Bangor University

DOCTOR OF PHILOSOPHY

Exploring the Cost, Long-Term Outcomes and Sibling Benefits of the Incredible Years (IY) Autism Spectrum and Language Delays Programme

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Award date:
2023

Awarding institution:
Bangor University

Link to publication

Bangor University

Prifysgol Bangor

Exploring the Cost, Long-Term Outcomes and Sibling Benefits of the Incredible Years (IY) Autism Spectrum and Language Delays Programme

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A thesis submitted to the School of Psychology, Bangor University, in partial fulfilment of the requirements of the degree of Doctor of Philosophy.

September 2023

This PhD was funded by a by a Knowledge Economy Skills Scholarship (KESS), supported by the European Social Fund (ESF) and by the Children's Early Intervention Trust Charity Ltd.

‘I hereby declare that this thesis is the results of my own investigations, except where otherwise stated. All other sources are acknowledged by bibliographic references. This work has not previously been accepted in substance for any degree and is not being concurrently submitted in candidature for any degree unless, as agreed by the University, for approved dual awards.

I confirm that I am submitting this work with the agreement of my Supervisor(s).’

‘Yr wyf drwy hyn yn datgan mai canlyniad fy ymchwil fy hun yw’r thesis hwn, ac eithrio lle nodir yn wahanol. Caiff ffynonellau eraill eu cydnabod gan droednodiadau yn rhoi cyfeiriadau eglur. Nid yw sylwedd y gwaith hwn wedi cael ei dderbyn o’r blaen ar gyfer unrhyw radd, ac nid yw’n cael ei gyflwyno ar yr un pryd mewn ymgeisiaeth am unrhyw radd oni bai ei fod, fel y cytunwyd gan y Brifysgol, am gymwysterau deuol cymeradwy.

Rwy'n cadarnhau fy mod yn cyflwyno'r gwaith hwn gyda chytundeb fy Ngoruchwyliwr (Goruchwylwyr)

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Summary

Autism spectrum disorder (ASD) is a life-long neurodevelopmental disability, characterised by impaired social communication, language delay and repetitive behaviour. Prevalence has increased dramatically over the past two decades, drawing attention to the needs of both children and their families. Children with ASD often have complex co-morbidities and challenging emotional and behavioural problems. Parents face difficulties far greater than those of raising a typically developing child and are at increased risk of mental health problems. The associated child emotional and behavioural problems are the main predictor of parental distress, more so than the core symptoms of ASD. Caring for a child with autism can impact negatively on quality of life, family adjustment and relationships. Siblings of children with ASD can also be at greater risk of social and/or emotional problems. Families can experience increased financial burden from possible loss of productivity, and costs of the child's additional needs or care requirements. Autism also has significant financial implications for public funded services.

This thesis evaluates the Incredible Years Autism Spectrum and Language Delays programme (IY-ASLD) ® following a randomised controlled trail (RCT) of its initial effectiveness and feasibility of delivery within existing specialist child services. Chapter 2 lays a foundation to the thesis by giving an overview of autism prevalence, characteristics and comorbidities, and the background to the familial and societal impact. Chapter 3 discusses behavioural parent training programmes that are effective in strengthening parent-child relationships, improving child behaviour and parent outcomes, and their potential benefits for families of children with autism. Chapter 4 describes the study design and methods adopted to examine the key elements of the thesis. Chapter 5 is a systematic review of the available economic evidence surrounding behavioural parenting programmes specifically targeted towards families of children with ASD. The chapter identifies a dearth of

economic evaluations, along with a need for standardised methodological approaches for the design and conduct of future trials. The first study (Chapter 6) is a detailed micro-costing of delivering the IY-ASLD® programme both within the trial and the likely cost of future roll-out within existing services. The cost of establishing and delivering the programme within the trial was lower than expected in comparison to costings of the other IY® programmes. This provides a useful framework to identify the costs of the programme for policy decision makers and to inform future cost-effectiveness evaluations of the programme. The second study (Chapter 7) explores the longer-term effects of the intervention from 18-month follow up data, to examine whether any post-intervention gains were maintained over time. Improvements to child behaviour, parent stress and depression, and reductions in ineffective parenting behaviour were maintained at 18 months, which although not significant, is encouraging. The final study (Chapter 8) looked at data gathered from siblings during the trial to explore whether the programme had wider-reaching benefits for other family members. No differences were observed between siblings from the intervention and control group following the programme. However, contrary to the literature, the majority of the siblings were not exhibiting child emotional or behavioural problems.

This was the first randomised controlled trial (RCT) to examine the long-term outcomes and sibling benefits of the Incredible Years® Autism Spectrum and Language Delays (IY-ASLD®) programme. This was also the first micro-costing of the newly developed programme, which can inform both policy decision makers and future cost-effectiveness evaluations. This thesis has explored the familial and societal impact of autism and highlighted the need for evidence-based parenting programmes to support families in this population. This study has concluded that the IY-ASLD® programme could be beneficial to this population, and promising long-term maintenance of relatively limited gains reported from the main trial (Williams, Hastings & Hutchings, 2020) warrants further investigations in a larger definitive trial. Findings from this thesis are valuable to inform the design of future research.

Chapter 1

General Introduction

Background

Autism, or autism spectrum disorder (ASD) refers to a broad range of neurodiverse conditions that share characteristic impairments in three core domains: social communication and interaction, language delay and restrictive or repetitive behaviour (American Psychiatric Association, 2013). The past two decades have observed a rapid growth in global prevalence, currently estimated to be at a record high of 1-1.5% of the population (Roman-Urrestarazu, van Kessel, Allison, Matthews, Brayne & Baron-Cohen, 2021; WHO 2022). Growing interest in autism research has informed strategic developments, and increased awareness of the familial and societal impact of autism (Pellicano, Dinsmore & Charman, 2013: NICE, 2013). The challenges associated with raising a child with ASD are multifaceted due to the many co-morbid physical and psychological health needs that frequently occur (Karst & Van Hecke, 2012; Kurzius-Spencer, 2018). Parents are at greater risk of elevated levels of stress and depression (Lecavalier, Leone & Wiltz, 2006), and report lower self-efficacy than parents of typically developing children and children with intellectual disabilities (Rezendes & Scarpa, 2011). Parents of children with ASD are also more likely to adopt maladaptive parenting behaviours which are known to exacerbate child behaviour problems (Maljaars, Boonen, Lambrechts, Van Leeuwen & Noens, 2014; Hutchings et al., 2007). Ineffective parenting can also impact negatively on the adjustment of typically developing siblings (Tomeny, Barry & Bader, 2012). Interventions based on behavioural principles such as the well-established Incredible Years® programmes are effective and cost effective in reducing conduct problems across populations (Bywater, Hutchings, Daley, Whitaker, Yeo, Jones, Eames & Edwards, 2009; O'Neill, D., McGilloway, S., Donnelly, Bywater & Kelly, 2013). These programmes improve parenting skills and promote child social and emotional competence (Hutchings et al., 2007), which frequently result in positive parent outcomes such as reduced stress and depression (Hutchings, Griffith, Bywater & Williams, 2017; Marcynyszyn, Maher & Corwin, 2011). Promising benefits for siblings have also been observed (Hutchings, Bywater, Daley, Gardner, Whitaker, Jones, Eames & Edwards, 2007). Although there are, currently, no treatments for the core symptoms of autism, support for parents in the form of group-led parenting programmes is recommended (NICE, 2013). The IY® Basic programme has been trialled with parents of children with ASD with encouraging findings. Roberts and Pickering (2010) led a small pilot study to assess the usefulness of the Basic programme for this population and found improvements in measures of parental mental health and child behaviour. However, the group was run with only eight parents, making it difficult to generalise findings. A larger sample also reported high satisfaction with the IY Basic® and demonstrated reduced parental stress in an acceptability trial of the programme for ASD, although there was no control group for comparison (Dababnah, Olson & Nichols, 2019; Dababnah & Parish, 2016a). Randomised controlled trials (RCTs) are stipulated as ideal in future research recommendations for more reliable evidence of programme effectiveness for this population (NICE, 2013). There is also a paucity of evaluations that test beyond child outcomes and explore the family factors of intervention effects (Karst & Van Hecke, 2012).

The Incredible Years® Autism Spectrum and Language Delays Programme

The Incredible Years (IY)® Autism Spectrum and Language Delays programme is a new addition to the IY suite with adaptations made to increase focus on coaching language development, social communication and imitation (Webster-Stratton, 2015). Emphasis is also placed on parental self-care and building a support group. Group sizes are smaller than other IY programmes, with no more than 6-8 parents recommended to attend. Parent-child relationships and broad developmental outcomes are targeted, for example communication, social and other adaptive skills, as well as behaviour problems. The programme is delivered in weekly sessions of 2 hours, with the programme developer making recent changes to the protocol extending the recommended delivery time to 14-16 sessions, as opposed to the original 12 weeks. Parents are introduced to positive parenting principles and shown video vignettes of the strategies being demonstrated in recordings depicting children on the autism spectrum. Facilitators engage parents into group discussion and encourage them to practice the skills in role play exercises, and home activities. The programme consists of eight topics as follows: 1. Child-directed narrated play; 2. pre-academic and persistence coaching; 3. social coaching; 4. emotion coaching; 5. developing imagination through pretend play; 6. promoting children's self-regulation skills; 7 using praise and rewards to motivate children; 8. effective limit setting and behaviour management.

The Parenting for Autism, Language, And Communication Evaluation Study (PALACES) Trial.

The charity Autistica funded a pragmatic randomised controlled trial (RCT) to evaluate the feasibility of delivering the IY-ASLD® programme within existing services in four sites across Northwest Wales. The specialist children's services are responsible for children with neurodevelopmental disorders such as ASD and attention deficit hyperactivity disorder (ADHD) and/or intellectual disabilities (ID). The multidisciplinary teams consist of child psychologists, paediatricians, specialist nurses and speech and language therapists who offer assessments, interventions and support to children with complex health needs and their families. Participants were 58 parents of children aged 3-8yrs with a recent, or strongly suspected diagnosis of ASD, who were already known to the specialist teams. Families were randomised to either intervention (to attend the programme immediately), or to a wait-list control group (to be offered the programme following completion of follow up data collection). The intervention was delivered in four groups, run concurrently across sites, between baseline and 6-month follow up data collection. Data collection was completed with both intervention and wait list control at baseline and post-intervention by researchers from the PALACES trial, who remained blind. Further data collection was conducted at 12 and 18-

month follow up with parents from the intervention arm only, for which the thesis author was responsible. A range of measures were administered including those covering child communication and adaptive skills, and child behaviour. Parent outcome measures included depression and parental stress questionnaires, and a measure of parenting style. Parenting skills were also assessed in a 10-minute observation of parent-child interactions (Please see Chapter 4: Design and Methods for more detailed information about the trial procedures). Initial effectiveness results showed reductions in child externalising behaviour problems and parental distress, and increased praise among parents in the intervention group. This was the first randomised controlled trial of the newly developed IY-ASLD® programme (Williams, Hastings & Hutchings, 2020).

Along with the Autistica funding for the main trial, a three-year PhD scholarship for the thesis author was awarded by the Knowledge Economy Skills Scholarships (KESS) and the Children's Early Interventions Trust (CEIT). It presented the opportunity to conduct further studies which form this thesis. Health economics measures and detailed costings of programme delivery will be explored to assess the feasibility of conducting future cost-effectiveness analyses, due to the importance of providing economic evidence of newly developed programmes for policy decision makers. The data from 12 and 18-month follow ups will be examined to assess whether any initial gains were maintained, or whether further improvements were observed. Due to the known impact of autism on wider family members, sibling adjustment will be assessed to explore any potential benefits of the programme for siblings. This is the first examination of cost, long term outcomes and possible sibling gains from the programme.

Aims and objectives of thesis

Exploring the cost and outcomes of the IY-ASLD® programme

The aim of this thesis was to further examine the newly developed Incredible Years® Autism Spectrum and Language Delays (IY-ASLD) programme following the PALACES trial to inform a larger definitive trial.

The main objectives of the study were:

1) To examine the economic impact of autism on families and public funded services, and to explore the existing economic evidence for similar parenting programmes in a systematic review.
2) To calculate accurate cost for programme delivery to determine the cost of the intervention during the trial, to inform future trial design and future roll-out of the programme.
3) To examine the potential long-term outcomes of the IY-ASLD® programme.
4) To explore the possible benefits of the IY-ASLD® programme for siblings of children diagnosed with ASD.
5) To reflect on key feasibility messages of novel studies to inform a future definitive trial.

Terminology

In this thesis, the terminology used when referring to autism and children identified as autistic will be "person-first", in which the person (e.g., child) appears before the condition (e.g., autism), such as "child with autism". Many autistic self-advocates and advocacy groups now prefer an identify-first form, such as "autistic child" (Kenny et al., 2016). In addition, autistic advocates have spoken about the desirability of using the terms "autism" and "autistic" rather than "autism spectrum disorder" (National Autistic Society, 2022). However, many professional journals, academic, scientific and professional organizations encourage the person-first form of identification (American Psychological Association [APA], 2013). At the time writing this thesis, terminological issues have not been settled, and although the author acknowledges the preference of groups of individuals with disabilities who prefer the person-first term, for the purpose of publication, terminology throughout the manuscript will be person-first, with the primary descriptors being autism spectrum disorder (ASD) and autism.

Structure of thesis

Due to the different elements included in this evaluation of the IY-ASLD® programme, this thesis is in the format of four separate papers. Therefore, there may be instances of repetition between chapters when defining certain key concepts or describing the programme.

This thesis consists of nine chapters in total, which are

Chapter 1 – General Introduction

Chapter 2 - Autism: Familial and societal impact

Chapter 3 - Parenting programmes for ASD

Chapter 4 – Study Design and Methods

Chapter 5 – Systematic review of the economic evidence

Chapter 6 – Micro-costing of programme delivery

Chapter 7 – Long term outcomes of the programme

Chapter 8 – Benefits of the programme for siblings

Chapter 9 – General Discussion

Chapter 2

Autism: Family and societal impact

Introduction

Autism is a lifelong, neurodevelopmental disorder defined in the Diagnostic and Statistical Manual (DSM-5) by significant and persistent deficits in social communication, social interaction impairments and restrictive or repetitive behaviours (APA, 2013). Communication problems typically manifest as difficulties with social initiation (beginning conversations or play with others), social reciprocity (taking turns in communication), synchrony (for instance meaningfully linking conversation to topic) and understanding and expressing appropriate non-verbal behaviours such as facial expressions (Baron-Cohen, Golan & Ashwin, 2009). Although the core symptoms of autism are consistent, they present differently in each individual and vary along a continuum of severity from very mild to severe (APA, 2013; Lord, Elsabbagh, Baird & Veenstra-Vanderweele, 2018). This chapter provides a context of autism and the many associated physical and mental health challenges. The familial and societal impacts of the disorder are also described to present the rationale for this thesis.

Diagnosis and prevalence

Due to the complex nature of autism spectrum disorder (ASD), diagnosis can be a lengthy and difficult process (Oswald, Haworth, Mackenzie & Willis, 2017; Lord & Jones, 2012). In the past, five pervasive developmental disorders were categorised separately for diagnostic purposes: Autistic disorder, Asperger’s Syndrome, Pervasive developmental disorder not otherwise specified, Child disintegrative disorder and Rett’s disorder (APA, 2000). However, changes were made to the most recent version of the DSM-5 to include all subtypes under the umbrella term of autism spectrum disorders (ASDs) (APA, 2013). Although diagnosis can be conducted reliably at age 2 years (Brett, Warnell, McConachie & Parr, 2016), the average age of diagnosis ranges between 3.1 and 7.5 years in population-based studies (Crane, Chester, Goddard, Henry & Hill, 2016; Karst & Van Hecke, 2012: Russell et al., 2022; Shattuch et al., 2009), and is much older for high functioning children (Mandell et al., 2005). Despite opinions that diagnostic revisions and epidemiological advances have led to the rapid increase in prevalence seen over the past two decades (Sharma, Gonda & Tarazi, 2018), the average time between parental contact with clinical services and formal diagnosis is 3.5 years (Crane et al., 2016). In the United Kingdom, this has prompted the implementation of integrated clinical initiatives to improve diagnostic services for children (Brett, Warnell, McConachie & Parr, 2016). Global prevalence estimates are currently at a record high of 1-1.5% of the population (Roman-Urrestarazu, et al., 2021; WHO 2022; Zeidan et al., 2022), with one in every 100 children in the UK believed to be on the autistic spectrum (Fombonne, MacFarlane & Salem, 2021). Males are consistently reported as being four times more likely than females to have ASD (APA, 2000; Baron-Cohen et al., 2011; Rubenstein, Wiggins & 2015). Lower reported incidences in girls have been attributed to a female phenotype of autism (Lockwood Estrin, Milner, Spain, Happé & Colvert, 2021), suggesting that their better ability to imitate superficial social skills can camouflage symptoms, making diagnosis more difficult (Allely, 2018; Dean, Harwood & Kasari, 2017). Based on this, girls are less likely to be referred for treatment and are often misdiagnosed (Haney, 2016; Lockwood et al., 2021; Young, Oreve & Speranza, 2018). The diagnostic process itself can be subject to further gender bias due to the conceptualisation of gender differences in behaviour (Clarke et al, 2021), and its focus on the externalising symptoms of autism, which are typically more prevalent in boys (Loomes, Hull & Mandy, 2017). Autism is more likely to manifest as internalising problems in girls, with an increased risk of diagnosed mood and/or eating disorders (Carpita, Muti, Cremone, Fagiolini & Dell’Osso, 2020), particularly among those with higher cognitive abilities (Solomon, Miller, Taylor, Hinshaw & Carter, 2012; Mandy, Chilvers, Chowdhury, Salter, Seigal & Skuse, 2012). Girls who are diagnosed with ASD therefore tend to have lower intellectual functioning and/or language delays (Dworzynski, Ronald, Bolton & Happé, 2012; Salomone, Charman, McConachie & Warreyn, 2016), which could explain why more girls are diagnosed with co-morbid conditions (Suckle, 2021).

Co-morbid health conditions

Co-morbidity is defined as the co-occurrence of two or more conditions or disorders (Mannion & Leader, 2013) and is frequently associated with ASD. Simonoff and colleagues (2008) found that 41% of children with ASD had co-occurring cognitive, physical, behavioural and/or mental health conditions, with ADHD, social anxiety and oppositional defiant disorder (ODD) being among the most frequently reported (Simonoff et al., 2008). It is estimated that over half of children with ASD have four or more co-existing conditions at any one time (Maskey, Warnell, Parr, Le Couteur & McConachie, 2013), which not only further complicates diagnosis (Thurm, Farmer, Salzman, Lord & Bishop, 2019), but also increases the risk of poorer psychosocial outcomes (Tonnsen et al., 2016). Autism co-occurs with intellectual disabilities (IDs) at very high rates of up to 75% (Matson & Shoemaker, 2009; Goldin, Matson & Cervantes, 2014; Kurzius-Spencer et al., 2018). Co-morbidities including Down syndrome, Fragile X and Prader-Willi syndrome (Totsika, Hastings, Emerson, Lancaster & Berridge, 2011; Matson & Shoemaker, 2009), can make the detection of other health problems difficult (Totsika, Hastings, Emerson, Lancaster & Berridge, 2011). Commonly associated health problems include seizure disorders, with the increased risk of epilepsy reported as being sevenfold in children with ASD (Thomas, Hovinga, Rai & Lee, 2017). Sleep disorders are also more prevalent and intense in children with ASD (Accardo & Malow, 2015; Mazurek & Sohl, 2016), with up to 80% of children exhibiting problems such as insomnia and night waking (Cortesi, Giannotti, Ivanenko & Johnson, 2010; Sivertsen, Posserud, Gillberg, Lundervold & Hysing, 2012). Self-injurious behaviours (SIBs) are also highly prevalent with up to 50% of individuals with ASD reported to engage in behaviours such as head banging, hair pulling, arm biting and/ or scratching during their lifetime (Soke et al., 2016; Waters & Healy, 2012). SIBs vary in severity, but impact on the physical health and quality of life of individuals and their families (Summers et al., 2017), and are the main cause of emergency medical treatment for this population (Soke et al., 2016). Antecedents often include underlying stress or frustration (Skegg, 2005), but others seem to be motivated by sensory seeking behaviours, related to pain or rhythmic head banging (Summers et al., 2017). Sensory traits are also common, and present in 96% of individuals with ASD in some manner (McCormick, Hepburn, Young & Rogers, 2016), including over or under responsiveness to touch, over-reactivity to smell sound or movement, and under-reactivity to pain (Ben-Sasson, Gal, Fluss, Katz-Zetler & Cermak, 2019; Klintwall et al., 2011; Marco, Hinkley, Hill & Nagarajan, 2011). Food selectivity or fussy eating can also be associated with sensory impairments, with children often having a distinct preference or dislike of certain food textures or tastes (Vissoker, Latzer, Gal, 2015). Unusual eating and feeding patterns are highly prevalent with ASD, often presenting during infancy (Holingue, Newill, Lee, Pasricha & Fallin, 2018). The most frequent problems are food refusal (Bandini et al., 2017), and/or limited food preferences (Baraskewich, von Ranson, McCrimmon & McMorris, 2021), which can be linked to the rigidity of thought that is a core characteristic of autism. The ingestion of non-food items, known as pica, is also disproportionally prevalent in children with ASD (Matson, Belva, Hattier, & Matson, 2011), as is anorexia nervosa among older children (Dinkler et al., 2021). Atypical eating behaviours have been associated with a higher incidence of gastrointestinal (GI) dysfunction which occurs within this population (Baraskewich et al., 2021; Holingue, Newill, Lee, Pasricha & Daniele Fallin, 2018). However, there is increasing research examining whether GI problems are part of the pathophysiology of autism, and not only a co-morbid condition (Coury et al.,2012). The prevalence and severity of GI problems are disproportionally higher when compared to typically developing children. Although rates differ significantly between studies, it is thought that up to 70% of children with ASD experience gastrointestinal problems that often merit costly clinical intervention not only to address the physical symptoms (Adams, Johansen, Powell, Quig & Rubin, 2011; Valicenti-McDermott et al., 2006) but also maladaptive behaviours which may occur as a result. Fulceri and colleagues (2016) examined possible associations between GI symptoms and internalising behaviours in children with ASD, reporting a significant relationship with increased anxiety (Fulceri et al., 2016), and depression (Ferguson, Dovgan, Takahashi & Beversdorf, 2019). Associations have also been observed between GI symptoms and externalising behaviours such as conduct problems (Babinská, Pivovarciova, Filcikova, Tomova & Ostatníková, 2016), irritability and hyperactivity (Chaidez, Hansen & Hertz-Picciotto, 2014). It is well documented that children with ASD are at greater risk of emotional and behavioural problems, and exploring the associated factors is a broad research area (Charman, Ricketts, Dockrell, Lindsay & Palikara, 2015; Chandler et al., 2016).

Child emotional and behavioural difficulties

It is estimated that at least 72% of children with ASD meet diagnostic criteria for co-occurring psychiatric disorders (Leyfer et al., 2006). Anxiety disorders are among the most common (Simonoff et al., 2008), presenting as separation anxiety, social anxiety and/or generalised anxiety disorder, in around 40% of children with ASD (van Steensel & Heeman, 2017). Anxiety diagnosis can also include specific phobias and compulsive disorders which often intensify the core symptoms of autism (Hallett et al., 2013). Depression can similarly exacerbate core ASD traits (Magnuson & Constantino, 2011), and is estimated to affect around 42% of children with ASD (Mayes, Calhoun, Murray & Zahid, 2011). Internalising conditions can present as somatic symptoms (Mazurek & Kanne, 2010); and/or behaviour problems, particularly in younger children (Mayes et al., 2011). Some studies report a greater incidence of internalising behaviour among children with higher IQ scores (Magnuson &

Constantino, 2011; Mazurek & Kanne, 2010; Hallett et al., 2013), suggesting that greater insight and awareness of their communication deficits may trigger internalising problems (DeFilippis, 2018), and that lower functioning children are less likely to express excessive worry (Hallett et al., 2013). Incidence of depression and anxiety could also be attributed to the challenges that children with ASD encounter with executive function and adaptive behaviour, given the important role they both play in mental health (Gardiner & Iarocci, 2018; Gilotty, Kenworthy, Sirian, Black & Wagner, 2002). Adaptive skills include the ability to carry out age-appropriate tasks in daily life such as feeding, personal hygiene, staying safe and building relationships with peers (Kanne et al., 2011). These difficulties with everyday functioning, that are core traits of autism, can be key predictors of social and behavioural outcomes. For instance, the metacognition of knowing and understanding social rules and peer interaction can present challenges for children with ASD. They are often ostracised within peer groups, placing them at greater risk of bullying and consequent poor mental health and wellbeing (Schroeder, Cappadocia, Bebko, Pepler & Weiss, 2014). School refusal can also be an outcome of social exclusion and bullying which can affect long-term academic attainment (Ochi, Kawabe, Ochi, Miyama, Horiuchi & Ueno, 2020). Perhaps the most frequently reported concomitant difficulties in the ASD literature are conduct problems. Children with disabilities are four times more likely to have behaviour problems than their peers (Stuttard et al., 2014), and these are often on a more severe level when compared to typically developing children (Matson, Wilkins & Macken, 2008). Hartley & Sikora (2009) reported that one third of children aged 1.5.to 5.8 years diagnosed with ASD had clinically significant behaviour problems. Behaviour challenges are the primary reason for intervention referral (Matson et al., 2008), however many more children with ASD exhibit problems below diagnostic thresholds (Williams, Hastings & Hutchings, 2020). Problem behaviours such as oppositional defiant disorder, conduct disorder and disruptive behaviour disorder exist across all populations, and the negative impact on the general health and wellbeing of children and those around them are extensively researched (Polanczyk, Salum, Sugaya, Caye & Rohde, 2015; Kaat & Lecavalier, 2013). Early onset can predict escalating problems in adolescence and adulthood if not addressed, such as antisocial or aggressive behaviour, property destruction and/or criminality, which can result in substantial societal costs (Romeo, Knapp & Scott, 2006; Mazurek, Kanne, & Wodka, 2013). More than 50% of children with ASD exhibit aggressive behaviour (Hanratty et al., 2015), which is reported as being the greatest cause of school disruption (Hastings & Brown, 2002) and in extreme cases, the main reason for placements in residential care (Abbott, Morris & Ward, 2000; Hodgetts, Nicholas & Zwaigenbaum, 2013). Although less severe, daily behaviour problems such as tantrums and persistent non-compliance, recurring from changes in the child's routine or environmental stimulus, are often reported by parents as being the most challenging due to their high frequency and substantial impact on family adjustment and wellbeing (O'Nions, Happé, Evers, Boonen & Noens, 2018).

Parental challenges

The challenges of raising a child with autism go beyond those of parenting typically developing children. Greater demands are placed on their time, with mothers reported to spend 50% more time with their ASD child than parents of neurotypical children (O'Nions et al., 2018). This could be due to the night waking of children with co-morbid sleep disorders, and the significantly reduced sleep reported by mothers (Meltzer, 2008). Rationale can also include their child's additional support needs during the day with daily tasks such as toileting, feeding (Thullen & Bonsall, 2017), communicating (Bonis, 2016), and/ or managing problem behaviours that require continuous supervision (Petrou, Soul, Koshy, McConachie & Parr, 2018). The latter is arguably the most demanding according to many parents, as their child's behaviour can interfere with daily family functioning (Maljaars et al., 2014), and prevent them from involving their child in conventional activities such as shopping, family and school events, and leisure activities (Walton, 2019). Parents report that fear of judgment from others about their child’s behaviour in public, and perceived stigma intensifies their reluctance to socialise (Woodgate, Ateah & Secco, 2008). Consequently, there are limited opportunities to form social relationships, and isolation is a frequently reported challenge for this population (Ludlow, Skelly & Rohleder, 2012; Karst & Van Hecke, 2012). Parents also describe feeling disconnected from family members following their child’s diagnosis (Nealy, O’Hare, Powers & Swick, 2012). A qualitative study reported parents feeling as though they were ‘living in a world of their own’, due to a lack of understanding and scaffolding from extended family (Woodgate et al., 2008). Minimal practical support also means that parents fulfil multiple roles, which exacerbate their beliefs that they are not doing enough to meet and manage their child’s needs (Ludlow et al., 2012). Emotional challenges are often heightened by parental feelings of inadequacy (Ludlow et al., 2012) and more importantly, poor parent-child relationships (Smith, Greenberg, Seltzer & Hong, 2008). Parents of children diagnosed with autism frequently express concerns about a disconnection with their child (Nealy et al., 2012), and often describe them as being hard to reach (Falk, Norris & Quinn, 2014). Their child’s unwillingness to engage (Crowell, Keluskar & Gorecki, 2019), adverse reactions to efforts of physical affection and lack of reciprocal interaction (Gau et al., 2010) are difficult for parents emotionally. As a result, parents of children with ASD consistently report lower self-efficacy than parents of typically developing children (Giallo, Wood, Jellett & Porter, 2013). Self-efficacy refers to the belief in their own ability to parent their child effectively and is of particular importance to parents of children with autism due to their specific challenges. They may be at risk of feeling increased helplessness as many of their child’s problem behaviours are, in fact, unchangeable. Core ASD symptoms, and lack of affective reciprocity with their child, could make them feel that they are not meeting their child’s emotional needs. Self-efficacy has been found to influence parental coping and problem-solving skills (Rezendes & Scarpa, 2011), and can be a strong predictor of ineffective parenting behaviour and mental health problems (Hastings & Brown, 2002).

Parental wellbeing

The complex challenges of raising a child with ASD can exhaust the coping skills of parents, putting them at increased risk of mental health difficulties (Hastings et al., 2005). As a result, the prevalence of mental health problems among this population is substantially higher when compared to parents of typically developing children (Barroso, Mendez, Graziano, & Bagner, 2018; Meltzer, 2011; Lecavalier, Leone & Wiltz, 2006; Padden & James, 2017), and children with disabilities including Down syndrome, Fragile X syndrome and other developmental delay disorders (Dabrowska & Pisula, 2010; Abbeduto, Seltzer, Shattuck, Krauss, Orsmond & Murphy, 2004; Hayes, & Watson, 2013). Parental anxiety presents long before a formal diagnosis is obtained (Voliovitch et al., 2021), with parents often noticing autism characteristics and developmental delays from as early as infancy (Karst & Van Hecke, 2012; Garrido, Carballo, Artis & Garcia-Retamero, 2018). Seeking a diagnosis for their child is stressful for parents (Elder, Kreider, Brasher & Ansell, 2017; Reed & Osborne, 2012). A qualitative study by Russell and Norwich (2012) reported parental descriptions of the process as being a ‘battle’ or ‘struggle’, illustrating the level of distress it can trigger. Receiving a diagnosis also has significant psychological repercussions, with many parents describing the event as being life changing (Banach, Iudice, Conway & Couse, 2010). Although around half of parents feel relief at gaining a formal diagnosis (Hutton & Caron, 2005), a range of other emotions are frequently reported including shock (Banach et al., 2010), sadness (Fernańdez-Alcántara et al., 2016), guilt (Ludlow, Skelly & Rohleder, 2012), and anger (Legg & Tickle, 2019). These are often emotions associated with grieving, and many parents convey a sense of loss or grief for the life they had envisaged for themselves and their families (Fernańdez-

Alcántara et al., 2016; Hutton & Caron, 2005). Post diagnosis is described as a peak time of stress; however, it is the long-term challenges that lead to significant psychological distress. Emerging research demonstrates that mental health problems among parents of children with autism, namely higher rates of stress, anxiety and depression (Meltzer, 2011; Rezendes & Scarpa, 2011; Totsika, Hastings, Emerson, Lancaster & Berridge, 2011), are associated with three key predictors; child behaviour problems, the severity of the child's ASD symptoms and social support. Associated child behaviour problems are frequently reported as being the most difficult aspect of parenting a child with ASD due to their persistent and pervasive nature (Benson & Karlof, 2009); and are the greatest predictors of elevated stress and depression (Lecavalier et al., 2006; Rezendes & Scarpa, 2011; Falk et al., 2014). Chronic stress has been associated with the elevated risk of depression and anxiety which is also higher in this population (Zablotsky, Anderson & Law, 2013). Symptom severity has not only been reported as a main predictor of elevated parental stress (Hastings et al., 2005), but was also correlated with levels of depression in a study where over three quarters of participating mothers were reporting clinical levels of depression (Jose, Gupta, Gulati, & Sapra, 2017), illustrating an interrelationship between mental health problems in this population. The lack of support depicted is also a predictor of stress and depression (Dabrowska & Pisula, 2010). Parental mental health difficulties are frequently associated with poorer parental functioning, and strongly influence parenting practices (Dissanayake, Richdale, Kolivas & Pamment, 2020). Elevated stress can reduce parents' ability to deal with challenging behaviour (Falk et al., 2014) and is known to diminish parents' ability to problem solve (Enea & Rusu, 2020). Parents with depression can be more critical, adopt more punitive discipline strategies and are less likely to interact with their child (Hastings, 2002; Hutchings et al., 2007). This could have implications not only for the diagnosed child, but throughout the family system.

Family impact

Wellbeing and relationships

Autism has a greater effect on families than any other childhood disorder (Gray, 2006). It is more difficult to examine the impact on fathers, as they are often misrepresented in ASD research (Gray, 2002; Cheuk & Lashewicz, 2016) due to increased focus on maternal involvement in child health and intervention research (Altiere & Von Kluge, 2009; Rankin, Paisley, Tomeny & Eldred, 2019). Fathers face more barriers to attending interventions (Flippin & Crais, 2011), and are less likely than mothers to seek support (Cheuk and Lashewicz, 2016). Parents of children with ASD experience more health-related problems (Stein, Foran & Cermak, 2011) and tend to neglect their own health needs to take care of their child (Kuhlthau et al., 2014). Both parents are at greater risk of elevated stress, with fathers thought to exhibit lower levels than mothers, who are often the primary carer (Karst & Van Hecke, 2012). The stress levels of fathers remain consistent, whereas maternal stress decreases as the child gets older (Karst & Van Hecke, 2012), possibly linked to increased acceptance. Fathers are thought to be more concerned about the stigma surrounding their child’s diagnosis (Gray, 2006; Shave & Lashewicz, 2016). Due to the demands placed on parents, particularly the mother (Sawyer, et al, 2010), there is less free time. Fathers reported fewer opportunities to socialise as being a disadvantage, and that they valued socialising with friends and colleagues to feel ‘normal’ (Seymour, Allen, Giallo & Wood, 2020). Less time is also available for investing in other relationships such as with partners or other children. High marital dissatisfaction is reported among couples who are raising a child with autism, more so from mothers (Lickenbrock, Ekas & Whitman, 2011). Divorce rates are higher when compared to those of typically developing children (Karst & Van Hecke, 2012), with some studies reporting parents of ASD children are twice as likely to end marriages than those of typically developing children (Hartley et al., 2010). Marital problems not only affect parents but can also have a negative impact on the whole family (Gau et al., 2012). Siblings of children with autism are reportedly at increased risk of psychosocial adjustment due to the challenges associated with the diagnosed child and their relationship with them (which is explored in more detail in Chapter 8). However, they could also be more vulnerable to mental health problems due to other risk factors within the family system such as environmental stress from marital conflict (Barak-Levy, Goldstein & Weinstock, 2010). Parents who experience relationship problems are also more likely to adopt more negative parenting practices (Camisasca, Miragoli & Di Blasio, 2016), therefore child behaviour problems among other children in the family could be a potential risk. Substance use has also been described as an increased risk for parents of children with ASD (Miles, Takahashi, Haber & Hadden, 2003), which could potentially contribute to further family dysfunction and marital conflict. Families of children with ASD present significantly lower quality of life rates than families raising neurotypical children (Vasilopoulou & Nisbet, 2016). This population consistently demonstrate lower subjective physical and mental health scores, as well as poor social functioning. In addition to concerns about their child's health and future care needs (Kheir et al., 2012), the economic burden that comes with caring for a child with autism can be a major influence on quality of life. The cost of raising a child with a disability is three times greater than that of a typical developing child (Järbrink, Fombonne & Knapp, 2003). Caring for a child with autism results in substantial demands on time and finances, causing further strain on health and relationships. Being in employment is shown to improve parental quality of life (Vasilopoulou & Nisbet, 2016), however parents of children with autism are less likely to be in employment and earn less due the demands of caring for their child (Cidav et al., 2012). Family composition also has an influence, particularly for mothers, with having more children associated with maternal quality of life (Dardas & Ahmad, 2014). Many individuals with autism have high support needs, and with fewer people with ASD now living in in high cost sheltered placements, many remain dependant on families or other support services for their entire lives (Howlin, Goode, Hutton & Rutter, 2004), which in turn has many cost implications for government bodies and families (Knapp, Romeo & Beecham, 2009).

Societal impact

Associated health needs and challenges mean that many individuals with ASD require lifelong support of some kind (Zeidan et al., 2022), resulting in financial burden not only for families but also from a public health perspective (Buescher, Cidav, Knapp & Mandell, 2014). Due to its early onset and lifelong persistence, autism is a cause of high service utilisation (Romeo, Knapp & Scott, 2006). Public health costs are categorised as being either direct, which include education, medical and health service costs (primary healthcare, hospital and emergency services, prescription medication, medical equipment and supplies and behavioural therapies); or indirect, which include productivity losses of the individual and their families due to autism (the value of lost work time, benefit costs, respite care or placement services, etc.). There is a paucity of accurate evidence with regards to the economic effect of ASD, due to the variance in reported global prevalence, disparity in healthcare systems (Dallman, Artis, Watson & Wright, 2021), and difficulty in differentiating between the cost of co-morbidities (Rogge & Janssen, 2019; Cidav, Lawer, Marcus & Mandell, 2013). However, individual life-time costs are currently estimated to be £0.92 million for someone without a co-morbid condition, rising to £1.5 million for someone with a co-occurring intellectual disability (Buescher, Cidav, Knapp & Mandell, 2014). Education is estimated as being a large component of cost (Rogge & Janssen, 2019) along with productivity losses of parents (Leigh & Du, 2015). The direct costs of autism are assumed to be at their highest during the early years and typically decline substantially after the child reaches the age of 8yrs (Ganz, 2007). This could be related to the high utilisation of medical and clinical services pre, during and immediately post intervention (Buescher et al., 2014).

The health care needs of individuals with ASD are complex and require a range of integrated services including health promotion care, rehabilitation services and collaboration with other sectors such as education, employment and social care (Myers, Johnson & Council on Children with Disabilities, 2007). Determining the financial and health service burden of autism through economic evaluation therefore is important to ensure sufficient allocation of funding, and adequate services for individuals with autism and their families across their life-course (Chiarotti & Venerosi, 2020).

Growing awareness of the financial impact of autism has increased focus within national programmes and on governmental agendas. The Autism act was initially introduced in 2009 to transform services for adults with autism and has shaped subsequent policies. The current national strategy in England builds on the ‘Think Autism’ (Department of Health, 2014) policy, which was developed to improve understanding and acceptance of autism within society and now extends focus to children. The national strategy for autistic children, young people and adults: 2021 to 2026 (Department of Education and Department of Health and Social Care, 2021) emphasises the importance of ensuring early diagnosis, and that necessary support and resources are available as soon as possible and across the lifetime of the individual and their families. The Welsh Government share a similar vision, and the Refreshed Autistic Spectrum Disorder Strategic Action Plan was published in response to what people with autism and their carers described as being important to them (Welsh Government, 2016). In addition to promoting awareness and available advice on autism, improvements to services and support have also been prioritised. A key development was the founding of the New Integrated Autism Service; a specialist team in every region of Wales providing support and advice in the community. Multi-agency working practices were introduced not only to improve child diagnosis and treatment, but also to ensure families have access to early intervention. The Integrated Family Support Service facilitates additional support that include advice and training for families and carers of children with ASD. Actions are based on good practice contained in the National Institute for Health and Care Excellence (NICE) guidelines (NICE, 2013), as are the government directives in England (Department of Education and Department of Health and Social Care, 2021). NICE guidelines specify that families of individuals with ASD should receive adequate assistance including the offer of respite provision, advice, training and support. Recommendations also state that parents, carers and siblings should also be offered an assessment of their own physical and mental health needs, including whether they have social and emotional support. Psychosocial behaviour-change interventions such as Applied Behavioural Analysis (ABA) and Early Intensive Behavioural Intervention *(*EIBI*)*, are included in the guidelines; however, interventions that aim to change child behaviour can have negative implications for families (Pennefather, Hieneman, Raulston & Caraway, 2018). Engagement in such interventions has been shown to amplify parental stress (Raulston, Hieneman, Caraway, Pennefather & Bhana, 2019), as can recruiting and maintaining a suitable team of therapists, (Grindle, Kovshoff, Hastings & Remington, 2009). A bidirectional relationship can occur between outcomes and parental wellbeing, with decreased intervention outcomes being observed if parents are reporting elevated stress and/or depression (Vitale, Schneider, Gardner, Alessandri & Marker, 2022). Parents have also reported added disruption to family routine (Grindle et al., 2009) and the process can often be financially burdensome (Dillenburger, Keenan, Gallagher & McElhinney, 2004). Pharmacological intervention such as antipsychotic medication are also included in the NICE guidelines, strictly as a last resort when other interventions have been unsuccessful or due to the severity of behaviour problems (National Institute for Clinical Excellence, 2013). However, it is estimated that 50% of children with ASD are medicated for non-core symptoms (Lamy & Erickson, 2018), presenting the problem of high cost due to the need for close medical supervision and provision of medication, and the increased risk of medical ill-effects (Matin et al., 2022). NICE guidelines largely promote preventative interventions such as parent training programmes in the first instance for behaviour problems, including for antisocial behaviour and conduct disorders (National Institute for Clinical Excellence, updated 2017). Group based programmes, that educate the parent to identify triggers and patterns of reinforcement, are recommended and have been shown to be effective in reducing early and emerging behaviour challenges for many populations (National Institute for Clinical Excellence, 2013). NICE guidelines also include recommendations for research and encourage the evaluation of interventions for parents of children with ASD. Using randomised controlled trials (RCTs) wherever possible, they encourage examining their potential effectiveness in improving parent and sibling stress, quality of life and child adaptive function. The value of parental support for this population is also reaffirmed by the World Health Organisation (WHO) which stresses the importance of early parenting interventions in their publication from the International Conference on Autism and Neurodevelopmental Disorders publication (WHO, 2017). Their recommendations have since included teaching parents the skills needed to help their child improve their social abilities, communication and behaviour through parent training programmes to optimise the healthy development, quality of life and overall wellbeing of individuals diagnosed with autism and their families (WHO, 2022).

