). This concept also forms part of tacit understanding whereby

> *"…the performer coordinates movements by dwelling in them as parts of his body while the watcher tries to correlate these moves by seeking to dwell in them from the outside"* (1966, p30)

This suggests that tacit understanding and kinaesthetic empathy may be intensified by drawing on personal experience. Bergson in Riley (2004) likewise discusses clarification and intensification of perception through attention and he refutes the belief that the *"eyes of the body, or those of the mind, cannot see more than they see"* stating that

> "*For hundreds of years, in fact, there have been men whose function has been precisely to see and to make us see what we do not naturally perceive. They are the artists"*. (Riley, 2004)

Jean also discussed that becoming a great artists meant letting go of any fixation towards hypermobile abilities,

> "*these are performances by dancers who have been able to let go of the hypermobility and let go of the technique, it is there it underpins what they do, but they are then free to explore, and quite honestly there are not too many dancers that have arrived at that, that is the difference between the Principle dancers, Great dancers and Soloists and Corp de ballet dancers…* (JE line 75-78, p2)

Indeed the initial reluctance from both bJean and Jannis to discuss the concept of hypermobility at all with the researcher is interesting and may tell us more about their understanding. Jannis initially declared that she had not heard of hypermobility however when further questioned she was very adept in describing and articulating the differentiating physical characteristics that are associated with a hypermobile aesthetic in dance. This suggests that Jannis potentially had a more tacit understanding of hypermobility (from

personal experience) without perhaps knowing or focusing on an explicit understanding (through learning or reading) about it. Jean also needed confirmation of her own tacit understanding of hypermobility and asked the researcher to clarify and explain what was meant by the term hypermobility before she also proceeded to readily discuss her experience of hypermobility in dance within a very similar context of the other three participants. Jean and Jannis were indeed the oldest and therefore most experienced dance masters of the group having worked as principle (and hypothetically hypermobile) dancers themselves before using this experiential knowledge when become expert teachers and dance masters. Their understanding of teaching dance and pedagogy was truly experiential, working with both vocational (trainee) and professional dancers in many contexts. Carolyn on the other hand had undergone professional development, formally training to become an osteopath and Pilates teacher since retiring from her own performance career, her understanding of hypermobility was therefore more objective although not exclusively explicit. This is evident in her approach to dealing with the dancer' instability, she discussed working with them on unstable surfaces knowing that this would help with their proprioception. Carolyn demonstrated that she understood the proximal and focal attributes of the hypermobile aesthetic and how this physically effected balance and stability however she did not have enough understanding to be able to articulate why this was so terrifying for them. This suggests that Carolyn's understanding of the tacit and distal aspects of how this instability feels to a hypermobile dancer was to a much lesser degree. A similar lack of understanding is also common across medical practitioners, hypermobility (and in particular for hEDS) is a condition that physiotherapists frequently and openly admit that they are at loss to know how best to help patients presenting with hypermobility (Hakim, A., & Grahame, 2003).

Furthermore a recent study showed that many physiotherapists (70.4%) declared a

need to know more about the condition and consequences and made requests regarding specific management suggestions (Rombaut et al., 2015a).

Understanding hypermobility includes not only the recognition of its bio-logical presentation but also the associated psychological traits and how these in turn may affect social elements. To this end, the results presented in the word-clouds gave indication that the participants did also have an awareness of some the psychological and social implications for the dancers they worked with. *Anxiety, disruption, emotions, feeling, insecurity, intelligence, self-regulation* and *expression* were words that were readily used in the interviews in association with the dancers that they recognised within the hypermobile aesthetic. Their understanding of hypermobility and associated psycho-social traits also correlates to the literature and current research (Carolina Baeza-Velasco, Bulbena, et al., 2018; Carolina Baeza-Velasco et al., 2015a; Bulbena et al., 2017b; Sinibaldi et al., 2015). The dance masters also highlighted the role of the *choreographer, feeling-desired, accepted* and *wanted* which suggest a more distal and tacit understanding of some of the social consequences of the hypermobile aesthetic in dance. Social implications of hypermobility within the dance environment have however yet to be properly researched and therefore do not feature in the literature. Generally the psych-socio implications of hypermobility such as anxiety and emotional fragility are reported and described as secondary complications and psycho-social issues (Simmonds and Kerr, 2007). Knight (2011) also explores the role of doctor/patient relationships within hypermobility and highlights the importance of trust. Being accepted as a patient with very specific needs is key within this relationship and the acceptance of the psycho-social symptoms of hypermobility is a critical part of this (Knight, 2011). Acceptance and trust were also a feature within the dance master interviews. For example, the issue of 'differing opinions' between dance masters and teachers about the hypermobile aesthetic with 'mixed messages' being given and beliefs on how to train hypermobile knees was a characteristic

feature in Jannis' interview. She discussed the confusion amongst teachers as to how a dancer could achieve the strength to control the hyperextension of the knee and how this is maintained (JA lines 13-21, p1). This is something that is readily debated in many dance teachers forums and particularly applies to the hyperextended knee in an open (gesture/*en l'air*) chain or closed chain (supporting) posture. Trust was also a feature in Carolyn and Michael's interviews however they discussed this from a more 'proximal' viewpoint than Jannis and Jean. For example Carolyn had an explicit understanding of the hypermobile anatomy however she still found it hard to understand why a hypermobile dancer should be 'unnecessarily insecure' and 'over-reliant' on her. The potential psycho-social implications and insecurities that remain part of the distal aspects of the hypermobile aesthetic are perhaps what Carolyn was overseeing because she was focusing on the proximal aspects such as the range of motion, strength and control acquired through hours of dance training. Similarly Michael also referred to the less experienced dancers and their need for guidance and mentoring in order to understand that if they used their hypermobile extensions without having the physical control then they would become vulnerable to physical injury and ultimately psychological distress.

Trust and acceptance were for an issue for Jean, she described her relationship as a pedagogue with some of the hypermobile dancers and how working on a trusting coach/performer relationship increased the dancers confidence stopping them becoming fixated so that they don't think that "*they have to lift their legs as high as thy possibly can"* (JA line 28-29, p1). Jean conferred how she sometimes found this relationship hard when dancers are encouraged by choreographers to exploit their hyperextension and ultimately be chosen for the work, she believed this type of dance bordered on "B*allet Sport"*. Foley and Bird (2010) likewise discuss that hypermobile dancers are often left not knowing if they are robust enough to make the grade or if their prolonged dedication and intensive training has

been negated because their body structures are genetically pre-determined to not cope with the brutal demands of a career in classical Ballet. When asked to comment on the mental wellbeing of the hypermobile dancers that they worked with (in relation to dancers who were not identified as hypermobile in their companies), Jannis initially thought that it was "*very far-fetched; sorry"* (JA line 69, p2) to even consider that there may be any difference. However she rapidly reflected on this statement and proceeded to describe how she could clearly distinguish the different mind-sets of those being encouraged to dance purely because of a the extra-ordinary physical attributes that go hand in hand with hypermobility as opposed to *"...really wanting to dance"* (JA line 79-80, p2). Jean also suggested that those who make it who are not hypermobile have a certain toughness and determination that is often not seen in the hypermobile dancer. Indeed she suggested that those who are pushed in to dance because of their hypermobile attributes are often the ones who give up before they get to the top of their game. Jean and Jannis' understanding of mental toughness and resilience in dancers suggest a deeper understanding of what it means to be hypermobile or not. Jean believed that over time, the intelligent dancers, the ones she also referred to as thinking dancers, developed a resilience to the 'emotional quandary' they experienced and developed the capacity to be great dancers .

The dance masters' perception of intelligence (or lack of) differed in degrees however this theme featured in the interviews as an integral part understanding hypermobility, they believed that an intelligent 'thinking dancer' could overcome the physical and psychological challenges they experienced and potentially turn them to their advantage. Jannis and Jeans' interpretation of the hypermobile aesthetic in this context may potentially evidence an alternative understanding of the data presented in the follow up longitudinal study by McCormack , Hakin, Briggs and Grahame (2009). In this study hypermobile dancers were tracked longitudinally through the upper school at the Royal Ballet, into and up the

company's hierarchical process. The findings showed that the higher the ranking in the company the less prevalence there was for hypermobility, McCormack et al (2009) suggest that the reason for this is that these dancers have been injured out of their careers before reaching the top however if we consider Jean and Jannis' interpretation then these dancers may have been selected for their hypermobile abilities and actually not have had the burning desire that is needed to reach the top.