Although all strategic frameworks recognise that autism is incurable, the general consensus is that early diagnosis followed by appropriate treatment can improve outcomes for most individuals and their families, and that social pragmatic approaches such as parent training programmes have the potential to improve family outcomes. Evidence is of great interest to families, health professionals and policy makers, therefore empirical evaluations of such programmes are invaluable.

Conclusion

- This chapter highlights the many physical and mental health challenges associated with ASD and the related familial and societal impacts, presenting the rationale for this thesis.
- The complex health needs and intense family care of a child with ASD are related to higher costs for both the family unit and the indirect costs for NHS and society.
- Growing awareness of the impact of ASD has driven governmental policy and programme changes, highlighting the importance of family support and access to early intervention.
- Parenting programmes based on behavioural principles have been shown to address two of the three key predictors of mental health problems in parents of children with autism: child behaviour problems and social support. Therefore, such programmes can be beneficial to this population.

Chapter 3

Parenting programmes for ASD

Introduction

Parenting a child with autism presents far greater challenges than those of raising a typically developing child (Lee, 2009). In addition to the many co-occurring health conditions, emotional and behavioural challenges are common, and problem behaviours often manifest as aggression, hyperactivity, and non-compliance (Simonoff et al., 2013; Crowell, Keluskar & Gorecki, 2019). Managing their child's co-occurring emotional and behavioural difficulties are frequently reported to be the most challenging aspects of parenting a child with ASD (O'Nions et al., 2018). The higher incidence of parental mental health problems observed among this population is a risk factor for ineffective parenting behaviours (Dittman, Farruggia, Palmer, Sanders & Keown, 2014).

Parenting behaviour

Parenting plays a significant role in the development and exacerbation of child behaviour problems (Gardner et al., 2006; Furlong et al., 2012). Certain parenting behaviours are known to contribute to the emergence of challenging behaviour such as parental inconsistency, high criticism, and harsh or punitive punishment (Hutchings et al., 2007). Parental wellbeing is a predictor of parenting style, with mental health problems such as elevated stress increasing the risk of maladaptive parenting (Maljaars et al., 2014). Hastings (2002) proposed a model of the relationship between parental mental health and child behaviour problems in families of children with disabilities. A reciprocal pattern of behaviour problems increases parental stress which affects the adoption of maladaptive parenting behaviours, thus further reinforcing the child's behaviour problems. Understanding and addressing parent behaviour is therefore fundamental in child behaviour management (Hastings, 2002). Parents of children with autism are at increased risk of negative parenting practices, due to the higher prevalence of parental mental health problems among the population (Totsika, Hastings, Emerson, Lancaster & Berridge, 2011). Maternal depression is a risk factor for withdrawn or harsh parenting (Goodman, Rouse, Connell, Broth, Hall & Heyward, 2011). Elevated stress is known to impair parental problem-solving ability (Kazdin & Whitley, 2003), and heighten reactivity (Martorell & Bugental, 2006). Increased stress also impacts upon parental self-efficacy, which is known to influence parent coping skills. Parents with higher self-efficacy view challenges as being less problematic and are more likely to adopt effective parenting techniques (Dissanayake, Richdale, Kolivas & Pamment, 2020). As well as being at increased risk of low self-efficacy (Giallo et al., 2013), parents of children with ASD tend to adopt more permissive parenting practices (Maljaars et al., 2014). Lax parenting with less limit-setting is strongly associated with child behaviour problems (Furlong et al., 2012). There are also indications that parents of children with ASD are less responsive to their child, possibly due to the social and affective reciprocity between parents and children on the autism spectrum (Boonen et al., 2015). It is well established that parents in this population have a greater need for scaffolding with regards to parenting support and practices than those of typically developing children (Rabba, Dissanayake & Barbaro, 2019). The World health Organisation’s resolution on autism spectrum disorders recommends that psychoeducation for parents and carers of children with ASD should be a priority, in order to provide them with knowledge and guidance to better understand their child’s diagnosis and associated challenges (WHO, 2014).

Parenting interventions

There are a growing number of targeted interventions that aim to improve the lives of families of children with autism through education and training, varying considerably with regards to their theoretical underpinning. Self-directed programmes such as mindfulness-based approaches (Hwang, Kearney, Klieve, Lang & Roberts, 2015), music therapy (Thompson, McFerran & Gold, 2014) and social inclusion programmes (McConkey, Cassin & McNaughton, 2020) can provide parents with much needed advice and support, often during the difficult period of post-diagnostic adjustment. Interventions can assume various approaches. Parent support programmes, typically have emphasis placed on knowledge transfer which directly benefits the parent by providing them with new information, practical knowledge, or skills (Dawson-Squibb, Davids, Harrison, Molony & de Vries, 2020; Matson et al., 2012). Alternatively, parent-mediated programmes are more strategic in nature, whereby parents are the agent of change instigating child outcomes, from which both benefit (Bearss, Burrell, Stewart & Scahill, 2015). Given the complexities of parenting a child on the autism spectrum, interventions specific to ASD are often a combination of both, and this merged modality is frequently referred to as parent education and training (PET) in the literature. Programmes can aim to improve social communication, such as the PACT (PArent-mediated social-Communication Therapy) programme, a clinic-based one to one intervention which has shown impressive improvements in child-initiated interaction with parents and expressive-receptive language (Pickles et al., 2016). Focus can also be on educational attainment as is the case with the TEACCH (Treatment of Autistic and Related Communication Handicapped Children) intervention, which assumes a ‘life-long’ approach to supporting families by initiating a parent-professional collaboration across all ages (Virues-Ortega, Julio & Pastor-Barriuso, 2013). With regards to school-aged children, a shared knowledge about the child’s unique way of learning helps parents and teachers work together in enhancing the child’s social development, communication and behaviour management within an educational context (Schopler, Mesibov & Hearsey, 1995). Parents are also provided with practical information, signposting to services and opportunities to meet with other parents (Virues-Ortega, Julio & Pastor-Barriuso, 2013). Although the programme is widely disseminated, there is limited robust evidence of effectiveness (Matson et al., 2012). Many consider more concentrated individual behaviour-change interventions such as Early Intensive Behavioural Intervention (*EIBI*) and Applied Behavioural Analysis (ABA), as being the most effective method of palliating the core symptoms of ASD (Dillenburger, Keenan, Doherty, Byrne & Gallagher, 2012; Matson et al., 2012). Although there is a consistent and strong evidence base for their success in changing targeted child problem behaviour and psychopathology, these are intense individual programmes that can mean additional stress for parents (Pennefather et al, 2018); and often necessitate a great deal of time and financial commitment as they command specialised professionals to implement them (McPhilemy & Dillenburger, 2013). Intensive behavioural interventions are also primarily focused on the child, and although practical benefits are observed by parents when therapy improves challenges such as toileting, feeding and managing behaviour (Grindle at al., 2009), the primary focus of ABA is rarely on parental outcomes. A recent review by Legg and Tickle (2019) highlighted the importance of parental needs among this population, which were considered as being emotional, informational and relational. The qualitative study highlighted the importance of parental information, and that support is vital for their acceptance of, and adaption to their child's diagnosis. A more recent scoping review of PET programmes specific to ASD was conducted by Dawson-Squibb and colleagues (2020) to explore programmes in relation to modalities of delivery, duration and overall objectives of the intervention. Thirty-seven studies across twenty countries (excluding the USA) were included, eight of which had been conducted in the UK. Of the 32 programmes represented, 31 focussed on strategies to manage or reduce problem behaviour, perhaps illustrating the impact of child challenging behaviour on parents. Although only eight aimed to provide parents with effective coping skills, over three quarters of the programmes sought to meet informational needs by providing strategies to help parents understand their child's social/ communication and behaviour difficulties. Emotional needs did not seem to be addressed to the same extent, with only one programme including parent and/or child emotional regulation strategies, and one with improving parent self-efficacy as a particular programme aim. The majority of the PET programmes identified were delivered in group format (Dawson-Squibb et al., 2020), which is encouraging when reflecting on the increased relational needs of parents in this population (Legg & Tickle, 2019), and the evidence-based recommendations made in the NICE guidelines that group led programmes should be offered to parents (NICE, 2013). The added benefit of group-based programmes in addressing family needs by promoting social and mutual support among parents, is well documented (Gardner et al., 2010; Furlong et al., 2013). Banach and colleagues (2010) reported that the most effective post-diagnosis support for parents of children with ASD were peer support groups, with those attending reporting substantial gains such as a sense of belonging and support through sharing their experiences. A recent review of parent-to-parent peer support groups conducted by Lee and colleagues (2023), recognised their importance in providing social, emotional and practical support. Although the shared experience of others was reported as being valuable towards parent coping strategies and adjustment, it was observed that most parents would prefer to learn strategies from professionals as opposed to fellow parents (Lee, Terol, Yoon & Meadan, 2023). The National Institute for Clinical Excellence (NICE) recommends that parents of children with autism should have access to the knowledge and competence of health care professionals during and following the diagnostic process (National Institute for Clinical Excellence, 2013), and group-led parent training programmes based on behavioural principles are recommended as first line treatment.

Behavioural parent training programmes

Parent training programmes founded on behavioural principles have consistently been shown to be effective in reducing dysfunctional parenting practices and improving child behaviour in populations at risk of conduct problems (Gardner et al., 2006; Hutchings et al., 2007). Teaching fundamental parenting skills not only improves child behaviour and wellbeing, but also results in positive parent outcomes (Gardner et al, 2006). Social learning theory

(Bandura, 1977) suggests that behaviours are learned through observation, and role modelling is a core component of many strategies taught in behavioural parent programmes. Patterson (1982) suggests that child behaviour problems are developed and maintained within the family by means of a coercive family process, primarily through the social learning process. A cycle of negative interaction can occur between parents and children when the parent uses ineffective parent behaviours, reinforcing similar behaviours in their children (Gardner et al., 2019). For example, a parent may raise their voice to gain the attention of a non-compliant child, which strengthens the parent behaviour if the child responds. This also models aggressive behaviour making it more likely that the child will act this way towards the parent when they seek parental attention. The parent and child then become caught in a coercive interaction cycle (Patterson, 1982; Patterson, 2002; Gardner et al., 2019). Behavioural parent training programmes aim to impact positive change by harnessing the parents' influence on the child by modelling positive behaviours (Bennett, Barlow, Huband, Smailagic & Roloff, 2013). Desired behaviours are reinforced by praise and/or reward, and unwanted behaviours are ignored (Gardner & Leijten, 2017). Programmes are typically delivered in group format through collaborative learning and discussion. Behavioural principles and effective parenting techniques are taught, such as engaging in child-led play to build positive relationships, giving praise and rewards to reinforce desired behaviours, and effective discipline strategies. Early intervention is most beneficial in addressing problem behaviours before they become entrenched, creating the best possibility of preventing later behaviour problems during adolescence and adulthood (Allen, 2011; Gardner et al., 2019), therefore programmes are often tailored for parents of younger children. Earlier intervention is also associated with a lower need for costly services as the child develops (O'Neill et al., 2013). Many of the key components and strategies taught in behavioural parenting programmes are found to be particularly relevant for parents of children with ASD, for example the focus on enhancing communication skills and strengthening relationships (Tarver et al., 2019). Due to the significant increase in prevalence and observed impact of autism on families and public health services, there is growing research examining the benefits of behaviour founded parent interventions to target the emotional and behavioural difficulties of children with autism and improving parenting skills (Postorino et al., 2017; Pellicano et al., 2013; Tarver et al., 2019). Programmes based on behavioural principles which incorporate the core theories of positive parenting are most effective (Hutchings & Lane, 2005), and in response, public and independently funded health authorities and charities support their roll out. Here within is a brief overview of the most common ASD specific behavioural parenting interventions being disseminated in the UK, to explore the modalities of programmes being delivered and their evidence of effectiveness.

Early Bird and Early Bird Plus

The Early bird programme is an autism specific parent-package developed in 1997 by the National Autistic Society for families of pre-school children aged up to 5yrs old. The Early Bird Plus is a later addition, designed for slightly older children between 4-10yrs, or those who have received a later diagnosis (National Autistic Society, 2010). Both versions of the programme are delivered in weekly 3hr sessions across 12 weeks, in a combination of group-led and individualised home visits. Six families typically attend each group and are supported during the difficult time immediately post diagnosis. The programme is structured to begin with informational sessions to help parents learn more about autism and associated social and communication deficits (Shields, 2001). The programme adopts a functional approach, and helps parents identify triggers to their child’s behaviour using the STAR (Setting, Triggers, Action, Results) method (Zarkowska & Clements, 2018). Time is also dedicated to helping parents understand their own way of communicating and learning more effective strategies to help manage their child’s difficulties. The programme is based around reassurance and support with a focus on building parent confidence through group discussions about parenting strategies. Groups are facilitated by two practitioners who need to have previous experience of working with children with autism (Shields, 2001). Early Bird has been widely disseminated to 27,000 families across 14 countries. Parents consistently report the programme's acceptability in routinely collected post-programme evaluations, with support from other parents being specified as the greatest benefit (Cutress & Muncer, 2014), however, there are very little pre-post studies using standardised parenting questionnaires (Pillay et al., 2011). A recent review by Dawson-Squibb and colleagues (2019) identified only lower-level evidence of programme efficacy, drawing attention to the lack of robust RCT studies assessing the programme and the need for more implementation science research to determine the feasibility of real-world delivery of programmes targeted towards this population (Dawson-Squibb, Davids & de Vries, 2019). Palmer and colleagues (2020) tested the feasibility of evaluating the delivery of Early Bird within routine clinical practice with the aim of informing a future trial to increase the evidence base for the program. Exploratory findings in the first known study of its kind, showed parent reported improvements in parent-child interaction and levels of parental stress. Recruitment and the acceptability of measures were also examined, which were positive and useful to inform further research (Palmer et al., 2020). Although measures of parental wellbeing and child behaviour were administered to assess their feasibility, no outcomes of effectiveness were reported.

Cygnet

The Cygnet programme was developed by the charity Barnardo's in the late 1990s as part of their parenting support provision. The intervention is similar to the Early Bird programmes in its approach, aiming to help parents understand autism and their child's different perspective and experience of the world, before supporting them to develop strategies and practical solutions to manage associated difficulties. Parents are also given the opportunity to meet others in similar situations and are signposted to useful services and organisations. The programme specifically targets the needs of families of newly diagnosed children aged 5-18yrs and is delivered in weekly 3 hr group sessions over six weeks. Up to six families can attend every group, with each being allocated two spaces so that both parents and other carers are given the opportunity to participate. An optional, informal follow up session is also held six weeks later. Groups are led by two trainers who must be qualified to degree level and have a minimum of 3yrs experience of working with families of children with autism (Barnardo’s, 2018). Delivery consists of a formal teaching component of didactic information sharing through power point slides, video clips followed by group discussions, and in group and homework activities. Although programme developers and those delivering the programme routinely report post-programme feedback showing the programme to be highly acceptable by parents (Barnardo’s, 2018; Robson, 2010), no independent evaluations have been conducted to date. Attempts have been made to assess parental competence as part of the feedback process, although none have included the use of standardised measures. There is also little research on the effectiveness of the programme. The first study to include standardised measures of effectiveness was a small non-randomised controlled trial conducted by Stuttard and colleagues (2016). Recruitment was good with 58 parents allocated to receive the intervention, and 62 to a wait-list control group, however the sample was under-powered to assess differences. Promising improvements were observed in child behaviour, along with significant improvements in parental sense of competence in favour of the intervention group. Conclusions were drawn that the programme could be useful to this population, and that the promising results justified a larger trial, with a longer term follow up of 6-months suggested to better assess programme outcomes. Although further research was recommended, a larger definitive trial has not been published as yet, therefore empirical evidence of effectiveness for the Cygnet programme is still lacking.

Stepping Stones -Triple P

Contrastingly, the Triple P Positive Parenting Programmes are a well-researched, evidence-based series which incorporate core positive parenting principles to promote children's social and emotional competence (Sanders, 1999). Five intervention levels of increasing intensity are available for families of children from birth to 16yrs, with different variants of the programme presented to include group delivery, individual sessions, and a hybrid model of both group and 1:1 sessions. The Stepping Stones: Triple P (SSTP) programme has been developed for parents of children aged 0-12yrs with a developmental disability, including ASD, Down Syndrome and Cerebral Palsy, and moderate to severe behavioural problems (Tellegen & Sanders, 2013). The Triple P parenting programmes upon which the SSTP is established have been rigorously researched and have an evidence rating of '2' in the Early Intervention Foundation (EIF) guidelines. The EIF is an independent charity that reports on effective early interventions by assessing the quality and strength of evidence and has categorised the SSTP programme as having preliminary evidence (evidence of improving a child outcome from a study involving at least 20 participants using validated measures). The aim of the SSTP programme is to help parents identify, observe and address problem behaviours. Techniques are based on empirically supported social learning theory principles, to target their child's specific problems. Topics focus on helping parents adapt to their child's disability, managing challenging behaviour and specific challenges (e.g., sleep disturbance, toilet training), and practicing self-care. The programme is delivered over 10 weeks, in a mixed format of groups (between 4-6 parents) and 1:1 sessions, consisting of interactive discussion, modelling, role play and feedback from facilitators (Tellegen & Sanders, 2013). Like all Triple P programmes, the SSTP has 5 levels of increasing intensity from very extensive (targets entire population) to narrow (targeting high risk children). Resources include detailed parent booklets, therapist manuals, videos and other materials for each level.

Ruane & Carr (2019) conducted a review examining the programme's effectiveness, which identified 16 international studies. Only controlled studies were included, enhancing the validity of the review. The analysis found the programme to be effective in improving behavioural outcomes in children with disabilities, and positive parent outcomes. However, the analysis was conducted with all five levels of programme intensity combined, therefore conclusions should be interpreted with caution, as they do not examine each level of the programme separately. Findings were promising, although it could be argued that the programme itself raises concerns with regards to programme fidelity. The programme is of 'flexible delivery', with parents able to choose the level of intensity and which components to complete (Tellegen & Sanders, 2013). Whilst flexibility can be an advantage for some parents, particularly when considering the idiosyncrasies of children with ASD, there could be a risk that the programme is not being delivered with fidelity. Although the programme begins with a comprehensive assessment of parent-child interaction and family needs to place dyads into groups with similar objectives; different developmental disorders can present varied challenges to address during group discussion, which could be challenging for facilitators to implement as intended. The literature repeatedly emphasises the complex and individual challenges of parents of children with ASD, thus group discussion to adequately resolve individual challenges could be difficult with numerous disorders to consider. Despite this, outcomes across studies were a promising indication that the programme was acceptable for this population (Ruane & Carr, 2019), and reinforced the findings of previous studies of the programme (Roux, Sofronoff & Sanders, 2013; Tellegen & Sanders, 2013). Only short-term evaluations of up to 3-months exist (Ruane & Carr, 2019), therefore there is no indication of possible long-term maintenance outcomes. There is also a lack of studies that included parental wellbeing, or wider-family outcomes, which was highlighted as a limitation, and an important focus for future research (Tellegen & Sanders, 2013). Over 70%

of the studies identified had been conducted in Australia (Ruane & Carr, 2019), where the programme has been developed, therefore robust evaluations of the programme need to be conducted in other countries and healthcare models to fully demonstrate generalisability of effectiveness.

Riding the Rapids

The Riding the Rapids: Living with autism or disabilities programme was developed in the UK by a specialist team of clinical psychologists in response to the lack of available programmes specifically targeted to meet the needs of families of children with ASD or intellectual disabilities (Todd et al., 2010). The programme aims to improve the emotional wellbeing and quality of life of children with disabilities and their families, using social learning principles to teach parents skills to manage child behaviour difficulties and sleep disturbance (Stuttard, Beresford, Clarke, Beecham, Todd & Bromley, 2014). The course is aimed at parents of children aged 3-11yrs, and consists of 10 weekly 2hr sessions, followed by a booster session three months post intervention. Similar to the forementioned programmes, Riding the Rapids is delivered in group format, and aims to help parents understand their child’s behaviour through functional analysis before supporting them to problem solve certain difficulties they may have with their child (Todd et al., 2010). Similar to the Early Bird programme, the first session focusses on using the STAR (Setting, Triggers, Action, Results) approach (Zarkowska & Clements, 2018) to help parents target a specific behaviour they find difficult to manage. Programme content is adapted to meet the needs of particular groups of children, but typically focuses on helping parents learn effective limit setting strategies, the use of communication techniques and tools (such as PECS), positive reinforcement and play. Programme content is delivered didactically by two facilitators, one of whom must be a clinical psychologist who conducts a behaviour analysis. Parents select a specific target behaviour and learning is focused on that behaviour throughout the course, which given the many co-morbid emotional challenges and behaviours associated with autism, could be limiting. Riding the Rapids places importance on parent outcomes more so than the other programmes, with wellbeing made a priority during sessions, with small treats such as magazines and toiletries given to parents to encourage self-care. Parents are also taught cognitive behavioural strategies for stress management. The programme is routinely disseminated as part of the Child and Adolescent Mental Health Service (CAMHS) learning disabilities teams' provision in the UK. Stuttard and colleagues (2014) conducted the first independent evaluation of the programme in a pragmatic, non-randomised control study with 48 parents receiving the intervention, and 28 placed in a control group. Standardised measures of child behaviour and parental sense of competence were administered and showed improvements in both outcomes in favour of the intervention group, which in addition to the high level of acceptability reported by parents, was a positive indication that the programme is beneficial to this population. Although focus is placed on parental wellbeing in the programme, no parental mental health measures were included in the study, which could be considered a limitation. However, intervention parents were followed up at 3month and 6month time points, which adds valuable evidence towards programme effectiveness. The programme was targeted towards ASD and learning disabilities, therefore again it could be difficult to meet the very specific needs of parents of children with ASD. The behaviour analysis approach is evidence based and shown to be effective. However, considering the complex co-morbidities associated with autism, choosing one behaviour to focus all learning on could be limiting. The literature has also suggested that focus on intensive behaviour change can be a stressor (Matson et al., 2012). This is especially relevant for parents of children with ASD, give the reported low rates of self-efficacy, and fear of failure to change the behaviour could impair intervention outcomes. The study concluded however that due to the lifelong impact of ASD and intellectual disabilities, longer term data would be beneficial to gather clearer evidence. The study determined a lack of robust evaluations of group-delivered parent programmes specific to parents of children with disabilities remains despite the continued development of more targeted interventions (Stuttard et al., 2014).

Caregivers Skills Training (CST)

One such example of a newly developed intervention is the Caregiver Skills Training (CST) package. The World Health Organisation (WHO) recently collaborated with international partners across twenty-one countries to develop an evidence-based programme specific to families of children with autism and other developmental disabilities, in response to an identified need for targeted psychoeducation for this population, particularly in low-income countries (Salamone et al., 2019; WHO, 2010). Based on the extremely well-researched principles of social learning theory, applied behaviour analysis and positive parenting, the programme teaches strategies to help caregivers develop their child's adaptive and communication skills and reduce problem behaviour (Tekola et al., 2022). The programme adopts a hybrid model of 9 weekly sessions and 3 individual home visits and uses a range of learning techniques to deliver the content including modelling, role-play, examples of positive practice and group discussion. The CST programme has also been piloted via telehealth delivery, as an alternative model with a view of removing barriers to accessibility (Montiel-Nava et al., 2022). However, it is important to consider that this removes the element most reported as being the biggest benefit; meeting and sharing experiences with other parents in the same situation, therefore this would need evaluating further. The existing studies of the programme focus only on the acceptability and feasibility of the programme, which is important for newly developed programmes before any further research can be considered to ensure they are being delivered as intended. The need to examine programme effectiveness is proposed as a future research direction in all the literature reviewed about the programme (Salomone et al., 2019; Tekola et al., 2022; Montiel-Nava et al., 2022) however, to date there have been no evaluations of effectiveness.

Conclusion

The dearth of data on the effectiveness of parent programmes for this population is well documented throughout the literature (Dawson-Squibb et al., 2020; Salomone et al., 2019; Tarver et al., 2019), despite the continued recommendations of their need and potential benefits for this population of parents (NICE, 2013; WHO, 2010; WHO, 2017; WHO, 2022). The needs of parents of children with autism are greater than other populations, and it is also important to remember the idiosyncratic nature of autism, which may reflect the wide range of interventions available, meaning the concept of parent training can apply to a variety of different treatments within the field of autism (Bearrs et al., 2015). It’s clear that not one programme will be suitable for all, however there are many common factors among the most widely distributed programmes that are shown to be effective. When evaluating any intervention, it is useful to include comparable programmes, taking factors such as structure, modality and purpose into account. It is also important to have robust, well-designed studies that can evaluate programmes within the context they are to be implemented in the real-world.

Key messages

- There is no treatment for autism. However, if parents are well-supported and are given the opportunity to develop skills to cope with the challenges of parenting a child with autism, they are more likely to have positive outcomes.
- This chapter highlights the recommendations that parents and carers should receive psychoeducation to provide them with information and support to help them understand and cope with their child’s diagnosis.

- Findings from the included studies are a positive indication that behavioural parent programmes are acceptable to parents of children with ASD.
- Although there is some evidence for programme effectiveness with regards to parental competence and satisfaction, there is a need to assess any intervention outcomes with regards to parental mental health.
- Given the lifelong nature of autism, it is also important to assess the longer-term outcomes of parent programmes, along with any wider-reaching benefits to other family members.
- It is particularly important to evaluate newly developed programmes to build their evidence base with regards to programme effectiveness for families, and economic viability to inform possible future implementation.
- There is a lack of replicability in the evaluation of parenting programmes specific to ASD, therefore further research is needed in this field.

Chapter 4

Design and Methods

Introduction

The PALACES trial was a pragmatic randomised controlled trial (RCT) conducted to evaluate the real-world feasibility of delivering the newly developed Incredible Years Autism Spectrum and Language Delays programme within existing NHS services (Williams, Hastings & Hutchings, 2020). The aim was to examine trial methods and procedures, and to examine whether it was possible to recruit and randomise parents to either intervention or wait-list control groups. Programme adherence and satisfaction were assessed, along with the level at which facilitators were able to deliver the programme with fidelity to the manual. Although the sample was not powered to test effectiveness, initial parent and child outcomes were explored.

This doctoral thesis is designed as a further exploration of the programme alongside the feasibility trial, to address outstanding questions with regards to taking the programme forward and to inform a larger definitive trial. Given the significant financial impact of autism on both families and publicly funded services (Buescher, Cidav, Knapp & Mandell, 2014; Knapp et al., 2009), the author's initial focus of additional research involved exploring the costs of programme delivery. Based on previous IY studies of long-term benefits of parent training programmes (Bywater et al., 2009; Webster-Stratton, Rinaldi & Reid, 2011), and the lifelong nature of autism (Zeidan et al., 2022), it was also of interest to examine any possible longer-term outcomes of the IY-ASLD programme. Furthermore, due to the known impact of autism on wider family members (Griffith, Hastings & Petalas, 2014), potential benefits of the programme for siblings were also explored. How this thesis has been informed by and extends from the feasibility trial is described in this chapter, along with the trial design and methods.

The Parenting for Autism, Language, And Communication Evaluation Study (PALACES).

The PALACES trial was conducted within four specialist NHS children's services across North and Mid Wales. These multidisciplinary teams consist of child psychologists, specialist nurses, paediatricians and speech and language therapists, who are responsible for the assessment, diagnosis and treatment of children with mild to moderate intellectual disabilities and/or neurodevelopmental disorders such as ASD and ADHD.

Methods

Participants and trial design

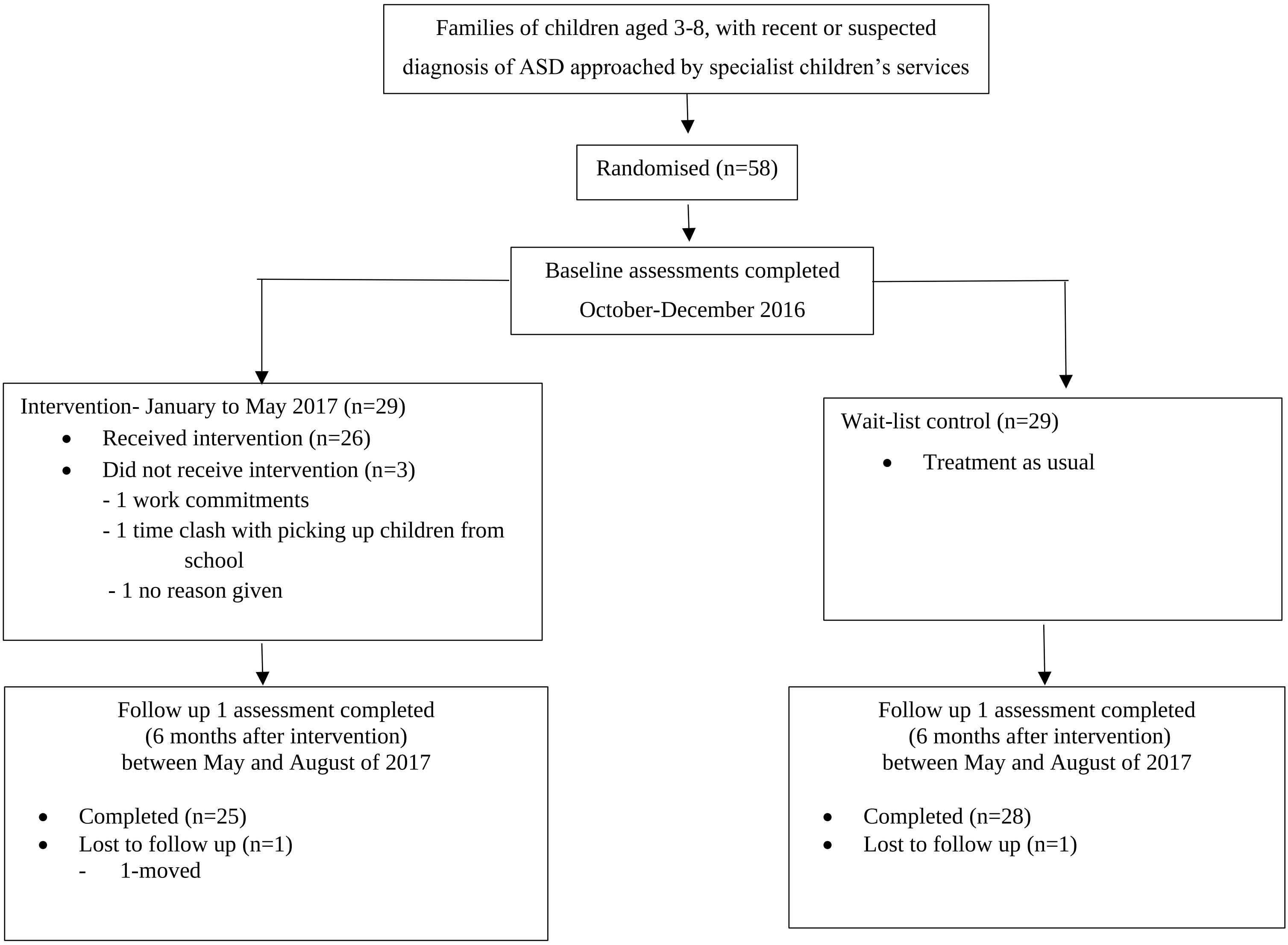
Trial participants were the primary caregivers of children aged 3-8 years, known by the service to have a recent or strongly suspected diagnosis of ASD. Exclusion criteria included a) caregiver did not have a good understanding of English; b) attendance of another parenting programme during intervention phase of trial; c) family was in crisis (e.g., child was at risk of residential placement); d) short term foster placement; e) child placed on child protection register; f) refusal to give participation consent. There were no exclusion criteria with regards to any co-occurring intellectual disability.

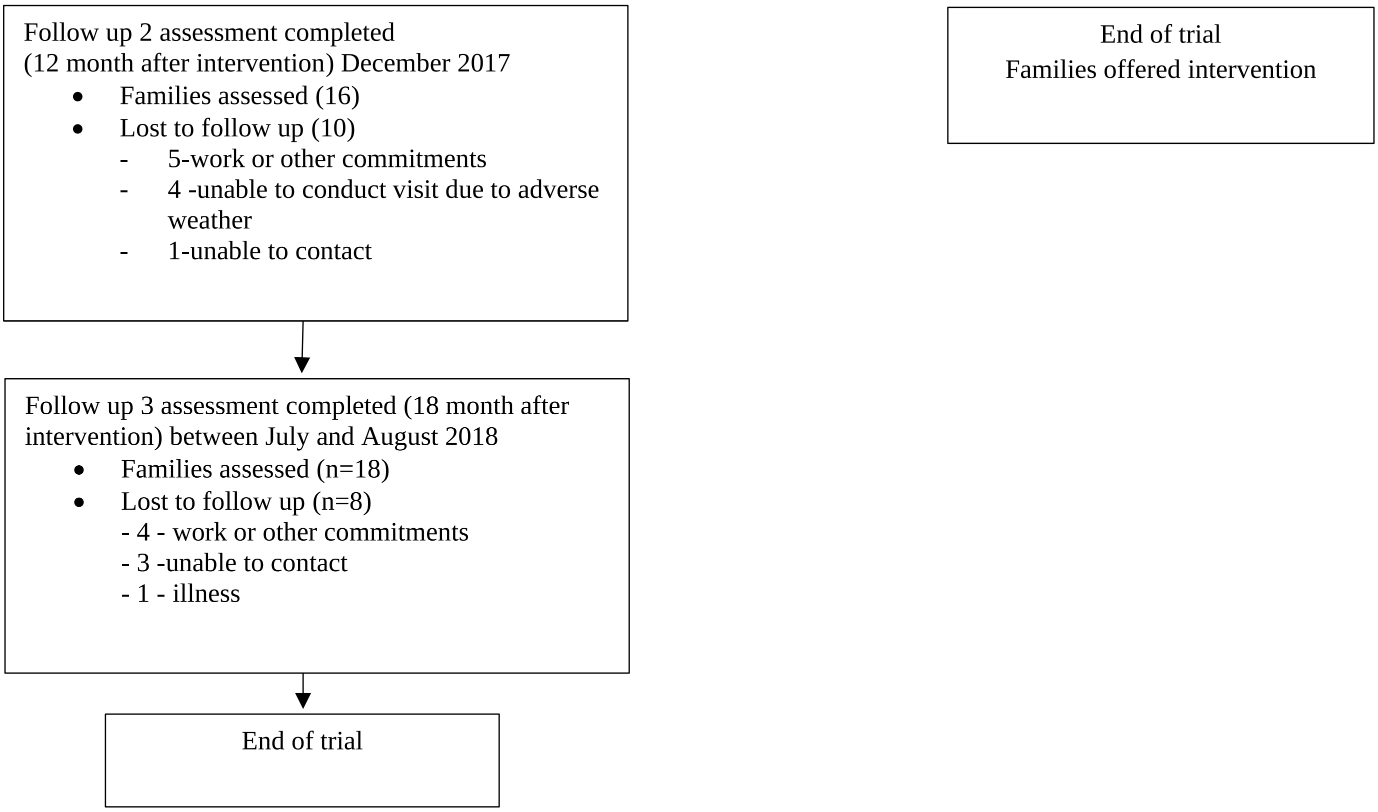
Recruitment and randomisation

Eligible families were recruited between October and December 2016. Specialist children's service practitioners contacted 65 parents already known to them to gauge their interest in participating in the trial. Of these, four were not eligible, and three declined to take part. Ultimately, fifty-eight parents were randomised into two groups; 29 to receive intervention immediately, and 29 into the wait-list control group. Families in the control condition received treatment as usual during the 6-month wait to receive the programme, meaning they continued to access any services they were already involved with, excluding any parent training interventions. Three participants withdrew prior to intervention delivery due to other commitments, therefore 26 received intervention (see Fig 4.1 for participant flow through the trial).

Parents who expressed an interest in participating gave verbal consent for their contact details to be shared with the research team, who then contacted parents directly within one week to discuss the project further. A home visit was arranged to ensure that caregivers had read the information sheet and to answer any arising questions. Written consent was obtained before any baseline measures were collected. The sample size was based on recommendations from the National Institute for Health Research (NIHR), who provide guidelines to inform research trial design. The determined number was thought to be adequate (according to NIHR guidelines) to test out all aspects of the feasibility trial, and to estimate a sample size for a further definitive trial.

Fig 4.1 Flow of participants through study:





Follow up 2 assessment completed
(12 month after intervention) December 2017
- Families assessed (16)
- Lost to follow up (10)
  - 5-work or other commitments
  - 4 -unable to conduct visit due to adverse weather
  - 1-unable to contact

Follow up 3 assessment completed (18 month after intervention) between July and August 2018
- Families assessed (n=18)
- Lost to follow up (n=8)
  - 4 - work or other commitments
  - 3 -unable to contact
  - 1 - illness

End of trial

End of trial
Families offered intervention

Randomisation and masking

Once baseline measures were collected, families were randomly allocated on a 1:1 ratio to either receiving the intervention immediately (IY-ASLD) or to wait-list control group (treatment as usual), who were subsequently offered the programme after 6-month follow up data collection. Randomisation was undertaken by the North Wales Organisation for Randomised Trials in Health and Social Care (NWORTH), who then informed the trial administrator of allocations. Each site facilitator was notified by the trial administrator, who then informed parents into which group they had been randomised via letter. All researchers involved in data collection and analysis remained blind to group allocation until after the six month follow up.

Materials

Intervention

Exploring the cost and outcomes of the IY-ASLD® programme

The IY-ASLD® programme is a group-based intervention specifically targeting the needs of parents of children with ASD (Webster-Stratton, 2015). The programme is divided into 12, weekly 2 hr sessions, although the programme developer has since recommended that it may take longer than this to complete the programme. During the PALACES trial however, the intervention was delivered over 12 weeks in all four centres to ensure consistency.

The programme focuses on parent-child relationships, along with a range of developmental outcomes such as language, social emotional and adaptive skills structured into the following eight topics; (1) child-directed narrative play; (2) pre-academic and persistence coaching; (3) social coaching; (4) emotion coaching; (5) developing imagination through pretend play; (6) promoting children's self-regulation skills; (7) using praise and rewards to motivate children; and (8) effective limit-setting and behaviour management. Programme leaders help parents to learn new skills by encouraging the group to watch and discuss video vignettes, facilitating role play activities to practice skills and setting homework tasks. Leaders encourage the use of these skills at home via weekly telephone calls between sessions. All centres delivered the intervention concurrently and according to the programme manual identically; however, one centre delivered some sessions on a one-to-one basis when parents had missed a session. Caregivers' partners were also given the opportunity to attend, of whom 11 attended at least one session.

Seven group leaders were trained (in November 2016) to deliver the IY-ASLD by the eighth leader, who was a certified IY trainer. Six facilitators were clinical psychologists, one was a mental health nurse and one a community nurse. Five had previous experience of working with children with ASD, of whom at least one was a co-facilitator in each group. Adherence to programme content was addressed by providing manuals and materials. Facilitators completed weekly checklists detailing which components had been covered and attended fortnightly supervision to enhance fidelity. Group sessions were recorded and discussed during fortnightly supervision. Delivery of the intervention took place between January and May 2017. The intervention was delivered concurrently across sites, between baseline and 6-month follow up data collection.

Measures

Parent and child factors were assessed using a combination of standardised questionnaires and direct behavioural observation, drawing on those validated in similar trials. Data collection was completed during home visits with both intervention and wait list control at baseline and 6-month follow up as part of the PALACES feasibility trial (Williams, Hastings & Hutchings, 2020). Data collection was conducted by the thesis author at 12-month and 18-month datapoints for the purpose of further exploration of the programme (see below). Measures were administered to primary caregivers from both intervention and control groups at baseline and at follow up one (6-months). Measures were administered to caregivers from the intervention group only at follow ups two and three (12 and 18-months). The intervention was delivered within the 6-month period between baseline and follow up one. See Table 4.1 below for measures and data collection timepoints.

Timepoint	**Baseline**	**Follow up 1 (6-month)**	**Follow up 2 (12-month)**	**Follow up 3 (18-month)**
Administered to	**Both conditions**	**Both conditions**	**Intervention group only**	**Intervention group only**
Measure				
Demographics	√			
Child Outcomes				
Child Behaviour Checklist	√	√	√	√
Social Communication Questionnaire	√	√	√	√
Vineland Adaptive Behaviour Scale	√		√	√
Parent Outcomes				
Parenting Stress Index	√	√	√	√
Beck Depression Inventory	√	√	√	√

Parenting Scale	√	√	√	√
Parent-child Observations	√	√	√	√
Sibling	√	√	√	√
SDQ				
Cost Diaries		√		

Table 4.1 Measures and timepoints collected

Demographics

Family demographic information covering a range of characteristics was gathered at baseline including parents age, gender, marital status, level of education and employment status of household (See Appendix A). The age, gender and diagnostic status (diagnosed vs. suspected) of the child diagnosed with ASD (index child) was established based on information provided by the specialist children's services involved in the trial. Parents were asked about the number of other children in the family, their age and gender. Sibling measures were completed about the sibling closest in age to the index child.