Overall the data suggests that there is a common consensus that dance masters do understand hypermobility in the dance environment to varying degrees. Their understanding of what hypermobility means within a dance context is evidently dependant on their explicit, implicit and tacit knowledge and experience of the condition. All dance masters in the study understand the hypermobile aesthetic, how this works and influences current trends in choreography and how this in turn drives the strategic planning of dance companies and theatres and ultimately the demand for this type of dancer. The dance masters opinions and views of the implications and meaning of being hypermobile were however varied and less objective. This was dependant on; tacit and personal experience of hypermobility and experience of teaching a range of hypermobile dancers, for example young dance students right through to apprentice and principle dancers.

So finally, what are the dance masters' beliefs about hypermobility? Similar to the dance masters' understanding of hypermobility, their beliefs of hypermobility also varied. Their beliefs surrounding the asset v liability debate for example was addressed and the general consensus was that both terms applied to varying degrees. The notion and debate for this thinking is also well contested in the literature (Foley and Bird, 2013; Larsson, Mudholkar, Baum and Srivastava, 1995; Larsson, Baum, Mudholkar and Kollia 1993; Grahame and Jenkins 1973). The most recent consensus is that within the dance environment it can be both needing careful management and a strategy for the future care and training of hypermobile

dancers (Foley & Bird, 2013b). Three (Jannis, Jean and Michael) of the participants did not acknowledge or refer to literature or bulletins that they had read when giving their opinions suggesting that their views were based on anecdotal experience alone. Carolyn was the only one to discuss a dance medicine conference where she had seen a paper presented about hypermobility and therefore had further objective insight, however they all confirmed and agreed with the notion that the preconceptions and values that the dancers themselves and those who train them have about hypermobility are important in determining if hypermobility can be articulated to become a dancer's strength as opposed to an 'Achilles heel'. The liability within hypermobility for professional dancers is that they have a prevalence of low levels of physical fitness, increased levels of fatigue, higher levels of stress and anxiety, including obsessive compulsions in comparison to their non-hypermobile colleagues (Foley and Bird, 2013; Grahame and Jenkins, 1972). These weaknesses were also confirmed by all four participants who each in their own way believe that they can recognize the hypermobile dancer through the physical weaknesses, lack of dynamics and ability to jump, alongside obsessive fixations, insecurities, and also poor intention to work hard to achieve and self-regulate.

The advantages of being a hypermobile dancer were also readily discussed by the participants, when asked how they perceived hypermobility in the dancers that they worked with Jean believed that hypermobility can easily be an asset if *"a dancer works to achieve it"* clarifying that she was not referring to achieving the increased flexibility that comes with hypermobility but achieving hypermobility as part of the control and technique a dancer requires to become great. She believed that a hypermobile dancer needs to understand the limitations that come with hypermobility and not go *"any further for fear that they will become injured".* Jannis also believed that hypermobility if understood can be a good thing, hypermobile dancers she explained *"can either use the mobility or not… they can always*

lower the legs whereas a non-hypermobile dancer can only achieve the extension they have". The common belief across all participants was that hypermobility had to be managed, and not only teachers and choreographers needed to understand the implications but also the dancers themselves as they were the ones who ultimately needed to manage their bodies intelligently. Using the 'varying degrees of understanding' (as described by Kelp 2015) and Polanyi's theory of tacit knowing as a means of understanding also play a role in the dance masters' beliefs about hypermobility. Polanyi theorizes indwelling and proposes indwelling as a means of interiorization and acceptance in relation to moral teaching and beliefs. He believes that "*to interiorize is to identify ourselves with the teachings in question"* and he pertains that *"…true knowledge lies in our ability to use it"* (Polanyi, 1966, p17). In the context of the dance masters then this would suggest that a dance masters' degree of understanding and their individual application of explicit, implicit and tacit knowledge about hypermobility will also influence their beliefs about it. The hypothesis that Jean and Jannis are themselves hypermobile may then in this sense suggest a more intense level of indwelling and interiorization and stronger beliefs about hypermobility as opposed to Carolyn and Michael who have a different less embodied experience of hypermobility.

Chapter 7 General discussion

7.1 Introduction

Each of the four studies provided data and evidence that are reported in this thesis. While each set of results and findings have been set out and discussed in separation, each establishing an individual discreet study, links between the four studies are established across the overall thesis and are discussed in this chapter. The picture that has emerged overall is one of an entangled and somewhat paradoxical positioning for hypermobility within the context of classical ballet dance.

It remains now in this final chapter to discuss the emerging juxtaposition of generalised joint hypermobility and to position this further in the context of the overarching aims of the thesis. These set out *to explore the health risk and experience of generalized joint hypermobility within a classical ballet narrative*. The rational for this was with a view to further understand to what extent GJH is known and understood in the classical ballet environment and to determine if GJH presents as a risk to dancers?

The lens applied to the study was biopsychosocial and throughout the development of the three studies a clearer representation of bio and psychosocial markers for GJH within classical ballet dance has emerged. Implications of these markers within the teaching, training and performance environment for young classical ballet dancers have also emerged. In addition evidence about what exactly the lived experience of GJH is within the context of classical ballet has also evolved, however as always, further questions have also materialised.

Building on the evolving literature whereby GJH was no longer isolated within biological domains, the application of a biopsychosocial approach to the research indeed helped to ease the emergence of new information about hypermobility in a classical ballet dance setting. Interestingly, much of the recent research for hypermobility in general actually

manifested by default following astute observations within clinical settings and data bases relating to mental health and wellbeing and the presentation of hypermobility was indeed a secondary condition in the clinical history of patients (C. Baeza-Velasco et al., 2011; Carolina Baeza-Velasco, Bourdon, et al., 2017b; Carolina Baeza-Velasco, Pailhez, Bulbena, & Baghdadli, 2015b; Pasquini et al., 2014). Similarly the initial draw to this research was also born through a prying need to understand why some dancers in the dance studio commonly operated in a different way?

At the onset the aspiration was that the findings from this research might inform just how GJH can potentially influence a dancer's career. In doing so this may also inform future practice in classical ballet and the art-form itself.

7.2 Synthesis of key findings

So what exactly have the three studies provided and added to our knowledge and understanding of this complex condition? Whilst not always categorical, the evidence that has emerged does support and build on the general consensus of GJH in recent clinical settings. That is that GJH is indeed more complex and much greater than the sum of its parts. For the dance environment, this thesis therefore provides alternative viewpoints towards the genetic disorder and heritable condition of hypermobility, potentially providing a foothold for further investigation and enquiry.

The findings of this investigation challenge the longstanding debate 'asset or liability' in dance (Marco Castori & Colombi, 2015; Foley & Bird, 2013a; Grahame & Jenkins, 1972; Rietveld, 2013) and the entangled position of hypermobility within classical ballet dance has potentially taken on a fresh perspective. So what does this mean? Thus far the debate has focused entirely on what hypermobility can or cannot bring to a dance artist. Evidence in this research suggests that classical ballet dance may well be the unexpected and stronger

counterpart acting as a protective factor towards the fragility that GJH brings to the dance environment.

At the onset, study I tackled understanding the mechanisms for correctly identifying and demonstrating the prevalence of hypermobility within dance. It also provided evidence towards musculoskeletal injury in dance disputing that hypermobility in the training environment could be named as a cause for concern within the vocational context. The fact is that musculoskeletal injury is part of dance just as it is part of any sport at an elite level (Novosel et al., 2019; Smith et al., 2015). Confirmation was provided early on in study I that signposting towards dance was indeed a fact to be seriously considered throughout the remaining exploration. The signposting and prevalence also fuelled the quest as a practitioner and researcher to understand GJH further. If GJH were to present in up to six of ten dancers in the studio then a teacher should know more and understand GJH better.

The emerging literature reviewed in this thesis also went beyond the musculoskeletal for hypermobility in general and suggested that the recurring 'asset liability' debate for dancers was perhaps short sighted and ill placed. This was also further confounded in study I that reported no significant relationship between GJH and injury prevalence within the specific vocational setting for classical ballet. Whilst these findings cannot be generalised for GJH within classical ballet and dance in general, they are representative of a cross sectional group of young dancers in full time training in Scotland.

Similarly, study II provided evidence to suggest that proprioception, one of the characteristic components that is characteristically in deficit in GJH (Fatoye, Palmer, Macmillan, Rowe, & van der Linden, 2009; Ghibellini, Brancati, & Castori, 2015), was potentially improved within the elite classical ballet training environment suggesting potential protective factors for GJH within a vocational classical ballet training. Additionally in study II it emerged that GJH whilst may be linked with increased anxiety however some

components of interoception (e.g. emotional awareness) may also provide enhanced emotional integrity that is beneficial to the aesthetics of dance.

The timely consortium on hypermobility (Castori et al., 2017) also provided clarity through its up-to-the-minute framework and positioning of joint hypermobility as a common and key feature within other more complex disorders. The downside within this reporting was to learn that the most common type of hypermobility (hypermobility type Ehlers Danlos syndrome hEDH) and the sub types of heritable spectrum disorders (GJH and HJH) that present within hEDS were frequently asymptomatic and therefore potentially also under-reported (Malfait et al., 2017). Therefore investment into discovering the genetic origins through DNA testing was channelled towards the seemingly more complex and life threatening forms of hypermobility, regrettably hEDH remains the only form of EDS that has yet to be genetically coded (Malfait et al., 2017; Castori 2017).

Psychosocial complications within hypermobility also appeared in the literature during the undertaking of this thesis, these were clearly evidenced in emerging publications (Bulbena et al., 2017b) which helped to inform the design and purpose of the studies II, III. Meanwhile at the back of the study design were many years of personal experience that the researcher really could not overlook. This was dealt with carefully using a researcher's memo that provided opportunity for objective and careful reflection throughout the research (Maxwell, 2013). The memo provided further evidence, albeit anecdotal, that dancers with hypermobility needed further investigation if this research were to provide understanding and clarity for the dance environment that went beyond the musculoskeletal.

Study II was quantitative and did provide insight despite the reality that some of the data was not fully conclusive. The proposals in the literature that aspects of body awareness (interoception and proprioception) pay a key role towards the experience of hypermobility were conferred. (Mallorqui--Bague et al., 2014; Núria Mallorquí-Bagué et al., 2016). The

data clearly demonstrated that dancers with hypermobility reacted more to cognitive overload resulting in anxiety whilst aspects of interoception, such as noticing, worrying and distracting were also significantly different to dancers who were not hypermobile. Whilst not conclusive the results presented credible support towards the proposed association of hyper-arousal, anxiety within bodily crisis (Toner et al., 2016) and the notions that dancers with hypermobility may have a window of tolerance (Ogden et al., 2006; Siegel, 2010) that is significantly different to other dancers. The proposition that Porges' vagal theory (Porges, 2017) may also provide insight into hypermobility was not conclusive as the HRV data could not be analysed. That said, it was not disproven and remains as a future challenge to the researcher. Interestingly, enhanced emotional awareness within hypermobility was conferred suggesting that this may be a reality that goes beyond the physical parameters of extreme range of motion for signposting hypermobility towards dance.

Evidence from study III in part quantified and conferred that the reality of hypermobility in dance was indeed different to hypermobility within the general population. It still remained a challenge however to understand how the altered parameters within hypermobility were actually experienced. Study three was therefore designed to provide insight in to the lived experience of hypermobility within dance. The dancers and teachers recruited for these final studies were experienced in their field yet not necessarily informed about hypermobility. The dancers interviewed were confirmed as hypermobile yet did not necessarily identify as hypermobile. This provided lived insight that was not contaminated by opinions or the 'labelling' that often comes with hypermobility. They gave honest, pragmatic and sometimes brutal accounts of the experience of being seemingly 'talented' merely because of how they were genetically wired. Discussions provided further evidence to suggest that the fight, flight and disassociated states proposed within the polyvagal theory (Porges, 2017) were indeed an all too readily experienced reality. The need for time and

space in order to feel safe were also common realities for these dancers. Coping with the biopsychosocial implications of their embodied experience of hypermobility had indeed developed resilience whilst their success was often envied by others as a constituent of their hyperextended limbs and ability to express a desired aesthetic. The interviews were retrospective in nature and signposted a need for mentorship and guidance at an early stage in a dancer's career that could potentially protect against the 'use' of their genetically different hypermobile physiques for creative exploration in choreographic practices.

The dance masters (some) in study III on the other hand, were in part in denial, refusing to believe that hypermobility could be anything more than a physical default that was 'yes' challenging to train yet signposted talent and potential. Others agreed that hypermobility was currently and regrettably over-recruited for in dance companies, with audiences and choreographers alike wanting to witness a spectacle and the extreme without realising the cost that extended beyond the bio markers of hypermobility. Extra time and support were not an option and the dancers in study III had clearly learned to cope, however at what cost was not clear.

Emerging from this research is consequently something that is potentially overlooked, that is that whilst hypermobility is clearly signposted towards dance, dance may also be an important protective factor for those with hypermobility. Whilst injury is known to be associated with the fragile tissue common in hypermobility (Bojan Bukva, Goran Vrgoč, Dejan Madić, Goran Sporiš, 2018; Day et al., 2011a; Teitz & Kilcoyne, 1998; Weber, Bedi, Tibor, Zaltz, & Larson, 2015), there is no evidence for example that the dancers with hypermobility in study I were any more susceptible to injury than the dancers who did not present with hypermobility. Likewise in study II balance was not in any apparent deficit for the participants with hypermobility whereas it is known to be an issue for those with

hypermobility in general (Scherper et al., 2013; Marulli, Harmon-Matthews, Davis-Coen, Willigenburg, & Hewett, 2017).

Finally, with regards to the overall research aims of the thesis, the three studies have provided evidence to suggest that while GJH may be a health risk and indicator of further complications and disorders in general, a specialist environment, such as the training and performance environment for vocational classical ballet dancers, is conducive and may also be protective for young dancers who present with GJH. Supplementary to this, the experience of hypermobility that is documented in study III clearly suggests that GJH in the classical ballet world is by no means understood, known or acknowledged and that the pseudonym of *Psyche* (the girl that was coveted but never understood) chosen for study III may well be the most appropriate name to currently attribute to GJH. The *Psyches,* or dancers with GJH, who withstand the professional environment for dance appear to develop coping mechanisms and thrive, the cost that they pay for this remains unknown just as how many dancers with GJH do not withstand the training is also unknown. Whilst new evidence pertaining to GJH in classical ballet has been generated in this thesis, further research is needed towards GJH within classical ballet and dance environments in general if it is to really be understood.

7.2.1 Reflection and personal lessons learned

I have added this paragraph to briefly indulge in my own reflection and personal journey within this research. Indeed, reflection is something that features significantly within the classical ballet environment, for dancers and practitioners and from start to finish reflection is the key to learning. It features as a mirror for perfecting skills and techniques to a means for professional and personal development. My journey and role in this environment continues, my awareness of hypermobility, initially inherent and contained in who I was, has grown to the point that my understanding of hypermobility is now, and will remain,

conclusively not only part of who I am but also a significant part of my practice. Some of the questions I had at the start of this journey have been answered, many more however have arisen. I now know a lot about hypermobility however I also know that there is much more to learn and even more to be discovered as the science around hypermobility develops. Hypermobility and dance have always been and will be strangely entangled, mutually compatible yet also complex. At a very basic level my understanding of this condition has provided insight to me personally with regards to my family. As a dancer, I wish that, I and others, had known more about hypermobility as this could have given insight to challenges I faced and the perceived ease with which I accomplished other skills. Alternatively, I believe that 'not knowing' was a construct of the resilience that I developed as a dancer. Now, as a practitioner, researcher and academic I hope that I can use this journey to guide others to understand and not be afraid or in awe of hypermobility.

7.3 Implications, recommendations and applications within dance practice.

As a longstanding teacher of classical ballet and dance in general and a teacher for trainee dancer teachers I feel compelled to make some unpretentious recommendations from the findings of this study.

1. GJH is clearly prevalent in classical ballet, Dance bodies and organisations for dance should therefore include evidence to raise awareness for GJH within the professional learning and certification they provide for teachers.
2. The desire for the hypermobile aesthetic remains as popular currency within dance. The companies, and directors should then also consider engaging further with emerging evidence that situates GJH within the biopsychosocial wellbeing of dancers and provide support and continual professional learning opportunities for their dancers, choreographers and dance masters.