Child Outcomes

Child Behaviour

Child behaviour difficulties were measured using the parent-rated Child Behaviour Checklist (CBCL) (Achenbach & Rescorla, 2000) (See Appendix B). The measure consists of two subscales: externalizing problems and internalizing problems, as well as a total score on which higher scores indicate more problem behaviours. Parents rate each item on a three-point scale from 0 (Not True) to 2 (Very True). A total score of above 63 indicates clinical levels of behaviour problems. The validated inventory has shown good internal consistency (Pandolfi, Magyar & Dill, 2009; Pandolfi, Magyar & Dill, 2012), and is routinely used in the assessment of co-occurring behaviour problems for children with ASD (Hoffmann, Weber, König, Becker & Kamp-Becker, 2016).

Child Social Communication

Child social communication behaviours were assessed using the Social Communication Questionnaire (SCQ; Berument, Rutter, Lord, Pickles, & Bailey, 1999), (See Appendix C) a validated 40-item measure developed from the Autism Diagnostic Interview-Revised (Le Couteur, Lord, & Rutter, 2003). Parents are asked to give a response of Yes or No to each question. All Yes responses are then summed to give a score between 0 and 40 with higher scores indicating more severe symptoms. A cut-off score of 15 can be used as an indication of possible ASD. There are two versions of the questionnaire that assess both lifetime and current behaviour; the latter was used for this study. The measure has demonstrated good internal consistency (Snow & Lecavalier, 2008) and performs well psychometrically in the assessment of ASD (Barnard-Brak, Richman, Chesnut & Little, 2016).

Child Adaptive Skills

Child adaptive skills were assessed using the Vineland Adaptive Behaviour Scales II Parent/Caregiver Rating Form (VABS; Sparrow, Balla, Cicchetti, Harrison, & Doll, 1984); (See Appendix D), a validated semi structured interview that assesses adaptive functioning in communication, socialisation and daily living skills. The overall adaptive behaviour standard score is used which has a population mean of 100 and standard deviation of 15, with higher scores demonstrating better adaptive skills. The cut-off used to indicate low levels of adaptive behaviour is <70. The measure is widely used for the diagnostic classification and treatment planning of ASD (Balboni, Tasso, Muratori & Cubelli, 2016), and has demonstrated good internal consistency (Miles et al., 2004)

Parent Outcomes

Parent mental health

Parental mental health was examined using two standardised questionnaires that have been consistently used in similar research.

Parental stress was assessed using The Parenting Stress Index-Short Form (Abidin, 1990); (See Appendix E) a 36-item inventory used to measure the stress experienced by parents of children up to the age of 12 years. Parents rate each item on a five-point scale from 1 (Strongly Disagree) to 5 (Strongly Agree) with higher scores indicating more stress. A cut-off score of 90 is used to signify clinical levels of stress. The measure has been widely used with a range of populations including those with ASD (Lecavalier, Leone & Wiltz, 2006), and has shown good internal consistency (Zaidman-Zait, Mirenda, Zumbo, Wellington, Dua & Kalynchuk, 2010).

The Beck Depression Inventory (BDI-II; Beck, Steer, & Brown, 1996) (See Appendix F), was administered to measure the level of parental depression. This validated questionnaire consists of 21-items assessing the severity of characteristic symptoms and attitudes typically associated with depression. Parents rate each item on a four-point scale with higher scores representing greater levels of depressive symptoms. Cut-off score guidelines for the BDI-II are given with the recommendation that thresholds be adjusted based on the characteristics of the sample, and the purpose for use. A total score of 0–13 is considered minimal range, 14–19 is mild, 20–28 is moderate, and 29–63 is severe. The measure has good test-retest reliability (r = 0.93), good convergent validity (r = 0.93) and high internal consistency (Beck, Steer & Brown, 1996).

Parenting Skills

The primary aim of the intervention was to improve parenting skills, which were assessed using the ‘Parenting Scale: A measure of dysfunctional parenting in discipline situations’ (Arnold, O’Leary, Wolff, & Acker, 1993), (See Appendix G). The 30-item self-report questionnaire contains three sub-scales measuring recognised factors associated with child behavioural difficulties of ineffective discipline: laxness, over-reactivity and verbosity. The 30 items also sum to create a total score of ineffective practices. Respondents rate their tendency to engage in certain behaviours on a seven-point Likert scale fixed between two alternative responses, for example, “When I tell my child not to do something” the response on the left is “I say very little” and on the right “I say a lot.” The scores are summed and averaged for each subscale, with higher scores representing more dysfunctional parenting practices. The measure has been used in several similar studies at the centre and internationally (Hutchings et al., 2007; Bywater et al., 2011; Gardner et al., 2006; Sanders, Baker & Turner, 2012), including with this population (Hutchings et al., 2016; Charman et al., 2021). The measure exhibits adequate internal validity (Rhoades & O'Leary, 2007), in addition to demonstrating significant correlations with observational measures of child problem behaviour (Arnold et al., 1993).

Parenting skills were also assessed with a 10-minute observation of parent–child interaction using categories from the Dyadic Parent–child Interaction Coding System (DPICS; Eyberg & Robinson, 1981), (See Appendix H). Parents were asked to engage in child-led play, which could be inside the house or outside in the garden, depending on the child’s preference. Parents could suggest activities, but it had to have been chosen by the child. Some subscales of the DPICS were grouped together to form the following behaviours to be coded: positive parenting (physical positive, positive affect), praise (unlabelled and labelled), social-emotional coaching, reflections, questions, and negative parenting (physical negative, negative commands). The frequency of each behaviour within the 10-min observation was coded meaning that higher scores represent a higher frequency of the behaviour. The DPICS has been widely used in previous studies of parenting programmes (Hutchings et al., 2017; Webster-Stratton, Rinaldi & Reid, 2011), and has shown good levels of reliability (Robinson & Eyberg, 1981). Inter-rater agreement, based on intraclass correlations, was very high (0.96 – 0.99).

Sibling Adjustment

The behavioural and emotional difficulties of siblings were assessed using The Strengths and Difficulties Questionnaire (Goodman, 1997) (See Appendix I), a globally recognised 25-item behavioural screening inventory for child psychopathology. Individual items (e.g., 'rather solitary, tends to play alone', 'generally obedient, usually does what adults request') are scored with a choice between three responses (0 = 'not true', 1 = 'somewhat true', or 2 = 'certainly true'). The measure consists of four problem focused subscales; emotional problems, peer problems, hyperactivity and conduct problems; and a pro-social behaviour scale. Results are calculated by summing scores on relevant items for the five subscales and a total problem score is the summation of the four problem scales. Questions 7, 11, 14, 21, and 25 are reverse scored prior to analysis. Strengths are identified as a result of high scores on the pro-social scale. Difficulties are ascertained by a high total score on the individual problem subscales and the summation of scores on the remaining four problem sub-scales. An overall score can also be calculated for internalising behaviour with the summation of scores from the emotional and peer problems subscales; and for externalising behaviour by summing scores for the hyperactivity and conduct problems subscales. The SDQ is a well-established, standardised measure of behaviour used in many studies of conduct disorder (Hutchings et al, 2007; Jones et al, 2008; Hutchings et al, 2011), and has previously been used to examine adjustment in siblings of children with autism (Cebula, 2012; Petalas et al., 2012). Two validated versions of the questionnaire were used depending on the child's age. There is a version for children aged 4-18, and a slightly modified version for children aged 2-4, whereby an item on reflectiveness *(Q.21)* is softened *('thinks things out before acting'/'can stop and think before acting')*, and two items on antisocial behaviour *(Q18 & Q22)* are replaced by items on oppositionality *('often lies or cheats'/ 'often argumentative with adults' & 'steals from home, school or elsewhere/ 'can be spiteful to others').* The measure exhibits good internal consistency for all subscales ranging from *($\alpha = 0.71$ to .82),* and high overall reliability *(α = .88).* Parents were asked to complete the questionnaire about a sibling in the family closest in age to the child with ASD. Measures were administered to primary care givers from both intervention and control groups at baseline and at follow up one (6 months later).

Programme Cost

A micro-costing of programme delivery was conducted from a public sector, multi-agency perspective alongside the trial. Data was gathered and extracted from previously established cost diaries (See Appendix J) which were completed weekly by group leaders (see Chapter 6 for full details).

Data analysis

Data from parent and child measures were checked for normality and found to be not normally distributed. All measures were therefore analysed using non-parametric test. Wilcoxon related-sample signed rank tests, a non-parametric alternative to t-tests, was used to compare differences in Median and ranges between data points (Baseline to six-month follow-up, and six-month to 18-month follow-up). (See Chapter 7 for further detail with regards to long term outcomes data analysis). Mann-Whitney tests were used to compare differences between sibling data from intervention and control groups post intervention (See Chapter 8 for further detail with regards to sibling data analysis). Missing data from individual items on questionnaires were dealt with based on the guidelines in questionnaire manuals, and in all cases, missing items were prorated. Some of the parents faced challenges engaging the child in a full 10-minutes of interaction, therefore only the first 5 minutes of the observations have been coded for all participants in this measure. Outcome effect sizes *(r)* are interpreted based on Cohen’s d (0.1-small, 0.3-medium, 0.5-large).

Ethical Approval

Ethical approval was granted by Bangor University Research Ethics Committee (Application number 2016-15768) and by the National Research Ethics Service of the National Health Service (Application No. 16/WA/ 0224).

Doctoral Thesis Design

The pragmatic nature of the feasibility trial afforded numerous opportunities to expand upon the evidence base of the newly developed programme. Due to the grant awarded to fund the research, steering group meetings were held quarterly throughout the year of planning and running the trial. Attendees consisted of the research team, staff from Early Intervention Wales Training and group leaders from each centre; all of whom contributed to decisions about cost gathering methods and discussed any issues with regards to programme delivery. The doctoral candidate held an integral role within the steering group, taking an active responsibility for decision making with regards to moving the trial forward, maintaining contact with participants following the intervention and facilitating longer term data collection. The author drew knowledge from this involvement to formulate further research questions which emerged from an amalgamation of own research interests, the reviewed literature and organically from the initial findings of the feasibility study. Four studies transpired that the author felt were important research questions to address about a newly developed programme, and that would be helpful in designing a larger definitive trial.

Given the substantial financial impact of autism both on family and publicly funded services, the author felt that expanding upon economic evidence was important. A systematic review of current literature around economic evidence for behavioural parenting programmes specific to ASD was conducted. The review aimed to establish the levels of existing research and examine the methodological issues to inform the design and conduct of future trials. A clear search strategy based on the Cochrane Systematic Review Guidelines (Structure of a Cochrane Review, 2018) was followed. The doctoral candidate completed the Cochrane

Interactive Learning ‘Conducting an Intervention Review’ training course prior to commencing the search, to ensure the review was conducted rigorously (See Chapter 5 for further detail). The systematic review was important to inform research questions in relation to the financial impact of autism, and any future economic evaluations of the newly developed programme. Trial-based economic evaluations are an important aspect of intervention science; with real-world RCT’s being the most accurate and effective means of assessment (Hughes et al., 2016), therefore an economic evaluation alongside the feasibility is robust. The doctoral candidate chose the second study to be a detailed micro-costing of delivering the IY-ASLD® programme both within the feasibility trial and the likely cost of future roll-out within existing services. Costings of programme delivery help inform the design of future definitive trials and policy decision makers and are a useful step towards further economic evaluations such as future cost effectiveness analyses. This was the first micro-costing of the programme to be undertaken. An established, detailed framework was followed to ensure a precise and rigorous analysis of the costs gathered in weekly cost diaries (see Chapter 6 for further details). Given the lifelong implications of autism, the author also considered the importance of examining the longer-term outcomes of the programme. It was deemed valuable to assess participant retention over time, and the feasibility of collecting longer term data in any future definitive trials. Data collection was therefore repeated at 12 and 18-months. Although the feasibility trial was not powered to assess programme effectiveness, the doctoral candidate was eager to expand upon the exploratory findings of the post intervention parent and child outcomes (see Table 4.1). for measures and data collection time points). For that reason, in addition to reporting on retention, long term follow up measures were explored to assess whether any gains were maintained or if new behaviours had emerged over time, by comparing medians across time points. The author thought this to be useful, as previous literature demonstrates that improvements to parenting behaviours are sometimes not immediate, but rather can be observed after some time of putting learning into practice (Furlong et al., 2013). In addition to any long-term benefits, the author felt that future research should explore the extent to which the programme may benefit other family members, given the considerable literature showing the family-wide impact of autism. The author decided to address this in the final study (Chapter 8), by examining measures to assess sibling adjustment. In addition to providing preliminary data of any wider-reaching programme benefits, the feasibility of gathering data from other family members will be helpful to inform future research proposals.

The four studies designed for this thesis utilise the robustness of the trial design and breadth of measures to expand upon the broader picture of potential benefits of the programme for this population of families. These are the first studies of accurate costs, long term outcomes and further family benefits of the IY-ASLD® programme and will contribute towards the evidence base for the newly developed programme and play a crucial role in informing future definitive trials.

Chapter 5

Systematic review of the economic evidence

Exploring the cost and outcomes of the IY-ASLD® programme

Introduction

Due to their complex needs, the cost of supporting children with ASD can be far greater than that of typically developing peers (Beecham, 2015). The estimated lifetime costs of supporting children with ASD in the UK is between £3.1 and £3.4 billion (Byford et al., 2015). Autism related costs can be categorised as follows; Direct medical costs, which include all health care needs (e.g., GP’s, inpatient and outpatient, prescriptions, physiotherapy etc); Direct non-medical costs (e.g., education, childcare and respite, behavioural therapies), and Caregiver productivity costs (Lavelle, Weinstein, Newhouse, Munir, Kuhlthau & Prosser, 2014). The highest proportion of annual public spending is accounted for by special education which is estimated to be £2546 (for children aged 2-3yrs), and £14,006 (for children between 4-17years) per capita, with costs higher if the child also has a co-morbid intellectual disability (Buescher, Cidav, Knapp, & Mandell, 2014). Families of children with autism use healthcare services more frequently than those of neurotypical children (Rogge & Janssen, 2019). A peak is observed during periods of child assessment and diagnosis, but generally the costs remain consistent and life-long for the individual with ASD (Horlin, Falkmer, Parsons, Albrecht & Falkmer, 2014). The greatest percentage (95%) of costs are paid for by public funding, with the remainder falling on families (Barrett et al., 2012), for example through loss of income or out of pocket expenses. Parents in this population are thought to be at a financial loss in comparison to those of typically developing children and work an average of seven hours less per week than those of children without additional needs (Buescher, Cidav, Knapp & Mandell, 2014). The majority of parent time is spent managing child emotional and behavioural challenges (Knapp, Romeo & Beecham, 2009), which is described as also being the main predictor of parental mental health problems (Dabrowska & Pisula, 2010). Failure to address child behaviour difficulties through early intervention can result in more severe, intractable problems (Willey, 2003), which can have long-lasting effects on both a family and societal level (Simonoff, Jones, Baird, Pickles, Happé & Charman, 2013). Knapp and colleagues (2009), report that the prevalence of autism is in fact higher than previously estimated, and although time trends have meant fewer children and people with autism live in residential placements then a few decades ago, there is still a greater level of life-long care needs which now often falls to parents (Knapp, Romeo & Beecham, 2009). Long-term prospects of children with autism are greatly improved due to earlier diagnosis and intervention, with special education often being provided at an earlier age of between 5-9yrs (Howlin & Moss, 2012). However, this results in greater costs for the additional learning support which is observed in this population (Barrett et al., 2012). Although improvements have been made to special education provision, children with ASD still generally maintain poorer outcomes when compared to their typically developing peers (Keen, Webster & Ridley, 2016), which is often due to inattention and/or behaviour problems (May, Rinehart, Wilding & Cornish, 2013). Poorer academic attainment can result in later disadvantage with regards to employment in later life (Chen, Leader, Sung & Leahy, 2015). The impact of autism can therefore be seen long term in wider domains such as education, employment family and other relationships, (Knapp et al., 2009). However, early intervention has been shown to predict better longer-term prospects for children at risk.

Improving parenting practices through parent training programmes have been shown to provide better outcomes for children at risk of conduct problems (Barlow & Coren, 2018). Behaviourally based parenting interventions have shown to be both effective and cost-effective in the prevention and treatment of early onset child behaviour problems (Furlong, McGilloway, Bywater, Hutchings, Smith & Donnelly, 2012). Due to the increasing evidence of their wide-reaching success, there is growing interest in the potential use of behavioural interventions to support families of children with ASD (Stuttard et al., 2014; Dababnah, Olson & Nichols, 2019; NICE, 2013). Significant improvements have been observed in child behaviour (Whittingham, Sofronoff, Sheffield & Sanders, 2009), and parental wellbeing (Dababnah & Parish, 2016). There are an increasing number of programme effectiveness studies for this population (Postorino et al., 2017; Pellicano et al., 2013; Tarver et al., 2019; Dawson-Squibb et al., 2020), however economic evaluations are also becoming an increasing part of health and social care research (Petrou & Gray, 2011). Growing specification for their inclusion in research grants is being observed, as well as in assessing whether current spending on interventions is the best use of public funding (Scheibel, Zane & Zimmerman, 2022). Health economic evaluations are typically conducted from a public spending perspective to inform the allocation of resources (Husereau et al., 2013), and are increasingly being applied to preventative interventions such as parenting programmes. Ideally, evaluations should be performed in conjunction with trials of effectiveness (Hughes et al, 2016). Demand for rigour in their methods is high, and standardised processes are frequently suggested (Drummond, Sculpher, Claxton, Stoddart & Torrance, 2015). Although the identification of costs and measurement in monetary value are standard across economic evaluations, the nature of consequence or effects are valued and measured using different methods. Cost effectiveness analysis is the preferred method of economic evaluation in the UK (Charles, Edwards, Bywater, Hutchings, 2013), however a brief description of the five main methods of economic evaluations are presented below in Table 2.

Table 5.1 Methods of economic evaluation

Economic evaluation method	Description
Cost-benefit analysis	Outcome and costs are valued in monetary terms as opposed to outcomes or health gains, for both the intervention being assessed and the alternative treatment.
Cost-minimisation analysis	Compares two interventions assumed to have the same outcome effects, to assess which is the least costly.

Cost-utility analysis	Health benefits or outcomes are measures in terms of Quality of Adjusted Life Years (QALYs) of the intervention being evaluated, and the alternative.
Cost-effectiveness analysis	Results are expressed as a ration of costs per unit of health outcome, which are usually represented as 'natural units' appropriate to the study. For example, in an evaluation of a parenting intervention, a child behaviour measure or parental wellbeing assessment tool would be appropriate.
Cost consequence analysis	A wide range of costs and effects (consequences) of an intervention are assessed and compared and reported separately. The analysis can include all types of effects (health or non-health related), for example in relation to caregivers, consequences can be health related (wellbeing, mental health), or non-health related (financial consequences, effect on employment).

Despite the significant economic impact that child conduct problems can have, O'Neill and colleagues (2013) highlighted a dearth of economic studies in relation to parenting interventions and conducted a rigorous evaluation of the Incredible Years Basic® programme. The evaluation ran alongside a pragmatic RCT that was addressing conduct disorders and health inequalities (O'Neill, McGilloway, Donnelly, Bywater & Kelly, 2013). Parent level data gathered from RCT's is the most valuable information for the cost effectiveness studies of parent programmes (Hughes et al., 2016). The cost-effectiveness of an intervention is presented as an incremental cost effectiveness ratio (ICER) representing the difference in cost between one treatment and a comparable alternative, divided by difference in outcome. Intervention gains are not expressed in monetary value but rather as a ratio of cost per unit of health outcome, which are typically expressed in 'natural units' appropriate to the study in question (Hughes et al., 2016). In this case, a measure of child behaviour was employed as a child outcome. Furthermore, O'Neill and colleagues (2013) conducted a cost benefit analysis during the trial, which is also a key component of intervention evaluation as it allowed for comparisons between the Incredible Years programme and treatment as usual. The evaluation concluded that the IY® Basic programme was a cost-effective intervention for reducing child behaviour problems and had the potential to yield favourable long term economic gains (O'Neill et al., 2013). Edwards and colleagues (2016) also completed a cost effectiveness of the Incredible Years Basic programme alongside a pragmatic trial of its effectiveness as part of a funded study that would inform implementation of the programme in local authorities, should it show to be cost effective (Edwards et al., 2016). Robust economic evaluations can therefore have a strong influence on policy deciders and public spending, particularly when conducted alongside trials of intervention delivery within existing services. However, the real-world nature of complex interventions such as parenting programmes can present research challenges, therefore adopting standardised methods of data collection which can be administered as part of empirical trials is crucial for the validity of findings (Goossens, Rutten-van Mölken, Vlaeyen & van der Linden, 2000). Edwards and colleagues (2016) ensured data validity for their study by using a previously established and tailored version of the Client Service Receipt Inventory questionnaire (Beecham et al, 2016), that had been used in a similar study to collect service utilisation data. A standardised framework was also used for detailed cost gathering during the trial, which had previously been developed to conduct a micro-costing of programme delivery cost (Charles, Edwards, Bywater & Hutchings, 2013). The Strengths and Difficulties (SDQ) questionnaire (Goodman, 1997) was administered as an outcome measure of effectiveness. Statistically significant improvements were observed across all three outcome measures, and the programme was found to be both clinically and cost-effective (Edwards et al., 2016). These were both well-executed economic evaluations, which conformed to the specific standards of The Drummond 10 item Checklist (Drummond et al., 1997), a well-established tool used in the appraisal of health economic studies to assess evaluations against criteria which constitute rigorous studies and provide a valuable framework for consistency throughout evaluations. Charles, Bywater & Edwards (2011) recognised a lack of consistent methods for economic appraisal in a review of behavioural parenting programmes for children at risk of conduct disorders. The cost evaluations they identified were sparce, missing the structured methods which adhere to those included in the Drummond checklist (Drummond et al., 1997). All studies identified used different methods to evaluate different programmes, making any comparisons difficult (Charles et al, 2011). The review highlighted a need for robust methods for cost effectiveness and presented recommendations of what future economic evaluations should include when conducted alongside randomised controlled trials (Charles, Bywater & Edwards, 2011). The 'real world' nature of health economic research can present challenges, particularly when evaluating complex interventions such as parenting programmes (Richards & Hallberg, 2015). The cost and outcome benefits following implementation can be accrued by numerous parties (Edwards et al., 2007), therefore evaluations of such programmes need to be conducted from a multi-agency approach. Programmes also vary widely with regards to theoretical approaches, intervention aims (e.g., to address child behaviour, wellbeing, language or communication), and/or means of delivery (self-directed/ interactive, individually delivered, group based), and are consequently difficult to compare. This is a key consideration when designing economic evaluations that include a comparator. As this review is being conducted to inform further research of the IY-ASLD® programme, the search will focus on group-led programmes, as recommended by NICE recommendations (National Institute for Clinical Excellence, 2013), which have the same characteristics as the IY-ASLD® programme, such as target child age, delivery method and intervention dose, to ensure comparisons can be made. It is also a key consideration before any economic evaluation can be considered, particularly those of cost-effectiveness, that the intervention has first of all demonstrated effectiveness (Hughes, 2016; NICE, 2013). A programme that does not achieve the required outcomes, cannot be cost-effective. Although the evidence for the IY-ASLD® is limited due to its recent development, the evidence of effectiveness for other Incredible Years programmes has been demonstrated among many populations (Bywater et al., 2011; Dababnah & Parish., 2016a; Garnder & Leijten., 2017). There is also strong evidence for other well-established parent programmes based on behavioural principles (Sanders et al., 2012). Therefore, the author has focused this search on programmes with the same behavioural underpinning an approach, which are delivered in group format, and have a similar dosage, which will put the findings into a similar context to that of the IY-ASLD®.

Aim

The aim of this systematic literature review is to establish the level of existing economic evaluations of behavioural parent education training programmes specific to families of children with ASD to identify any gaps in the current literature. Any associated methodological issues arising from the literature will also be considered, to help inform the design and conduct of future trials and/or economic evaluations of the Incredible Years Autism Spectrum and Language Delays programme.

Method

Search strategy (See Appendix K)

A systematic search was conducted to identify literature relating to health economic evidence of parenting interventions for families with children on the autism spectrum based on the Cochrane systematic review guidelines (Structure of a Cochrane Review, 2018). Titles, abstracts and papers were searched for using the search terms: Autism, Autism Spectrum Disorder, ASD, parenting, parent interventions, parent training, parent programmes, cost, cost effectiveness analysis, cost benefit analysis, cost utility analysis, cost consequence analysis, cost minimisation analysis, unit cost, economics, health economics and economic evaluation.

The following electronic databases were searched: Applied Social Science Index and Abstracts (ASSIA), PsycINFO, CINAHL, Pubmed and Medline.

The following inclusion criteria were used in selecting studies for this review.

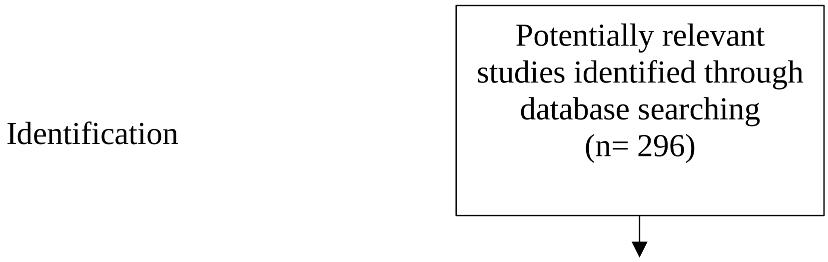
1. a general review article of the health economic evidence of behavioural parenting programmes for families of children with autism.
2. a partial or full economic evaluation of behavioural parenting programmes tailored towards families of children on the autism spectrum.
3. programme aimed at parents of children aged 3 to 8 yrs.
4. programme is of collaborative, group-led format.

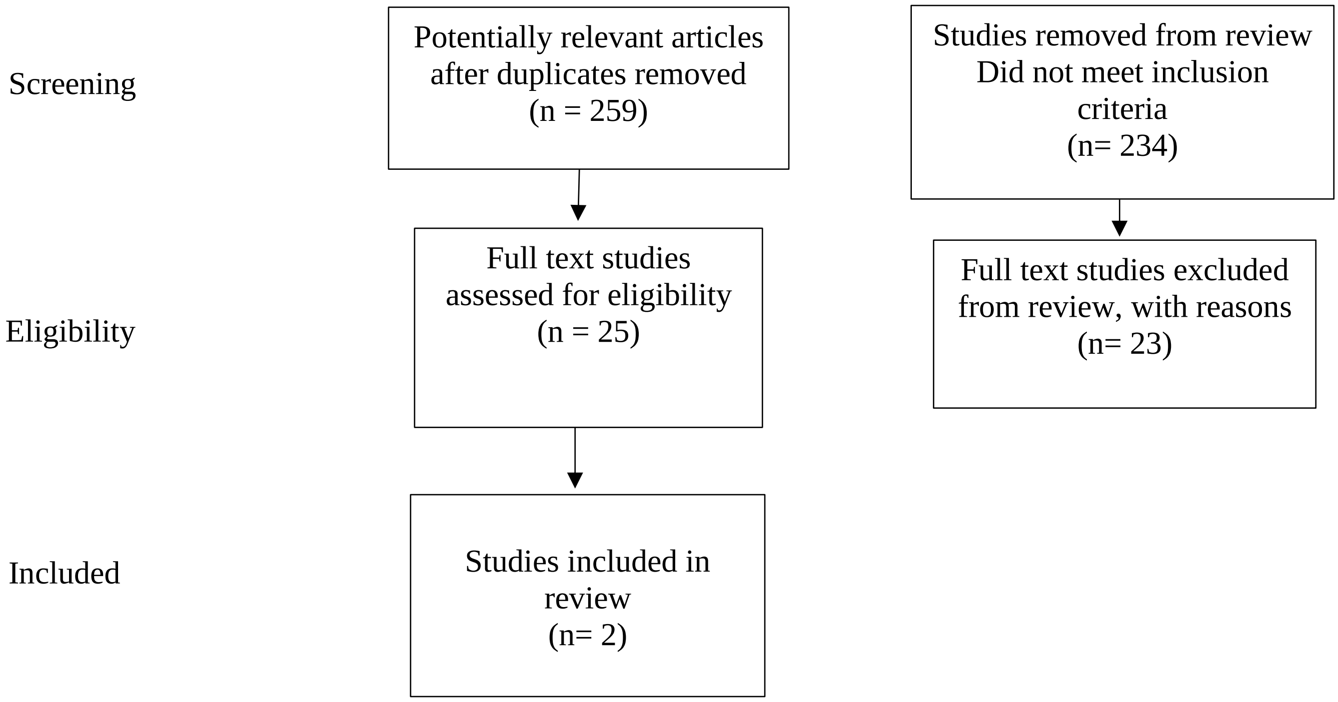
Exclusion criteria for the selection of studies were as follows.

1. published in a language other than English.
2. programme was not specifically related to ASD.
3. programme not founded upon behavioural principles.
4. was a clinical effectiveness paper that did not include economic evidence.
5. evaluation is of 1:1 or online programmes.

A time range of 10 years (since 2009) was specified for database searches. This constraint was decided upon due to the rapidly changing evidence and developments within the field of ASD, to ensure that literature was current. An additional filter was added to only include studies involving children aged from birth to 12yrs. The electronic search was initially conducted in November 2018 and updated in February 2020.

Figure 5:1. Flow chart outlining paper selection





Study selection

The initial search yielded 296 potentially relevant articles. Duplicated records were removed, which was conducted with the assistance of an academic support librarian, reducing the number to 259 potentially relevant articles for initial screening. The author used a systematic approach to screen remaining articles at title and abstract level in accordance with inclusion/exclusion criteria. Articles were categorised as follows; 1) programme not targeted for ASD, 2) clinical effectiveness, 3) programme format, 4) Child age (adolescent), 5) irrelevant intervention (e.g., sleep, CBT, mindfulness), 6) diagnostic article, 7) other. A further 234 articles were removed, leaving a total of 25 potentially relevant articles for full text review. The same categories were used to exclude/include studies and eligibility was discussed in supervision. Two articles met the criteria were selected for inclusion in the review.

Results

A total of 296 potentially relevant studies were identified through database searching. Following the removal of duplicates, and studies that did not meet inclusion criteria from the screening of abstracts, the full texts of twenty-five studies were assessed for eligibility. The most common reasons for rejection were that studies did not include an economic evaluation and were studies of programme effectiveness. Papers were also excluded if the interventions being evaluated were not group-led behavioural parent education training programmes (for example any individual behaviour change programmes, or programmes focussed on methods such as music therapy or mindfulness). Only two studies were identified to have any relevance and to meet the inclusion criteria in any way (Stuttard, Beresford, Clarke, Beecham, Todd & Bromley, 2014; Stuttard, Beresford, Clarke, Beecham & Morris, 2016). Both are presented in Table 5:1 to illustrate how they were assessed against the Drummond checklist (1997). The Drummond checklist is a well-established measure used in health economics to assess economic evaluations against criteria that constitute a robust study (Drummond et al., 1997).

Drummond criteria	**Stuttard et al., 2014**	**Stuttard et al., 2016**
Was a well-defined question posed in an answerable form?	×	×
Was a comprehensive description of the competing alternatives given?	√	√
Was the effectiveness of the programmes or services established?	√	√
Were all the important and relevant costs and consequences for each alternative identified?	×	×
Were costs and consequences measured accurately in appropriate physical units?	×	×
Were costs and consequences valued credibly?	×	×
Were costs and consequences adjusted for differential timing?	×	√
Was an incremental analysis of costs and consequences of alternatives performed?	×	×

Was allowance made for uncertainty in the establishments of costs and consequences?	×	×
Did the presentation and discussion of study results include all issues of concern to users?	×	×

Table 5.2 Paper appraisal using Drummond checklist (Drummond et al., 1997)

The two included studies had been conducted by the same research team two years apart, therefore similarities were expected in the design and methodological approach of the trials. Both were evaluations of the effectiveness of group-led parenting interventions specific to parents of children with ASD, with cost gathering conducted as part of both trials. Table 5.3 displays the key characteristics of the studies.

Study	**Riding the Rapids: Living with autism or disability—An evaluation of a parenting support intervention for parents of disabled children**	**An evaluation of the Cygnet parenting support programme for parents of children with autism spectrum conditions.**
Authors	Stuttard, L., Beresford, B., Clarke, S., Beecham, J., Todd, S., & Bromley, J. (2014).	Stuttard, L., Beresford, B., Clarke, S., Beecham, J., & Morris, A. (2016).
Age range of programme	3-11yrs	5-18yrs
Target population	Children with intellectual disabilities and/or ASD	Children with a diagnosis of ASD
Structure	Group-led, didactic information, role-play, group discussions. Functional analysis conducted to identify a targeted behaviour-change goal.	Group-led teaching element (power point presentations), video examples, small and whole group exercises and discussion.
Dose	2hr Weekly sessions delivered over 10 weeks (Total 20hrs) Optional booster/ follow up session after 3 weeks	3hr Weekly sessions delivered over 6 weeks (Total 18hrs) Informal follow up six weeks following programme
Study design	Pragmatic, non-randomised control trial	Pragmatic, two-centre, non-randomised control trial
Participants	Parents of children diagnosed with ASD and other disabilities. n = 48 (intervention) n = 28 (comparison)	Parents of children with a diagnosis of ASD n = 35 (intervention) n = 33 (control)
Measures	Child behaviour (ECBI) Parental sense of competence (PSOC)	Child behaviour (ECBI) Parental sense of competence (PSOC)
Length of follow up	Baseline and post intervention with both intervention and control.	Baseline, 6-week, and 3-month follow ups with both intervention and control.

	3-month and 6-month with intervention group only.	Baseline and post intervention with both intervention and control. 3-month and 6-month with intervention group only.6-month with intervention group only.
Outcomes	Parental sense of competence Child behaviour Achievement of behaviour change goal	Parental sense of competence Child behaviour Achievement of behaviour change goal
Comparator	Treatment as usual	Wait list control

Table 5.3. Illustrates trial characteristics, and intervention differences

Although the studies did not comprise of economic evaluations as specified in the search criteria or meet those included in the Drummond checklist to constitute a robust evaluation, both did include monetary costings of the intervention as part of the trial. It was therefore decided that both should be included for discussion in the review as there were learning opportunities from doing so.

The first study recognised is an evaluation of the Riding the Rapids: Living with Autism or Disability programme (previously described in further detail in Chapter 3). Stuttard and colleagues (2014) examined the effectiveness of the programme in improving child behaviour, and parental competence. The study adopts a pragmatic design, after recognising a lack of research that evaluates disability specific programmes within routine services. The programme is delivered by two facilitators, with the stipulation that the leader should be a clinical psychologist, due to the functional analysis of child behaviour that is part of the programme. Service providers were responsible for recording and sharing staff qualifications and pay grades, along with participant numbers, venue costs, and ‘other’ costs which included materials and resources, refreshments, and participant incentives. Although this information was provided to the research team, costs have not been reported separately in the article. The largest proportion of costs were accounted for by staff time, which collectively included, planning, travelling, delivering and debriefing. Recruitment procedures for the trial were not reported in the publication, although the programme developer recommends that parents can either self-refer or be referred to the programme by an education or teaching practitioner within routine services. It is therefore unclear whether staff time in this trial includes recruiting parents to the group. Having staff time categorised clearly would improve the way that data could be used for projected costs of programme delivery, for instance to assess the possibility of training staff of different grades to reduce costs in future groups. The study reports high rotational rates of delivery, with around eight groups delivered within every 15-month period in routine services. For the purpose of the trial, parents on the referral database were recruited to form a control condition for the trial, with the only comparator being 'treatment as usual'/no intervention. The control group was non-randomised and were instead allocated to groups due to geographical reasons, which is a limitation in an empirical evaluation. During the trial, the programme was delivered by the same facilitators that deliver routinely, increasing the likelihood that the programme was delivered with high fidelity, as the facilitators were knowledgeable and experienced. The mean cost of delivering the programme was calculated as being £3,225, although this was reported ambiguously, it is presumed as being per group for the 10-week duration. There was a substantial difference in the range between lowest costs of £2,582, and highest cost of £4,200 per block of delivery, which was dependant on the number of parents attending, pay grade of facilitator, and staff time including set up, travel. However, none of these details were recorded separately, therefore it is difficult to allocate spending to certain categories or to make any future roll-out calculations as they cannot be identified individually. Face to face contact was recorded as accounting for larger proportion of staff expenditure, but again this is difficult to compare without accurate recording of costs. Venue-hire or childcare costs were not included, as these were provided free of charge by the school associated with delivery. Furthermore, no cost comparison to an alternative intervention was included, nor any reference made to the running costs of similar interventions which is a limitation of the study. The author acknowledges it was not possible to conduct a cost effectiveness analysis with the available data, however outcome measures were administered as part of the trial and were found to be acceptable by parents, which is useful for future evaluations.

The same research team also conducted the second study included in the review. Stuttard and colleagues (2016) led an evaluation of the Cygnet programme (discussed in further detail in Chapter 3) which also provided delivery costs (Stuttard et al., 2016). Parents self-refer to the Cygnet programme through the charity Barnardo's, the programme developers, therefore recruitment information was not reported in the study. Up to six parents were allocated to each group delivery of the programme, with partners also invited to attend taking the maximum numbers of attendees up to 12, thus similar to the Riding the Rapids group numbers. There was a high rate of delivery, with seven or eight rounds of groups completed every 8-15 months. This could indicate that staff are routinely trained, however cost of training is not mentioned in the costings. The evaluation was conducted as part of routine delivery of the programme, with service delivery cost data gathered alongside the trial. Groups are led by two facilitators following accreditation. Although no professional qualification (e.g., clinical psychologist) is specified as is the case for the Riding the Rapids programme, the 'lead trainer' must possess a relevant university degree, at least three years of working with individuals with autism, and experience of facilitating group training sessions. Similar requirements in relation to experience are necessary for co-trainers. As well as assessing programme acceptability through qualitative feedback, and programme effectiveness in terms of child behaviour outcomes and parental competence, an additional aim of the evaluation was to provide preliminary costs of programme delivery. Costs were gathered by service co-ordinators, and again included facilitator information, participant numbers and resource costs associated with delivery such as programme materials and refreshments. Professional qualifications and grades of staff involved in each session was recorded, along with time of setting up, planning, debriefing and travel. Trainers themselves recorded parent attendance to calculate numbers and recorded their time and service in ‘unit costs’, which were sourced using the Curtis Unit Costs of Health and Social Care Compendium (2010), a well-established method of identifying costs for health economic evaluations. The programme is manualised, therefore is more likely to be delivered with fidelity, however there is no mention of supervision or additional training for facilitators. Improvements were observed in child behaviour and parental self-confidence. Although the desired sample size was achieved, missing data and drop-out rates meant that the final sample was under-powered for the child behaviour measure, therefore these outcome changes should be interpreted with caution due to the lack of comparison. In order to conduct a valid cost-effectiveness study, further evidence of programme effectiveness would need to be determined in a definitive trial. Mean costs of delivering the programme were calculated as being £2,390 per group, over the six weeks. However, the range between costs varied considerably from between £1,190 and £3,460 but with no clear breakdown of costs to assess differentials. Nevertheless, an improvement from the previous study of Riding the Rapids is that a breakdown of costs per session was calculated, therefore if the number of attendees were obtained, a cost per child could be determined which is useful information to provide to any stakeholders. The mean cost per session was £185 when led by social work staff from a third sector organisation. The evaluation did note some differences in staff costs per session when comparing differently skilled facilitators. Service cost differences were presented between those anchored within social services, and a group led by the Child and Adolescent Mental Health Service at a cost of between £360 and £490, which shows how costs can change across different contexts, and the importance of calculating differential costs. There was no reference to childcare, but course materials and room hire were stated to be accountable for 10% of costs. There was also no measure of service utilisation which would constitute as a full valuation economic evaluation. No cost comparison was made with regards to any other programme, including the previous costing of Riding the Rapids conducted by the team, which could be considered a shortfall.

Discussion

This review was conducted to establish the level of economic evaluations of parenting programmes targeted towards parents of children with ASD and has highlighted a dearth of evidence within the literature. The systematic search was comprehensive with regards to the evaluation methods applied, with the four most common techniques included in the search (cost-benefit, cost-minimisation, cost-utility, cost-effectiveness, cost-consequence and cost-minimisation analyses). However only two studies were identified as having included any element of costs within the study. There is no set minimum number of studies to include in a systematic review, and appraising and synthesising two to three studies can constitute a sound review (Peters et al., 2015). Ideally, a greater number would result in a richer narrative, however the identification of only two studies in the searches illustrates the lack of available evidence, thus answers an important research question. The author does acknowledge that the search focused only on including group-based programmes which were founded on behavioural theories and targeted toward a particular age group of children, therefore was restrictive. The most successful economic evaluations run alongside RCT's and include both a control arm and a comparator (Hughes et al., 2016), for which purpose the interventions should be similar. This provides a rationale for the decision to only include like for like interventions in this review, as the aim was to assess the level of existing evidence for comparable programmes.