7.4 Limitations

As with any research, there are always limitations, the cross sectional study in study I for example surveyed young dancers for both injury and GJH. The participants recruited for the study were representative of the dance environment with more than double the number of females to males, this however meant that the sample was not homogenous and this may have affected the findings. Maturity was also not considered within this initial study I which may have also been detrimental to the findings. This was mitigated in study II following further reading and research, there were however other limitations within study II, The main limitation was the technology chosen to record the HRV which unfortunately did not record the data as planned. This is a common problem when using equipment that is manufactured for sports in dance. Rarely does such equipment take into consideration the physiques of young dancers which are commonly very petite. Study III did have a more homogenous representation of male and female dancers in the participant group, the dance master sample however was not as equal meaning that there was only one male and three female participants. Whilst the participants were very experienced dance masters providing excellent information power in the sample it remains that there was only one male in the group. Overall and in hindsight there could have been other and possibly more efficient ways of investigating the health risk and experience of dancers with GJH. The study however commenced at a time when the literature about GJH was very indistinct. More recent classification and criteria relating to GJH has now enabled further clarification that would have enhanced studies I and II greatly.

7.5 Future Directions

The findings from this study suggest that whilst hypermobility appears to be signposted towards dance, dance itself appears to be a protective factor for the dancers with GJH. A future direction for both researchers and practitioners would be to devise practical methods to

support teachers and dancers alike to better understand hypermobility within the dance environment

An additional consideration remains for the research undertaken here to be replicated for other activities. Whilst this study was specific to the dance environment, hypermobility also remains a prevalent body type in other sports and activities that require extreme range of motion, such as for example gymnastics. Hypermobility is also prevalent within other performing artists such as for example musicians. There is scope therefore for some of the methods used in this study to be replicated in the future and applied across other disciplines

7.6 Conclusion

Finally and to conclude, this thesis provides evidence of the multi-faceted nature of GJH within the classical ballet context. Evidence supports that GJH is prevalent in young classical dancer however it appears that GJH is not associated with injury, possibly due to the protective feature of their dance training. It is also highlighted that are further aspects that intersect the psychosocial domain of body awareness and anxiety in dancers with GJH. This study however provides clear evidence to suggest that GJH signposts dancers towards dance for both physical and emotional capacities. It also provides evidence towards altered body awareness including enhanced body mind connections and increased anxiety within GJH. Finally, whilst the dancers and dance masters clearly recognised the physical experiences of GJH they demonstrated disconnect between the psychosocial aspects they discussed and the implications of these with GJH. This brings new insight to the experience of GJH that potentially has wider implications for dance teaching learning and performance whilst also potentially influencing the creative process for dance. This may further inform practice to provide insight in to the psychosocial ramifications of this condition and a healthier environment for dancers with GJH. This is particularly noteworthy within the education for dancers and dancer teachers and potentially other aesthetic sports where GJH is prevalent.

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Appendix 1

a) Study I

Participant Informed consent forms

Study I Part 1
A study to determine the reliability and stability of Beighton score measures

THE UNIVERSITY *of* EDINBURGH
Moray House School of Education

The participant informed consent form for parents of dancers under 18

I am an academic researcher at the University of Edinburgh. I would like to invite your child to take part in a small reliability study that forms part of my Doctoral degree. This form provides you with information about the study and the researcher Wendy Timmons. Please read the information below and ask any questions you might have before deciding whether or not to take part.

The aim of the study
To determine the reliability and stability of measures of flexibility

What the study involves

1. As part of the annual screening that takes place at your dance school your child will be asked to take part in a short study whereby elements of their flexibility that includes ham string flexibility, knee and elbow extension, and flexibility in the hands (thumb and fifth finger) will be measured and recorded for this research.
2. These measures are part of the annual screening however for this study they will be taken by two different researchers (physiotherapist and dance teacher) two times. This may delay your annual screening slightly but no more than 30 min.
3. Confidentiality

- The information that your child provides in this process will be **confidential** and only used for this research purpose.
- Only my supervisor at the university and myself (contact details given below) will access this information. Significant others such as your dance teachers and peers **WILL NOT** see the information to identify you as individual.
- The data related to the flexibility screening that your child is involved in will be destroyed after the completion of the study, and will be no longer available to access.

Your child's participation during the study is entirely **voluntary**. Your child does not have to answer any questions they do not want to. Your child can also stop participating at any time and there will be no loss of benefits.

Please check below if you understand and are willing to allow your child to participate in the study.

- ☐ I understand the purpose of the study.
- ☐ I am aware that I can withdraw at any time without any penalty.
- ☐ I agree to participate in the study as outlined to me.

Signed
Printed name
Consent taken by
Date

Please feel free to contact with either my supervisor or myself if you have any questions or concerns.

Researcher
Name: Wendy Timmons

Supervisor
Name: Professor John Sproule

E-mail: Email:

Study I Part 1
A study to determine the reliability and stability of Beighton score measures

THE UNIVERSITY of EDINBURGH
Moray House School of Education

The participant informed consent form for dancers under 18

I am an academic researcher at the University of Edinburgh. I would like to invite you to take part in a small reliability study that forms part of my Doctoral degree. This form provides you with information about the study and the researcher Wendy Timmons. Please read the information below and ask any questions you might have before deciding whether or not to take part.

The aim of the study
To determine the reliability and stability of measures of flexibility

What the study involves

1. As part of the annual screening that takes place at your dance school you will be asked to take part in a short study whereby elements of their flexibility that includes ham string flexibility, knee and elbow extension, and flexibility in the hands (thumb and fifth finger) will be measured and recorded for this research.
2. These measures are part of the annual screening however for this study they will be taken by two different researchers (physiotherapist and dance teacher) two times. This may delay your annual screening slightly but no more than 30 min.
3. Confidentiality

- The information that you provide in this process will be **confidential** and only used for this research purpose.
- Only my supervisor at the university and myself (contact details given below) will access this information. Significant others such as dance teachers other than the researcher and peers **WILL NOT** see the information to identify you as individual.
- The data related to the flexibility screening that you are involved in will be destroyed after the completion of the study, and will be no longer available to access.

Your participation during the study is entirely **voluntary**. You does not have to answer any questions they do not want to. You can also stop participating at any time and there will be no loss of benefits.

Please check below if you understand and are willing to allow your child to participate in the study.

- ☐ I understand the purpose of the study.
- ☐ I am aware that I can withdraw at any time without any penalty.
- ☐ I agree to participate in the study as outlined to me.

Signed
Printed name
Consent taken by
Date

Please feel free to contact with either my supervisor or myself if you have any questions or concerns.

Researcher
Name: Wendy Timmons
E-mail:

Supervisor
Name: Professor John Sproule
Email:

Study I Part 2
Surveying the prevalence of hypermobility and musculoskeletal injury in young pre-vocational dancers within a Scottish context

THE UNIVERSITY of EDINBURGH
Moray House School of Education

The participant informed consent form for parents of dancers under 18

I am an academic researcher at the University of Edinburgh. I would like to invite your child to take part in a project that forms part of my Doctoral degree. This form provides you with information about the study and the researcher Wendy Timmons. Please read the information below and ask any questions you might have before deciding whether or not to take part.

The aim of the study
To survey the prevalence of hypermobility and musculoskeletal injury in young pre-vocational dancers within a Scottish context

What the study involves

1. Your child will be asked to take part in a 30 min screening whereby elements of their flexibility that includes ham string flexibility, knee and elbow extension, and flexibility in the hands (thumb and fifth finger) will be measured and recorded.
2. Your child will then be asked to fill in an injury survey that will require them to recall injuries during the past year of their dance training. The researcher will be on hand while they do this to answer any questions the pupils may have with regards to the survey.

Confidentiality

- The information that your child provides in this process will be **confidential** and only used for this research purpose.
- Only my supervisor at the university and myself (contact details given below) will access this information. Significant others such as your dance teachers and peers **WILL NOT** see the information to identify you as individual.
- The data related to the screening and injury survey that your child is involved in will be destroyed after the completion of the study, and will be no longer available to access.

Your child's participation during the study is entirely **voluntary**. Your child does not have to answer any questions they do not want to. Your child can also stop participating at any time and there will be no loss of benefits.

Please check below if you understand and are willing to allow your child to participate in the study.

- ☐ I understand the purpose of the study.
- ☐ I am aware that I can withdraw at any time without any penalty.
- ☐ I agree to participate in the study as outlined to me.

Signed
Printed name

Please feel free to contact with either my supervisor or myself if you have any questions or concerns.

Researcher
Name: Wendy Timmons
E-mail: Wendy.Timmons@ed.ac.uk

Supervisor
Name: Professor John Sproule
Email:

Study I Part 2
Surveying the prevalence of hypermobility and musculoskeletal injury in young pre-vocational dancers within a Scottish context

THE UNIVERSITY of EDINBURGH
Moray House School of Education

The participant informed consent form for dancers under 18

I am a dance teacher and academic researcher at the University of Edinburgh. I would like to invite you to take part in a project that forms part of my Doctoral degree. This form provides you with information about the study and the researcher Wendy Timmons. Please read the information below and ask any questions you might have before deciding whether or not to take part.

The aim of the study
To survey the prevalence of flexibility and musculoskeletal injury in young pre-vocational dancers within a Scottish context

What the study involves

1. You will be asked to take part in a 30 min screening whereby elements of your flexibility that include ham-string flexibility, knee and elbow extension, and flexibility in the hands (thumb and fifth finger) will be measured and recorded.
2. You will then be asked to fill in an injury survey that will require you to recall injuries during the past year of your dance training. The researcher will be on hand while they do this to answer any questions you may have with regards to the survey.

Confidentiality

- The information that you provide in this process will be **confidential** and only used for this research purpose.
- Only my supervisor at the university and myself (contact details given below) will access this information. Significant others such as your dance teachers and peers **WILL NOT** see the information to identify you as individual.
- The data related to the screening and injury survey that you are involved in will be destroyed after the completion of the study, and will be no longer available to access.

Your participation during the study is entirely **voluntary**. You do not have to answer any questions you do not want to. You can also stop participating at any time and there will be no loss of benefits.

Please check below if you understand and are willing to allow your child to participate in the study.

- ☐ I understand the purpose of the study.
- ☐ I am aware that I can withdraw at any time without any penalty.
- ☐ I agree to participate in the study as outlined to me.

Signed
Printed name
Person taking consent
Date

Please feel free to contact with either my supervisor or myself if you have any questions or concerns.

Researcher
Name: Wendy Timmons
E-mail: Wendy.Timmons@ed.ac.uk

Supervisor
Name: Professor John Sproule
Email:

Study I Part 2
Surveying the prevalence of hypermobility and musculoskeletal injury in young pre-vocational dancers within a Scottish context

THE UNIVERSITY of EDINBURGH
Moray House School of Education

The participant informed consent form for the dance school

Researcher: 0131 6516596
Supervisor: Professor John Sproule,

I am an academic researcher at the University of Edinburgh. I would like to invite your school to involve in a study that forms part of my doctoral thesis. This study is the first of three and aims to investigate flexibility and musculoskeletal injury in young dancers

Background of the study
In theory dancers who are very flexible are often signposted to dance training because of their ability to meet the aesthetic demands in dance, increased flexibility is also often associated with a condition of the connective tissue known as hypermobility. Musculoskeletal injury is also often common within vocational dance students because of the intensity and nature of their training. This study aims to determine if there is a relationship between hypermobility and injury in young vocational dancers

What is involved in the study?
1. Pupils would be asked to take part in a 30 min screening whereby elements of their flexibility that includes ham string flexibility, knee and elbow extension, and flexibility in the hands (thumb and fifth finger) will be measured and recorded.
2. Pupils will then be asked to fill in an injury survey that will require them to recall injuries during the past year of their dance training. The researcher will be on hand while they do this to answer any questions the pupils may have with regards to the survey.

Possible benefits from the study
At the end of the overall study recommendations will be provided for teachers and participants. These approaches will allow pupils to pursue their passion and achieve goals in a healthy way which would have positive influences on their development across the life span.

Data Protection
Any information collected during this research will be confidential and used for this research purpose only. No information could lead to the identification of either the school or any dancer. No identifiable personal data will be published or shared with any other organisation.

Risks and inconveniences that participants might incur
As the study requires the questionnaire and the interview, there will not be any physical harms. Participants might find some questions uncomfortable to answer in which case they will have a right to stop answering at any time they wish.

Withdrawal from study
Your child's participation in the study is entirely **voluntary**; that means he/she or the school can withdraw at any stage of the project without being penalised or disadvantage in any way. Participants and their parents also have the same right; in addition, participants are free to stop answering the questionnaires and the interview when they do not want to.

Signature: _____
Date: _____
School: _____
Address: _____
School telephone number: .
E-mail address: _____

b) Study II

Study II
A case study exploring body awareness indicators of young dancers within a vocational setting in Scotland

THE UNIVERSITY of EDINBURGH
Moray House School of Education

The participant informed consent form for dancers under 18

I am a dance teacher and academic researcher at the University of Edinburgh. I would like to invite you to take part in a project that forms part of my Doctoral degree. This form provides you with information about the study and the researcher Wendy Timmons. Please read the information below and ask any questions you might have before deciding whether or not to take part.

The aim of the study

This study is to explore the body awareness of young dancers

What the study involves

1. You will be asked to take part in a 45 min screening whereby elements of your flexibility that include ham-string flexibility, knee and elbow extension, and flexibility in the hands (thumb and fifth finger) will be measured and recorded.
2. You will then be asked to complete three other tests whilst you are connected to a monitor that records your heart rate and skin conductance (one is worn like a lycra crop top and has a sensor inside it and the other is placed on a finger on the had you have free for the tapping tests)
    a. The first involves you tapping two fingers on an ipad screen, this is to determine your coordination
    b. The second tapping test you will also be asked to complete is to measure your spatial and visual memory, this is again on a screen
    c. The third tests is a simple balance test standing on one leg with your eyes open and then closed.
3. Finally you will fill in a survey questionnaire that will ask questions about the way you perceive your body. The researcher will be on hand while you do this to answer any questions you may have with regards to the survey.

Confidentiality

- The information that you provide in this process will be **confidential** and only used for this research purpose.
- Only my supervisor at the university and myself (contact details given below) will access this information. Significant others such as your dance teachers and peers **WILL NOT** see the information to identify you as individual.
- The data related to the screening and injury survey that you are involved in will be destroyed after the completion of the study, and will be no longer available to access.

Your participation during the study is entirely **voluntary**. You do not have to answer any questions you do not want to. You can also stop participating at any time and there will be no loss of benefits.

Please check below if you understand and are willing to allow your child to participate in the study.

- ☐ I understand the purpose of the study.
- ☐ I am aware that I can withdraw at any time without any penalty.
- ☐ I agree to participate in the study as outlined to me.

Signed
Printed name
Person taking consent
Date

Please feel free to contact with either my supervisor or myself if you have any questions or concerns.

Researcher
Name: Wendy Timmons
E-mail: Wendy.Timmons@ed.ac.uk

Supervisor
Name: Professor John Sproule
Email:

Study II

A case study exploring body awareness indicators of young dancers within a vocational setting in Scotland

THE UNIVERSITY of EDINBURGH
Moray House School of Education

The participant informed consent form for parents of dancers under 18

I am a dance teacher and academic researcher at the University of Edinburgh. I would like to invite your child to take part in a project that forms part of my Doctoral degree. This form provides you with information about the study and the researcher Wendy Timmons. Please read the information below and ask any questions you might have before deciding whether or not to take part.

The aim of the study

This study is to explore the body awareness of young dancers

What the study involves

1. Your child will be asked to take part in a 45 min screening whereby elements of your child's flexibility that include ham-string flexibility, knee and elbow extension, and flexibility in the hands (thumb and fifth finger) will be measured and recorded.
2. You will then be asked to complete three other tests whilst you are connected to a monitor that records your heart rate and skin conductance (one is worn like a lycra crop top and has a sensor inside it and the other is placed on a finger on the had you have free for the tapping tests)
    a. The first involves your child tapping two fingers on an ipad screen, this is to determine your coordination
    b. The second tapping test your child will also be asked to complete is to measure your spatial and visual memory, this is again on a screen
    c. The third tests is a simple balance test standing on one leg with your eyes open and then closed.
3. Finally your chid will fill in a survey questionnaire that will ask questions about the way your child perceives their body. The researcher will be on hand while they do this to answer any questions you may have with regards to the survey.