Both included trials adopted a pragmatic approach, with programmes being evaluated as part of routine delivery, however, there were limitations in design with regards to randomisation. Parents were allocated to groups geographically; therefore, the study cannot be considered an RCT, which is the ideal for the evaluation of complex interventions such as parenting programmes (Hughes et al., 2016). With regards to the Drummond checklist criteria, although both studies were clearly described as being evaluations of programme effectiveness, neither included a clearly defined question in relation to economic outcomes, which is an important stipulation. According to Drummond and colleagues (1997), robust economic evaluations should comprise of a competing alternative intervention, even in RCT's; however, the only comparator in both studies was 'treatment as usual' which limits the possibility of calculating any true cost difference or thorough differential analysis. Both studies evaluated programme effectiveness, however positive outcomes should be interpreted with caution due to the small sample sizes, therefore further evaluations should be conducted with larger samples to determine definitive evidence before further economic analysis. Relative costs and consequences were not identified accurately, or at least not reported as such making future replication and/or performing of sensitivity analyses difficult.

The studies show that it is possible to gather cost data alongside an evaluation of programme effectiveness, and doing so within existing services is a strength. However, the appraisal of both has emphasised the importance of adopting robust methods to ensure rigour. Although is it useful to have programme costs, aggregated figures are not helpful beyond the trial. If, however, detailed micro costs had been recorded, these could be generalised for other purposes such as informing further research, and to provide important expenditure information for services and policy deciders.

The largest proportion of costs in both trials was staff time, although information about the qualifications and pay grades were not reported. Both studies report how staff time was split into travel, preparation and programme delivery, however none of these are shared, therefore further assessment of staff costs resulting from this trial would be difficult. There is also no mention of staff training, ongoing support or supervision. Such 'set-up' costs are important not only for economic evaluations, but also for service providers to access for projected costs. Staff training and the purchasing of resources are one off expenditures, which would then reduce during future roll-out. The facilitators delivering the programme in both trials were experienced, however should service providers decide on training less qualified staff to deliver the programme, accurate costs would be needed to conduct a sensitivity analysis of projected costs. Supervision would be necessary to support them to ensure the programme is still delivered with fidelity. Programme fidelity, and implementation are important factors to consider when designing a larger trial with health economic questions to be addressed, therefore more accurate details about the costs of staff training and ongoing supervision may need to be a consideration to ensure the programme is being delivered as intended before any cost effectiveness studies are conducted. Although venue hire or childcare costs were not included in the costings for both trials, these are also important considerations. Even though free creche facilities were provided as part of both trials, this is not always the case. Childcare can often be a barrier to parents being able to access parenting support, therefore it is a key consideration when planning either trails or routine delivery of parenting programmes. Costs were gathered by either facilitators or service co-ordinators, with no involvement from researchers mentioned. In order to ensure accuracy, cost gathering should be a collaborative task as the personnel involved in service provision have knowledge about finance with regards to staffing and training, the group leaders know more about ‘hand’s on’ spending, such as refreshments and participant incentives, and researchers are able to assess that information is being gathered in a way that will return functional data which can be applied to different contexts. Therefore, to ensure good practice, all personnel should be involved in the design of robust data gathering methods.

Although both studies (Stuttard et al., 2014; Stuttard et al., 2016) included a control group, they were not RCT studies. Nevertheless, both trials were pragmatic in design, which is a strength in any economic evaluation as it examines how an intervention works in real life conditions and is the most robust measure of effectiveness (Hughes et al., 2016). Both studies were missing vital elements of the Drummond et al (1997) checklist which was used for quality appraisal of identified studies, therefore neither can be considered a valid economic evaluation, and should rather be regarded as costings of programme delivery. It was agreed however that both studies would be included in the review, due to the learning attained from the studies, which are relevant to any future costings or economic evaluations of the IY-ASLD® programme. Both studies show that it is possible to gather cost data alongside a trial evaluation of effectiveness, however only overall costs were recorded rather than accurate, broken-down costs which is a limitation (Stuttard et al., 2014; Stuttard et al., 2016). This also highlights that although reporting delivery costs during trials of programme delivery is informative; in order to be useful for any future research or planning for prospective intervention expenditure on programme roll-out, a more structured, accurate breakdown of micro costs is needed. This systematic review has not only highlighted the lack of economic research conducted in this field, but the appraisal of the two studies identified have highlighted the absence of standardised cost gathering methods. Neither followed a detailed framework such as the standardised method of micro-costing developed by Charles et al (2013). Such approaches are recommended to bring transparency to the evaluation methods not only for validity, but also for the ease and accuracy of additional and future calculations allowing for robust comparison between different programs and/or interventions. A standardised cost gathering measure could also be considered useful for future studies as a potential means of comparing programmes that differ in structure in the inclusion criteria. A future search strategy could include more varied formats of programmes and instead have the use of standardised data collection methods and frameworks as an inclusion criterion to widen the search and be more inclusive. On reflection, the search could have been less restrictive by adopting a scoping review method to seek an overview of a potentially larger and more diverse body of evidence. This wider search may have been more suitable and could have included other programme formats such as individualised interventions, ABA and more intense interventions. Nevertheless, by conducting the search in this way, we have identified the very limited economic evidence, and that the very few attempts at increasing the evidence fall short with regards to the consistency in methods.

Lessons learned

- This review has highlighted a lack of economic evaluations across the growing literature with regards to parenting programmes targeted towards families of children with autism.
- Although the two studies included in the review consisted of costings of the programmes, there is an identified need for a standardised framework for the costing and economic evaluations of parenting interventions in order for them to be more accurate towards informing grant funding applications, further research trials and future programme roll-out.
- Detailed costings of programme delivery alongside evaluations of effectiveness are a useful step towards further economic evaluations such as future cost effectiveness analyses.

Summary

Parenting programmes are effective in reducing child behaviour problems and dysfunctional parenting practices. Improved wellbeing outcomes for children at risk of conduct disorders and their families have also been observed (Gardner et al., 2006; Hutchings et al., 2007; Mingebach et al., 2018). Group parent programmes have been shown to be a more cost-effective way of addressing child behaviour problems (Edwards, Jones, Berry, Charles, Linck, Bywater & Hutchings, 2016). There is increasing evidence showing their potential benefits in improving outcomes for children with ASD and their families, a population with high service utilisation (Romeo, Knapp & Scott, 2006). However, a lack of economic evaluations running alongside trials of effectiveness has been identified in this review. Given the increased interest in improving outcomes for children with autism and their families on governmental agenda, it is important to provide economic evidence alongside that of effectiveness. It is important that policy makers are equipped to make informed decisions on which interventions to implement within services to allow them to assess at what cost.

Chapter 6

Micro-costing of programme delivery

Introduction

Autism spectrum disorder (ASD) is a life-long neurodevelopmental disability characterised by impaired social interaction, language delay and repetitive behaviour (National Autistic Society, 2018). It is well documented that communication problems often diagnosed alongside ASD are strong predictors of externalising maladaptive behaviour such as aggression, defiance and inattention (Hartley, Sikora & McCoy, 2008). Children on the autism spectrum are at increased risk of developing mental health disorders (Simonoff et al., 2008), and are four times more likely to demonstrate behaviour problems than typically developing peers (Stuttard et al., 2014). Parents of children with autism face many challenges due to the complexity of their child’s needs; however, conduct difficulties and child emotional problems are consistently reported as being their greatest cause of distress (Totsika et al., 2011). Hard to manage behaviours can impact negatively, not only on parental mental health but also on that of partners and siblings (Ludlow et al., 2012). Individual and familial quality of life can be influenced with additional strain placed on relationships, family leisure time and employment (McStay et al., 2014). Furthermore, financial burden is often placed on these families, not only from lost productivity but also due to out of pocket payments for additional services (Knapp et al., 2009). Essential access to specialist health or intervention services and educational support also give rise to high direct public costs (Lavelle, Weinstein, Newhouse, Munir, Kuhlthau & Prosser, 2014). According to Flanders and colleagues (2007), autism places a greater financial burden on public services than any other childhood illness or intellectual disability (Flanders, Engelhart, Pandina & McCracken, 2007). Lifetime societal cost of supporting an individual with autism is estimated as being £0.98 million, increasing to £1.6 million when co-occurring with another intellectual disability (Buescher et al., 2014 (costs inflated to 2017; Curtis 2017)). Ganz (2007) reported that across a life-time trajectory, a large proportion of this is accounted for during the first eight years of life on behavioural therapies, child/respite care and special education.

Although autism is incurable, support and effective interventions have been shown to improve outcomes for children with autism and their families (Russa, Matthews & Owen-DeSchryver, 2015). Behavioural and socialisation therapies are effective in improving developmental and adaptive aspects in some children (Makrygianni & Reed, 2010); however, the National Institute for Health and Care Excellence (NICE, 2013) recommends that research should focus on group-based parent-training interventions that help parents learn skills to deal with behavioural challenges before they become problematic (National Institute for Health and Clinical Excellence (NICE), 2006; Todd et al., 2010).

Parenting Programmes

Behavioural parent training programmes such as the Incredible Years® (IY) series have been widely researched over many decades and demonstrate effectiveness for both treatment and prevention of child conduct problems and associated social and emotional problems (National Institute for Health and Clinical Excellence (NICE), 2006). Strong evidence also shows improvements in parental mental health and stress levels (Bywater et al., 2009; Dababnah & Parish, 2016; Hutchings et al., 2017).

The IY parenting programmes are generally delivered by two group leaders in 2-hour collaborative weekly sessions, lasting between 8-12 weeks (Webster-Stratton and Reid, 2018). Components include group led discussion, role-play, and video modelling of strategies to enhance skills to manage problem behaviour and teach alternatives. Despite extensive understanding of programme benefits, Todd et al., (2010) highlighted that previous IY programmes were not tailored to address the specific parenting challenges for children with Learning Disabilities (LD) or ASD. This was reflected in other evaluations of the IY Basic programme when delivered to families of children with ASD. Parents reported that although the programme improved many family issues not directly related to the child, such as marriage and personal feelings; they found the video vignettes difficult to relate to as they did not feature children with ASD (Dababnah & Parish, 2016). When exploring necessary adaptions to the IY programmes for suitability for families of children with autism, McIntyre (2008) suggested including more information about ASD in the programme content, particularly concerning communication difficulties and challenging behaviour. Subsequently, the IY Autism and Language Delays Programme was published as a new addition to the IY suite with increased focus on coaching language and developmental outcomes including communication, social skills and behaviour problems. Adaptions involved the inclusion of video vignettes of children on the autism spectrum (Webster-Stratton, 2015).

Economic Evaluations of parenting programmes

In the UK, parenting programmes are typically delivered through health and social care services, provided through a predominantly tax funded system (Charles, Edwards, Bywater & Hutchings, 2013). Budgets are usually set by the Government, with commissioners allocating funds to local services. Accurate and detailed cost data for new programmes are therefore needed to provide policy makers with appropriate information to decide whether they justify the financial investment. Although there is no economic evidence for the newly developed IY Autism programme as yet, evaluations of more established IY programmes have been undertaken.

Edwards, Céilleachair, Bywater, Hughes & Hutchings (2007) performed a cost effectiveness analysis of the IY Basic programme alongside a pragmatic Randomised Control Trial (RCT) (Hutchings et al., 2007) with positive results. The intervention was delivered by health visitors across 11 Sure Start areas, to parents of children at risk of developing conduct disorder. The total cost of establishing and running the programme over 12 weeks was calculated as being £20,801.29, equating to £1733.43 per child with 12 parents attending (inflated to 2016/2017 costs (Curtis, 2017)). Costs were divided into recurrent and non-recurrent costs, so that future programme roll-out costs could be calculated. Once costs such as initial training and the purchase of programme materials were removed, roll-out costs were estimated to decrease to £1430.33 per child with 12 parents attending the group (inflated to 2016/2017 costs (Curtis,2017)), further increasing its value for money (Edwards et al. 2007).

More recently, an economic evaluation of the IY Basic programme delivered as part of the Brighter Futures initiative in Birmingham, UK realised a greater incremental cost-effectiveness ratio (ICER). Although still cost-effective, mean costs per child were higher, at £1768 (Edwards et al., 2016 (inflated to 2016/2017 costs (Curtis, 2017)) per child based on 12 parents attending. Alongside possible inflation costs, the increase was thought to be essentially due to the high costs of training/supervising less experienced staff as group leaders. This prompted the reconfiguration of services and roll out of the programme by Birmingham City Council; significantly reducing delivery costs and demonstrating the importance of economic evidence for interventions (Edwards et al., 2016).

Due to the potential impact on policy making, it is therefore imperative that economic evaluations are thorough. Accurate costs of an intervention at micro level are essential before any further economic analysis, such as cost effectiveness or cost benefit analysis can be performed. Micro-costing is a cost estimation method which involves direct enumeration of every cost associated with an intervention (Phillips, 2008), and is a valuable economic tool if conducted accurately (Drummond et al., 2015). In an effort to ensure transparency and accuracy from health economic researchers, a standardised framework was formulated by Charles et al., (2013) providing a step-by-step description of the Micro-costing process. The IY Toddler programme was used as a working example, and costs were calculated within three “real world” contexts, which included the initial set up of the programme within Flying Start areas in Wales, UK. Subsequent delivery costs were also calculated along with set up and delivery costs within a research trial. The overall cost of establishing the programme within a research trial was £10916.59. Roll-out costs were calculated as being less than in previous economic evaluations, at £717.37 (inflated to 2016/2017 costs (Curtis, 2017) per child with 10 parents attending the group, once initial training and supervision had been undertaken. Groups were facilitated by one health visitor and one childcare practitioner, which may explain the difference in cost to other evaluations (Edwards et al., 2007; Edwards et al., 2016). Detailed methods of how costs were gathered, labelled and divided into categories provided a useful template for future micro-costings. This model ensures that all expenditures are accounted for, such as initial training and group set up cost, which were omitted from previous examples of micro costings of similar interventions (O'Neill et al., 2013). Parenting programmes are complex interventions from which the cost and benefits can be accrued by multiple agencies (Edwards et al., 2007); therefore, a standardised framework from which to assess this accurately is a valuable tool. Autism also has a widespread impact across many different services and agencies, and in the current economic climate, there is a growing need for detailed economic evidence of interventions that may improve outcomes for individuals and their families.

This is the first micro-costing of the newly developed IY Autism Spectrum and Language Delays programme. The main aim of the study is to calculate full and accurate costs of delivering the programme during the feasibility trial, in order to inform a full economic costing (fEC) for a larger definitive trial. The micro-costing will also afford vital cost data for future economic evaluations of the programme, such as a cost effectiveness analysis, and provide policy makers with essential data to inform wider implementation and roll-out.

Method

This micro-costing was conducted from a public sector, multi-agency perspective alongside a pragmatic pilot randomised controlled trial (RCT) evaluating the newly developed IY programme for parents of young children with autism (Williams, Hastings, Charles, Evans & Hutchings, 2017). The main trial was funded by autism research charity Autistica, with further analysis of the programme including detailed micro-costing by a Knowledge Economy Skills Scholarship (KESS), supported by the European Social Fund (ESF) and the Children's Early Interventions Trust (CEIT). The sample consisted of 58 families who were randomly allocated to either the intervention condition (IY Autism programme) or to a wait-list control condition. The children were aged between 3 and 8 years, with a diagnosis, or strongly suspected diagnosis, of autism. Parents were recruited through specialist children's services in four centres across Northwest and mid Wales.

IY Autism and Language Delay programme Format

A maximum of 10 parents were allocated to each group of which an average of 5 parents attended weekly 2-2.5hr parent programme sessions for 12 weeks. Both parents were given the opportunity to attend the session, but generally only the primary caregiver attended. Participating families completed assessments, which included measures of parental well-being, child behaviour and development, and observations of parent-child interactions at baseline and follow-up. Two trained leaders introduced the structured sequence of topics within the groups, involving discussion, video examples of the strategies being presented, role-play and home activities.

Group Leaders

The group leaders were a consultant psychologist and child psychologists, a social services well-being officer and nurse, who were all highly experienced in providing direct clinical input to support parents of children with significant communication problems and resulting behaviour challenges. Six leaders had previously completed the IY pre-school basic training, a prerequisite of the Incredible Years Autism Programme at the time of the trial. Both leaders who had not completed the basic training did so before attending the Incredible Years ASD training delivered by Early Intervention Wales Training as part of the funded trial.

Micro-Costing

Parenting programmes such as the IY series are complex interventions (Craig et al., 2008); therefore, the cost and outcome benefits can be accrued by numerous parties; hence the multi-agency approach of this analysis.

The costs of the IY Autism parenting programme were calculated in four contexts.

1) Initial set up and delivery of the programme over 12 weeks within a research trial (which include initial training, supervision etc.)

2) Research costs (as above) with additional home visits (circumstances such as rurality and the complex health needs of one child meant that in some instances, home visits were deemed necessary).

3) Roll-out costs- the subsequent delivery of the IY Autism programme as part of normal service delivery (with initial training and supervision already undertaken, materials purchased and lower paid group leaders facilitating the sessions).

4) Roll-out costs (as above) taking into account proposed changes to programme protocol (amendments to training accreditation and programme running time extended)

The micro costing framework used was based on standard methods of cost gathering and previous examples of micro-costing of IY programmes (Charles, Edwards, Bywater & Hutchings, 2013; Drummond et al., 2015; Edwards et al. 2007). As much data as possible were obtained on each aspect of delivery costs. All elements of cost were labelled individually and given their own unit cost. Costs were summed to give subtotals for each category (set-up costs, delivery costs). Subtotals were then summed to give an overall cost of the programme. Costs were divided into the following components based on the real world process necessary to set up and deliver the programme: 1) Set-up costs (e.g., initial training costs, supervision, and set-up before the start of the programme) and 2) Delivery costs (e.g., the unit delivery costs of running the group such as time recruiting, home visits, telephone calls, room preparation, session delivery time, rental of halls, refreshments and crèche facilities).

Cost gathering

Due to the grant awarded to fund the trial, steering group meetings were held quarterly throughout the year of planning and running the trial. Attendees consisted of the research team, staff from Early Intervention Wales Training and group leaders from each centre; all of whom contributed to decisions about cost gathering methods and discussed any issues with regards to programme delivery.

The mean unit delivery costs of running the group such as recruitment, home visits, telephone calls room preparation, session delivery time, refreshments, rental of halls and crèche facilities were extracted from cost diaries *(See Appendix I).* The diaries were completed weekly by both consenting leaders of the four intervention groups. Minor modifications were made to cost diaries previously developed with group leader focus groups by Edwards et al. (2007). The categories were listed in a spreadsheet with a column for each week of programme delivery. Group leaders were asked at the start of the RCT to consent to completing a cost diary and were given paper copies to complete at the end of each session. Group leaders were requested to give as much detail as possible about the length of time spent on each task (eg.travel to group session, room preparation and time spent running the group). Activities such as the purchase of raffle prizes, photocopying and writing materials were listed as examples under the heading of ‘other costs’ in the diary to guide and inform leaders. There was a high level of compliance with regards to diary entries, with minimal missing data retrieved via email to group leaders following programme completion. Diaries from all four centres were returned to the administrator at the Centre for Evidence Based Early Intervention (CEBEI), Bangor University at the end of programme delivery (12 weeks). The cost data were then entered into a Microsoft Excel spreadsheet by the first author. With the exception of one centre, group leaders attended the same fortnightly supervision, and therefore reported the same time taken to attend. All but one of the centres had access to a free venue, therefore this venue rental cost was reported separately, and the sum referenced for future roll-out calculations. The same centre also differed with regards to making home visits to parents who could not attend the group due to rurality and the complex health needs of their child. The costs of the additional home visits have been added separately in Table 2. There was also the exception of a one-off taxi fare for one parent to attend a session, which was listed under ‘other costs’. The purchase of sundries such as raffle prizes given as motivators to parents were also recorded as ‘other costs’. With regards to costs such as room and session preparation time, outside contact with families, refreshments and administration costs, the group leaders reported a range of time taken and costs; therefore, a mean was calculated to provide average costs across the groups and presented in the tables. Costs not retrieved from diaries were gathered from additional sources, for example, salaries of group leaders, group materials and costs of training. The first author requested the NHS salary bands of all group leaders, along with those of future group leaders so that comparisons could be made between research and roll-out costs. The hourly wage presented in the tables shows the mean wage for group leaders, upon which the calculation of costs for all staff-related tasks in programme delivery is based. The purchase costs of programme materials, initial training and trainers’ wages to deliver supervision were all obtained from the local IY training agency. All intervention costs were averaged and divided by the number of participants attending to calculate average cost per family. The number of parents allocated to each group in this trial ranged from 7 to 9, with an average of 5 parents attending each session. Costs are therefore shown for 5 attendees per session.

Supervision was held fortnightly for two hours throughout programme delivery by the third author, an accredited IY mentor at the time. Although costly, supervision was considered essential to ensure implementation fidelity of the newly developed programme and to provide delivery support as the programme was being delivered by leaders new to the programme.

Crèche facilities were also considered an essential cost to include as childcare problems can prevent some parents from attending (Hutchings, Gardner & Lane., 2004). Childcare facilities were offered during the research trial but were not required by any of the group members. However, for accurate prediction of roll-out costs, these costs (taken from previous evaluations and inflated to cost year 2016/17) have been included in the predicted calculation of future costs.

Two group leaders did not attend supervision, as the leader from that centre was a highly experienced and accredited programme trainer; therefore, supervised her co-leader throughout programme delivery. Consequently, the mean costs of travelling to and from supervision were calculated based on the time reported by the remaining six leaders.

Sensitivity analysis

Sensitivity analysis was applied to establish costs based on 10 parents in each group in addition to the average 5 attending each group in the trial. The IY series developer recommends that the maximum number of parents attending each group does not exceed ten (Webster-Stratton, 2015). The rationale for this being that more concentrated support is needed by parents of children with autism in role play practices and home activity feedback, due to the complexity of the challenges that they may be facing. Calculations were therefore based on these criteria.

Future costs were also calculated to account for proposed changes to the programme protocol, which means the programme will run for 14 weeks in future. This was following feedback that group leaders were finding it difficult to work through the programme content, and parent feedback indicating that more time spent on group discussion would be beneficial. The IY developer has also made changes to the group leader training procedure. It is no longer a requirement that leaders complete the IY basic parent group leader training beforehand, resulting in a stand-alone three-day IY Autism training at a cost of £500, eliminating the cost of the previously required three-day IY basic training.

Results

Table 6:1 Total costs and costs per child of set up and delivery of the Incredible Years ASD Parenting Programme over 12 weekly sessions within a research trial with two clinical psychologists as group leaders

Type of cost	Units	Unit cost (£)	Total cost (£) based on 5 per group	Total cost (£) based on 10 per group
Set up costs:				
Initial training costs:				
Programme materials	1 pack of IY ASD materials	£922.80 (including Value Added Tax)	£922.80	£922.80
IY Toddler book for each parent	1 book	£16	£80	£160
Training course fee	2-day training	£480.00 per leader (including Value Added Tax)	£960 for 2 leaders to attend training	£960 for 2 leaders to attend training
Wages for 2 group leaders to attend training	2-day training (7hrs each)	£316.96 per leader	£633.92	£633.92
Travel to/from training	Varied depending upon group leader location 44 miles mean round trip	£17.60 per leader/per day (40p a mile)	£70.40	£70.40
Fee for fortnightly supervision throughout programme	½ day (6 x sessions)	£225.00 (flat rate for trainer wages to deliver)	£1350	£1350
Wages for group leaders to attend supervision	½ day (6 x sessions)	£90.56 per leader	£1086.72 for 2 leaders attending	£1086.72 for 2 leaders attending
Travel to/from supervision	Varied depending upon group leader location 44 miles mean round trip	£17.60 per leader/per day (40p per mile)	£211.20	£211.20
Subtotal:			**£5315.04**	**£5395.04**
Delivery costs:				
Time recruiting families	13 hrs 18 minutes per centre	£301.11	£301.11 in direct wages	£602.22 in direct wages

(Phone calls and letters)				
Room prep time for 2 leaders	32 minutes	£23.99 for 2 group leaders	£287.88 in direct wages to prep room for 12 weeks	£287.88 in direct wages to prep room for 12 weeks
Session prep time for 2 leaders	1 hr 56 minutes	£87.39 for 2 group leaders	£1048.68 in direct wages to prepare group session for 12 weeks	£1048.68 in direct wages to prepare group session for 12 weeks
Group time	120 min per week for 2 leaders	£90.56	£1086.72	£1086.72
Travel time to group (round trip)	36 mins per leader	£27.16	£325.92	£325.92
Contact time with families outside session	12 hrs 18 minutes per centre	£278.47	£278.47	£556.94
Food/ catering costs	£1.72 a week	£1.72 per centre	£20.64	£41.28
Venue hire *	£15.00 a week	£15.00	£180.00	£180.00
Other costs		£13.89	£13.89	£13.89
Subtotal:			**£3543.31**	**£4143.53**
Overall costs:				
Costs of establishing and running parenting group over a 12-week program:				
Total:			**£8858.35**	**£9538.57**
Cost/child based on 5 parents per group			**£1771.67**	
Cost/child based on 10 parents per group				**£953.86**

▲Venue hire – all but one centre had access to a venue free of charge, therefore not included in research costs. Venue hire has been included in future roll-out costs (Table 6.3)

•Average hourly rate of group leaders in research trial- £22.64/hr

***** None of the parents included in the research trial required the offer of childcare, therefore these have not been added. The estimated cost of childcare has been included in future roll-out costs (Table 3)

Table 6:2 Total costs and costs per child of set up and delivery of the Incredible Years ASD Parenting Programme over 12 weekly sessions delivered within a research trial with two clinical psychologists as group leaders, with home visits conducted when necessary (see discussion)

Type of cost	Units	Unit cost (£)	Total cost (£) based on 5 per group	Total cost (£) based on 10 per group
Program set up and delivery costs (from Table 1) Subtotal;			£8858.35	£9538.57
Additional home visits;				
No. of home visits	15 over 12 weeks			
Travel to home visits (average round trip)	19.5 miles/.40p per mile	£7.80	£117.00	£234.00
Time at home visit	2hrs for 1 group leader	£45.28	£543.36	£1086.72
Total:			**£9518.71**	**£10,859.29**
Cost/child based on 5 parents per group with home visits when necessary			**£1903.74**	
Cost/child based on 10 parents per group with home visits when necessary				**£1085.93**

Table 6:3 Total costs and costs per child of delivery of the Incredible Years ASD Parenting Programme over 12 weekly sessions delivered by one clinical psychologist and one nurse as programme roll-out. Initial training has been undertaken and programme materials purchased (see Table 1)

Type of cost	Units	Unit cost (£)	Total cost (£) based on 5 per group	Total cost (£) based on 10 per group
Delivery costs;				

IY Toddler book for each parent	1 book	£16	£80	£160
Wages for group leaders to attend supervision	½ day (1 x session per term)	£69.88 per leader	£139.76 for 2 leaders attending	£139.76 for 2 leaders attending
Engagement and recruitment of families	13 hrs 18 minutes per centre	£232.35 per centre	£232.35 in direct wages	£464.70 in direct wages
Room prep time for 2 leaders	32 minutes	£18.51 for 2 leaders	£222.21 in direct wages to prep room for 12 weeks	£222.21 in direct wages to prep room for 12 weeks
Session prep time for 2 leaders	1 hr 56 minutes	£67.43 for 2 leaders	£809.16 in direct wages to prepare group session for 12 weeks	£809.16 in direct wages to prepare group session for 12 weeks
Group time	120 min per week for 2 leaders	£69.88 for 2 leaders	£838.56 in direct wages to deliver group	£838.56 in direct wages to deliver group
Contact time with families outside session	12 hrs 18 minutes per centre	£214.88	£214.88	£429.76
Food/ catering costs	£1.72 a week	£1.72	£20.64	£41.28
Venue hire	£15.00	£15.00	£180.00	£180.00
Provision of crèche facilities (salary of crèche staff)	£119.73 per week	£119.73 per week	£1436.76 per programme	£1436.76 per programme
Other costs (raffle prizes etc.)		£13.89	£13.89	£13.89
Potential admin (e.g., photocopying)		£6.99 per week	£83.88	£83.88
Overall costs of running parenting group over a 12-week program: Total:			**£4272.09**	**£4819.96**
Cost/child based on 5 parents per group			**£854.42**	
Cost/child based on 10 parents per group				**£481.99**

*Calculations based on cost data from trial
▲ Average hourly rate of group leaders for future roll-out calculated as £17.47/hr
●Although programme materials have previously been purchased, additional costs may be incurred for photocopying resources (e.g., home activity sheets for parents) inflated costs from previous economic evaluations used for cost prediction

Table 6:4 Total costs and costs per child of delivery of the Incredible Years ASD Parenting Programme delivered by one clinical psychologist and one nurse as programme roll-out. Initial training costs for new leaders included. Programme materials have been purchased (see Table 1) Following proposed changes to programme protocol (programme running time extended to 14 weeks)

Type of cost	Units	Unit cost (£)	Total cost (£) based on 5 per group	Total cost (£) based on 10 per group
Training course fee	Stand-alone 3-day course	£500	£500	£500
Wages for 2 group leaders to attend training	3-day training (7hrs each)	£733.74	£733.74	£733.74
Delivery costs;				
IY Toddler book for each parent	1 book	£16	£80	£160
Wages for group leaders to attend supervision	½ day (1 x session per term)	£69.88 per leader	£139.76 for 2 leaders attending	£139.76 for 2 leaders attending
Engagement and recruitment of families	13 hrs 18 minutes per centre	£232.35 per centre	£232.35 in direct wages	£464.70 in direct wages
Room prep time for 2 leaders	32 minutes	£18.51 for 2 leaders	£259.14 in direct wages to prep room for 14 weeks	£259.14 in direct wages to prep room for 14 weeks
Session prep time for 2 leaders	1 hr 56 minutes	£67.43 for 2 leaders	£944.02 in direct wages to prepare group session for 14 weeks	£944.02 in direct wages to prepare group session for 14 weeks
Group time	120 min per week for 2 leaders	£69.88 for 2 leaders	£978.32 in direct wages to deliver group	£978.32 in direct wages to deliver group
Contact time with families outside session	12 hrs 18 minutes per centre	£214.88	£214.88	£429.76

Food/ catering costs	£1.72 a week	£1.72	£24.08	£48.16
Venue hire	£15.00	£15.00	£210.00	£210.00
Provision of crèche facilities (salary of crèche staff)	£105.75 per week	£105.75 per week	£1480.50 per programme	£1480.50 per programme
Other costs (raffle prizes etc.)		£13.89	£13.89	£13.89
Potential admin (e.g., photocopying)		£6.18 per week	£86.52	£86.52
Overall costs of running parenting group over a 12-week program: Total:			**£5897.20**	**£6448.51**
Cost/child based on 5 parents per group			**£1179.44**	
Cost/child based on 10 parents per group				**£644.85**

Discussion

This micro-costing provides costs for the newly developed IY Autism programme delivered within a pragmatic research trial and as part of normal service delivery, highlighting differential costs depending on delivery, context and setting. Using a previously standardized framework (Charles et al., 2013), delivery costs were calculated in the following context; 1) Research costs (Which include initial training, supervision etc.); 2) Research costs with additional home visits (circumstances such as rurality and the complex needs of one child meant that in some instances, home visits were deemed necessary); 3) Roll-out costs - the subsequent delivery of the IY Autism programme as part of normal service delivery (with initial training and supervision already undertaken, materials purchased and lower paid group leaders); 4) Roll-out costs with proposed changes to programme protocol (changes to training protocol and programme running time extended).

Table 1 shows the reported costs of setting up and delivering the programme within a research trial based on weekly cost diaries completed by the four group leaders. Overall costs of establishing the programme with five parents per group was £8858.35, equivalent to £1771.67 per child. Calculations were made based on five parents, as this was the average number recruited or attending per week in each group. A sensitivity analysis was conducted to also determine the costs of running the programme with ten parents attending, as it was deemed feasible to engage more parents into each group if a longer recruitment period was possible. On the recommendation of the programme developer, and following discussions with the third author, an experienced IY trainer and facilitator, this was considered as the maximum number of attendees per group without compromising programme fidelity (Webster-Stratton, 2015). Increasing the number of parents to ten would result in a greater set up cost of £9538.57 but would yield a smaller cost per child of £953.86, almost half the cost of a smaller group.

In addition to the benefits of social support, one important objective of group led training programmes is to reduce demands on staff time having to travel into communities to deliver them. Getting the parents to attend groups in centralised locations contributes to reducing these costs, however due to the rurality of one of the centres (Powys), and the complex health needs of one of the children, home visits were deemed necessary. These costs have been calculated separately (Table 2) to give an accurate account of research delivery costs, and to inform similar frameworks in the future. Once travel costs and additional group leader time had been added, the overall set up costs increased by £660.36 when home visits were conducted, increasing costs per child to £1903.74 (with 5 parents needing additional visits).

Using the same approach as Edwards et al (2007) costs were divided into non-recurrent and recurrent items, making it easier to calculate future roll-out costs. A large proportion of set up costs included the initial outlay for training, programme materials and fortnightly supervision, which service managers and decision makers would need to consider when introducing the programme. As well as the exclusion of resources and training, differences in roll-out costs included staff salaries. Group leaders who delivered the programme as part of the research trial were experienced and highly paid staff, on NHS salary bands ranging from band 4 to band 8d (£21,00-£73,812), giving a mean hourly rate of £22.64/hr. Following consultation with all centres about ongoing plans to take the programme forward, it was advised that future groups would be facilitated by one nurse (in most cases NHS band 4), and one Psychologist (NHS band 8a). Predicted roll-out estimates were therefore based on these proposed salaries, giving a mean hourly rate of £17.47. Costs of the intervention as part of normal service delivery once training had been undertaken, materials purchased and with proposed changes to facilitators decreased considerably to £4272.09 (£854.42 per child), with 5 parents attending. A sensitivity analysis was performed to show that cost per child decreased further to £481.99 with ten parents attending each group (see Table 3), potentially increasing cost effectiveness.

With regards to taking the programme forward, proposed protocol changes made by the programme developer suggest that future programmes should run for 14 weeks. This came as a result of parent and facilitator feedback, expressing concern that working through the additional content may be difficult should any parents have particularly difficult challenges. Although more costly, the longer running period ensures that group leaders can support parents experiencing unique challenges using the programme's content, ensuring its full effectiveness. Changes have also been made to the training protocol, which means that group leaders no longer have to complete the IY Basic training before undertaking the IY Autism training. The stand-alone training will result in a lower cost of training new or inexperienced facilitators, by £380 per leader. A sensitivity analysis was conducted to calculate the costs of the programme following the protocol changes, which established an increase to £1179.44 per child with five parents attending. Increasing participant numbers to the proposed maximum number of ten per group would decrease this to £644.85 per child.

In comparison to economic evaluations of other IY programmes (Edwards et al., 2007; Charles et al., 2013; Edwards et al., 2016), the cost of establishing and running the trial of the IY Autism was unexpectedly lower than other programmes. This may be due to differences in the trials, such as a greater number of leaders being trained, the use of crèche facilities (Charles et al., 2013; Edwards et al., 2016), and higher catering and venue hire costs (Edwards et al., 2007).

All four groups in the research trial were facilitated by Psychologists within local authorities, where venues were readily available free of charge. All but one of the centres had access to a free venue, therefore venue costs are based on the fee recorded by said centre (£15 per week). Venue hire costs were reported as being considerably higher in previous evaluations (around

£60/week). It is therefore important to consider that venue hire during future implementation could exceed this cost. It was also noted in the cost diaries that refreshments were sometimes provided using supplies left over from previous training groups/meetings within the local authorities, minimising catering costs in this instance.

Other notable differences included the reduced recruitment time for this programme compared to others. This was considered to be due to pre-existing contact between families and specialist child services due to the process of obtaining a diagnosis of ASD. Clerical support was also included in other evaluations (Edwards et al., 2013; Edwards et al., 2016) although not recorded by group leaders in this trial. This could have been influenced by the continuous communication between group leaders, researchers and University staff during regular steering group meetings, resolving any administration queries.

Additional costs include ongoing supervision to ensure programme fidelity. This is deemed essential to running the programme effectively, particularly as it is planned to roll out future programmes with lower paid, and possibly less experienced facilitators. Reducing on-going cost, while maintaining the expertise and competence of group leaders was an issue discussed at length. Centres that will continue to run the programme will provide supervision once every term, to be held with a clinical psychologist (one of the group leaders in the trial). This eliminates travel time and expenses to group supervision sessions, meaning that future roll out calculations only include staff time.

Another potential high cost is the provision of childcare facilities. All the children in this trial were attending school either part or full-time throughout programme delivery, which resulted in lesser need for crèche facilities in comparison to trials of the IY Toddler and Basic programmes (Edwards et al., 2016; Charles et al. 2013). Along with travel costs, difficulties arranging childcare can present as a barrier to parents attending groups. Providing childcare when necessary is therefore a crucial cost to consider for roll-out, to ensure uptake and completion of the programme. For that reason, costs needed to be calculated but were done so based on similar costs from previous costings with an inflation calculation applied (Charles et al., 2013).

Group leaders in this trial had previously undertaken the basic training, however this may not be the case in future roll-out. Training costs need to be considered as a capital cost. Fore mentioned changes to the training protocol mean that a one-time cost of £500 for a 3-day stand-alone training results in an overall future reduction of £350 per group leader.

Lessons from sensitivity analysis

A sensitivity analysis was performed to test varying assumptions of costs (group size, setting, and proposed changes to programme protocol). This established how the costs of the IY Autism programme were affected by increasing the number of group participants from 5 to 10. The cost per child fell when group size increased, which has implications for any future cost-effectiveness analyses i.e. a higher cost-effectiveness ratio would result from increasing numbers. However, it is important to keep in mind the recommendation of 10 parents in each group as a maximum.

The programme was originally 12 weeks, and the research trial was administered as such. Changes were made to ensure programme fidelity; therefore, it was important to calculate the increased cost of these additional sessions. Predictive calculations were also necessary to illustrate costs following proposed changes to training protocol, which result in reduced capital costs to train new group leaders.

The trial was conducted in rural North Wales. More urban settings may have lower travel costs, and more facilities, but higher salary and venue charges – which would need to be considered by local authorities and service managers wishing to implement the programme in their locality.

Strengths

Service managers and decision makers who are considering a new programme to be introduced to their list of services require cost information to establish whether their budget would support the set-up, delivery and roll-out of such a programme. Any lack of detail or underestimation of costs could result in a flawed cost-effective analysis with considerable implications. Accuracy was ensured in this instance by the adherence to the standardised framework Charles et al., (2013). Using a previously developed and standardised framework ensured a precise micro-costing of the IY Autism programme.

As recommended by Charles et al., (2013), methods of gathering costs and resource use data were developed by the research team following consultation with service staff and intervention providers, which occurred during regular steering group meetings throughout the planning and running of the trial. All group leaders were therefore clear on the cost gathering procedure, resulting in high compliance and minimal missing data. Any issues were discussed in steering group meetings ensuring data recording was consistent and accurate. The difficult task of accounting for every cost associated with a service was made easier by the use of well-established cost diaries, which also reduced the need for estimates.

The highest-cost item is group leader time to recruit parents and deliver the programme. Unlike previous evaluations, this micro-costing includes roll-out costs based on predicted lower paid facilitators, giving a more realistic calculation of “real world” and “roll out” costs.

Sensitivity analyses were conducted to test assumptions made in micro-costing by varying costs depending on group size, context and other adjustable variables such as proposed changes to programme and training protocols. This resulted in a helpful projection of delivery costs for the newly developed programme within a range of settings. Calculations of future roll-out costs are valuable to decision makers, and this analysis is particularly informative due to the close communication between researchers and intervention facilitators.

Detailed roll-out strategies identifying proposed staff salaries and supervision arrangements were discussed, giving pragmatic calculations as opposed to figures solely based on the research trial.

Limitations

Using the previously developed framework (Charles et al., 2013) highlights context-bound cost issues; for example, leader travel costs to attend fortnightly supervision and conduct additional home visits. During the trial, groups were run in rural Wales, U.K., where long distances travelled suggest that presented costs may be higher than in urban settings. This is an important consideration for policy with regards to facilitating supervision and prompted discussion about each location having an accredited leader who could provide supervision in their local areas. Consideration could also be given to reducing costs by conducting the sessions virtually, a means that has proven successful during the COVID pandemic.

Although viewed as a strength, the use of diaries as a primary method of cost gathering had potential risks. There was the possibility of leaders missing hidden costs or being unable to identify specific categories of costs such as administrative expenses. This potential risk was minimised by listing certain activities as examples under each category, and by involving group leaders in the development of the cost diaries. Any disparities were also discussed in the steering group meetings.

With regards to hidden costs, there was no method of assessing opportunity costs, such as parents having to take time away from work or other commitments to attend groups. This should be addressed in future evaluations, as 12-14 weeks is a prolonged commitment, and can mean financial losses for the family. The small number of participants could also be considered a limitation, and prospective trials could explore how increasing participant numbers maintain current assumptions.

Although the majority of costs are from accurate figures recorded during the trial, the costs of venue hire, and childcare provision are largely estimated. Although inflation calculations were applied to increase accuracy, these figures could potentially differ.

Implications

This evaluation has provided a direct measure of accurate cost involved in delivering the newly developed IY Autism programme as part of a feasibility trial. A detailed framework, previously developed and used in similar trials, was adhered to ensuring that all expenditures are accounted for when undertaking a full economic costing (fEC) for a larger, definitive trial. The data is precise and categorised in such detail that it can be meaningfully applied to grant funding applications for costing programme delivery under different contexts or in different geographical locations, making this micro-costing a valuable contribution to further research. The study provides crucial, detailed cost data for grant funding applications, and the design of a future definitive trial. This micro-costing also contributes towards identifying the costs and benefits of the programme for policy deciders (providing costs and benefits of different options) for future implementation and provides precise information for any future cost-effectiveness or cost-benefit analysis. Economic evaluations provide both researchers and policy makers with essential data to inform further research, wider implementation and roll-out.