Confidentiality

- The information that you provide in this process will be **confidential** and only used for this research purpose.
- Only my supervisor at the university and myself (contact details given below) will access this information. Significant others such as your dance teachers and peers **WILL NOT** see the information to identify you as individual.
- The data related to the screening and injury survey that you are involved in will be destroyed after the completion of the study, and will be no longer available to access.

Your participation during the study is entirely **voluntary**. You do not have to answer any questions you do not want to. You can also stop participating at any time and there will be no loss of benefits.

Please check below if you understand and are willing to allow your child to participate in the study.

- ☐ I understand the purpose of the study.
- ☐ I am aware that I can withdraw at any time without any penalty.
- ☐ I agree to participate in the study as outlined to me.

Signed
Printed name
Person taking consent
Date

Please feel free to contact with either my supervisor or myself if you have any questions or concerns.

Researcher
Name: Wendy Timmons
E-mail: Wendy.Timmons@ed.ac.uk

Supervisor
Name: Professor John Sproule
Email:

c) Study III

Study III. Retrospective: the lived experience of professional classical ballet and contemporary dance.

THE UNIVERSITY *of* EDINBURGH
Moray House School of Education

The participant informed consent form for the dancers over 18 years of age

INFORMATION ABOUT THE RESEARCH

This is a small study that is part of a larger research project that is being undertaken at the University of Edinburgh by Wendy Timmons. In this part of the project we would like to find out more about the lived experience of professional dancers from classical ballet and contemporary dance practice.

WHAT WILL HAPPEN

If you agree, the principle investigator (Wendy Timmons) will contact you to arrange a convenient time and place to interview you in order to find out more about your wellbeing throughout your career. The type of questions you will be asked will be about injuries and illnesses you sustained during your career and also about performance demands that you were asked to meet whilst performing and developing new choreographic works.

WHAT WE WILL NEED YOU TO DO

Following the interview the researcher will transcribe your interview removing all identification so that the data is anonymous, the transcript will then be sent to you to read. You will be asked if the comments you made during the interview are all correct and in context, if this is the case then the researcher will use this data in the project. You will also be sent a short questionnaire to complete and return following the interview
If you do not agree with the accuracy of the transcription then adjustments will be made and the transcript will be sent back to you for further confirmation of accuracy.
We may also send you a questionnaire to complete as part of the study , this will be completed by e mail and returned to the researcher , your personal data will be protected and kept confidential at all times.

PARTICIPANTS' RIGHTS

1. You may decide to stop being a part of the research study at any time without providing us with any explanation. You have the right to ask that any data you have supplied up to that point be withdrawn/destroyed.

2. You have the right to omit or refuse to answer or respond to any question that is asked of you.

3. You have the right to have your questions about the procedures answered. If you have any questions as a result of reading this information sheet, you should ask the researcher before the study begins.

BENEFITS AND RISKS

4. There are no known benefits or risks for you in this study.

COST, REIMBURSEMENT AND COMPENSATION

Your participation in this study is voluntary – we thank you for your time.

CONFIDENTIALITY/ANONYMITY

The data we collect may contain some personal information, for example all data relating to your identity and work place(s) will be made anonymous and the coding will be held under a secure password by the principle investigator for this project, your identity will not be shared. Once we have done this, the data will be held until the research and publication process for this project is complete. During this period it will be password protected and your identity will be concealed.

FOR FURTHER INFORMATION

Please contact research supervisor, Prof John Sproule
A summary of our findings will be made available to you electronically in August 2018
Written Consent:

□ I have understood all the information on this sheet and am happy to participate in this study

Name: Date:
Consent taken by Date

Study III

Retrospective: the lived experience of professional classical ballet and contemporary dance.

THE UNIVERSITY of EDINBURGH
Moray House School of Education

The participant informed consent form for the dance masters (over 18 Yr.)

INFORMATION ABOUT THE RESEARCH
This is a small study that is part of a larger research project that is being undertaken at the University of Edinburgh by Wendy Timmons. In this part of the project we would like to find out more about your own experience of working with professional dancers from classical ballet and contemporary dance practice.
WHAT WILL HAPPEN
If you agree, the principle investigator (Wendy Timmons) will contact you to arrange a convenient time and place to interview you in order to find out more about your experience as a dance master. The type of questions you will be asked will be about your experience of working with dancers across your career, we may ask you to recall specific events in order that you are able to give some examples however at no point will we ask you to disclose any identification for the dancers that you recall or discuss.
WHAT WE WILL NEED YOU TO DO
Following the interview the researcher will transcribe your interview removing all identification so that the data is anonymous, the transcript will then be sent to you to read. You will be asked if the comments you made during the interview are all correct and in context, if this is the case then the researcher will use this data in the project. You will also be sent a short questionnaire to complete and return following the interview
If you do not agree with the accuracy of the transcription then adjustments will be made and the transcript will be sent back to you for further confirmation of accuracy.
PARTICIPANTS' RIGHTS

1. You may decide to stop being a part of the research study at any time without providing us with any explanation. You have the right to ask that any data you have supplied up to that point be withdrawn/destroyed.

2. You have the right to omit or refuse to answer or respond to any question that is asked of you.

3. You have the right to have your questions about the procedures answered. If you have any questions as a result of reading this information sheet, you should ask the researcher before the study begins.

BENEFITS AND RISKS
There are no known benefits or risks for you in this study.
COST, REIMBURSEMENT AND COMPENSATION
Your participation in this study is voluntary – we thank you for your time.
CONFIDENTIALITY/ANONYMITY
The data we collect may contain some personal information, for example all data relating to your identity and work place(s) will be made anonymous and the coding will be held under a secure password by the principle investigator for this project, your identity will not be shared. Once we have done this, the data will be held until the research and publication process for this project is complete. During this period it will be password protected and your identity will be concealed.

FOR FURTHER INFORMATION
Please contact research supervisor, Prof John Sproule
A summary of our findings will be made available to you electronically in August 2018

Written Consent:
□ I have understood all the information on this sheet and am happy to participate in this study

Name: Date:
Consent taken by: Date:

Appendix 2

BEIGHTON SCORE ADAPTED FROM NIDIMS

Equipment needed; *Goniometer, massage bed/mat*

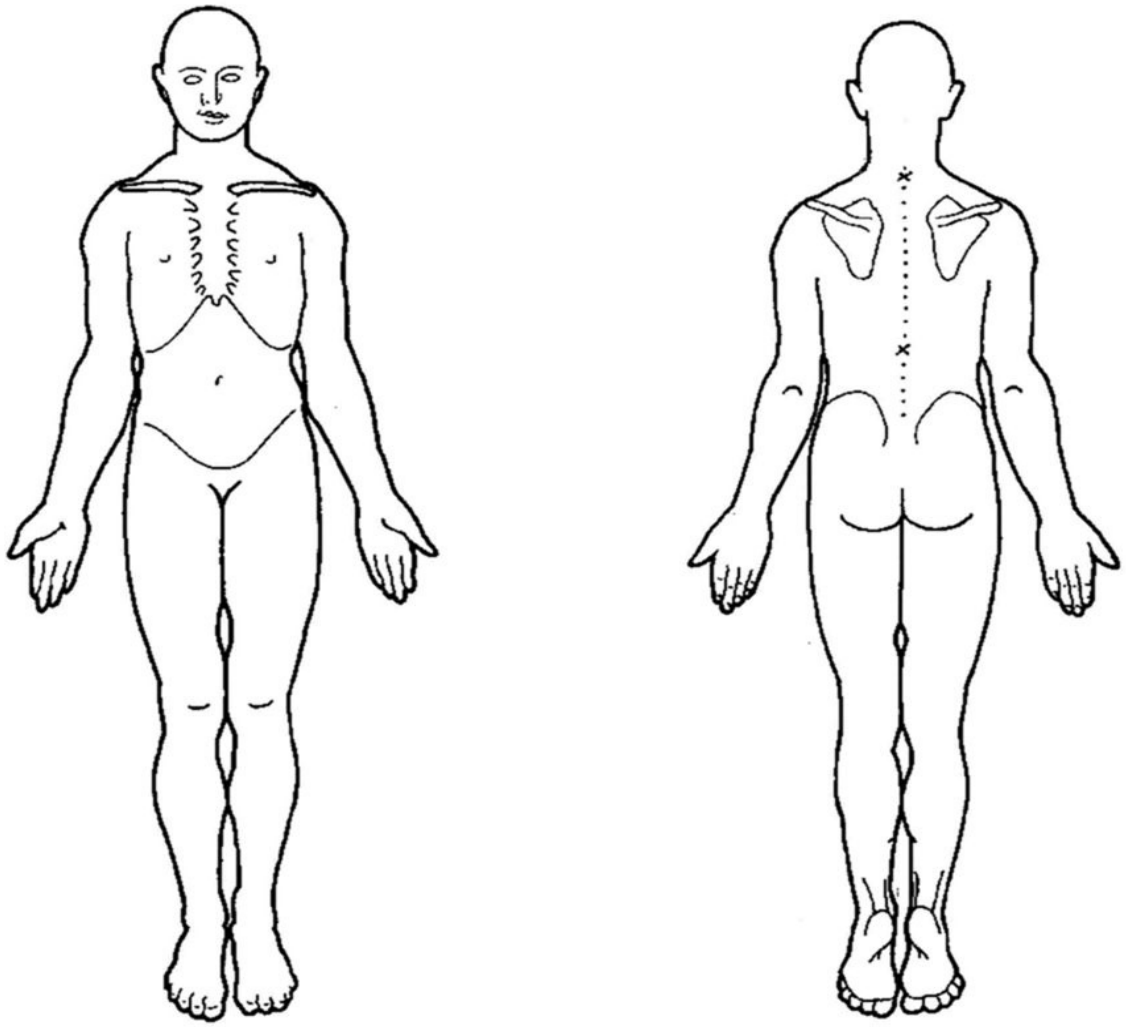
- Before the participant begins ensure their clothing allows access/visibility to their elbows and knees. Explain to the participant that you are going to be looking at joint hypermobility during these tests.
- Explain to the participant that if they feel any pain during the movements to notify and you can stop the assessment.