Key Messages

- This is the first detailed costing of a new parenting programme specifically for parents of children with autism.
- The economic evaluations of new interventions following standardised methods are important to inform future research trials, programme roll-out to scale and further economic evaluations.

- Communication between researchers and service/intervention minimises missing data and contributes towards an accurate account of costs.
- The importance of fidelity, including supervision must be emphasised, and needs to be included in all future estimations despite the additional cost. Exploring ways of minimising costs is important (e.g., each centre having an accredited person local to them to oversee supervision of group leaders, possible facilitation of supervision sessions virtually).
- Sensitivity analysis is valuable to test assumptions and explore how increasing numbers (whilst still maintain programme fidelity) can increase cost effectiveness.
- Costs have been divided into distinct categories (e.g. recurrent and non-recurrent costs, training, resources, staff time and salaries), thus more informative to calculate full economic costs of future trials and roll-out, under different contexts.

Chapter 7

Long term outcomes of the programme

Introduction

The core characteristics of autism spectrum disorder (ASD) are impaired functioning in areas of communication and social interaction, and restricted or repetitive behaviours or interests (American Psychiatric Association, 2013). An estimated one in every 100 children in the UK are on the autistic spectrum (National Autistic Society, 2022) with a higher prevalence among males (Adak & Halder, 2017). Children with ASD frequently have co-morbid physical and mental health conditions (Maskey, Warnell, Parr, Le Couteur & McConachie, 2013), and are at increased risk of emotional and behavioural problems by comparison with typically developing peers (Newcomb & Hagopian, 2018). It is well established that this population exhibits elevated levels of both externalising (hyperactivity, aggression, impulsiveness, self-injury) and internalising (anxiety, depression, sleep disturbances) behaviours (Ahearn, Castine, Nault & Green, 2001; McClain, Mills & Murphy, 2017; Mannion & Leader, 2013). Co-occurring emotional and/or behaviour problems can be multifaceted, with several studies reporting rates as high as 50-75% of children exhibiting multiple forms of problem behaviour (Charman, Ricketts, Dockrell, Lindsay & Palikara, 2015; Petrou, Soul, Koshy, McConachie & Parr, 2018), with aggression, impulsiveness, hyperactivity and noncompliance among the most prevalent (Simonoff et al., 2008; Kanne & Mazurek, 2011; O’Nions et al., 2018). Typically, problems persist into adolescence and adulthood resulting in a host of difficulties within educational, familial and social domains (Simonoff et al., 2013; Knapp et al., 2009). Not only does this impact negatively on the child’s acquisition and performance of daily living skills, but parents are also more likely to encounter higher levels of psychosocial adversity (Krakovich, McGrew, Yu & Ruble, 2016), due to the unique and often overwhelming challenges they face (Clauser, Ding, Chen, Cho, Wang & Hwang 2021; Enea & Rusu, 2020). Parents of children with ASD experience disproportionate levels of stress and depression in comparison to those of neurotypical children (Griffith, Hastings, Nash & Hill, 2010; Kissel & Nelson, 2016; Schnabel et al.,2020), this occurs to varying degrees based on specific diagnosis and severity of key symptoms (Hayes & Watson, 2013; Mello, Rivard, Morin, Patel & Morin, 2021). High prevalence of parental mental health problems has been ascribed to various factors including the diagnostic process itself (Gabriels, Cuccaro, Hill, Ivers & Goldson, 2005), feeling unsupported and socially isolated post-diagnosis (Banach et al., 2010), and uncertainty about the future impact of the diagnosis on family function (Falk et al., 2014). However, parental distress among parents of children with autism is predominantly attributed to managing the co-occurring child emotional and behavioural problems (Lecavalier & Wiltz, 2006; Hayes & Watson, 2013). Many parents report such difficulties as being the most challenging aspect of their parenting experience; more so than those associated with the core characteristics of ASD (Petrou et al., 2018), or the high level of practical support required by their child (Kissel & Nelson, 2016).

Parental mental health problems can affect the ability to parent effectively (Gross, Shaw, Burwell & Nagin, 2009). Elevated stress can reduce parental responsiveness (Gerstein, Crnic, Blacher & Baker, 2009) and impair the parent’s ability to use effective discipline and coping strategies (Shawler & Sullivan, 2017; Clauser et al., 2021). Maternal depression is also an important determinant of family functioning and is a known predictor of low self-efficacy (Albanese, Russo & Geller, 2019). Belief in their own parenting ability is critical for parents as it can alter their assessment of situations as being less problematic and help them to feel better equipped to deal with challenges (Bloomfield & Kendall, 2012). Self-efficacy is also a recognised mediator of parental stress (Raikes & Thompson, 2005) with parents of children with ASD at greater risk of lower perceived self-efficacy and having poor parent-child relationships (Rezendes & Scarpa, 2011; Dissanayake et al., 2020). This can result in increased use of ineffective parenting strategies (Albanese et al., 2019). Maljaars and colleagues (2013) observed that parents of children with autism are more likely to adopt lax parenting behaviours, with less rule setting and discipline than parents of typically developing children. Ineffective parenting practices, including inconsistent and lax discipline, are strongly associated with increased risk of child behaviour problems and long-term difficulties (Nelson, Stage, Duppong-Hurley, Synhorst & Epstein, 2007; Furlong et al., 2012), and can result in a coercive cycle of behaviours within the dyad if not addressed through effective intervention (Patterson, 2002; Hastings, 2002). Improving parental self-efficacy, mental health and parent-child relationships can therefore be important protective strategies to overcome the development and maintenance of problem behaviours (Albanese et al., 2019; Leijten et al., 2019; Sanders & Woolley, 2005).

Based on guidance from the National Institute for Health and Care Excellence (NICE), behaviourally based parenting programmes are recommended for both prevention and early treatment of child behaviour problems in both typically developing children and those with ASD (NICE, 2013; NICE, 2021). Parent training programmes offer many benefits to families of children with ASD, as their potential to improve parental psychosocial wellbeing can be twofold (McConachie & Diggle, 2007; Matson, Mahan & Matson, 2009; Stadnick, Stahmer & Brookman-Frazee, 2015). Although their primary focus is to increase parenting skills to support their child’s physical and emotional development, parents are also provided with strategies that can directly improve their own psychological functioning. This is particularly true of interventions founded on cognitive-behavioural approaches, making them the most effective interventions both in clinical and community settings (Gardner, Burton & Klimes, 2006; Hutchings & Lane, 2005).

The Incredible Years (IY)® programmes are a series of developmentally appropriate group based parenting interventions founded upon social learning theory (Webster-Stratton, 2011), that have been shown to be effective in a large number of randomised controlled trials across multiple settings (Bywater et al., 2009; Bywater et al., 2011; Hutchings et al., 2017;

Jones, Daley, Hutchings, Bywater & Eames, 2008; Morpeth et al., 2017). Their aim is to improve child behaviour through improving parenting quality and parent-child relationships using a collaborative, group-based approach (Webster-Stratton, 2015). Parents are encouraged to recognise and rehearse positive parenting skills, and to identify effective problem solving and coping strategies. The group members discuss video examples of positive parenting approaches, practice what they see in role play exercises and complete homework activities to reinforce their learning (Webster-Stratton, 2011). The IY Basic® programme is the most widely delivered and researched of the IY series, with considerable evidence showing it to be an equally effective treatment and prevention of conduct disorder (CD) and attention deficit hyperactivity disorder (ADHD) for various populations (Bywater et al., 2009; Bywater et al., 2011; Hutchings et al., 2017; Leijten et al., 2018; Leijten et al., 2019). Programme outcomes have consistently included increased positive parenting behaviour, reduced parental stress and depression, less frequent child behaviour problems and enhanced child social competence (Menting, de Castro & Matthys, 2013). The IY Basic® is also one of 11 'Blueprint' interventions approved by the Centre for Violence Prevention due to its extensive evidence base including in randomised control trials (RCT's), long term follow-up studies and independently replicated trials (Mihalic, Fagan, Irwin, Ballard & Elliot, 2002; Webster-Stratton & Reid, 2018).

Although the Incredible Years® programmes are delivered over a comparatively short time (generally 12 weeks), previous research has established that gains made in this time are maintained longer term (Bywater et al., 2009; Overbeek et al., 2021; Webster-Stratton, Rinaldi & Reid, 2011). This is an important consideration given that a recent review of parent programme effectiveness concluded not only that further research is required to establish their efficacy for parents of children with additional needs, but also to determine whether their effects are maintained over time (Barlow & Coren, 2018). Webster-Stratton and colleagues (2011) evaluated the long-term adjustment of children whose early onset conduct problems had been treated with IY programmes 8-12 years earlier. Overall findings showed that the children of parents who had attended during their early years (aged 3-7), exhibited less severe conduct problems in adolescence than expected; although conclusions were reliant on predicted outcomes based on childhood trajectories rather than comparison to any control conditions (Webster-Stratton, Rinaldi & Reid, 2011). Scott, Briskman & O’Connor (2014) examined the long-term benefits of the IY Basic programme from two high-quality RCT’s of the intervention as an early prevention for young children at risk of later conduct problems. The study examined whether improvements made during initial trials with children aged 3-7 on conduct disorder symptoms and antisocial character traits had any persisting effects into adolescence. Families allocated to the intervention group showed a large reduction in child behaviour problems and significant improvements in directly observed parenting behaviour, which were maintained at 18 months. Longitudinal follow ups conducted up to ten years post intervention, showed that although improvements were maintained in child antisocial behaviour and parental warmth, no differences were found in directly observed parenting style. It was concluded that although parent training programmes could be beneficial in the prevention of child behaviour problems both in the short- and longer-term, booster courses might be required to maintain changes in parenting behaviour and to address new developmental challenges as children moved into adolescence (Scott et al., 2014). The study carried important implications for public health with regards to calculating the benefits to the future societal costs of problem behaviour but highlighted the need for further research to establish definitive long-term benefits of the programme. The study also lacked any measures of parental wellbeing outcomes, which could be considered a limitation. Benefits to parental mental health were explored in addition to long-term efficacy by Bywater et al. (2009), in a randomised control trial of the IY Basic programme with 153 families from disadvantaged areas. Post intervention improvements were observed on child behaviour, positive parenting practices and parental stress and depression. Although other studies show a typical pattern of intervention outcomes diminishing across subsequent follow ups (Scott et al., 2014; Overbeek et al., 2021), these gains were maintained with no loss of effectiveness at further 12 and 18 month follow ups (Hutchings et al., 2007; Bywater et al., 2009), implying that booster courses were not always necessary as inferred in longer-term studies (Scott et al., 2014).

In addition to children with conduct disorders and high-risk families, the IY Basic has been the focus of studies with other populations. The programme was delivered to families of children with ASD within a newly established NHS service promoting the wellbeing of children and young people (Roberts & Pickering, 2010). An evaluation of the service had identified the need for parental support following a marked increase in referrals from families with complex neurodevelopmental disorders. Parents who were experiencing behaviour management difficulties were invited to attend, with participating children either having received a diagnosis of ASD or being under assessment. The IY Basic programme was chosen as it was well established within local services, and both the behavioural practitioner and clinical psychologist facilitating the groups were experienced in its delivery. It was anticipated that the components of the programme, for instance the focus on emotion coaching, social communication, and behaviour management techniques would be particularly relevant. Post-intervention measures found improvements in parental well-being, including reduced stress, and improvements in child behaviour. Although the pilot was conducted with only eight parents, and no control group for comparison, the results were helpful to inform further research (Roberts & Pickering, 2010). Notwithstanding the promising results demonstrating the efficacy of IY Basic in its original format in this pilot trial with this population, Dababnah and Parish (2016) undertook a feasibility study using a tailored version of the Basic programme for parents of children with ASD. Adaptations to the programme included increased focus on emotion coaching and self-regulation skills, and additional time spent discussing parental stress and the unique play behaviours of children with autism. Positive effects were reported in improved child behaviour, and the study also explored parental wellbeing outcomes. Significant reductions were observed in parental stress, replicating the promising parent outcomes from previous trials of the programme with children primarily referred for conduct disorders (Bywater et al., 2009). Dabanah and Parish (2016) provided good evidence for the effectiveness of the IY Basic but made recommendations to increase its suitability to this population. Although feedback was generally positive, parents felt that the video vignettes, which are integral to the programme, were dated and not representative of children on the autistic spectrum. The study also had limitations, in that there was no control group to ascertain whether gains were due to the intervention rather than other factors. However, the IY Basic® programme was found to be generally acceptable to parents of young children with autism (Dabanah & Parish, 2016; Dababnah et al., 2019).

In 2015, the Incredible Years Autism Spectrum and Language Delays Programme (IY-ASLD®) was developed as a new addition to the IY series that specifically targets parents of children with autism. It uses the same core delivery processes as the IY Basic® programme but with increased focus on coaching language and communication skills and includes video examples depicting children with ASD. Imitation and pretend play techniques are introduced to help develop the child's empathy and social skills. Greater emphasis is placed on parental self-care along with the importance of building a support group; therefore, although smaller groups are recommended (6-8 parents), partners or other family members are also encouraged to attend. Hutchings and colleagues (2016) conducted an initial pilot study of the newly developed programme, delivering it for the first time in the UK, to a group of nine parents. As well as exploratory measures of parental well-being and child behaviour, course evaluation questionnaires and semi-structured interviews were completed to establish parents’ response to the programme. High satisfaction scores were received reflecting the positive parental feedback from previous research (Dababnah & Parish, 2016). In contrast, parents rated the video content of the new programme as being very helpful because they depicted children on the autism spectrum, suggesting that the changes made had increased relevance. Parents also reported the group discussions and meeting other parents in the same situation as beneficial. The main limitation of the study was the small sample size and absence of a comparison group. The study also relied heavily on parent reported measures, which can be subject to bias (Hutchings et al., 2007). Nevertheless, the positive feedback and encouraging results lay a solid foundation for a larger study.

Williams, Hastings and Hutchings (2020) subsequently ran a small randomised controlled trial to examine the feasibility of delivering the IY-ASLD® programme within existing child services in Wales. The PALACES (Parenting for Autism, Language and Communication Evaluation Study) trial was designed to explore the acceptability of the newly developed programme by examining implementation fidelity, programme adherence and retention, and the satisfaction of parents and facilitators. Assessing these aspects of programme delivery are essential before being able to reliably evaluate the effectiveness of an intervention in definitive trials. Feasibility outcomes were operationalised in terms of participant recruitment and programme attendance. Programme acceptability was assessed with a parent-reported end of programme questionnaire. Feedback was gathered about teaching format and group leaders, the parenting strategies learned and overall satisfaction. A high proportion of parents completed the questionnaire, reporting very high satisfaction levels for teaching format and facilitator skills. All attendees reported that they would recommend the programme to other parents. Programme attendance was good, with 73% of parents attending eight or more sessions, further indicating that the programme is acceptable to this population. The most common programme benefit reported was meeting other parents and shared experiences. Feasibility studies also aim to test the methods and procedures that will be relevant to larger studies of programme effectiveness, which includes testing measures. A range of standardised parent and child measures were administered, including those to assess parental well-being, and child behaviour and adaptive skills. As part of the feasibility study, data collection was conducted at baseline, and at 6-month follow up. Although there were no significant differences in any outcomes at 6-month follow-up, exploratory analyses of programme effectiveness showed promising improvements in favour of the intervention group including moderate effect sizes in increased parental praise. Small effect size benefits also occurred in other observed parenting behaviours such as increased coaching, observed questions and reduced laxness. Reductions were also seen in measures of parental distress and child externalising behaviours (findings presented in Williams, Hastings & Hutchings, 2020). Preliminary findings at 6-month follow up were encouraging and warrant further research in a larger trial. Many feasibility questions were also addressed with regards to planning a future trial. Short term evaluations are typical in studies of parent training programmes, however longer-term evaluations are important to determine whether any changes are maintained over time (Leijten et al., 2019). It is therefore necessary to also consider the feasibility of assessing parents for some time after the programme has ended. This is especially pertinent bearing in mind that some parenting programmes which were previously endorsed as blueprints for violence prevention, have been downgraded due to lack of evidence of long-term effectiveness (World Health Organisation, 2013). Long-term monitoring is especially relevant to ASD specific interventions, given the lifelong nature of the disorder. Based on previous evidence of long-term outcomes of IY® programmes (Hutchings et al., 2007; Bywater et al., 2009), this study will examine whether the IY-ASLD® can also yield longer term benefits. It is suggested that any future trials of the programme should include long-term follow up with parents, therefore it was deemed valuable to assess participant retention over time, and the feasibility of collecting longer term data in future definitive trials. This study will further examine the feasibility of the measures, and participant retention by engaging parents in longer term data collection. The study aims to explore whether the promising gains observed at 6-months during the PALACES trial were maintained, and/or whether any new behaviours were seen to emerge over time to further explore the real-world value of the IY-ASLD® programme.

Method

Please see Chapter 4: Methods and Design for the full details of trial methods, and further information on the intervention and battery of measures administered.

A range of parent and child measures that had been administered at baseline and at 6-month follow up during the PALACES trial, were re-administered at 12-month and 18-month follow up to the intervention group of parents only. Participant retention rates were calculated at both timepoints, along with an inspection of missing data to further consider the acceptability of measures and inform any potential concerns about retention in future trials.

Parent outcomes

Parenting behaviours were examined with the Arnold O'Leary Parenting Scale (Arnold, O'Leary, Wolff, & Acker, 1993) which contains three subscales (laxness, verbosity and over-reactivity) and a total score. A behavioural observation of parent-child interaction was also conducted to assess frequencies of parental praise, coaching, reflections, questions and negative parenting practices (DPICS; Eyberg & Robinson, 1981). Only the first 5 minutes of the parent-child interaction were coded due to some parents experiencing challenges in engaging the child for the full 10 minutes.

Parental mental health was assessed using the Beck Depression Inventory (BDI-II; Beck, Steer, & Brown, 1996), and the Parent Stress Index (Abidin, 1990), which consists of four subscales (parental distress, parent-child dysfunctional interaction, difficult child, defensive responding) and a total score.

Child Outcomes

Child measures included the Child Behaviour Checklist (CBCL) (Achenbach & Rescorla, 2000), which consists of two subscales (externalising and internalising) as well as a total score. Child adaptive skills were measured using the Vineland Adaptive Behaviours Scale (VABS; Sparrow, Balla, Cicchetti, Harrison, & Doll, 1984), which assesses communication, socialisation and daily living skills, as well as a total score. Social communication skills were assessed using the Social Communication Questionnaire (SCQ; Berument, Rutter, Lord, Pickles, & Bailey, 1999).

Data analysis

Long term data were checked for normality and found to be not normally distributed. All measures were therefore analysed using a Wilcoxon related-sample signed rank test; a non-parametric alternative to t-test. Median and ranges were compared to determine any differences between data points (Baseline to six-month follow-up, and six-month to 18-month follow-up). Missing data from individual items on questionnaires were dealt with based on the guidelines in questionnaire manuals, and in all cases, missing items were prorated. Some parents faced challenges engaging the child in a full 10-minutes of interaction, therefore only the first 5 minutes of the observations were coded for all participants. Outcome effect sizes *(r)* were interpreted based on Cohen’s d (0.1-small, 0.3-medium, 0.5-large).

Analytic approaches

Two analytical approaches were taken with the long-term data. Firstly, the targeted gains that were observed as part of the exploratory findings of the PALACES trial were followed up at 18-months to examine whether those gains had been maintained longer term. These included reductions in child externalising behaviour, the lax parenting subscale of the parenting scale, and the parental distress subscale of the Parenting Stress Index. Promising improvements had also been observed in observed parental praise, coaching and asking questions. Wilcoxon related sample signed rank tests were performed to assess for differences between post-intervention at 6-months, and 18-months for all targeted gains to assess whether these gains had been maintained over time.

Secondly, exploratory assessments were conducted on all outcomes to examine where changes were occurring, if any, across time points. Analysis was run to compare 6-month data with 18-month data to examine whether any new gains that were not evident at 6-months had emerged. Differences were also compared between baseline and 18-months to examine any changes between baseline and final follow up.

Results

Sample Characteristics

Eighteen parents participated in data collection at 18-month follow up. Children in the intervention group were predominantly male (73%, *n* = 19), with a mean age of five and a half (*M* = 67.12 months) at baseline with a diagnosis of ASD (88%, *n* = 23). The majority (88%, *n* = 23) of children had scores of <70 on the VABS adaptive behaviour standard score, and more than three quarters (76%, *n* = 19) scored ≥63 on the CBCL indicating elevated child behaviour problems. Participating parents were predominantly female (88%, *n* =23), with a mean age of 37 years old at baseline. The majority were reporting elevated levels of stress (69%, *n* = 18), but only a minority (33%, *n* = 8) scored between 14-28 (mild to moderate) on the depression inventory at baseline. Over half (53%, *n* =14) left school before the age of 17 years. Only a small minority were younger than 20 years old at birth of first child (8%, *n* = 2). There was employment in most households (77%, *n* = 20).

Table 7:1. Baseline Demographics

	(*n* = 26)

Child age (months)	67.12 (15.19)
< 6 years	*17(65.40)*
≥ 6 years	*9(34.60)*
Child gender	
Male	*19 (73.08)*
Female	*7 (26.92)*
Diagnosis status	
ASD	*23 (88.46)*
Suspected	*3 (11.54)*
CBCL total score	71.12 (8.50)
T > 63 (n = 25)	*19 (76)*
T ≤ 63 (n = 25)	*6 (24)*
VABS adaptive behaviour standard score	59.50 (11.00)
< 70	*23 (88.46)*
≥ 70	*3 (11.54)*
Parent age (years)	36.77 (7.64)
Parent gender	
Male	*3 (11.54)*
Female	*23 (88.46)*
Teenage parent	
< 20 years birth of first child	*2 (7.70)*
≥ 20 years birth of first child	*24 (92.30)*
Education	
< 17 years left school	*14 (53.85)*
≥ 17 years left school	*12 (46.15)*
Unemployment	
No employment in household	*6 (23.08)*
Employment in household	*20 (76.92)*

Retention

Retention was high (96%) at 6-month follow up, falling significantly at 12-month follow up to 64% before improving to 69% at 18-month assessments. Of the eight families lost to the final follow up, five (62.5%) had left education before the age of 17. Those lost to follow up included three of the four fathers randomised to the intervention group.

Targeted gains

The gains that were observed post intervention in the PALACES trial were further examined at 18-months to see whether they had been maintained over time. Between six and 18-month follow-up, praise was significantly lower *(T = 20, p = .074, r = -0.06)*, as was coaching *(T = 8.5, p = .029, r = -0.08)*. Therefore, despite the increases observed post intervention, the frequencies of both parental praise and coaching decreased between 6-month and 18-month follow up, returning to similar levels observed at baseline. An increase in the frequency of asking questions was also seen at 6 months follow up, however a non-significant reduction was observed between 6-months and 18-months *(T = 45, p = .638, r = -0.02),* although frequencies did remain higher at 18-months than they were at baseline.

Non-significant reductions were observed in the Laxness subscale between six-month and 18-month follow-up *(T = 92.5, p = .760, r = 0.00).* Child externalising behaviour was non-significantly lower between six-month and 18-month follow-up *(Externalising; T = 45.4, p = 0.81, r = -0.04).* There was no statistical difference in parental distress between 6-months and 18-months *(T = 69.5, p = .938, r = 0.02),* however levels remained lower than baseline.

Exploratory findings

Parent outcomes

Parental mental health:

Between baseline and 18-month follow-up, there was a non-significant reduction in the total score of parenting stress *(T = 40.5, p = .268, r = -0.03)*, difficult child *(T = 36, p = .172, r = -0.04)* and defensive responding *(T = 43, p = .333, r= -0.03)* subscales. There was a non-significant increase in parent-child dysfunctional interaction *(T = 62, p = .550, r = 0.01),* and a significant reduction in parental distress *(T = 31, p = .309, r = - 0.03).* Between 6-months and 18-months, a non-significant increase was observed in the parent-child dysfunctional interaction subscale *(T = 94, p = .178, r = 0.03),* and in defensive responding *(T = 70, p = .917, r = 0.00).* Median scores on four out of the five subscales were lower at 18-month follow-up than at baseline *(see Table 2 for median scores).*

Parental depression was assessed using the Beck Depression Inventory (BDI-II; Beck, Steer, & Brown, 1996). There was a non-significant reduction in BDI scores between baseline and 18-month follow-up *(T = 43.5, p = .571, r = - 0.02).* A non-significant reduction was also observed between six-month and 18-month follow-up *(T = 47, p = .729, r = -0.01).* The median scores were lower at 18-month follow-up than at baseline *(see Table 2 for median scores).*

Parenting practice:

There was a non-significant reduction in all subscales between baseline and 18-month follow-up *(Laxness: T = 43, p = .334, r = -0.03; Over-reactivity: T = 41, p = .161, r = -0.04; Verbosity: 70.50, .776, -0.00; Total score: T = 43, p = .196, r = -0.04).* A decrease was observed in *Over-reactivity: T = 49.5, p = .200, r = -0.0; and Verbosity: T = 94, p = .407, r = 0.02)* between 6-months and 18-months, and a non-significant increase observed on the total score *(T= 88.5, p = .896, r = 0.00)*. Median scores were lower at 18-month follow-up than at baseline on all subscales *(see Table 2 for median scores).*

Parent-child observation:

Between baseline and 18-month follow up, a significant reduction was observed in positive parenting *(T = 1.5, p = <0.05, r = -0.11*), and reflections *(T = 6, p = <0.05, r = -0.05).* There was a non-significant reduction in negative parenting *(T = 21.5 p = .304, r = -0.04),* and parental praise *(T = 20.5, p = .475, r = -0.05)*; and a non-significant increase in questions *(T= 37, p = .875, r = - 0.00)*. Between six-month follow-up and 18-month follow-up positive parenting was again significantly lower *(T = 5, p = 0.05, r = -0.10).* Negative parenting was non-significantly lower *(T = 17.5, p = .091, r = -0.06),* as was reflections *(T = 29, p = .436, r = 0.03).* Medians were lower between baseline and 18-month follow up on three categories: positive parenting, reflections and negative parenting. Two of the categories had higher medians between baseline and 18-month follow-up (praise and questions). There was no statistical change between medians of coaching between baseline and 18-month follow up *(see Table 7.2 for median scores).*

Child outcomes

Child behaviour:

Child behaviour was assessed using the Child Behaviour Checklist [CBCL] (Achenbach & Rescorla, 2000). There was no change in Total scores between 18-months and baseline *(T = 33, p = .070, r = - 0.05);* there was no statistical difference on total scores between six-month and 18-month follow-up *(T = 39, p = .396, r = - 0.02).* Scores on both subscales of the CBCL were significantly lower at 18-month follow-up than at baseline *(Externalising; T = 37.5, p = < .05, r = -0.05: Internalising; T = 22, p= < .05, r = - 0.06);* and non-significantly lower between six-month and 18-month follow-up *(Externalising; T = 45.4, p = 0.81, r = - 0.04: Internalising; T = 64, p = .836, r = 0.00).* Median scores were lower at 18-month follow up than at baseline, apart from the Total scores, which remained the same (see Table 2 for median scores).

Child Social Communication:

Child social communication was assessed using the Social Communication Questionnaire [SCQ] (Berument et al., 1999). There was a non-significant reduction between baseline and 18-month follow-up *(T = 28, p = .068, r = -0.05),* with no statistical difference between six-month and 18-month follow up *(T = 44.5, p = .614, r = - 0.01).* Median scores were lower at 18-month follow-up than at baseline but had slightly increased from the 6-month follow-up *(see Table 7.2 for median scores).*

Child adaptive skills (VABS):

Child adaptive skills were assessed using the Vineland Adaptive Behaviour Scales II Parent/Caregiver Rating Form (VABS; Sparrow, Balla, Cicchetti, Harrison, & Doll, 1984). Total adaptive skills scores were significantly lower at 18-months than at baseline *(T = 9, p=*

.001, r = -0.09) indicating a worsening of adaptive skills. There was also a significant reduction on the socialisation sub-scale between the two time points *(T = 12, p = .002, r = -0.08)*. Scores on the communication sub-scale were non-significantly lower at 18-months *(T = 33.5, p = .740, r = -0.05)*. There was a non-significant increase on daily living skills subscale *(T = 94.5, p = .170, r = 0.04)*. Median scores were lower at 18-month follow-up than at baseline for three of the subscales and higher for the daily living skills subscale *(see Table 7.2 for median scores)*.

Table 7:2. *Long-term maintenance effects for all outcome measures*

		BL		**6-mo FU**		**18-mo FU**		**BL to 18-mo FU**		**6- to 18-mo FU**
	n	***Median (range)***	***n***	***Median (range)***	***n***	***Median (range)***	***n***	**T (*p*, r)**	***n***	**T (*p*, r)**
Child behaviour										
CBCL externalising	25	65.00 (48.00)	25	63.00 (45.00)	18	61.50 (29.00)	18	37.50 (.036, -0.05)	18	45.40 (.081, -0.04)
CBCL internalising	25	71.00 (27.00)	25	66.00 (33.00)	18	64.00 (30.00)	18	22.00 (.031, -0.06)	18	64.00 (.836, 0.00)
CBCL total	25	71.00 (40.00)	25	70.00 (37.00)	18	71.00 (37.00)	18	33.00 (.070, -0.05)	18	39.00 (.396, -0.02)
Child social communication and adaptive skills										
SCQ total	24	25.00 (28.00)	24	19.50 (26.00)	18	21.50 (29.00)	17	28.00 (.068, -0.05)	17	44.50 (.614, -.0.01)
VABS communication	26	62.50 (59.00)	/	/	18	51.50 (75.00)	18	33.50 (.740, -0.05)	/	/
VABS daily living	26	56.50 (55.00)	/	/	18	59.50 (64.00)	18	94.50 (.170, 0.04)	/	/
VABS socialisation	26	57.00 (34.00)	/	/	18	50.00 (39.00)	18	12.00 (.002, -0.08)	/	/
VABS total	26	56.00 (50.00)	/	/	18	49.00 (50.00)	18	9.00 (.001, -0.09)	/	/
Parenting practices										
PS laxness	24	2.73 (3.55)	24	2.64 (3.55)	18	2.59 (2.90)	24	43.00 (.334, -0.03)	18	92.50 (.760, 0.00)
PS over-reactivity	24	1.90 (2.20)	24	1.90 (2.00)	18	1.55 (1.50)	24	41.00 (.161, -0.04)	18	49.50 (.200, -0.03)
PS verbosity	24	3.14 (3.00)	24	2.57 (2.71)	18	2.79 (3.90)	24	70.50 (.776, -0.00)	18	94.00 (.407, 0.02)
PS total	24	2.82 (2.50)	25	2.54 (2.23)	18	2.40 (2.30)	24	43.00 (.196, -0.04)	18	88.5 (.896, 0.00)
Parental mental health										
PSI total	25	97.00 (80.00)	24	92.00 (78.00)	17	90.00 (81.00)	16	40.50 (.268, -0.03)	16	71.50 (.856, 0.00)
PSI Defensive Responding	25	20.00 (19.00)	25	17.00 (21.00)	17	17.00 (21.00)	16	43.00 (.333, -0.03)	17	70.00 (.917, 0.00)
PSI Parental Distress	25	31.00 (34.00)	25	28.00 (34.00)	17	29.00 (36.00)	16	31.00 (.309, -0.03)	17	69.50 (.938, 0.02)
PSI PCDI	25	29.00 (22.00)	25	29.00 (22.00)	17	30.00 (25.00)	16	62.00 (.550, 0.01)	17	94.00 (.178, 0.03)
PSI Difficult Child	25	38.00 (31.00)	24	35.50 (32.00)	17	35.00 (29.00)	16	36.00 (.172, -0.04)	16	52.00 (.649, -0.01)
BDI	24	9.00 (33.00)	25	9.00 (25.00)	16	6.00 (26.00)	15	43.50 (.571, -0.02)	15	47.00 (.729, -0.01)
Parent-child observation										
Positive Parenting	24	8.50 (54.00)	24	6.50 (80.00)	14	2.50 (13.00)	13	1.50 (.003, -0.11)	14	5.00 (.005, -0.10)
Praise	24	4.00 (15.00)	24	8.50 (23.00)	14	4.50 (19.00)	13	20.50 (.475, -0.05)	14	20.00 (.074, -0.06)
Coaching	24	0.00 (27.00)	24	3.50 (16.00)	14	0.00 (6.00)	13	6.00 (.173, -0.05)	14	8.50 (.029, -0.08)
Reflections	24	2.00 (16.00)	24	2.00 (20.00)	14	1.00 (13.00)	13	6.00 (.046, -0.05)	14	2.00 (.436, 0.03)
Questions	24	12.50 (36.00)	24	18.00 (35.00)	14	16.00 (32.00)	13	37.00 (.875, -0.00)	14	45.00 (.638, -0.02)
Negative Parenting	24	1.00 (10.00)	24	1.00 (15.00)	14	0.00 (13.00)	13	21.50 (.304, -0.04)	14	17.50 (.091, -0.06)

Discussion

The PALACES trial was the first RCT of the newly developed Incredible Years Autism Spectrum and Language Delays (IY-ASLD®) programme, designed to examine the feasibility of delivering the programme successfully within existing services. Exploratory findings from the trial had observed promising improvements in parenting behaviours, parental stress and child behaviour (Williams et al., 2020). The aim of this study is to further explore feasibility questions following the initial pilot, including the achievability of retaining parents for longer term data collection and to explore the longer-term outcomes to inform a larger, definitive trial. The main objectives with regards to exploratory measures were a) to determine whether improvements observed post-intervention were maintained longer term, and b) to assess whether any new improvements had emerged after parents had been using the learnt strategies for a longer period of time. Among the 26 who attended the programme, parents were predominantly mothers which is consistent with the literature. The majority of children had a confirmed diagnosis of ASD, and most were male, which was predicted given the higher prevalence of ASD among boys. Retention of intervention parents was high at 6-month follow up, but fell at 18-months to 69%, which despite the decrease, is still a good percentage. The reasons for drop out from follow up were incidental and included work or other commitments such as child pick up times, or illness rather than wanting to dis-engage from the trial. The indication that parents are willing to commit to long term participation is encouraging for future research.

Targeted analyses were conducted to examine whether the gains observed at 6-month follow up were maintained. The post-intervention improvements in parental praise and coaching observed at 6-months had not been maintained at 18-month follow up, with frequencies returning to baseline levels following the initial improvements. A medium effect size in increased praise had been observed at 6-months suggesting that parents were praising their child more after learning how to use praise as a reward in the programme. However, praise is a ‘social reinforcer’, and there is evidence to suggest that children with autism do not respond to praise in the same way as typically developing children (Bayat, 2011). It is possible therefore, that although parents were using praise as a reinforcement for desired behaviour initially, they may not have continued to do so if it was not an effective strategy. There could also be a social reason for the reduction is coaching, as its success is very much dependant on back-and-forth interaction between parent and child. There were several children with language delays or who were non-verbal within the sample, which could be a reason for less opportunities to coach language and emotions. However, there is also the possibility that children in the sample were not engaging in communication during the observation due to the associated preference for solitary play. This would mean less opportunities for parents to praise and coach during the observed interaction. There were also changes to the frequency of asking questions across time points, which could be attributed to child language difficulties and the preference to play alone. An increase in asking questions had been observed in post intervention assessments during the PALACES trial, which although is not generally the goal of child-led play, was considered a positive during interpretation of outcomes as it was indicative of the parent trying to engage with the child, although parent-led. Follow up analysis showed the frequencies of questions did reduce between 6-month and 18-month timepoints, which again may be due to the lack of reciprocal communication, as it is widely recognised that children with ASD respond less often to communication attempts than typically developing children. Changes in parenting behaviours had also been observed post-intervention, with a reduction in laxness among intervention parents. At 18-month follow up, there was a further reduction which is encouraging, especially given that the literature shows a greater risk of lax parenting among parents of children with ASD (Maljaars et al., 2014). However, it should be acknowledged that the self-report measure used to assess parent behaviours could be subject to bias, either from exaggerated responses and/or treatment expectation. There is also the risk that parents are responding in a way they think is desirable to the researcher. However, these reductions could be consistent with other positive changes observed for instance in child externalising behaviour, which had decreased at 18-month follow-up, following a previous reduction at 6-months. Child externalising behaviour is shown to be exacerbated by lax parenting (Tarver, Daley, Lockwood & Sayal, 2014), therefore, these further improvements in behaviour could be an indication that parents were using more effective discipline techniques following the programme. Despite not screening for child behaviour problems, high levels were reported at baseline with 76% scoring within the clinical range which is consistent with the research showing increased risk of behaviour problems in children with ASD (Newcomb & Hagopian, 2018). There is also a well-documented relationship between child externalising behaviour and parental distress which had also reduced following the programme at 6-months. Although no further reductions were observed at 18-months, levels of parental distress remained lower than at baseline which is a promising long-term result considering the increased levels of stress among this population of parents.

As is good practice following feasibility studies, further exploratory analyses were conducted to explore whether there were any other changes or new gains following the programme. Baseline levels of parental stress were high in comparison to population expectation, with 66% of parents reporting levels of stress above the PSI cut off score of 90. This is consistent with the extensive literature involving parental stress and ASD (Clauser et al., 2021; Griffith et al., 2010). As well as the maintenance of gains that were observed in parental distress at 6-months, non-significant reductions were seen in the total score and on the difficult child subscale at 18-month follow up. There was a non-significant reduction in defensive responding, a scale that assesses the likelihood of parents answering in a favourable way to minimise any problems or stress to researchers. However, scores were not low enough to justify concern that parents were not honest in their responses. This means that overall, parental stress levels had improved at final follow up, which is encouraging. There were also improvements to levels of parental depression. Contrary to other research involving this population (Schnabel et al., 2020), depression scores among the sample were not elevated, with a minority (33%) scoring mild to moderate levels of depression on the BDI at baseline. Parental depression scores had reduced at 18 months, from both baseline and six-month follow-up, although not significantly. Long-term median scores for both parent well-being measures have maintained at lower than baseline, which is encouraging. Given that social isolation and the feeling of being unsupported were identified as parental mental health risk factors, it should also be considered that these improvements could have resulted from the social aspect of attending the programme. Group discussions could contribute towards normalising child behaviour and daily challenges, thus reducing parental anxieties through shared experiences. However, improvements in parental wellbeing measures were maintained at 18-month follow up, strengthening the argument that gains were due to the improved coping strategies learned in the programme as the group meetings did not continue after the 12-week programme ended; although the research team did not know if any social relationships had developed outside the formal group. Final follow up median scores were also lower than baseline on the parenting scale. As well as the maintenance gains on the laxness subscale that were observed in the PALACES trial, continuing reductions were observed in all subscales of the measure. Although non-significant due to the sample size, the results are encouraging. Consideration must be given again to the fact that the measure is self-reported, however it could be regarded that behavioural observations of parent-child interaction helps in counteracting bias. Non-significant reductions were observed in negative parenting behaviours, which was encouraging. However, the frequencies of positive parenting behaviours also decreased between baseline and 18-month follow up. Although an increase in praise had been observed post intervention at 6-month follow up, reductions ensued at final follow up. Decreases were also seen in coaching and reflections at final follow up, which again could be explained by the fact that the children had limited speech for the parent to respond to. The DPICS is reliant on reciprocal parent-child interaction, however, some parents struggled to maintain the engagement of their child for the full 10 minutes of child-led play. It is well established that children on the autistic spectrum find it difficult to participate in shared imaginative play, which often results in solitary play behaviours. The characteristic communication difficulties exhibited by some children with ASD could influence the findings from the observations, as some children had limited language or were non-verbal. This could impact on the frequencies of reflections, due to the child not speaking, and could also be the reason for the increase in observed questions due to the parent's attempts to communicate with the child. The way in which the positive parenting subscale is coded may also have influenced findings, due to 'physical positive' and 'positive affect' being included as criteria. It is well-established that children with ASD can have sensory challenges and may not like to be touched, or communicative difficulties such as making eye-contact. This could explain the low frequency of positive parenting incidences during the observation and may suggest that an alternative observational tool would be more suited to this population due to such wide-ranging difficulties. Since many parents found engaging their child in child-led play for the full 10 minutes challenging, it could also be favourable for researchers to specify an activity for the dyad to participate in during the observation for future research. These changes could potentially reduce the amount of missing observational data which occurred during this study. There may also be an alternative observational measure more suitable for this population. Charman et al. (2021) recently employed a newly developed observational tool in a pilot RCT of a targeted parenting programme for children with ASD aged 4-8yrs, with positive results. The Observation Schedule for Children with Autism–Anxiety, Behaviour and Parenting (OSCA–ABP) (Palmer et al., 2021) assesses more relevant behaviours such as aggression, frustration, and destructive behaviour, and have specific tasks that the parent and child participate in during observation. This could also be an improvement as some parents chose diverse activities during the observations, some of which involved little or no interaction, or were difficult to code using the DPICS.