- Left and right 5th metacarpal-Ask the participant to stand in parallel and to place their left arm horizontally on a table, with their palm facing the floor. Take their little finger and extend it. Repeat on the right side. **Test is TRUE if finger can extend further than 90 degrees.**
- Left and right thumb-Ask the participant to stand in parallel and place their left arm in front of them, flexed. Ask them to flex and rotate their wrist and try to touch their left thumb to their forearm, using their right thumb for assistance. Demonstrate to the participant if necessary. Repeat on the right side. **Test is TRUE if thumb can touch forearm.**
- Left and right elbow-Ask the participant to stand in parallel and place their left arm horizontally in front of them, with the palm facing upwards. Assistant to hold participants arm above and below elbow, gently rotate the lower arm (supination) and then (gently) passively stretch the arm. Place the goniometer axis on the lateral side of elbow condyle, keeping the stationary arm in line with the humerus and the moving arm along the ulna (n.b. the goniometer will not be horizontal). Repeat on the right side. **Test is TRUE if more than 10 degrees.**
- Forward flexion-Ask the participant to stand with their feet in parallel and to flex forward from the hips, trying to place their palms on the ground to the side of their feet, with fingers pointing backwards. Record the position reached in accordance with the following rating:
    1. Finger tips touching shins (No for hypermobility)
    2. Finger tips touching floor (No for hypermobility)
    3. Palms flat on floor in front of feet (Yes for hypermobility)
    4. Palms flat on the floor beside the feet, finger tips facing forward (Yes for hypermobility)
    5. Palms flat on the floor behind the feet, heels of hands in line with the heels of feet and finger tips facing backward (Yes for hypermobility).
- Left and right knee-Ask the participant to lay down on a massage bed (or mat if bed not available). Ask them to lay relaxed. The assistant will (gently) passively stretch the knee by holding above and below the knee. Place the goniometer on the lateral side of the knee, with the stationary arm along the femur and the moving arm along the lateral side of the lower leg (NB the goniometer will not be horizontal). Repeat on the right side. **Test is TRUE if more than 10 degrees**.

Appendix 3
Retrospective musculoskeletal injury survey

Injury History retrospective for the past school year

Please mark areas where an injury has been sustained.
Please number each injury and complete the details in the following table.



Please list details of injuries that have affected your dance training and/or performance as per the above diagram

		Injury 1	Injury 2	Injury 3
How long did the injury affect your dance related performance; **7-28 days** (MILD), **29 – 85 days** (MODERATE) or **>84 days** (SEVERE)? Please specify the number of days.				
Was this the first/only episode or a recurrent injury? (please circle as appropriate)		First/ Recurring	First/ Recurring	First/ Recurring
Date your injury occurred (month/year)				
Area of the body where the injury occurred (e.g. ankle, knee, spine)				
Which side of the body was your injury?				
Where/how did your injury occur?				
Did you continue dancing with your injury?				
If no, whose decision was it to sit from dance activity? (self/ physiotherapist/ teacher/ choreographer)				
Was your injury diagnosed by a licensed healthcare professional? (e.g. Doctor, Physiotherapist etc.)				
Diagnosed by a licensed healthcare professional	**If yes,** who did you see and what did they diagnose your injury as?			
	If yes, did they describe your injury as "traumatic" (an injury resulting from a specific identifiable event) or "overuse" (an injury caused by repeated micro-trauma without a single identifiable event responsible)?			
Not diagnosed by a licensed healthcare professional	**If no,** what do you think your injury may have been? (e.g. muscle strain)			
	If no, why did you not receive any treatment?			

		Injury 4	Injury 5	Injury 6
How long did the injury affect your dance related performance; **7-28 days** (MILD), **29 – 85 days** (MODERATE) or **>84 days** (SEVERE)? Please specify the number of days.				
Was this the first/only episode or a recurrent injury? (please circle as appropriate)		First/ Recurring	First/ Recurring	First/ Recurring
Date your injury occurred (month/year)				
Area of the body where the injury occurred (e.g. ankle, knee, spine)				
Which side of the body was your injury?				
Where/how did your injury occur?				
Did you continue dancing with your injury?				
If no, whose decision was it to sit from dance activity? (self/ physiotherapist/ teacher/ choreographer)				
Was your injury diagnosed by a licensed healthcare professional? (e.g. Doctor, Physiotherapist etc.)				
Diagnosed by a licensed healthcare professional	**If yes,** who did you see and what did they diagnose your injury as?			
	If yes, did they describe your injury as "traumatic" (an injury resulting from a specific identifiable event) or "overuse" (an injury caused by repeated micro-trauma without a single identifiable event responsible)?			
Not diagnosed by a licensed healthcare professional	**If no,** what do you think your injury may have been? (e.g. muscle strain)			
	If no, why did you not receive any treatment?			

Appendix 4

Researcher memo

Link to pebble pad

Appendix 5
Measurement protocol for PVH

Measurements Protocols https://www.usask.ca/kin-growthutility/phv_ui.php

Need to measure and record the following: body mass (kg), standing height (cm) and sitting height (cm)

A. Body Mass – Weigh subject with minimal clothing and with shoes removed.

1. Check the scale is reading zero
2. Ask subject to stand on the centre of scales, without support and with their weight distributed evenly on both feet – record body mass to nearest 0.1 kg.
3. Ask subject to step off the scale
4. Repeat steps 1 to 3
5. If the 2 measurements differ by more than 0.4 kg then repeat steps 1 to 3
6. If two measurement record the average value. If three measurements record the median value.

B. Standing Height – Use the stretch stature method. Stature is the maximum distance from the floor to the vertex of the head (Figure 1). The vertex is defined as the highest point on the skull when the head is held in the Frankfort plane (Figure 2). This position is when the imaginary line joining the orbitale to the tragion is perpendicular or at a right angle to the long axis of the body as shown in Figure 2. Subject is measured with shoes removed.

1. Ask subject to stand with back, buttocks and heels against a stadiometer. Subject's feet should be together and flat on the floor.
2. Place subject's head in the Frankfort plane (Figures 1 & 2). Place your hands far enough along the line of the subjects jaw to ensure that upward pressure is transferred through the mastoid processes.