Promising improvements were observed with regards to child behaviour with the decrease in externalising difficulties observed at 6-months being maintained at final follow up. Further exploration of child behaviour outcomes showed that scores for both externalising and internalising subscales of the measure were significantly lower between baseline and long-term follow-up, and non-significantly lower between six and 18-months, indicating that child behaviour problems had continued to reduce following the intervention, which is promising. This could be attributed to the changes in parenting behaviours, such as better discipline strategies and the improvements which have been observed in parental mental health, known to be a risk factor for child problem behaviour. Child outcomes also included social communication problems. Baseline scores on the social communication questionnaire were relatively low among the sample, however they were above the cut-off of 15 used in the diagnostic process; therefore, there were no concerns that the measure was not suitable for the population. A decrease in social communication problems were observed between baseline and 18-month follow up, which is encouraging particularly considering that there were children in the sample who had significant language delays or were non-verbal. Only minimal improvements were seen on the adaptive skills measure (VABS; Sparrow, Balla, Cicchetti, Harrison, & Doll, 1984), with scores for both total adaptive, and socialisation subscales significantly lower at follow-up. There was also a non-significant reduction in communication subscale. The high prevalence of language and communication difficulties within the sample could account for minimal gains. There was a non-significant increase in the daily living skills scores. However, this is possibly due to developmental advances rather than the intervention; as the programme does not focus on improving the living skills assessed by this subscale such as feeding, toileting and self-care. It must also be remembered that impaired adaptive and social skills are key characteristics of ASD, thus intractable, therefore it may be ambitious to see gains from a relatively short parent-mediated programme. The measure was not distributed during the first follow-up visit at 6-months as no developmental advancements were expected during the brief time since baseline measures were completed. Again, baseline scores were indicative of clinical problems with 88% scoring below the clinical cut off, indicating the likelihood of co-occurring intellectual disabilities. However, this is difficult to determine as there was no formal measure of IQ, which is a limitation of the study. Nonetheless, the low baseline scores indicate that a large proportion of the sample may have had difficulties in functioning independently and acquiring adaptive skills at the expected rate over time. Given the evidence that ASD severity can influence the level of parental mental health problems, for future trials it could be useful to have a specific measure of ASD symptoms as well as the diagnostic status of the child included in baseline demographics in future studies so that this relationship could be examined. Overall, the findings from long-term follow up is positive, with improvements seen in parental mental health, self-reported parenting behaviours and child behaviour. The outcomes observed immediately post-intervention have been maintained without refresher or booster sessions. Although results should be interpreted with caution due to the small sample size, a further definitive trial can be justified, building upon the limitations of this study.

Strengths and limitations

This was a multicentre, pragmatic RCT trial which resulted in the evaluation of the intervention delivery within a real-world setting by existing services, therefore the population was representative of people living in real-life conditions. Both recruitment and programme delivery were conducted within existing services, and considerable effort was made to ensure that the programme was delivered with fidelity to maximise the reliability of the assessment of the programme. The need for long term evaluations of parenting programmes has been highlighted in previous literature and is important to show the real value of interventions. This is particularly true for newly developed programmes; therefore, this study makes an important contribution to exploring meaningful gains for families of children with ASD and society.

The trial has some limitations. Notably, the sample size was small and not powered for an evaluation of programme effectiveness. There was also no definitive way of knowing whether parents had received any other intervention or parenting support prior to 18 follow up, as the inclusion criteria in the trial protocol requested that parents did not complete any other programmes during the intervention phase only. The absence of a control group at long term follow-up is also a limitation; however, this would be difficult to rectify given the ethical considerations of restricting the support that parents could seek during the extensive period of time between intervention and long term follow up. The outcome measures were predominantly parent-reported, and it is well-established from previous evaluations of similar interventions that parental expectations of treatment outcome can bias responses (Hutchings et al., 2007). Although the use of both self-report and behavioural observations is a strength it could be argued that the observational measure adopted for the study was unsuitable for this population. However, all parent-report outcome measures were validated, reliable tools that had been used extensively in previous parent training evaluation studies, and with the ASD population. There was minimal missing data on the questionnaires, suggesting that generally, they were found to be acceptable by parents.

The improvements in child behaviour, parental stress and depression, and ineffective parenting style were maintained 18-month follow-up, which although not significant, is encouraging evidence that this programme can be beneficial to families of children with ASD. However, further research with a larger sample is needed to be conclusive about programme effectiveness. Future longitudinal studies, along with long term health economics evaluations could also examine whether any gains are maintained further into adolescence and adulthood.

Implications

- This is the first long term evaluation of the IY Autism Spectrum and Language Delays programme, demonstrating the possibility of engaging this population into a longer-term trial, with good retention at 18-months.
- Findings from long-term evaluations can be helpful in identifying the need to deliver booster or additional sessions, or to assess any additional support required. This could be particularly important to consider when supporting parents of children with autism as it is a lifelong disorder, and the unique and diverse challenges they are faced with.
- The fact that some of the positive intervention effects were maintained 18 months after the end of intervention, even in the absence of continuing contact or booster sessions, is promising and warrants including long term assessment in future definitive trials.
- Assessments of long-term benefits of programmes are important to inform policy deciders of the possible long-term outcomes of programme, particularly alongside any future evaluations of cost-effectiveness, so that policy deciders can calculate the long-term investment benefits of the programme.

Chapter 8

Benefits of the programme for siblings

Introduction

Autism is a life-long condition characterised by repetitive behaviours and interests, and difficulties in verbal and non-verbal communication and social interaction (American Psychiatric Association, 2013) that can have a profound impact on the family unit (Petrou et al., 2018). It is well documented that parents of children with autism experience higher levels of psychological distress than those of typically developing (TD) children (Schnabel et al., 2020), however increasing attention is being drawn to the wider impact of autism spectrum disorder (ASD) on the well-being of other family members. Sibling interactions play an important role in children’s early development (Bontinck, Warreyn, Van der Paelt, Demurie & Roeyers, 2018). Within the family system, siblings are usually regarded as being a source of companionship, emotional support and first social interactions (McHale, Updegraff & Whiteman, 2012; Smorti & Ponti, 2018). However social communication and establishing relationships can be a core challenge for children with ASD meaning that they may not communicate in the conventional way (Roemer, 2021). Children who have a sibling with ASD are at increased risk of socioemotional and mental health difficulties (Cassel, Messinger, Ibanez, Haltigan, Acosta & Buchman, 2007; Griffith, Hastings & Petalas, 2014), and are up to four times more likely to exhibit language and developmental delay in comparison to the general population (Marrus et al., 2018; Green, 2013). Findings are mixed with regards to different risk factors that contribute towards maladjustment within families of children with ASD and have been attributed to genetic and/or environmental factors. Siblings of children with ASD are known to be at increased risk of broader autism phenotype (BAP), meaning they themselves could be demonstrating traits resembling autism, but at a milder, sub-clinical level (Petalas et al., 2012; Pisula & Ziegart-Sadowska, 2015). Environmental factors associated with psychosocial dynamics within the family include the level of parental well-being (Quintero & McIntyre, 2010), the severity of symptoms that the child diagnosed with ASD exhibits (Pilowsky, Yirmiya, Doppelt, Gross-Tsur & Shalev, 2004), and the sibling's understanding of the disorder (Coffman, Kelso, Antezana, Braconnier, Richey & Wolf, 2021). However, the main focus of the review of literature for this study is the impact of ASD on sibling relationships.

Children with autism and their siblings reportedly spend less time with each other than typical developing dyads (Jones et al., 2019), with less intimacy and warmth observed than in other sibling interactions (McHale, Updegraff & Feinberg, 2016). The reciprocal nature of social play and conversation can be challenging for children with ASD resulting in more negative interactions with siblings and poorer quality relationships (Petalas et al., 2012; Jones et al., 2019). Although there is less conflict between children with ASD and their siblings, compared to those with other intellectual disabilities (Hastings & Petalas, 2014), siblings of children with ASD often report the bluntness or disregard shown towards them as being upsetting (O'Brien, Cuskelly & Slaughter, 2020), which can impact negatively on their mental health over time. Children with autism often engage in aggressive behaviours (Green, 2013), and frustrations due to poor social and communication skills are frequently an antecedent to problems including tantrums, aggression and self-injury (Kaminsky & Dewey, 2002; Jones et al., 2019), which can cause great distress to observing siblings (Ferraioli & Harris, 2009). There is also an increased likelihood that typically developing siblings may imitate these maladaptive behaviours through social learning (Tomeny, Barry & Bader, 2012; Jones et al., 2019), putting an extra burden on parents. Negative sibling and parent interaction are also frequently reported (Orsmond & Seltzer, 2007), and have been associated with attention seeking behaviour due to feeling overlooked within the family (Verté, Roeyers & Buysse, 2003), and the perception of parental favouritism towards the diagnosed child (Tsao et al., 2012). Sibling behaviour problems can also manifest in higher levels of attentional problems (Tsao, Davenport & Schmiege, 2012). A review by Giallo and colleagues (2012) reported that between 20-30% of siblings of children with ASD met clinically high levels of hyperactivity, as well as for overall difficulties and conduct problems on a parent-reported measure of child strengths and difficulties (Giallo, Gavidia-payne, Minett & Kapoor, 2012). However, there is conflicting evidence, and a recent study of the association between the characteristics of autism and sibling adjustment showed no significant differences in levels of externalising behaviour and/or social problems between siblings of children with ASD and the control group (Rixon, Hastings, Kovshoff & Bailey, 2021). Measures of sibling relationship and the sibling's strengths and difficulties (Buhrmester & Furman, 1990; Goodman, 1997) were completed by both parents and siblings, thus gathering separate responses as a comparison was a strength. Findings echoed those previously observed by Hastings and Petalas (2014), who reported no differences in externalising behaviour problems. Conclusions emphasised that interpretation of findings should be done with caution due to the relatively small sample size. This is an important consideration for any study but may be of upmost importance when conducting research with the ASD population given the idiosyncratic nature of the disorder, and the individual circumstances of families. A significant increase was observed in the peer problems subscale of the Strengths and Difficulties measure (Hastings & Petalas, 2014), and despite reservations about the reliability of sample size, conclusions about the increased risk of internalising behaviours are less equivocal than that of externalising problems since these findings are consistently replicated (Lovell & Wetherell, 2016; Macks & Reeve, 2007; Petalas, Hastings, Nash, Lloyd & Dowey, 2009; Thomas, Reddy & Sagar, 2015).

The emotional problems experienced by families of children with autism are often classed as being the most distressing factor in raising a child with autism (Petrou et al., 2018; Leedham, Thompson & Freeth, 2020). Frequently described sibling problems include higher prevalence of depression and anxiety (Lovell & Wetherell, 2016), which can be linked to feeling isolated and lonely (Thomas, Reddy & Sagar, 2015). Because children with ASD often have severe social deficits, this can often lead to little or no social contacts outside the family (Nealy, O'Hare, Powers & Swick, 2012) which impacts negatively on family members, such as in restricted life experiences (Angell, Meadan & Stoner, 2012), and a poorer quality of life (Jones et al., 2019). Children with autism often engage in behaviours such as obsessive ritualistic behaviours and accommodating their inflexible daily routines can be challenging for siblings (Cridland, Jones, Magee & Caputi, 2014), and affect the family climate as a whole (Petalas et al., 2012). The lack of socialisation that siblings may experience can affect how they form relationships with others. Siblings of children with ASD are not only more likely to experience peer problems, but also at greater risk of being bullied, and of others mocking their siblings with ASD. Children may encounter negative reactions to their sibling from the public and their peers (Thomas, Reddy & Sagar, 2015), and report increased feelings of embarrassment about their sibling (Orsmond & Seltzer, 2007) which impacts upon their interaction with others.

Having a sibling with ASD is not always difficult or negative. Outcomes such as increased pro-social behaviour and greater empathy for others are frequently reported (Orm, Haukeland, Vatne, Silverman & Fjermestad, 2022; Pilowsky et al., 2004; Walton, 2016). It is well documented that prosocial behaviour in children can be an important protective factor against future mental health problems (Orm et al., 2022), and promotes general well-being, academic attainment and peer acceptance (Memmott-Elison, Holmgren, Padilla-Walker & Hawkins, 2020). Siblings of children with ASD therefore may be more accepting of their siblings, but also of others in general. Acceptance of their sibling’s condition was identified as an important protective factor against internalising problems along with time spent alone with parents, interaction with other siblings and a supportive and inclusive environment (Leedham, Thompson & Freeth, 2020). Understanding protective factors and the bidirectional nature of family relationships is important when evaluating interventions for positive family outcomes. For instance, increased empathy although positive, can also increase the risk of sibling internalising behaviour due to their worries about their sibling, and the effect of the disorder on other family members. Siblings may worry about the increased stress and burden their parents may be experiencing (Karst & Van Hecke, 2012), and older children may have worries about the future care needs of their sibling, and their role as potential caregivers (Heller & Arnold, 2010). Evidence suggests that siblings of children with ASD are more aware of parental distress and there is increasing evidence to suggest that parent focused interventions targeting children with behavioural difficulties could benefit siblings due to their effectiveness in improving parental mental health (Hutchings et al., 2007; McGilloway et al., 2009).

From a family systems perspective, something which impacts upon one individual can have a pervasive effect on others. It is well established that parenting skills and positive parent-child relationships contribute to children becoming well-adjusted (Barlow et al., 2016), and that teaching effective parenting strategies is a protective factor against child mental health and behaviour problems (Williams & Hutchings, 2018; Webster-Stratton & Reid, 2018). It is also well-documented that parents of children with ASD are more likely to have lower levels of parental limit-setting (Boonen et al., 2015), parenting self-efficacy (Rezendes & Scarpa, 2011) and higher levels of stress (Salomone et al., 2018) and may benefit greatly from interventions to improve parenting skills. Behaviourally based parent training programmes have long been recommended as both treatment and prevention not only for child conduct problems, but also for the challenging behaviour associated with ASD (National Institute for Health and Care Excellence [NICE], 2015). Until recently attention has focused on the effectiveness of such programmes solely for the parent and referred child (Tsao et al., 2012), however, there is increasing evidence showing that, for children with conduct problems, benefits can also impact siblings (McGilloway et al., 2009). In their RCT of the Incredible Years Basic® programme with children at risk of child behaviour problems, Hutchings et al. (2007) administered the Eyberg Child Behaviour Inventory [ECBI] (Eyberg & Pincus, 1999) to both the index child and the sibling closest to them in age. Parents from the intervention group not only reported significant reductions in behaviour problems for the index child at follow-up, but also in the perceived intensity of problems with siblings. Findings were similar in a RCT conducted by Gardner and colleagues (2006) with a combination of the ECBI and direct observations showing significant intervention effects. However, instead of child closest in age, the parent completed the ECBI with the child they saw to be most difficult in mind. Although the sample size was small, results were deemed encouraging and strongly suggested that more than one child per family could benefit from a group parenting intervention (Gardner et al., 2006). A possible critique, however, is that the ECBI is a problem focused questionnaire, and when considering family focussed research with parents of children with intellectual disabilities and/or neurodevelopmental disorders, this should be an important consideration. Increasing emphasis is being placed on the language used in research involving these populations (Hastings, 2003), stipulating that both capabilities and concerns are regarded (APA). It may therefore be more appropriate to consider a measure consisting of more positively framed questions that assess strengths as well as problems when designing research involving children with ASD and their families. With the rapid increase in prevalence over the past decades (Adak & Halder, 2017), and the impact of ASD on families becoming more apparent, it is more important than ever to address the need for family-focused early interventions to support this population and to explore their possible wider-reaching benefits. Dababnah and Parish (2016) conducted an acceptability study of the Incredible Years Basic® programme; one of the most well established and researched behavioural parenting programmes (Bywater et al., 2011), with parents of children with ASD. Minor amendments were made to the programme to make it more relatable, such as increased focus on improving communication, and more emphasis on coping skills for parents under stress. Although measures were only completed to assess parent outcomes, qualitative interviews were conducted to gain information on family dynamics. Many parents indicated that they were able to use the programme strategies with their other children and reported reduced sibling problems following the programme. Exploring the wider benefits of the programme during exit interviews was a definite strength of the study and adds to the pre-existing evidence for the programme, however formal measures administered to siblings would give a clearer indication of its benefits. The study concluded that although the programme seemed acceptable, there is a need for evidence-based programmes exclusively for parents raising children with ASD, and for further evaluations of their effectiveness (Dababnah & Parish, 2016).

The Incredible Years Autism Spectrum and Language Delays programme (IY-ASLD) is a recently developed addition to the suite of Incredible Years (IY) programmes. It follows the same core components as the IY Basic programme but was developed specifically for parents of young children with autism and language delay with increased focus on communication, emotional regulation, and promoting friendship skills and empathy through pretend play. The PALACES (Parenting for Autism, Language and Communication Evaluation Study) trial was designed to explore the acceptability of the programme by examining recruitment and retention, implementation fidelity and parent and facilitator satisfaction. Although the main aim of the trial was to explore the feasibility of delivering the programme within existing services (See further details about the trial in Chapter 4: Design and Methods), measures of child behaviour and parental wellbeing were administered to examine exploratory outcomes of effectiveness. Due to the considerable literature suggesting a greater risk of psychosocial maladjustment among siblings of children with autism and the previous evidence of sibling benefits following parent training programmes (Gardner et al, 2006; Dababnah & Parish, 2016), it was decided to also administer sibling measures. Given the wider reaching impact of autism on the family it is important to examine any additional familial benefits, thus useful to examine the possibility of including sibling measures in evaluations of the programme. The feasibility of gathering sibling data in a pragmatic trial was therefore tested in this study, using a well-established measure previously used with this population. Not only was the Strengths and Difficulties Questionnaire thought suitable due to its widespread application withing the research area, but as previous studies have shown (Petalas & Hastings, 2014; Rixon, Hastings, Kovshoff & Bailey, 2021), the measure assesses many of the frequently reported emotional and behavioural problems such as hyperactivity and difficulties with peers, as well as positive strengths such as prosocial behaviours. It was hypothesised that parents would find it acceptable to complete the sibling measure, and that improvements would be observed in the emotional and behavioural symptoms of other children in the families of parents who attended the IY-ASLD programme. The objectives of this study were therefore to a) assess the feasibility of gathering sibling data in a pragmatic trial of a parenting intervention, and b) to explore any improvements in the adjustment of siblings among the intervention group.

Method

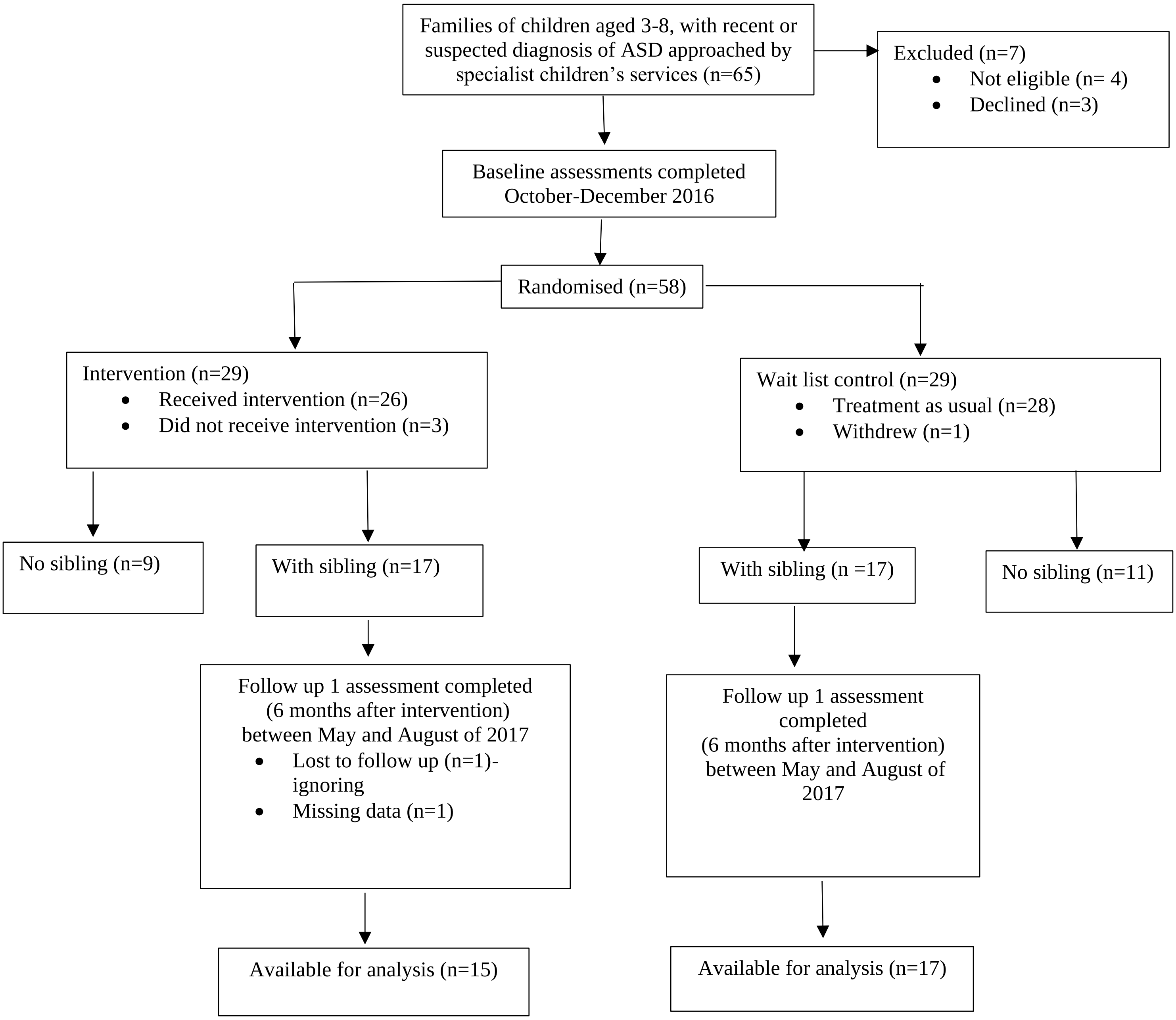
Please see Chapter 4: Methods and Design for the full details of trial methods, and further information on the intervention.

Participants

Trial participants were parents of children aged 3-8 years, with a recent or strongly suspected diagnosis of ASD. Participants for this study were drawn from the full trial sample and consisted of parents from both intervention and control groups who had multiple children (See Chapter 4 for full details). Parents were asked about the number of other children in the family, their age and gender. Seventeen families in each group had multiple children. Sibling measures were completed about the sibling closest in age to the index child.

The flow of participants included in this study are illustrated below.

Fig 8.1. Flow of participants through the study



Measures

Outcome measure- Sibling Strengths and Difficulties ***(See Appendix H)***

The behavioural and emotional difficulties of the sibling were assessed using The Strengths and Difficulties Questionnaire (Goodman, 1997), a globally recognised 25-item behavioural screening inventory for child psychopathology. Individual items (e.g., ‘rather solitary, tends to play alone’, ‘generally obedient, usually does what adults request’) are scored with a choice between three responses (0 = ‘not true’, 1 = ‘somewhat true’, or 2 = ‘certainly true’). The measure consists of four problem focused subscales; emotional problems, peer problems, hyperactivity and conduct problems; and a pro-social behaviour scale. Results are calculated by summing scores on relevant items for the five subscales and a total problem score is the summation of the four problem scales. Questions 7, 11, 14, 21, and 25 are reverse scored prior to analysis. Strengths are identified as a result of high scores on the pro-social scale. Difficulties are ascertained by a high total score on the individual problem subscales and the summation of scores on the remaining four problem sub-scales. An overall score can also be calculated for internalising behaviour with the summation of scores from the emotional and peer problems subscales; and for externalising behaviour by summing scores for the hyperactivity and conduct problems subscales (cut off thresholds for each subscale are shown below in Fig.2). The SDQ is a well-established, standardised measure of behaviour used in many studies of conduct disorder (Hutchings et al, 2007; Jones et al, 2008; Hutchings et al, 2011), and has previously been used to examine adjustment in siblings of children with autism (Cebula, 2012; Petalas et al., 2012). Two validated versions of the questionnaire were used depending on the child’s age. There is a version for children aged 4-18, and a slightly modified version for children aged 2-4, whereby an item on reflectiveness *(Q.21)* is softened *(‘thinks things out before acting’/’can stop and think before acting’)*, and two items on antisocial behaviour *(Q18 & Q22)* are replaced by items on oppositionality *(‘often lies or cheats’/ ‘often argumentative with adults’ & ‘steals from home, school or elsewhere/ ‘can be spiteful to others’).* The measure exhibits good internal consistency for all subscales ranging from *(α = 0.71 to .82),* and high overall reliability *(α = .88).*

Table 8.1. Cut off thresholds for each subscale of the SDQ

SDQ Subscale	**Close to Average (80% of population)**	**Slightly raised (/lowered) (10% of population)**	**High (/low) (5% of population)**	**Very high (/very low) (5% of population)**
Emotional problems	0-3	4	5-6	7-10
Conduct problems	0-2	3	4-5	6-10
Hyperactivity	0-5	6-7	8	9-10
Peer problems	0-2	3	4	5-10
Prosocial	8-10	7	6	0-5
Externalising	0-7	8-10	11-13	14-20
Internalising	0-3	4-7	8-10	11-20
Total difficulties	0-13	14-16	17-19	20-40

Data analysis

Differences in baseline characteristics between conditions were assessed with an analysis of variance (ANOVA) of all categorical variables including mean ages of parents and children, levels of parent stress and child behaviour problems *(see Table 1).* Differences in nominal variables (e.g., gender, diagnostic status) were assessed using a Chi-square test. A non-parametric alternative (Mann-Whitney) was used to assess differences in the BDI data which were not normally distributed. Means and standard deviations for each subscale were calculated for the whole sample. The subscale categories were collapsed from four (*see Fig 2.*) to two (close to average and slightly raised = low; high and very high = high) for reporting due to small sample size (Emotional, Conduct, Hyperactivity, Peer, Prosocial, Externalising, Internalising and Total difficulties). As assumptions for normality were not met, a Mann-Whitney test was used to analyse post-intervention outcomes for each condition. Missing data were dealt with based on the guidelines in the questionnaire manual for the SDQ (Goodman, 1997). If more than two items were missing on a subscale, it would be deemed invalid. If two or less items were missing on subscale, missing responses were prorated as ‘0’. There were two complete sets of missing data from intervention group in post-intervention follow up (therefore available for post-intervention- analysis: intervention (n=15); control (n=17)). There were no individual item responses missing. Outcome effect size *(r)* was calculated by converting z-scores (r = z-scores / total of observations), with interpretation based on Cohen’s d (0.1-small, 0.3-medium, 0.5-large).

Results

Sample characteristics

Children in the whole sample (*n* = 34) were mostly male (55%, *n* = 18), with a mean age of just under 6 and a half years old (*M* = 6.47 yrs) at baseline *(see Table 1)*. They were the siblings of children referred to the initial PALACES trial, of whom 85% (*n* = 29) had a confirmed diagnosis of ASD. The majority of the referred (index) children were male (65%, *n* = 22), with a mean age of 5 and a half at baseline (*M* = 70.74 months). Most of the referred (index) children had elevated child behaviour problems (68%, *n* = 23), and were scoring below the standard score of 70 on the VABS adaptive scale (91%, *n* = 31), and above the cut off of 15 for the SCQ (88%, *n* = 30) indicating a high prevalence of communication difficulties. Participating parents were predominantly female (88%, *n* = 30) with a mean age of 36 years. The majority were reporting elevated levels of stress (71%, *n* = 24), with a minority scoring between 14-28 (mild to moderate) on the depression inventory at baseline (12%, *n* = 4). Over half (56%, *n* = 19) had left full time education before the age of 17, and the majority were 20 years of age or older at the birth of their first child (88%, *n* = 30). There was employment in 76% of households. Most parents (91%, *n* = 31) were married or co-habiting, and 32% (*n* = 11) had more children in addition to the index child and sibling included in this study. There were no significant differences between intervention and control groups at baseline.

Table 8:2. Baseline characteristics of whole sample, showing mean scores and standard deviations for complete sample, and differences between control and intervention group

	n	ALL		TAU, WL Control		Intervention	*p*
Child Age (years) *Sibling, M (SD)*	*34*	6.47 *(2.84)*	*17*	6.89 *(2.80)*	*17*	6.06 (2.90)	.406
Child Gender (sibling)	*33*		*16*		*17*		
Male, n (%)		18 *(55)*		7 *(44)*		11(65)	.376
Female, n (%)		15 *(45)*		9 *(56)*		6 (35)	
Child Age (months) *Index, M (SD)*	*34*	70.74 *(15.09)*	*17*	69.59 *(15.26)*	*17*	71.88 (15.30)	.664
Child gender (index)	*34*		*17*		*17*		
Male, n (%)		22 *(65)*		11 (65)		11(65)	
Female, n (5)		12 *(35)*		6 (35)		6 (35)	.640
Diagnostic status	34		*17*		*17*		
ASD, n (%)		29 *(85)*		14(82)		15(88)	
Suspected, n (%)		5 *(15)*		3(18)		2(12)	.500
Child behaviour, communication and adaptive skills (index child)	34		*17*		*17*		
CBCL, M (SD)		68.32 *(9.92)*					
T > 63, n (%)		23 *(68)*		11*(65)*		12 *(70)*	
T ≤ 63, n (%)		11 *(32)*		6 *(35)*		5 *(30)*	.165
SCQ, M (SD)		22.82 *(6.78)*					
<15, n (%)		4 *(12)*		4 *(24)*		0 *(0)*	
≥15, n (%)		30 *(88)*		13 *(76)*		17 *(100)*	.166
VABS, *M (SD)*		57.16 *(8.38)*					
< 70, M (SD)		31*(91)*		16 (94)		15 *(88)*	
≥ 70, M (SD*)*		3 *(9)*		1 (6)		2 *(12)*	.779
Parent Age, *M (SD)*	34	36.00 *(6.44)*	*17*	35.76 (6.40)	*17*	36.24 (6.66)	.835
Parent Gender	34		*17*		*17*		
Male, n (%)		4 *(12)*		1(6)		3(18)	
Female, n (%)		*30 (88)*		16 (94)		14 (82)	.301
Marital status	34		*17*		*17*		
Single parent, n (%)		3 (9)		1 (6)		2 (12)	
Married/cohab, n (%)		31 (91)		16 (94)		15 (88)	.257
Living in poverty	34		*17*		*17*		
Yes		22 *(65)*		`10 *(59)*		12 *(71)*	.190
No		12 *(35)*		7 *(41)*		*5 (29)*	
Number of children in household	34		*17*		*17*		
Index and sibling only, n (%)		23 *(68)*		14 (82)		9 (53)	
More than 2 children, n (%)		11 *(32)*		3 (18)		8 (47)	.252
Parent age at birth of 1st child, M (SD)	34	26.68 *(5.93)*	*17*	28.18 (6.40)	*17*	25.18 (5.16)	
<20 yrs at birth of 1st child		*5 (15)*		3(18)		2 (12)	
≥20 yrs at birth of 1st child		*29 (85)*		14 (82)		15 (88)	.143
Education	34		*17*		*17*		
< 17 yrs left school, n (%)		19 *(56)*		10 (59)		9 (53)	
≥17 yrs left school, n (%)		15 *(44)*		7 (41)		8 (47)	.841
Unemployment	34		*17*		*17*		
No employment in household, n (%)		7 *(20)*		5 (29)		3 (18)	
Employment in household, n (%)		27 *(80)*		12 *(71)*		14 *(82)*	.605
Parental Wellbeing	34		*17*		*17*		
PSI		96.68 *(19.02)*					
<90, n (%)		10 *(29)*		6 *(35)*		4 *(24)*	
≥90, n (%)		*24 (71)*		11*(65)*		13 *(76)*	.590

BDI,	32	9.38 *(8.73)*	*16*		*16*		
<20 (minimal-mild), n (%)		28 *(88)*		15 *(94)*		13 *(81)*	.254
≥20 (moderate-severe), n (%)		4 *(12)*		1 *(6)*		3 *(19)*	

Sibling Strengths and Difficulties

Baseline. Median scores for the whole sample were calculated for each subscale. The median score for the emotional subscale was 2, with scores ranging from 0-8. A minority (18%, *n* = 6) of siblings scored within the high/very high threshold for the emotional problems subscale (between 5-10), with the remaining 82% (*n* = 28) scoring between 0-4 falling into the close to average/slightly raised category. The same median score was calculated for the conduct problems subscale (*Mdn* = 2) with scores again ranging between 0-8. Ten siblings (29%) scored in the high/ very high threshold for the subscale, with the majority of siblings (71%, *n* = 24) scoring within the close to average/slightly raised category. The median score for the hyperactivity subscale was slightly higher at 4, with scores ranging between 0-10. A minority of 8 (24%) scored within the high/very high threshold, and the remaining 76% (*n* = 26) falling into the close to average category. The median score for peer problems was 0 (range 0-7), with 88% (*n* = 33) scoring falling into the close to average category. The externalising category is a summation of the conduct and hyperactivity scores. The median score was 6, with scores ranging from 0-17. A minority of 35% (*n* = 12) fell into the high/very high category for externalising problems. The internalising subscale is a summation of scores for the emotional and the peer problems subscales. The majority of the sample (85%, *n* = 29) scored between 0-7, falling into the close to average category. The median score was 4, ranging from 0-19. Total difficulties were calculated by adding all the scores from the problem subscales together. A minority of 24% (*n* = 8) scored in the high/ very high category. Prosocial median scores were at the top end of the close to average category (*Mdn* = 9, range 3-10), with a minority of 18% (*n* = 6) falling in to the low/very low category for prosocial skills (see Table 8.2).

Table 8:3 SDQ subscale scores for whole sample at baseline (n = 34)

SDQ Subscale and cut off scores	**Median *(min-max)***	***N (%)*** ***(n=34)***
Emotional	2 *(0-8)*	
High/ very high *(5-10)*		6 *(18)*
Close to average/ slightly raised *(0-4)*		28 *(82)*
Conduct	*2 (0-8)*	
High/ very high *(4-10)*		10 *(29)*
Close to average/ slightly raised *(0-3)*		24 *(71)*
Hyperactivity	4 *(0-10)*	
High/ very high *(8-10)*		8 *(24)*
Close to average/ slightly raised (0-7)		26 *(76)*
Peer	0 *(0-7)*	
High/ very high *(4-10)*		4 *(12)*
Close to average/ slightly raised *(0-3)*		30 *(88)*
Prosocial	9 *(3-10)*	
Close to average/ slightly lowered *(7-10)*		28 *(82)*
Low/ very low *(0-6)*		6 *(18)*
Externalising	6 *(0-17)*	
High/ very high *(11-20)*		12 *(35)*
Close to average/ slightly raised *(0-10)*		*22 (65)*
Internalising	*4 (0-19)*	
High /very high (8-20)		5 *(15)*
Close to average/ slightly raised *(0-7)*		29 *(85)*
Total	9.5 *(0-23)*	
High/ very high *(17-40)*		8 *(24)*
Close to average/ slightly raised *(0-16)*		22 *(65)*

Pre-post changes in sibling SDQ. There were no significant differences between control and intervention group scores on any of the subscales of the SDQ at follow up (see Table 3.)

Table 8:4 Post-intervention outcomes. Mean scores of SDQ subscales for control and intervention groups pre- and post-intervention (Baseline and 6-month follow up)

	Control		**Intervention**		
	BL (n=17)	**FU (n=17)**	**BL (n=17)**	**FU (n=15)**	**FU**
Subscales	*Mdn, (min-max)*	*Mdn, (min-max)*	*Mdn (min-max)*	*Mdn (min-max)*	*U (z, p, r)*
Emotional symptoms	3, *(0-8)*	3, *(0-9)*	2, *(0-5)*	3, *(0-10)*	122.00 (-.210, .852, -6.56)
Conduct problems	2, *(0-8)*	1, *(0-7)*	2, *(0-8)*	2, *(0-9)*	132.50 (.193, .852, 6.03)
Hyperactivity scale	5, *(0-10)*	5, *(0-10)*	3, *(0-10)*	3, *(0-10)*	100.00 (-1.05, .313, -0.03)
Peer problems	0, *(0-4)*	0, *(0-5)*	1, *(0-7)*	2, *(0-6)*	151.00 (.932, .390, 0.03)
Total difficulties	11, *(0-20)*	10, *(3-23)*	7, *(0-23)*	10, *(0-30)*	120.00 (-.284, .794, -8.88)
Internalising	8, *(0-20)*	4, *(0-10)*	7, *(0-19)*	5, *(0-16)*	130.50 (.114, .911, 3.56)
Externalising	8, *(0-13)*	5, *(0-15)*	4, *(0-17)*	4, *(0-16)*	110.50 (-.645, .526, -0.02)
Prosocial	9, *(4-10)*	9, *(6-10)*	9, *(3-10)*	9, *(5-10)*	127.50 (.000, 1.00, 0.00)

Discussion

This is the first study to explore the possible benefits of the newly developed Incredible Years Autism Spectrum and Language Delays (IY-ASLD) programme for siblings of children diagnosed with autism. Of the 58 families who participated in the PALACES trial (Williams, Hastings & Hutchings, 2020), 34 parents had multiple children, and were therefore asked to complete the Strengths and Difficulties Questionnaire (SDQ) with the sibling closest in age to the child diagnosed with ASD in mind. Seventeen had been randomised to the intervention group at the beginning of the trial, and coincidently the same into the control group (see Design and Methods chapter). The rationale for exploring sibling outcomes was firstly, the increased risk of psychosocial maladjustment within the population, and also that previous trials of parenting interventions have reported benefits for children other than the index child (Gardner, 2006, Dababnah & Parish, 2016). Not only was the feasibility of gathering sibling data tested in this study, but preliminary outcomes were also explored with a view of informing the design of future trials.

The siblings involved in the study were just over six years old, with no significant difference in gender distribution. However, the majority of index children were male, corresponding to the extensive evidence of higher prevalence of ASD among boys. Only five of the index children were under ongoing assessment, with most of the sample in receipt of a confirmed diagnosis prior to enrolment onto the trial. The high proportion of diagnosis within the sample is corroborated by majority scores of below the diagnostic cut off for the Vineland Adaptive Behaviour Scale (VABS), and above the cut off for problems on the SCQ (Social Communication Questionnaire) in the sample at baseline. In addition to communicative and adaptive deficits, most children were scoring above the cut off on the Child Behaviour Checklist (CBCL), inferring behaviour problems among the index children. This was to be expected as behaviour problems are frequently associated with the disorder, however the mean was not significantly higher than the cut off score, suggesting that behaviour difficulties were moderate across the sample. Parents were mostly mothers, as is typical of parenting intervention research. The mean age for parents in the sample was 36 years, and a vast majority were married or co-habiting. A small minority had given birth to their first child before the age of 20, resulting in a low number of teenage parents among the sample. Just over half of the parents had left school before the age of 17. Although the majority of the sample were classed as living in poverty, there was employment in 80% of the households. This conflicts with the usual literature within this population that frequently cites high unemployment rates and greater loss of productivity among parents in this population, due to the many barriers that may prevent them from working (Buescher, Cidav, Knapp & Mandell, 2014). Families in the sample could therefore be experiencing financial hardship due to any additional services and out of pocket expenses required for their child's needs, even though they are in employment. The fact that most of these parents are in employment could be conducive to the small incidence of depression within this sample, as financial burden and social isolation are often presented as risk factors of the increased mental health difficulties experienced by parents in this population. Only five parents in the whole sample scored above the cut off on the depression scale at baseline, although a majority were reporting elevated levels of stress, corresponding to the literature. The measures used to gather baseline characteristics were well established, standardised measures previously shown to be acceptable by the sample (Williams, Hastings & Hutchings, 2020), which is a strength of the study.

The outcome measure was the Strengths and Difficulties questionnaire (SDQ), a standardised and widely used measure in the screening and assessment of behavioural problems and the evaluation of outcomes. Parents were instructed to complete the measure about the siblings closest in age to index children in the trial. Whole sample median scores were calculated to examine scores for each subscale of the measure (Emotional, Conduct, Hyperactivity, Peer problems, Total problems, Externalising, Internalising behaviour, Prosocial skills) at baseline. Contrary to the literature, whole sample scores for all problem subscales were low. The majority of siblings scored within the close to average/ slightly raised categories for each problem subscale, meaning they were scoring within the same banding as ninety percent of the general population. This means that the sample in this trial were not exhibiting maladjustment difficulties, conflicting with preponderant literature (Cassel et al., 2007; Griffith et al., 2014). However, the sample did show similar results to other studies in terms of high prosocial behaviour (Orm et al., 2022; Pilowsky et al., 2004; Walton, 2016). Prosocial scores are reversed on the measure, meaning that a higher score on the subscale is positive. A majority of the sample scored within the close to average/slightly lowered banding. The median score across the whole sample was 9, one less than the maximum possible score, therefore the sample was scoring highly on the subscale. The importance of prosocial behaviour during childhood is well documented. Acknowledging these skills as protective factors against psychosocial difficulties is important when understanding the sample, and it is also important to focus and commend the positive functioning of this population. No differences were observed between the intervention and control conditions post intervention; however, this is understandable given the absence of emotional and behavioural problems within the whole sample at baseline. The sample were not exhibiting maladjustment; therefore, it is not possible to detect any differences between groups, or improvements following the intervention. The sample may be too small to detect any differences, and a larger sample would be required for a definitive trial to ensure representation of population norms with regards to sibling adjustment. It is also possible that the Strength and Difficulties questionnaire itself does not reflect the difficulties experienced by siblings, as the measure is parent-reported and therefore may be subject to bias. Parents may be under-reporting problems due to either comparison to the child with ASD with regards to problem behaviour, and/or fear of judgment about their child’s behaviour problems. There is also evidence to suggest that siblings are at greater risk of maladjustment when the child with ASD has significant behavioural difficulties, which was not the case with the majority of diagnosed children in the trial.

Further aspects of family climate could have been assessed to better understand why the siblings from this sample differed from that expected from existing literature. For instance, the severity of ASD symptoms has been shown to influence the level of family psychosocial maladjustment. Although the VABS and SCQ were administered to assess the adaptive and communicative abilities of the index child, the nature and severity of problems are not easily identifiable with these measures. This could be significant to examine when considering the internalising problems of the sibling. For example, they may be more distressed by a sibling who exhibits aggressive or self-injurious behaviours in comparison to one that has severe mobility problems or is non-verbal. Symptom severity could also be an influencing factor for parental wellbeing, which could also in turn affect the sibling’s psychosocial adjustment, given the importance of the inter-relationship between the wellbeing of different members of the family. The lack of gauging symptom severity, or a formal measure of co-occurring intellectual disabilities, should therefore be noted as a limitation. Nevertheless, useful information was gathered in the demographic questionnaire that allows us to consider factors that may have influenced the sample and can inform future research. For example, a high proportion of parents were working, which is unusual for this population. The level of parental depression was also low in comparison to that typically reported for this population, as was the low incidence of single parents. It could be argued that these personal and social relationships provide support and reduce the feeling of isolation frequently reported by parents of children with ASD, emphasising the importance of social support as a protective factor against psychosocial maladjustment. This could also be said for the children in the sample. Child internalising problems reported in the literature, were often associated with isolation and loneliness, however only a minority of the sample had other siblings apart from the child with ASD. The low scores on the peer problems scale indicate the ability to form positive friendships and supportive social relationships outside the home, which could be a protective factor against internalising problems, explaining the low scores on this subscale. It would be of great interest to further examine how family demographics and environmental variables influence the severity of, or act as a protective factor against, sibling maladjustment. Odds ratio analyses could be performed to examine any trends; however, the sample was significantly under powered to perform this analysis. For future research, a much larger sample would be required in order to examine any relationships between having a sibling with ASD and any strengths and difficulties. As well as psychosocial maladjustment, it was identified in the literature that siblings of children with ASD are also at increased risk of language and developmental delay (Marrus et al., 2018; Green, 2013), however this was not examined in the current study. For future research, it may also therefore be beneficial to administer the VABS or the SCQ to participating siblings to assess their social communicative and adaptive skills, or a more specific measure of language delay to assess any baseline problems and changes due to intervention. This could be an added element in the evaluation of the transferrable benefits of the IY-ASLD programme to siblings, given the emphasis on language development strategies within the programme.

All measures used in the trial were validated, reliable tools that have been routinely used with parents of children with ASD and/or in other evaluations of parenting programmes. However, a notable limitation is that the only outcome measure was a parent-reported questionnaire which could be subject to bias (Hutchings et al., 2007), particularly due to the problem focused nature of the questions. A way of controlling for this in future research would be to also administer a teacher version of the SDQ. As well as providing an independent comparison, teachers may also be better placed to observe any difficulties, such as peer problems and inattention. Although there were no elevated levels of internalising behaviour among siblings in the group, it is important to consider the vast evidence for internalising problems within the literature. For future trials, it may be useful to consider the suitability of the SDQ as a measure of this. Scores for the internalising subscale are calculated with the summation the emotional and peer scales. There are only five general questions within the emotional subscale and if, as is the case with this sample, children do not have peer problems, it is possible for emotional problems to perhaps go undetected. It could therefore be beneficial to administer a more specific measure of child mental health in future research, such as the Child anxiety and depression scale [RCADS] (Chorpita, Moffitt & Gray, 2005). Direct observations could also be an addition in future trials as a further empirical measure, with researchers remaining blind to condition allocations for added robustness. Given the importance of family interactions in the socialisation of children, and as protective factors for maladjustment, this method of data collection could be a useful research tool. However, if direct observations were to be introduced as an additional measure, it would be important to map the observation activity carefully, due to the challenges some children with ASD experience with reciprocal play.

The main limitation remains to be the small sample size, and in order to conduct a definitive evaluation of the programme's benefits for siblings, further research is needed with a larger sample using the effect sizes from this trial. When calculating a sample size that is sufficiently powered to detect differences in the future, it is important to consider that the sample for this study was drawn from a larger sample in a RCT, but only included families with multiple children. This meant that the sample was drawn from an already small data set, which should be a consideration for future power calculations. It may also be necessary to consider the idiosyncratic nature of autism and the diverse differences that already exist within the population, which have been observed in this trial. It may be necessary therefore, to recruit an even greater sample than required for meaningful interpretation. The presence of a control group is a great strength of this trial, as is the pragmatic nature of the RCT and programme delivery. The sample was recruited from families already known to and delivered within existing services, therefore resulting in a real-world evaluation of the programme. Retention was high, with measures for all but one case gathered at follow up which is encouraging for future trials. There was also minimal missing data, allowing optimal analysis, and strongly suggests that the measures were acceptable to parents. This study contributes to the literature about the psychosocial adjustment of siblings of children with ASD, and the differences within the population. The inclusion of sibling data is also a useful element of the evaluation of the newly developed programme, and a valuable addition to the evidence base of the benefits of family interventions for this population.

Implications

- This is the first study to examine the sibling outcomes of the IY Autism Spectrum and Language Delays programme to explore its wider-reaching benefits to other family members.
- This research is timely, particularly in light of the rapid rise in the prevalence of ASD, and the increased awareness of the need to support families.
- The study has been useful in exploring the inconsistencies in related literature and differences in population and contributes to the literature surrounding the wider reaching impacts of ASD
- This study is a valuable contribution to the research into the acceptability and effectiveness of parenting programmes to improve sibling outcomes as well as referred children.

Chapter 9

General Discussion

Thesis outline and objectives

The aim of this thesis was to further examine the newly developed Incredible Years® Autism Spectrum and Language Delays (IY-ASLD) programme following the PALACES trial. The thesis builds on the multicentre, pragmatic randomised controlled trial (RCT) which examined the feasibility of delivering the programme to parents of children aged 3-8-years with Autism Spectrum Disorders (ASDs) within existing services. The main objectives of this thesis were to a) examine the economic impact of autism on families and public funded services and explore the existing economic evidence for similar parenting programmes in a systematic review; b) calculate accurate cost for programme delivery both during the trial and prospective roll-out to inform future research and prospective roll-out of the programme; c) examine the feasibility of retaining participants to gather longer-term data, and explore the long-term outcomes of the IY-ASLD® programme; d) test the feasibility of gathering sibling data, and assess the possible benefits of the IY-ASLD® programme for siblings, and e) reflect on key feasibility messages of novel studies to inform a future definitive trial.

The initial review of literature illustrated the impact that raising a child with ASD can have on parental mental health and wellbeing and on the psychosocial adjustment of other family members, particularly siblings. The economic impact on both families and public health services were recognised, highlighting how UK governmental agenda and treatment guidelines have been influenced with the increasing awareness of ASD. This background knowledge has contributed to the development of the studies for this thesis, the findings from which will inform the design and formulation of research questions for future research.

The growing body of evidence illustrated the many challenges that families in this population face, and the impact that having a child with autism can have not only on parents, but also on wider family members and the home environment. However, our sample demographic did not correspond to the literature in many ways. For instance, although most parents were reporting elevated levels of stress, which was expected given the disproportionately high prevalence of mental health problems reported within this population; the sample were not reporting high incidence of depression at baseline. Although over half the parents had left school before the age of 17, there was employment in more than three quarters of households, which is also contrary to the literature. The fact that most of these parents are in employment could be conducive to the small incidence of depression within this sample, as financial burden and social isolation are often presented as risk factors of the increased mental health difficulties experienced by parents in this population. Only five parents in the whole sample scored above the cut off on the depression scale at baseline, although a majority were reporting elevated levels of stress, corresponding to the literature. The majority of the sample were either married or co-habiting, opposing the literature, which typically reports an increased risk of relationship break-down and divorce among parents of children with ASD (Karst & Van Hecke, 2012). This could perhaps be an influence on the unexpectedly low number of parents with elevated depression, in that parents had the emotional support of a partner. The sample was small and perhaps not fully representative of population norms, therefore the differences in sample characteristics may affect the way the results from the studies are interpreted and compared to existing research.

Key findings from studies

Although there is increasing evidence for the effectiveness of parent programmes, little research was found to have been conducted into their cost-effectiveness. Economic evaluations are important to inform policy and practice decisions about which interventions to implement, at what cost and with what benefit. The systematic review searched the current literature, focusing on published economic evidence for parenting programmes specific to ASD. Only two relevant articles were selected for inclusion in the review, highlighting the dearth of evidence. Both were costings of programme delivery as opposed to economic evaluations, but were appraised in accordance with the Drummond et al., (1997) to illustrate the critical elements of a robust economic evaluation. Although many would argue that the inclusion of only two studies does not constitute a robust systematic review, a great deal was learned from doing so. Not only did the minimal numbers of relevant literature reflect the poor evidence base for the economic outcomes of ASD specific programmes, but also left wider scope to focus on the critical appraisal of the two studies and the interventions themselves in comparison to the IY-ASLD® programme. Methodological issues within the research were considered, resulting in a greater understanding of which standardised methods would be useful to ensure robust evaluations. These findings lay a foundation to the micro-costing chapter of this thesis, following the observation of a lack of standardised methods within the field which allow for empirical and replicable research. On reflection, a scoping review may have been less restrictive and would have enabled the inclusion of a greater variety of interventions specific to ASD. However, this thesis aimed to inform future trials, therefore it was important that the interventions identified were comparable in theoretical underpinning, dose and delivery format to that of the IY-ASDL®.

The micro-costing was conducted from a public sector, multi-agency perspective, which is a strength when costing complex interventions such as parenting programmes as there are numerous stakeholders. This was the first cost evaluation of the IY-ASLD® programme, providing a direct measure of accurate delivery costs. Overall costs were calculated for running the programme during the trial, with sensitivity analyses conducted to establish how costs are affected in different scenarios (e.g., increasing participant numbers, facilitators with lower salary grades), and to calculate future roll out costs of the programme in real-world settings. Costs were calculated in four contexts: Initial set up and delivery of the programme over the 12 weeks of the research trial, accounting for cost of training and purchase of materials etc. (£1771.67/per child with 5 parents per group). The cost during the research trial with additional home visits included when deemed necessary for certain families, e.g., in rural areas, or specific child health needs etc (1903.74/per child). The cost of programme delivery within normal services, with initial facilitator training already undertaken, and resources purchased and with lower paid facilitators (£854.42/per child). Finally, the roll-out costs were calculated accounting for proposed changes to programme protocol to extend the running time to 14 weeks (£1179.44/per child) as advised by the programme developer and based on leader and parent feedback. The accurate breakdown of costs in different delivery set-ups illustrate how useful a standardised costing framework can be with regards to projection and service planning. The cost of running the programme was unexpectedly lower than other Incredible Years group programmes of a similar length, which could be due to differences such as higher/lower utilisation of childcare facilities and catering costs, and a greater number of facilitators being trained in previous trials of other Incredible Years® programmes. Recruitment costs were also lower, due to families already having existing contact with the specialist children's service and were already know to the group facilitators. Strengths of the study included the use of a previously developed and standardised framework to ensure precise costing. Cost gathering methods were decided upon collaboratively between researchers and group facilitators, ensuring that all unexpected costs were considered, resulting in a rigorous process. Unlike other evaluations, this micro-costing included rollout costs based on predicted lower paid facilitators, giving a more pragmatic projection of costs. The micro-costing increases availability of economic information on parenting programmes for families of children with ASD, which is important given the financial impact of autism, and lack of current economic evaluations of targeted interventions for this population. Accurate and detailed cost data for new programmes is necessary to inform service managers and decision makers of costs before investment in intervention roll-out. Detailed cost information is also useful for the full economic costings required for grant applications and the design of future trials. Accurate costs gathered in this trial can be used alongside future larger evaluations of effectiveness, to calculate a cost effectiveness evaluation. Although the systematic review did not identify any full economic evaluations of similar ASD specific programmes for comparison, previous evaluations have reported other programmes from the IY® series as being a cost-effective preventative intervention; implying that the IY-ASLD® could also be cost effective, as it was found to be even less expensive to deliver. However, to fully assess cost effectiveness, definitive programme effectiveness and long-term follow ups would need to be conducted. Charles et al. (2011) suggested a need to follow-up families for 10 or 15 years to assess long-term clinical and cost effectiveness and allow population of wider economic models. Therefore, it is important to design research that provides longitudinal data, by exploring the long-term outcome of newly developed programmes in particular.

The thesis author identified the importance of longer-term evaluations of parenting interventions, and therefore conducted the first study to examine the longer-term outcomes of the IY-ASLD® programme. The aim of this study was not only to assess the feasibility of gathering longer term data from participating families, but also to investigate whether there were any longer-term outcomes from attending the IY-ASLD programme. Preliminary results showed promising gains during the PALACES trial, which were examined for maintenance over time in this study. Data was also explored to see whether any new benefits had emerged over time. Improvements to both parental stress and child behaviour had continued despite the provision of additional booster or refresher sessions, which is encouraging. Improvements were also observed in parental depression, despite the low incidence at baseline. This may pose the question that perhaps greater improvements would have been observed had there been increased levels among the sample and is a consideration for further research with regards to calculating power for future research. Although social communication problems were lower at 18 months than at baseline, gains were minimal, and unexpected reductions were observed on the adaptive skills subscale. It is important to consider that adaptive and communication difficulties are core traits of autism, and are therefore difficult to alter, particularly with such a short, parent-mediated intervention. More importantly, changing ASD patterns of behaviour is not the aim of the programme, but to improve parental skills and wellbeing and address collateral child difficulties. Short term evaluations of parent programmes are typical, however due to the life course of autism, and the unalterable behaviour traits that are associated, it is also useful to examine longer-term benefits of interventions, and this novel study contributes toward the important evidence base. The retention of intervention parents was good at 18-month follow up considering the period of time that had elapsed since baseline and is encouraging to note that parents were still willing to engage in the study long term. The parent reported gains observed at six months in child behaviour and the improvements to parental mental health were promising, and further indication that the programme could be beneficial for this population. It was emphasised however that all findings should be interpreted with caution due to the lack of power in the sample to detect differences.

The sample for the final study was smaller still, due to the participants being drawn from the already small data set to focus only on parents with multiple children, to conduct the first exploration of possible benefits to siblings of children diagnosed with ASD. The author designed the final study due to the growing evidence showing that siblings are at greater risk of psychological distress and behaviour problems than typically developing children (Lovell & Wetherell, 2016; Macks & Reeve, 2007). Similar evaluations of behavioural parent programmes have demonstrated observed benefits for siblings as secondary outcomes (Hutchings et al., 2007). The Strengths and Difficulties Questionnaire (Goodman, 1997) was chosen as the most appropriate measure due to its well-established use with this population, and relevance of variables being measured. Parents were asked to complete the SDQ about the sibling closest in age to the index child. However, as was suggested by Hastings and colleagues (2014), if there are multiple children, it may be more meaningful to consider the sibling with the most behavioural concerns rather than asking parents to complete the measure with the sibling closest in age to the index child in mind. No differences were observed in baseline characteristics between conditions. Whole sample scores at baseline were low, suggesting there were no significant emotional or behaviour challenges, again contrary to the literature. Median scores on the prosocial scale were high, as was expected from reviewed literature. No sibling differences were observed between the intervention and control groups between pre- and post-intervention. However, it is important to note that there were no elevated socioemotional problems in either sample at baseline, therefore there was no real opportunity or necessity for change. The sample size was small, which is the main limitation of the study. However, the RCT design of the study is a strength, as the presence of a control group enhances the internal validity of the study and has the potential to assess programme effectiveness more robustly. Using the SDQ could also be considered a strength due to its well-established use and global validity, although it’s parent-reported method could be subject to bias. A teacher version of the measure also exists, therefore for future research, this could be incorporated so that the child’s educator completes an independent measure. This was the first study to explore the potential benefits of the IY-ASLD® programme for siblings of children diagnosed with autism. The research is important given the dramatic rise in the prevalence of ASD, and the growing interest in the wide-ranging impact of autism on the family system. The studies conducted in this thesis are novel, and collectively make an important contribution to the evidence base for not only this newly developed programme, but also to the literature with regards to the usefulness of parent training programmes for this population.

Thesis strengths and limitations

The main limitation was the small sample size, and for the 18-month follow-up the absence of a control group for comparison at long-term follow-up. However, it would be unethical to restrict services including a potentially beneficial intervention to parents of children with a diagnosed condition. The emphasis placed on parent reported measures was also highlighted as a limitation due to the risk of bias, however the direct observations of parent-child interaction was identified as an advantage and could compensate for any potential bias when assessing changes in parent behaviour. The pragmatic RCT design of the trial is a strength of the study and is representative of real-world conditions as the sample were recruited and received the programme from staff within existing services. Both evaluations of effectiveness and economic implications are therefore true to real-world implementation. This was the first study to examine the costs of delivering the newly developed Incredible Years Autism Spectrum and Language Delays Programme, providing accurate costs for service managers and policy decision makers with regards to costs of implementation and roll-out of the programme within existing services. The costing is also useful for future economic evaluations of the programme, such as cost-effectiveness alongside larger definitive trials. The thesis also provides the first long term evaluation of the programme, by looking at data from 18-month follow ups. Short term evaluations are typical of parenting programmes, however establishing longer-term gains is important for possible cost benefit evaluations and with regards to the long-term impact of autism. Assessing any long-term benefits can possibly strengthen the support offered to families. The thesis also includes the first exploration of potential benefits the programme may have for other family members, therefore assessing the wider impact of the programme. Important lessons have been learned to inform future studies, for example of exploring benefits for partners in prospective studies. The feasibility trial was a RCT, thus the gold standard in evaluating the effectiveness of interventions. The exploration conducted therefore are done within pragmatic, ‘real world’ conditions, and although findings are minimal, they contribute to the programme’s evidence base, which is of particular importance with regards to newly developed programmes. The pragmatic design of the trial is also a great advantage as it examines feasibility, service delivery and its effectiveness within a real-world context. The programme was delivered by highly skilled facilitators, some of whom had previous experience of delivering other IY programmes, and all of whom of supporting parents of children with a diagnosis of autism and were therefore knowledgeable about their specific challenges. Although group leaders were highly skilled, all attended fortnightly supervision with a certified IY programme trainer which perhaps was the greatest strength with regards to ensuring that the programme was delivered as intended i.e., with fidelity. This also ensured they were able to share any concerns about the groups, without the involvement of the researchers who remained independent of programme delivery. Collaboration between the research team, group facilitators, and intervention providers was consistent throughout trial development in regular steering group meetings, ensuring a multidisciplinary approach to trial design. This also contributed towards minimising missing data with regards to developing measures such as cost diaries, resulting in an accurate and full account of delivery costs. Programme attendance was good, as was retention in the research trial, suggesting that both the programme and research methodology were acceptable to this population. Considerable effort was made throughout to ensure that the programme was delivered with fidelity to maximise the reliability of the programme, and its evaluation. This was done by ensuring all facilitators completed the accredited programme training pre-intervention. Adherence to the prescriptive manual was also ensured, along with the completion of detailed session checklists at the end of each session. Parents were also asked to complete feedback questionnaires at the end of the 12-week programme. Adding to validity, the intervention and research teams were independent of the programme development. Randomisation was undertaken independently by the North Wales Organisation for Randomised Trials in Health and Social Care (NWORTH), and researchers remained blind to condition allocation until six-month data collection had been completed, therefore a strength with regards to the sibling data. Parents from the intervention group were offered the programme immediately after follow-ups were complete, therefore limiting their wait time for support.

Despite their importance, longer term follow-up studies of parent programmes are rare. Long-term evaluations are important to understand behaviour changes over time and can reflect the real-world value of the intervention, therefore the study of longer-term outcomes included in this thesis is a useful examination of potential maintenance effects for the families in the intervention condition and a contribution to the evidence base for the programme. Exploring potential benefits for siblings is also a strength of this study, given the growing evidence of the impact of raising a child with autism on the psychosocial adjustment of wider family members, particularly siblings. The thesis also carries implications for public health, with the accurate micro-costing able to inform service managers and policy deciders about costs of future implementation of the programme, which is a strength. The exploration of health economic measures being administered alongside a pragmatic trial is valuable for future evaluations, as is the accurate costing of the programme, which can not only inform service mangers and policy deciders of cost before investment but can also identify tools for future cost-effectiveness analyses. The economic evaluations of new interventions following standardised methods are important for future implementation and roll-out of programmes.

The reliance on self-report measures could be considered a limitation, although all those employed were standardised and valid measures. It is well documented however, that self-report measures can be at risk of bias due to anticipation of effect (Hutchings et al., 2007: Furlong et al., 2012), although that is unavoidable when participants receive an intervention, as it is impossible for them to remain blind. Parents could also be reluctant to divulge personal feelings or information in self-report measures, particularly with regards to mental health questionnaires. Parental responses to measures of child behaviour and/or adaptive skills could also be subject to inaccurate or false answers, particularly if the questions in the measure are specific to certain behaviours, or are problem focused. The additional inclusion of the DPICS as a direct observational measure of parent-child interactions is a strength, however questions were raised with regards to the suitability of this measure, which could be considered a limitation, and area of improvement for future research. Some parents found engaging the child in a 10-minute parent-child interaction or activity challenging, therefore, only the first five minutes of observed frequencies were coded. It could also be argued that the categories of the DPICS are unsuitable for this population due to some of the categories, for example positive parenting, which included physical affect as a subcategory, and the disabilities of some children meant the categories were unsuitable as they were non-verbal for instance. It is well documented that up to 96% of children diagnosed with ASD can have sensory difficulties (Ben-Sasson, et al., 2019), therefore some of the sample may have been averse to physical touch, limiting the parent's opportunity to show positive parenting behaviours. The influence of ASD symptom severity on parental wellbeing outcomes was also reported in the literature review, therefore a lack of a formal measure of ASD severity could be considered a limitation. Although the VABS was administered as a measure of adaptive skills, a child could have physical impairments but no cognitive delay, and vice versa. IQ tests are regularly used to differentiate between low and high functioning autism in clinical assessments, therefore their use could be worthwhile in future research. Another possible limitation is the absence of a specific parental self-efficacy measure, to explore the relationship identified in many studies with parental wellbeing.

Future research

The present study has established strong foundations for future research. The most evident being the need for a larger definitive RCT, which would be better powered to assess effectiveness. The idiosyncrasies of ASD have been highlighted with regards to the broad spectrum of co-morbidities that can occur with autism, and the varied challenges that parents of children with a diagnosis of autism face. This could be an important consideration with regards to future implementation of the programme, so that it is delivered with adequate time spent on the additional support that parents in this population may require. The programme developer has made changes to the programme protocol with this in mind, suggesting that the programme should run for 14 weeks, to work through the content and allow adequate time for in-depth group discussion and support. The need for additional time was not observed during the trial, however the facilitators during the trial were highly experienced as both Incredible Years® trainers and autism practitioners, which may not be the case in future roll-out. Differences in family challenges, and child needs could also be an important consideration for future research, particularly with regards to calculating an adequate sample size to power effectiveness analyses in relation to different challenges. It could be argued that due to the diversity of co-morbidities and continuum of symptom severity, it would be harder to make comparisons within samples and between conditions. Therefore, a larger sample, including parents with a broad range of challenges and with specific challenge may be needed to detect meaningful outcomes. Some of the categories in the DPICS may also have been unsuitable for some of the participating children as there were several non-verbal children, therefore coding frequencies of reflections, for example, were not appropriate. Some parents also struggled to engage their child in the full 10-minutes of child-led play, therefore only the first 5 minutes had been coded, resulting in limited instances of the measured behaviours. Perhaps for future trials, giving the child a choice of prescribed activities would be more successful with regards to engagement, and easier to code than free play which varied between the dyads. There is also the overall question as to whether the DPICS is suitable for this population, considering the inclusion of coding physical affects, and social interactions such as eye contact and parent reflection. This has been questioned in other research, prompting the development of alternative measures which could be more suitable for future trials. The Observation Schedule for Children with Autism–Anxiety, Behaviour and Parenting (OSCA–ABP) (Palmer et al., 2021) was developed as an objective measure of child emotional and behavioural problems to supplement parent reported outcomes in similar research. The variables coded are perhaps more relevant to children with autism than the reciprocal interactions observed in the DPICS, with frequencies of aggression, irritability, hyperactivity an anxiety coded instead. Parent behaviours of laxness and over-reactivity are also observed, which may negate the need for a separate parenting scale in future trials and make it easier to find relationship trends between parenting practices and child behaviour. Thus far, the newly developed measure has demonstrated good variance and sensitivity to change, with reliable coding of both parent and child behaviours reported. For a recent trial, 83 parents participated, with the sample size adequate to achieve 80% power. This could therefore be a favourable alternative for future research and help inform a power calculation of adequate sample size for a larger definitive trail. With regards to measures, the remainder were deemed reliable and acceptable to parents, therefore those that demonstrated promising improvement but with small effect sizes, such as the Beck Depression Inventory, could also help determine an appropriate sample to detect change in a future trial. The addition of a measure of IQ and/or intellectual disability as aforementioned, could be beneficial for more accurate analysis since the challenges of having a child with more severe additional needs potentially non-verbal child are very different from those of a high functioning and highly verbal child. A family satisfaction questionnaire was administered during the trial with the aim of exploring any benefits to partners from the programme. However missing data was extremely high, with poor return of responses. Due to the high percentage of employment in the households, which is contrary to the literature (Knapp et al., 2009; Buescher, 2014), many partners were out at work during the data collection home visits. The questionnaires were therefore left with the primary carer, with researchers requesting that the measures were completed at a convenient time and returned to them. However, the number of responses were extremely low, for unknown reasons. The majority of partners were male in the sample, and it is well documented that fathers are harder to engage in both parenting interventions and in research (Flippin & Crais, 2011). There was also a misunderstanding with regards to some families, whereby the primary carer had completed, and returned the questionnaire on the partners behalf, making the responses invalid. It was therefore not possible to include the data in the analysis. This is unfortunate due to the known influence of family and relationship dissatisfaction on child adjustment (Camisasca, Miragoli & Di Blasio, 2016). For future research, it could be more beneficial to administer an electronic version of the Family APGAR, either through a direct link to the questionnaire or via email to the partner, they could then be completed independently of partners and returned directly to researchers, reducing the risk of biased or false responses. Number of responses may be higher if the offer of electronic completion was more convenient. Future research could also include the analysis of structured interviews conducted with intervention parents and group facilitators during the feasibility trial, to gather their thoughts and insights about the programme, which could inform further development of the programme and future implementation. Another important future direction is to expand upon the economic evidence for the programme by way of a full economic evaluation. This would need to not only include a much larger sample, but also following evidence of programme effectiveness. The review of literature conducted for this thesis will inform the choice of comparative treatment, and findings from the micro-costing will be invaluable for informing the economic evaluation. A measure of service utilisation would also be a necessary addition in order to calculate any analysis of cost benefit.

The potential for further exploration of the IY-ASLD programme is plentiful. This thesis has made important contributions with regards to the background research, resolution of feasibility questions and the provision of preliminary evidence to inform the design of a larger definitive trial. The studies included have been novel, and rigorously conducted and add valuable findings towards enhancing the programme’s evidence base.

Conclusions

- This thesis has emphasised the need for parent programmes to support families of children with autism. The acceptability of the programme and retention in the research indicates that parents from this population are likely to engage in such interventions.
- The thesis has highlighted a paucity of economic evaluations with regards to behavioural parent programmes for families of children with ASD, and a need for economic evaluations to be conducted alongside those of effectiveness.
- The micro-costing evaluation has provided a direct measure of accurate cost involved with delivering the newly developed IY Autism programme. This contributes towards a framework for identifying the costs and benefits of programmes for policy deciders (costs and benefits of different options) for future implementation and provides information for future cost-effectiveness analysis.
- The thesis has conducted the first examination of long-term effectiveness. Although not significant, the promising gains observed in the feasibility trial were maintained without any booster sessions. This warrants further research in a larger trial.
- The thesis has also resulted in the first exploration of programme benefits to wider family members by assessing sibling outcomes. Although no differences were

observed, the study has answered important questions that will inform the design of a future study.

- Many lessons have been learned from this thesis with regards to accurate costings of programme delivery, acceptability of measures, and preliminary outcomes of long term and sibling data. This thesis has contributed to the existing literature with regards to the usefulness of psychosocial education programmes based on behavioural principles for parents of children with autism and provides valuable lessons which will inform a larger definitive trial.
- The studies included are novel, empirical studies that not only lay a robust foundation for further research by the author, but also provide a strong basis for replicable studies by other researchers, contributing to developing the research within this field.

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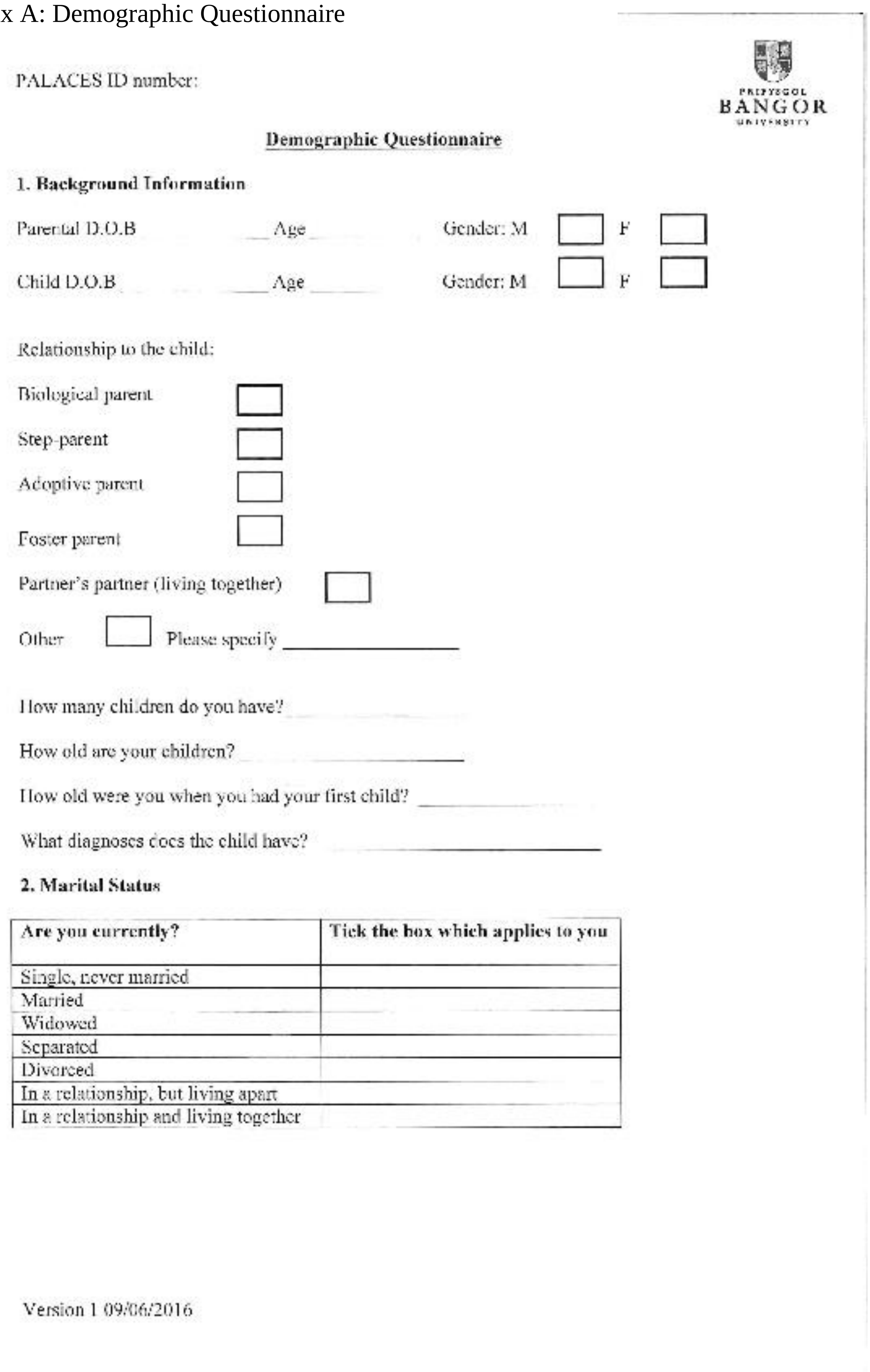
Appendices

Appendix A

Appendix A: Demographic Questionnaire



PALACES ID number:

PRIFYSGOL BANGOR UNIVERSITY

Demographic Questionnaire

1. Background Information

Parental D.O.B ___ Age ___ Gender: M ☐ F ☐

Child D.O.B ___ Age ___ Gender: M ☐ F ☐

Relationship to the child:

Biological parent ☐

Step-parent ☐

Adoptive parent ☐

Foster parent ☐

Partner's partner (living together) ☐

Other ☐ Please specify ____________

How many children do you have? ________

How old are your children? ________

How old were you when you had your first child? ________

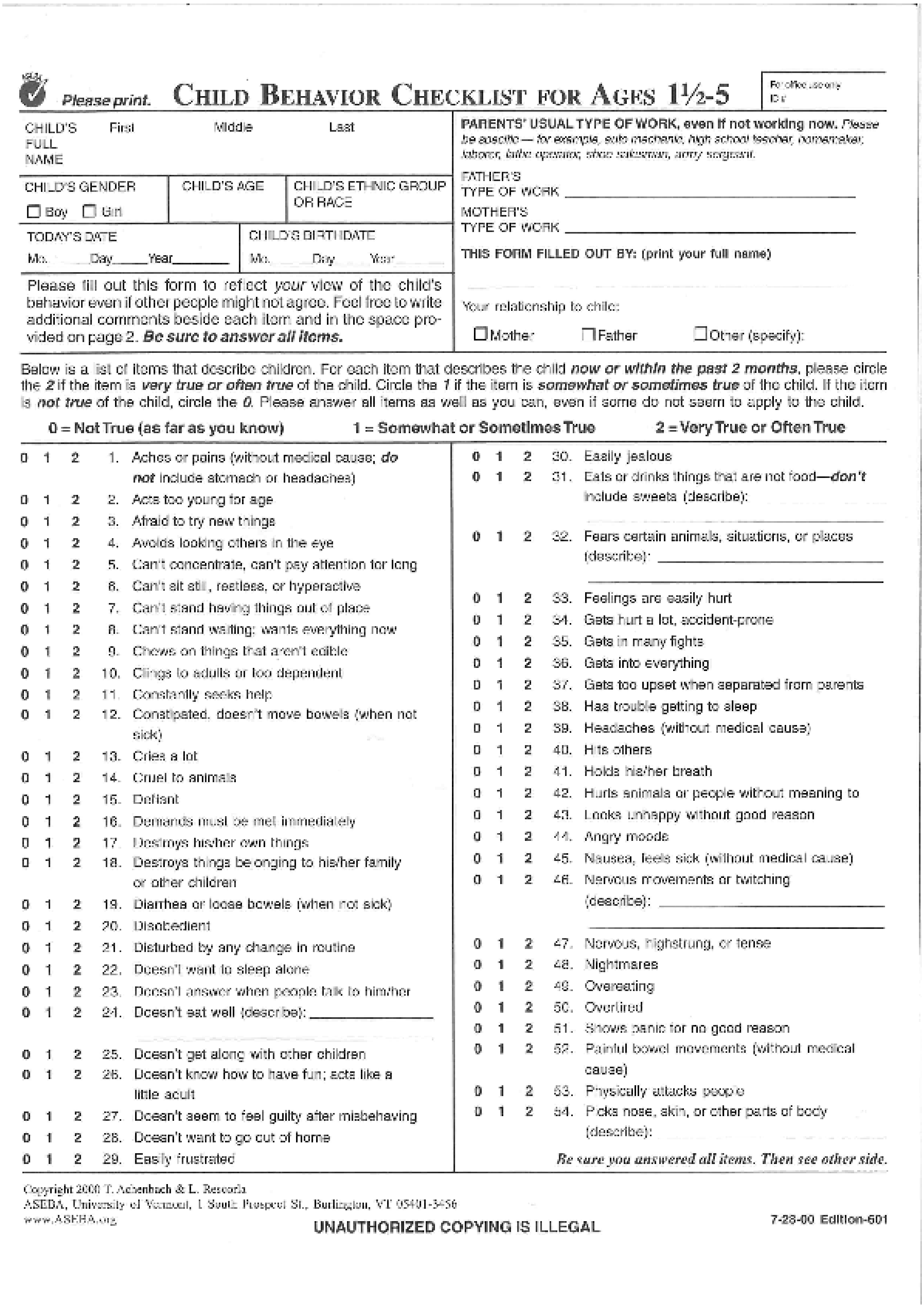
What diagnoses does the child have? ________

2. Marital Status

Are you currently?	**Tick the box which applies to you**
Single, never married	
Married	
Widowed	
Separated	
Divorced	
In a relationship, but living apart	
In a relationship and living together	

Version 1 09/06/2016

Appendix B



Please print. **CHILD BEHAVIOR CHECKLIST FOR AGES 1½-5**

For office use only
ID #

CHILD'S FULL NAME First Middle Last

CHILD'S GENDER ☐ Boy ☐ Girl

CHILD'S AGE

CHILD'S ETHNIC GROUP OR RACE

TODAY'S DATE Mo.____ Day____ Year____

CHILD'S BIRTHDATE Mo.____ Day____ Year____

Please fill out this form to reflect *your* view of the child's behavior even if other people might not agree. Feel free to write additional comments beside each item and in the space provided on page 2. ***Be sure to answer all items.***

PARENTS' USUAL TYPE OF WORK, even if not working now. *Please be specific — for example, auto mechanic, high school teacher, homemaker, laborer, lathe operator, shoe salesman, army sergeant.*

FATHER'S TYPE OF WORK ________________

MOTHER'S TYPE OF WORK ________________

THIS FORM FILLED OUT BY: (print your full name)

Your relationship to child:
☐ Mother ☐ Father ☐ Other (specify):

Below is a list of items that describe children. For each item that describes the child ***now or within the past 2 months***, please circle the ***2*** if the item is ***very true or often true*** of the child. Circle the ***1*** if the item is ***somewhat or sometimes true*** of the child. If the item is ***not true*** of the child, circle the ***0***. Please answer all items as well as you can, even if some do not seem to apply to the child.

0 = Not True (as far as you know) 1 = Somewhat or Sometimes True 2 = Very True or Often True

0 1 2 1. Aches or pains (without medical cause; ***do not*** include stomach or headaches)
0 1 2 2. Acts too young for age
0 1 2 3. Afraid to try new things
0 1 2 4. Avoids looking others in the eye
0 1 2 5. Can't concentrate, can't pay attention for long
0 1 2 6. Can't sit still, restless, or hyperactive
0 1 2 7. Can't stand having things out of place
0 1 2 8. Can't stand waiting; wants everything now
0 1 2 9. Chews on things that aren't edible
0 1 2 10. Clings to adults or too dependent
0 1 2 11. Constantly seeks help
0 1 2 12. Constipated, doesn't move bowels (when not sick)
0 1 2 13. Cries a lot
0 1 2 14. Cruel to animals
0 1 2 15. Defiant
0 1 2 16. Demands must be met immediately
0 1 2 17. Destroys his/her own things
0 1 2 18. Destroys things belonging to his/her family or other children
0 1 2 19. Diarrhea or loose bowels (when not sick)
0 1 2 20. Disobedient
0 1 2 21. Disturbed by any change in routine
0 1 2 22. Doesn't want to sleep alone
0 1 2 23. Doesn't answer when people talk to him/her
0 1 2 24. Doesn't eat well (describe): ____________
0 1 2 25. Doesn't get along with other children
0 1 2 26. Doesn't know how to have fun; acts like a little adult
0 1 2 27. Doesn't seem to feel guilty after misbehaving
0 1 2 28. Doesn't want to go out of home
0 1 2 29. Easily frustrated
0 1 2 30. Easily jealous
0 1 2 31. Eats or drinks things that are not food—***don't*** include sweets (describe):
0 1 2 32. Fears certain animals, situations, or places (describe): ____________
0 1 2 33. Feelings are easily hurt
0 1 2 34. Gets hurt a lot, accident-prone
0 1 2 35. Gets in many fights
0 1 2 36. Gets into everything
0 1 2 37. Gets too upset when separated from parents
0 1 2 38. Has trouble getting to sleep
0 1 2 39. Headaches (without medical cause)
0 1 2 40. Hits others
0 1 2 41. Holds his/her breath
0 1 2 42. Hurts animals or people without meaning to
0 1 2 43. Looks unhappy without good reason
0 1 2 44. Angry moods
0 1 2 45. Nausea, feels sick (without medical cause)
0 1 2 46. Nervous movements or twitching (describe): ____________
0 1 2 47. Nervous, highstrung, or tense
0 1 2 48. Nightmares
0 1 2 49. Overeating
0 1 2 50. Overtired
0 1 2 51. Shows panic for no good reason
0 1 2 52. Painful bowel movements (without medical cause)
0 1 2 53. Physically attacks people
0 1 2 54. Picks nose, skin, or other parts of body (describe):

Be sure you answered all items. Then see other side.