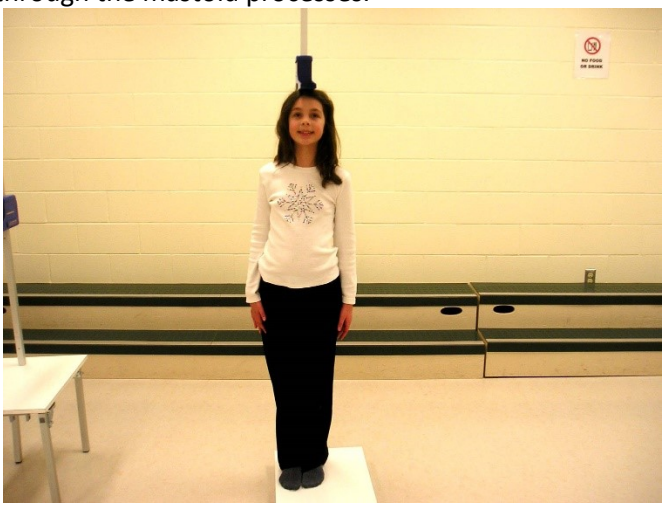


Figure 1: Measuring stretch stature Figure 2: Position of the head in Frankfort plane

3. Instruct subject to take and hold a deep breath. While keeping the head in the Frankfort plane apply gentle upward lift through the mastoid processes. At the same time place the headboard firmly down on the vertex, crushing the hair as much as possible. Ensure that the feet do not come off the ground and that the position of the head is maintained in the Frankfort plane.
4. Record measurement at the end of the subject's deep inward breath – record stature to the nearest 0.1 cm.
5. Ask subject to step away from the stadiometer
6. Repeat steps 1 to 4
7. If the 2 measurements differ by more than 0.4 cm then repeat steps 1 to 4
8. If two measurement record the average value. If three measurements record the median value.

C. Sitting height – Use the stretch stature method. Sitting height is the maximum distance from the vertex to the base of the sitting surface (Figure 3).

1. Seat subject on a measuring box or level platform (of known height) with their hands resting on their thighs

Figure 3: Measurement of sitting height

2. Instruct subject to take and hold a deep breath. While keeping the head in the Frankfort plane (Figure 2) apply gentle upward lift through the mastoid processes. At the same time place the headboard firmly down on the vertex, crushing the hair as much as possible. Ensure the subject does not contract the gluteal muscles nor push with the legs.
3. Record measurement at the end of the subject's deep inward breath – record sitting stature to the nearest 0.1 cm.
4. Ask subject to step off the box and away from the stadiometer
5. Repeat steps 1 to 4
6. If the 2 measurements differ by more than 0.4 cm then repeat steps 1 to 4
7. If two measurement record the average value. If three measurements record the median value.

8. If using a floor stadiometer the observed height minus the box / platform height is the sitting height.

D. Leg Length = Standing Height (cm) - Sitting height (cm)

Ross WD, Marfell-Jones MJ. Kinanthropometry. In MacDougall JD, Wenger HA, Green HJ, eds. *Physiological Testing of the High-Performance Athlete*, pp 223-308. Champaign, Illinois: Human Kinetics Books, 1991.

Appendix 6
Multidimensional Assessment of Interoceptive Awareness (MAIA)

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Multidimensional Assessment of Interoceptive Awareness

Permission and Copyright

Although the MAIA survey is copyrighted, it is available without charge and no written permission is required for its use. This assumes agreement with the following as a consequence of using a MAIA survey:

- Please refer to the survey using its complete name – Multidimensional Assessment of Interoceptive Awareness - and provide the appropriate citation.
- Modifications may be made without our written permission. However, please clearly identify any modifications in any publications as having been made by the users. If you modify the survey, please let us know for our records. We recommend including entire subscales when selecting items from the MAIA to retain the psychometric features of these subscales (rather than selecting items from subscales).
- If you translate the MAIA into another language, please send us a copy for our records.
- If other investigators are interested in obtaining the survey, please refer them to the source document (PLoSONE 2012, and www.osher.ucsf.edu/maia/) to assure they obtain the most recent version and scoring instructions.

Scoring Instructions

Take the average of the items on each scale.
Note: Reverse-score items 5, 6, and 7 on Not-Distracting, and items 8 and 9 on Not-Worrying.

1. **Noticing:** Awareness of uncomfortable, comfortable, and neutral body sensations Q1_____ + Q2_____ + Q3_____ + Q4_____ / 4 = _____

2. **Not-Distracting:** Tendency not to ignore or distract oneself from sensations of pain or discomfort Q5(**reverse**)_____ + Q6(**reverse**)_____ + Q7(**reverse**)_____ / 3 = _____

3. **Not-Worrying:** Tendency not to worry or experience emotional distress with sensations of pain or discomfort Q8(**reverse**)_____ + Q9(**reverse**)_____ + Q10_____ / 3 = _____

4. **Attention Regulation:** Ability to sustain and control attention to body sensations 11_____ + Q12_____ + Q13_____ + Q14_____ + Q15_____ + Q16_____ + Q17_____ / 7 = _____

5. **Emotional Awareness:** Awareness of the connection between body sensations and emotional states Q18_____ + Q19_____ + Q20_____ + Q21_____ + Q22_____ / 5 = _____

6. **Self-Regulation:** Ability to regulate distress by attention to body sensations Q23_____ + Q24_____ + Q25_____ + Q26_____ / 4= _____

7. **Body Listening:** Active listening to the body for insight Q27_____ + Q28_____ + Q29_____ / 3= _____

8. **Trusting:** Experience of one's body as safe and trustworthy Q30_____ + Q31_____ + Q32_____ / 3= _____

Below you will find a list of statements. Please indicate how often each statement applies to you generally in daily life.

.	Circle one number on each line					
	Never					**Always**
1. When I am tense I notice where the tension is located in my body.	0	1	2	3	4	5
2. I notice when I am uncomfortable in my body.	0	1	2	3	4	5
3. I notice where in my body I am comfortable.	0	1	2	3	4	5
4. I notice changes in my breathing, such as whether it slows down or speeds up.	0	1	2	3	4	5
5. I do not notice (I ignore) physical tension or discomfort until they become more severe.	0	1	2	3	4	5
6. I distract myself from sensations of discomfort.	0	1	2	3	4	5
7. When I feel pain or discomfort, I try to power through it.	0	1	2	3	4	5
8. When I feel physical pain, I become upset.	0	1	2	3	4	5
9. I start to worry that something is wrong if I feel any discomfort.	0	1	2	3	4	5
10. I can notice an unpleasant body sensation without worrying about it.	0	1	2	3	4	5
11. I can pay attention to my breath without being distracted by things happening around me.	0	1	2	3	4	5
12. I can maintain awareness of my inner bodily sensations even when there is a lot going on around me.	0	1	2	3	4	5
13. When I am in conversation with someone, I can pay attention to my posture.	0	1	2	3	4	5
14. I can return awareness to my body if I am distracted.	0	1	2	3	4	5
15. I can refocus my attention from thinking to sensing my body.	0	1	2	3	4	5
16. I can maintain awareness of my whole body even when a part of me is in pain or discomfort.	0	1	2	3	4	5

Please indicate how often each statement applies to you generally in daily life.

	Circle one number on each line					
	Never					**Always**
17. I am able to consciously focus on my body as a whole.	0	1	2	3	4	5
18. I notice how my body changes when I am angry.	0	1	2	3	4	5
19. When something is wrong in my life I can feel it in my body.	0	1	2	3	4	5
20. I notice that my body feels different after a peaceful experience.	0	1	2	3	4	5
21. I notice that my breathing becomes free and easy when I feel comfortable.	0	1	2	3	4	5
22. I notice how my body changes when I feel happy / joyful.	0	1	2	3	4	5
23. When I feel overwhelmed I can find a calm place inside.	0	1	2	3	4	5
24. When I bring awareness to my body I feel a sense of calm.	0	1	2	3	4	5
25. I can use my breath to reduce tension.	0	1	2	3	4	5
26. When I am caught up in thoughts, I can calm my mind by focusing on my body/breathing.	0	1	2	3	4	5
27. I listen for information from my body about my emotional state.	0	1	2	3	4	5
28. When I am upset, I take time to explore how my body feels.	0	1	2	3	4	5
29. I listen to my body to inform me about what to do.	0	1	2	3	4	5
30. I am at home in my body.	0	1	2	3	4	5
31. I feel my body is a safe place.	0	1	2	3	4	5
32. I trust my body sensations.	0	1	2	3	4	5

[i] Former Etoile of the PO and guest artist RB, also confirmed to have Hypermobility (Knight, 2013)