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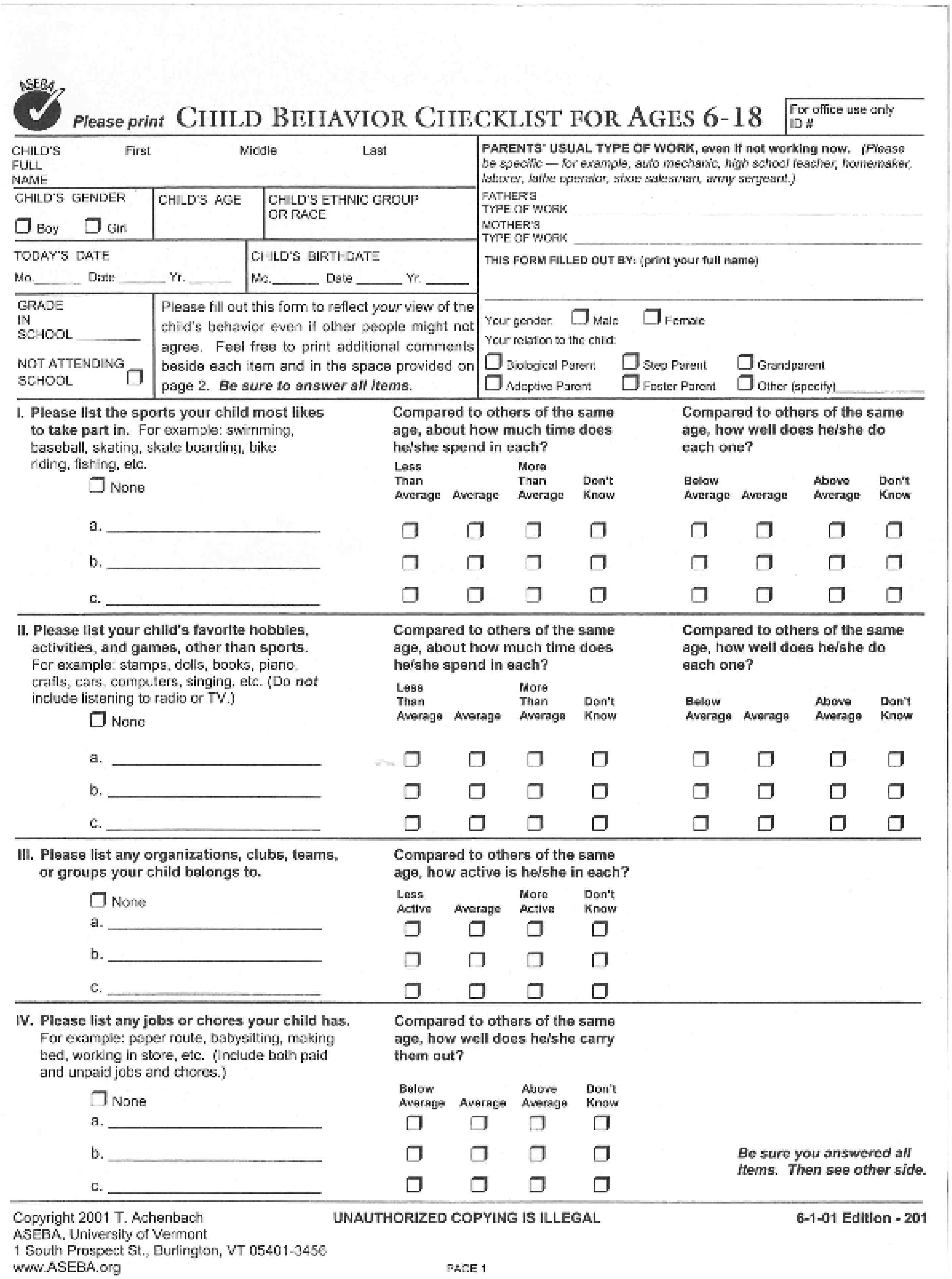
7-28-00 Edition-601



ASEBA

Please print **CHILD BEHAVIOR CHECKLIST FOR AGES 6-18**

For office use only
ID #

CHILD'S FULL NAME First Middle Last

CHILD'S GENDER ☐ Boy ☐ Girl

CHILD'S AGE

CHILD'S ETHNIC GROUP OR RACE

TODAY'S DATE Mo.______ Date ______ Yr. ______

CHILD'S BIRTHDATE Mo.______ Date ______ Yr. ______

GRADE IN SCHOOL ______

NOT ATTENDING SCHOOL ☐

Please fill out this form to reflect *your* view of the child's behavior even if other people might not agree. Feel free to print additional comments beside each item and in the space provided on page 2. ***Be sure to answer all items.***

PARENTS' USUAL TYPE OF WORK, even if not working now. *(Please be specific — for example, auto mechanic, high school teacher, homemaker, laborer, lathe operator, shoe salesman, army sergeant.)*

FATHER'S TYPE OF WORK ______

MOTHER'S TYPE OF WORK ______

THIS FORM FILLED OUT BY: (print your full name)

Your gender: ☐ Male ☐ Female

Your relation to the child:

☐ Biological Parent ☐ Step Parent ☐ Grandparent

☐ Adoptive Parent ☐ Foster Parent ☐ Other (specify)______

I. Please list the sports your child most likes to take part in. For example: swimming, baseball, skating, skate boarding, bike riding, fishing, etc.

☐ None

	Compared to others of the same age, about how much time does he/she spend in each?				**Compared to others of the same age, how well does he/she do each one?**			
	Less Than Average	**Average**	**More Than Average**	**Don't Know**	**Below Average**	**Average**	**Above Average**	**Don't Know**
a. ______	☐	☐	☐	☐	☐	☐	☐	☐
b. ______	☐	☐	☐	☐	☐	☐	☐	☐
c. ______	☐	☐	☐	☐	☐	☐	☐	☐

II. Please list your child's favorite hobbies, activities, and games, other than sports. For example: stamps, dolls, books, piano, crafts, cars, computers, singing, etc. (Do *not* include listening to radio or TV.)

☐ None

	Compared to others of the same age, about how much time does he/she spend in each?				**Compared to others of the same age, how well does he/she do each one?**			
	Less Than Average	**Average**	**More Than Average**	**Don't Know**	**Below Average**	**Average**	**Above Average**	**Don't Know**
a. ______	☐	☐	☐	☐	☐	☐	☐	☐
b. ______	☐	☐	☐	☐	☐	☐	☐	☐
c. ______	☐	☐	☐	☐	☐	☐	☐	☐

III. Please list any organizations, clubs, teams, or groups your child belongs to.

☐ None

	Compared to others of the same age, how active is he/she in each?			
	Less Active	**Average**	**More Active**	**Don't Know**
a. ______	☐	☐	☐	☐
b. ______	☐	☐	☐	☐
c. ______	☐	☐	☐	☐

IV. Please list any jobs or chores your child has. For example: paper route, babysitting, making bed, working in store, etc. (Include both paid and unpaid jobs and chores.)

☐ None

	Compared to others of the same age, how well does he/she carry them out?			
	Below Average	**Average**	**Above Average**	**Don't Know**
a. ______	☐	☐	☐	☐
b. ______	☐	☐	☐	☐
c. ______	☐	☐	☐	☐

Be sure you answered all items. Then see other side.

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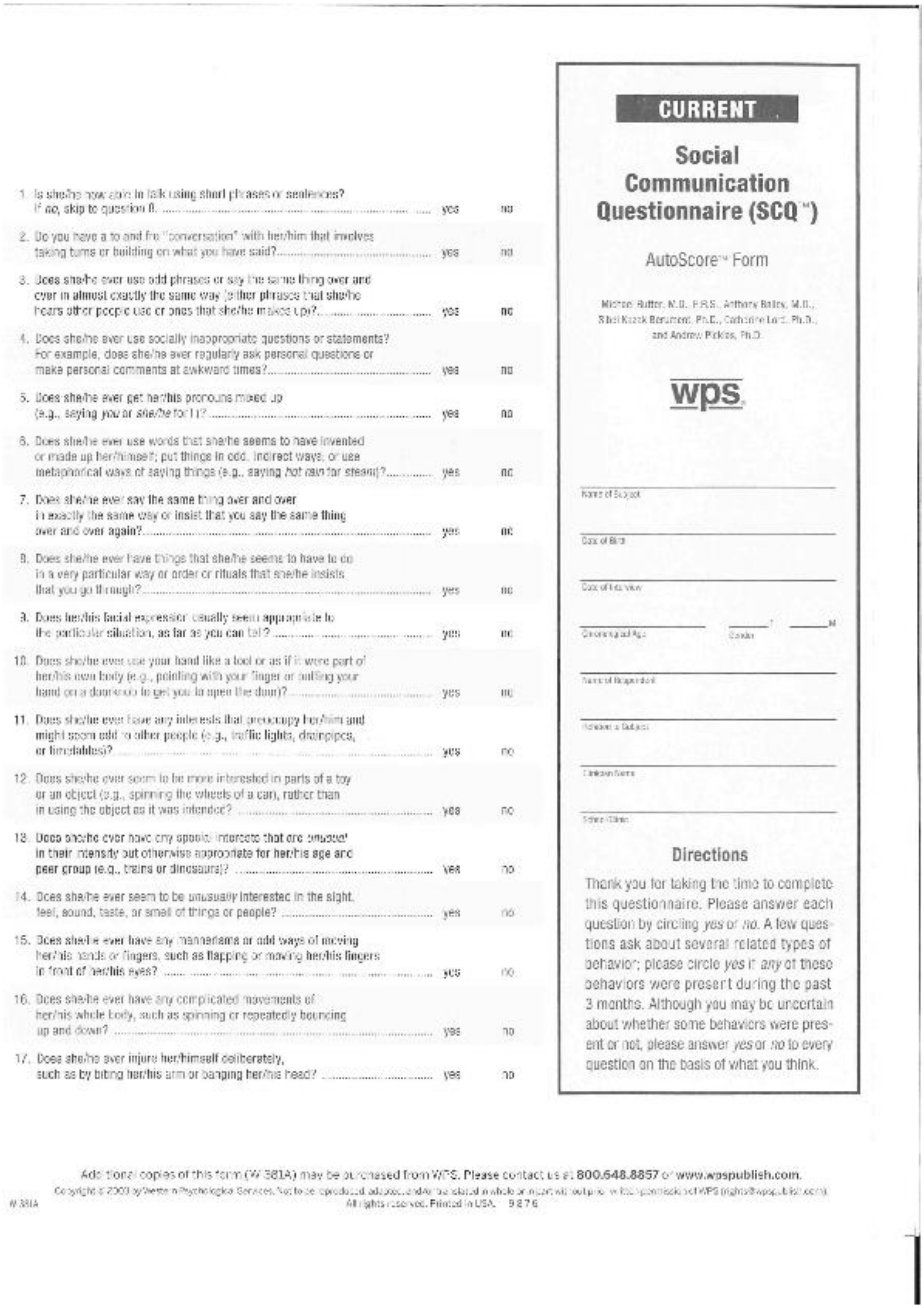
PAGE 1

Appendix C



1. Is she/he now able to talk using short phrases or sentences? If *no*, skip to question 8. .... yes no

2. Do you have a to and fro "conversation" with her/him that involves taking turns or building on what you have said? .... yes no

3. Does she/he ever use odd phrases or say the same thing over and over in almost exactly the same way (either phrases that she/he hears other people use or ones that she/he makes up)? .... yes no

4. Does she/he ever use socially inappropriate questions or statements? For example, does she/he ever regularly ask personal questions or make personal comments at awkward times? .... yes no

5. Does she/he ever get her/his pronouns mixed up (e.g., saying *you* or *she/he* for I)? .... yes no

6. Does she/he ever use words that she/he seems to have invented or made up her/himself; put things in odd, indirect ways; or use metaphorical ways of saying things (e.g., saying *hot rain* for steam)? .... yes no

7. Does she/he ever say the same thing over and over in exactly the same way or insist that you say the same thing over and over again? .... yes no

8. Does she/he ever have things that she/he seems to have to do in a very particular way or order or rituals that she/he insists that you go through? .... yes no

9. Does her/his facial expression usually seem appropriate to the particular situation, as far as you can tell? .... yes no

10. Does she/he ever use your hand like a tool or as if it were part of her/his own body (e.g., pointing with your finger or putting your hand on a doorknob to get you to open the door)? .... yes no

11. Does she/he ever have any interests that preoccupy her/him and might seem odd to other people (e.g., traffic lights, drainpipes, or timetables)? .... yes no

12. Does she/he ever seem to be more interested in parts of a toy or an object (e.g., spinning the wheels of a car), rather than in using the object as it was intended? .... yes no

13. Does she/he ever have any special interests that are unusual in their intensity but otherwise appropriate for her/his age and peer group (e.g., trains or dinosaurs)? .... yes no

14. Does she/he ever seem to be unusually interested in the sight, feel, sound, taste, or smell of things or people? .... yes no

15. Does she/he ever have any mannerisms or odd ways of moving her/his hands or fingers, such as flapping or moving her/his fingers in front of her/his eyes? .... yes no

16. Does she/he ever have any complicated movements of her/his whole body, such as spinning or repeatedly bouncing up and down? .... yes no

17. Does she/he ever injure her/himself deliberately, such as by biting her/his arm or banging her/his head? .... yes no

CURRENT

Social Communication Questionnaire (SCQ™)

AutoScore™ Form

Michael Rutter, M.D., F.R.S., Anthony Bailey, M.D.,
Sibel Kazak Berument, Ph.D., Catherine Lord, Ph.D.,
and Andrew Pickles, Ph.D.

WPS

Name of Subject ____________________

Date of Birth ____________________

Date of Interview ____________________

Chronological Age ____________________ Gender ______F ______M

Name of Respondent ____________________

Relation to Subject ____________________

Clinician Name ____________________

School/Clinic ____________________

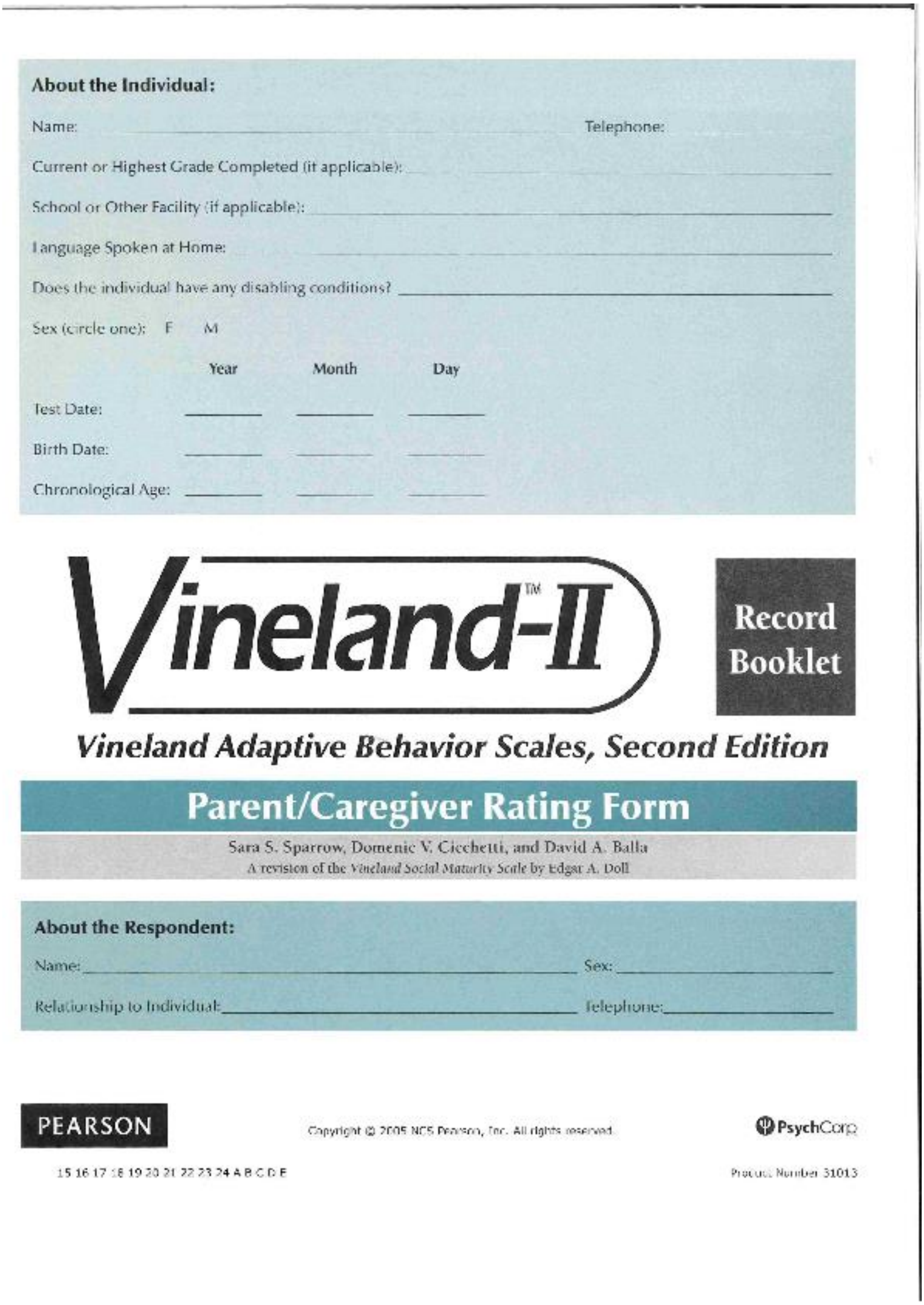
Directions

Thank you for taking the time to complete this questionnaire. Please answer each question by circling *yes* or *no*. A few questions ask about several related types of behavior; please circle *yes* if *any* of these behaviors were present during the past 3 months. Although you may be uncertain about whether some behaviors were present or not, please answer *yes* or *no* to every question on the basis of what you think.

Additional copies of this form (W-381A) may be purchased from WPS. Please contact us at **800.648.8857** or **www.wpspublish.com**.

W-381A

Appendix D



About the Individual:

Name: ____________________ Telephone: __________

Current or Highest Grade Completed (if applicable): ____________________

School or Other Facility (if applicable): ____________________

Language Spoken at Home: ____________________

Does the individual have any disabling conditions? ____________________

Sex (circle one): F M

	Year	Month	Day
Test Date:			
Birth Date:			
Chronological Age:			

Vineland-II™

Record Booklet

Vineland Adaptive Behavior Scales, Second Edition

Parent/Caregiver Rating Form

Sara S. Sparrow, Domenic V. Cicchetti, and David A. Balla
A revision of the *Vineland Social Maturity Scale* by Edgar A. Doll

About the Respondent:

Name: ____________________ Sex: __________

Relationship to Individual: ____________________ Telephone: __________

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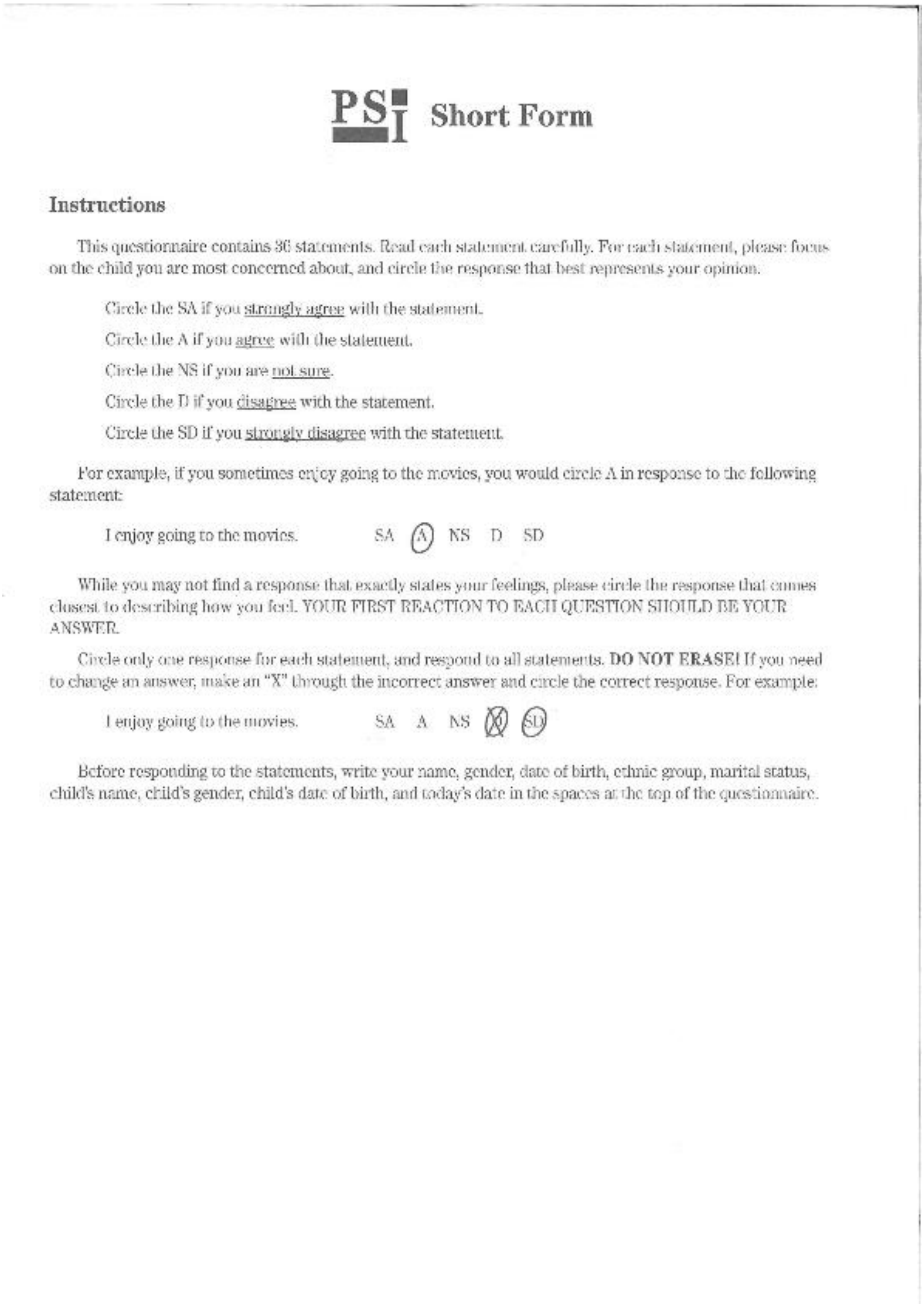
Appendix E



PSI Short Form

Instructions

This questionnaire contains 36 statements. Read each statement carefully. For each statement, please focus on the child you are most concerned about, and circle the response that best represents your opinion.

Circle the SA if you strongly agree with the statement.

Circle the A if you agree with the statement.

Circle the NS if you are not sure.

Circle the D if you disagree with the statement.

Circle the SD if you strongly disagree with the statement.

For example, if you sometimes enjoy going to the movies, you would circle A in response to the following statement:

I enjoy going to the movies. SA [✓ A] NS D SD

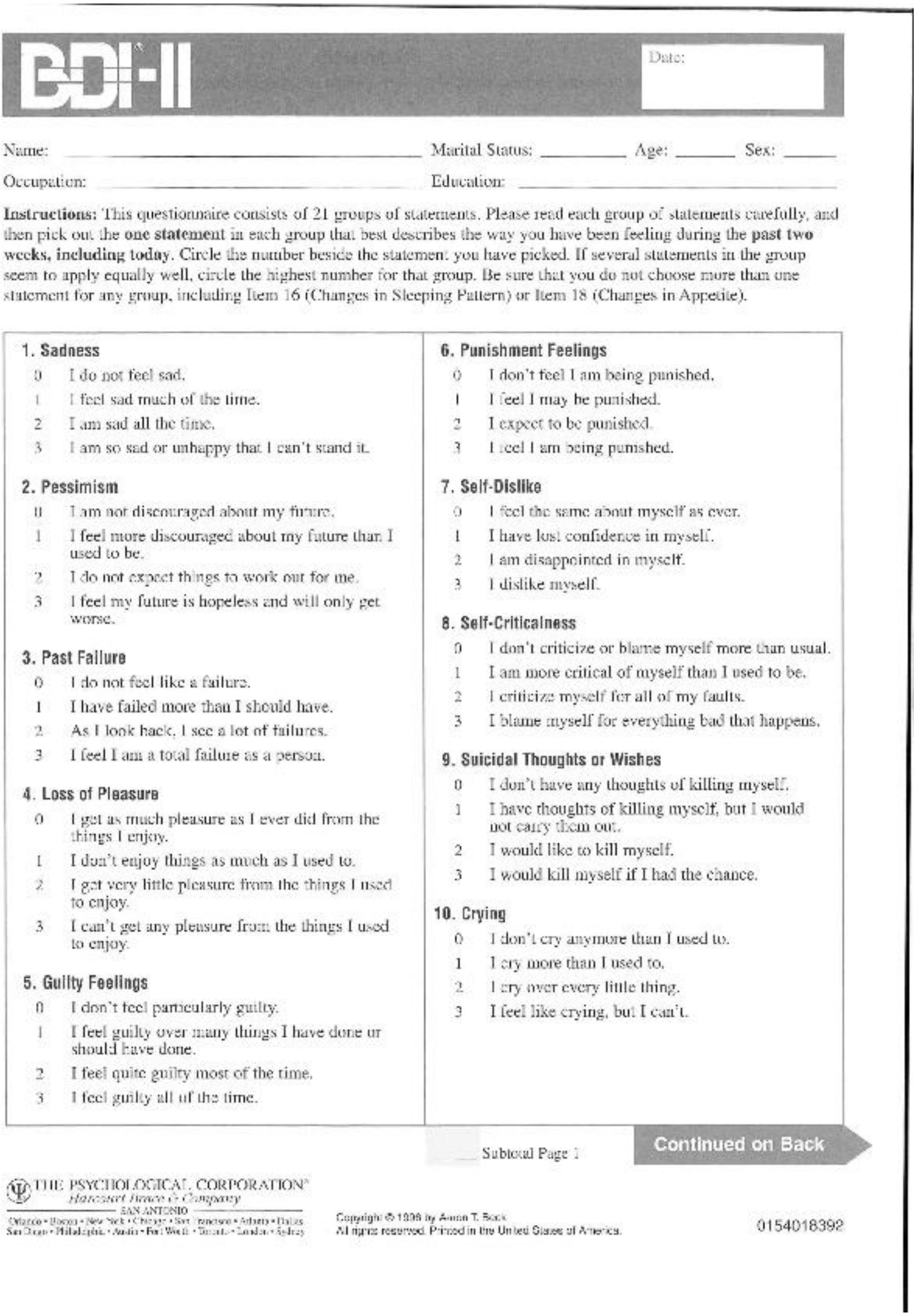
While you may not find a response that exactly states your feelings, please circle the response that comes closest to describing how you feel. YOUR FIRST REACTION TO EACH QUESTION SHOULD BE YOUR ANSWER.

Circle only one response for each statement, and respond to all statements. **DO NOT ERASE!** If you need to change an answer, make an "X" through the incorrect answer and circle the correct response. For example:

I enjoy going to the movies. SA A NS ~~D~~ [✓ SD]

Before responding to the statements, write your name, gender, date of birth, ethnic group, marital status, child's name, child's gender, child's date of birth, and today's date in the spaces at the top of the questionnaire.

Appendix F



BDI-II

Date:

Name: ______________________ Marital Status: ________ Age: ______ Sex: ______

Occupation: ______________________ Education: ______________________

Instructions: This questionnaire consists of 21 groups of statements. Please read each group of statements carefully, and then pick out the **one statement** in each group that best describes the way you have been feeling during the **past two weeks, including today**. Circle the number beside the statement you have picked. If several statements in the group seem to apply equally well, circle the highest number for that group. Be sure that you do not choose more than one statement for any group, including Item 16 (Changes in Sleeping Pattern) or Item 18 (Changes in Appetite).

1. Sadness

0 I do not feel sad.
1 I feel sad much of the time.
2 I am sad all the time.
3 I am so sad or unhappy that I can't stand it.

2. Pessimism

0 I am not discouraged about my future.
1 I feel more discouraged about my future than I used to be.
2 I do not expect things to work out for me.
3 I feel my future is hopeless and will only get worse.

3. Past Failure

0 I do not feel like a failure.
1 I have failed more than I should have.
2 As I look back, I see a lot of failures.
3 I feel I am a total failure as a person.

4. Loss of Pleasure

0 I get as much pleasure as I ever did from the things I enjoy.
1 I don't enjoy things as much as I used to.
2 I get very little pleasure from the things I used to enjoy.
3 I can't get any pleasure from the things I used to enjoy.

5. Guilty Feelings

0 I don't feel particularly guilty.
1 I feel guilty over many things I have done or should have done.
2 I feel quite guilty most of the time.
3 I feel guilty all of the time.

6. Punishment Feelings

0 I don't feel I am being punished.
1 I feel I may be punished.
2 I expect to be punished.
3 I feel I am being punished.

7. Self-Dislike

0 I feel the same about myself as ever.
1 I have lost confidence in myself.
2 I am disappointed in myself.
3 I dislike myself.

8. Self-Criticalness

0 I don't criticize or blame myself more than usual.
1 I am more critical of myself than I used to be.
2 I criticize myself for all of my faults.
3 I blame myself for everything bad that happens.

9. Suicidal Thoughts or Wishes

0 I don't have any thoughts of killing myself.
1 I have thoughts of killing myself, but I would not carry them out.
2 I would like to kill myself.
3 I would kill myself if I had the chance.

10. Crying

0 I don't cry anymore than I used to.
1 I cry more than I used to.
2 I cry over every little thing.
3 I feel like crying, but I can't.

____ Subtotal Page 1

Continued on Back

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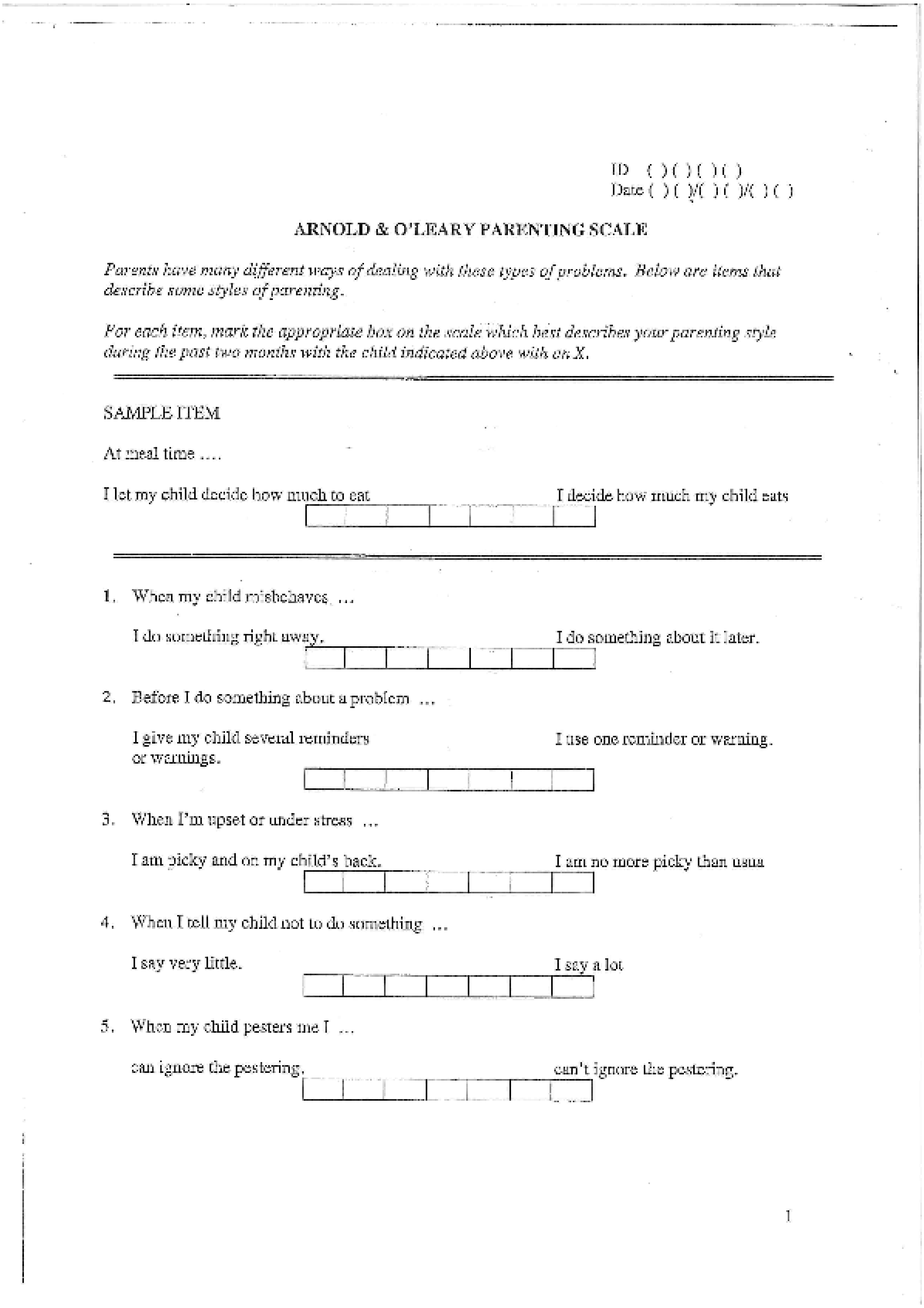
Appendix G



ID ( ) ( ) ( ) ( )
Date ( ) ( )/( ) ( )/( ) ( )

ARNOLD & O'LEARY PARENTING SCALE

Parents have many different ways of dealing with these types of problems. Below are items that describe some styles of parenting.

For each item, mark the appropriate box on the scale which best describes your parenting style during the past two months with the child indicated above with an X.

SAMPLE ITEM

At meal time ....

I let my child decide how much to eat ☐☐☐☐☐☐☐ I decide how much my child eats

1. When my child misbehaves ...

   I do something right away. ☐☐☐☐☐☐☐ I do something about it later.

2. Before I do something about a problem ...

   I give my child several reminders or warnings. ☐☐☐☐☐☐☐ I use one reminder or warning.

3. When I'm upset or under stress ...

   I am picky and on my child's back. ☐☐☐☐☐☐☐ I am no more picky than usua

4. When I tell my child not to do something ...

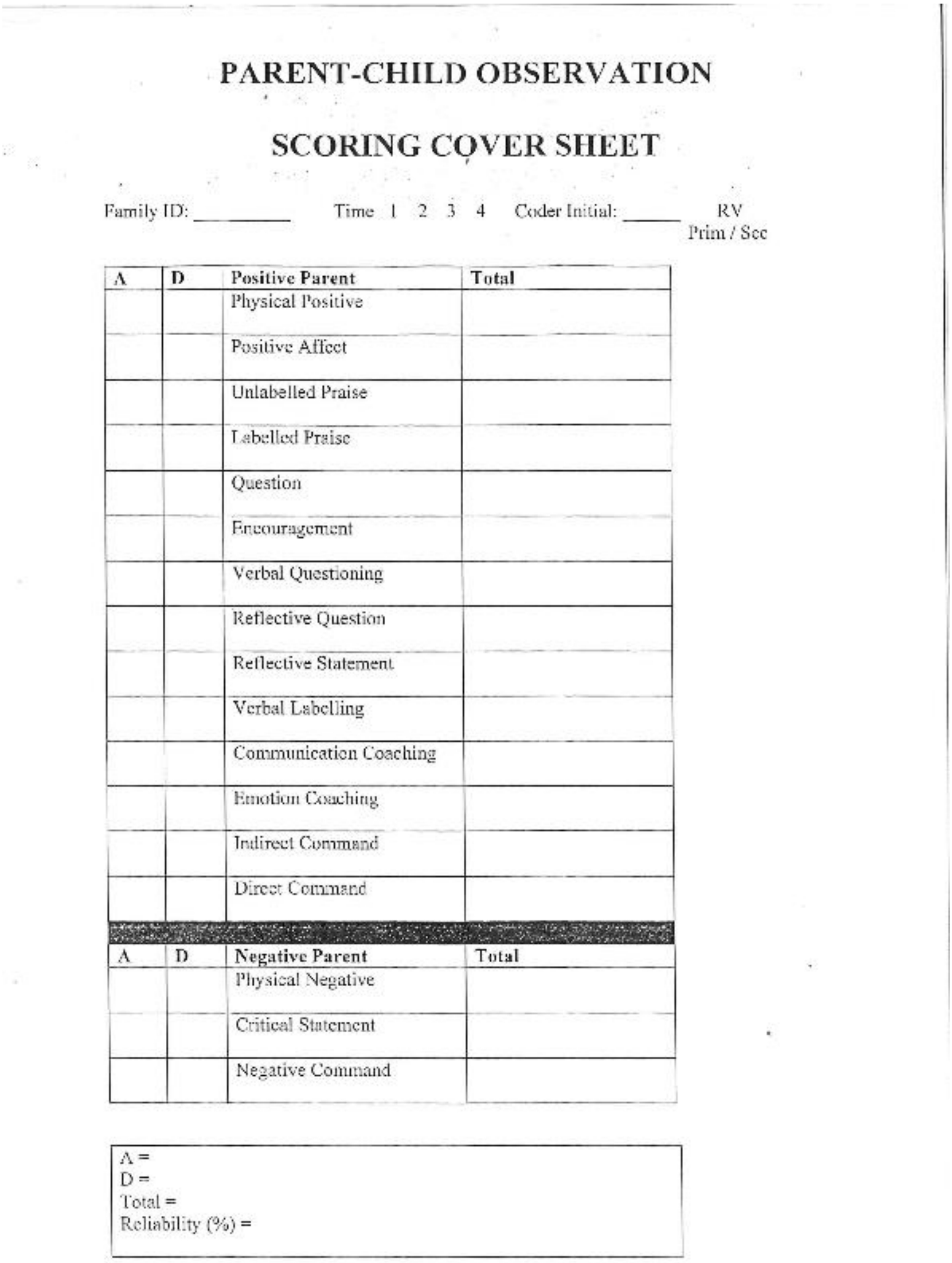
   I say very little. ☐☐☐☐☐☐☐ I say a lot

5. When my child pesters me I ...

   can ignore the pestering. ☐☐☐☐☐☐☐ can't ignore the pestering.

1

Appendix H



PARENT-CHILD OBSERVATION

SCORING COVER SHEET

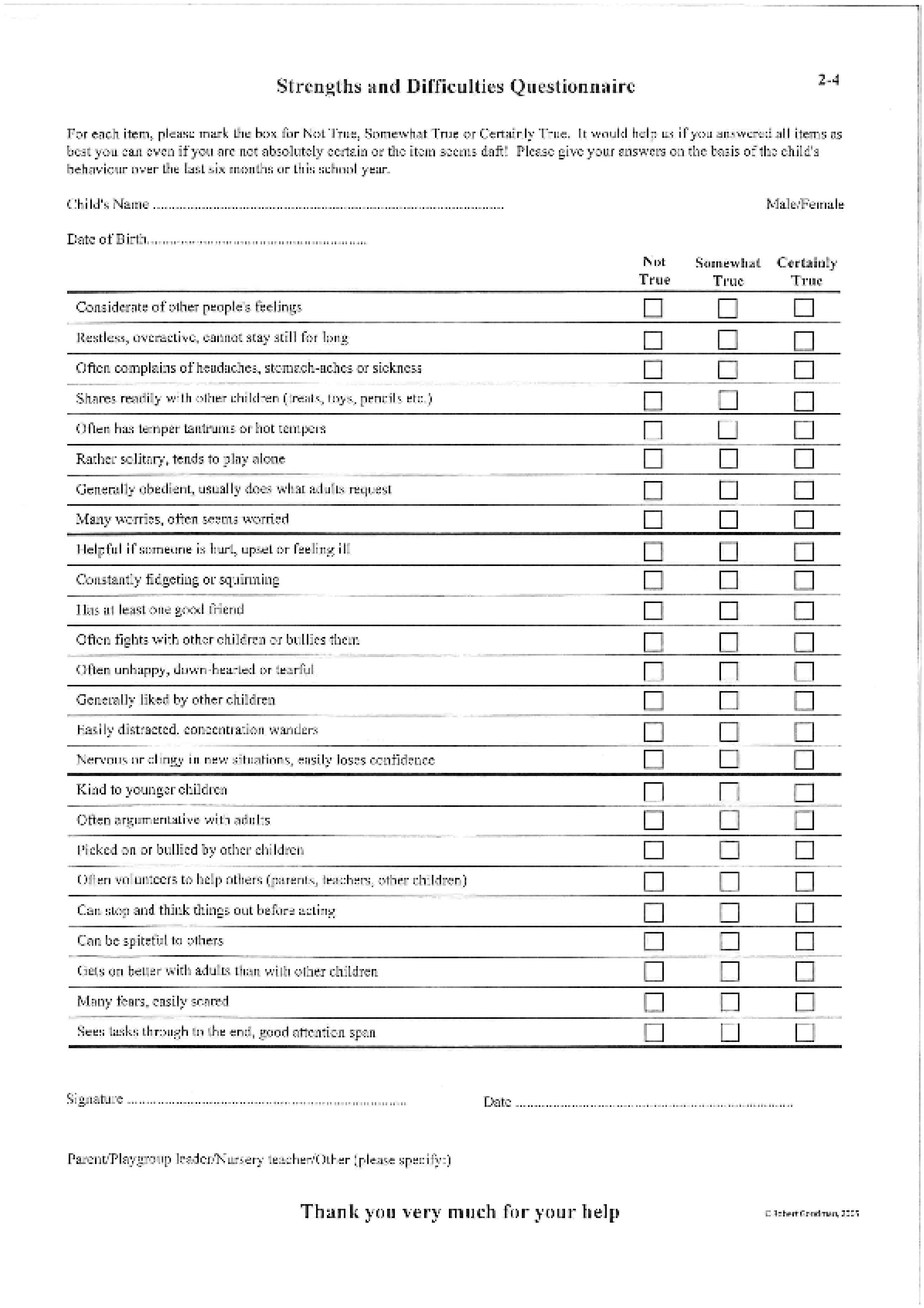
Family ID: _________ Time 1 2 3 4 Coder Initial: _____ RV
Prim / Sec

A	D	**Positive Parent**	**Total**
		Physical Positive	
		Positive Affect	
		Unlabelled Praise	
		Labelled Praise	
		Question	
		Encouragement	
		Verbal Questioning	
		Reflective Question	
		Reflective Statement	
		Verbal Labelling	
		Communication Coaching	
		Emotion Coaching	
		Indirect Command	
		Direct Command	

A	D	**Negative Parent**	**Total**
		Physical Negative	
		Critical Statement	
		Negative Command	

A =
D =
Total =
Reliability (%) =

Appendix I



Strengths and Difficulties Questionnaire

2-4

For each item, please mark the box for Not True, Somewhat True or Certainly True. It would help us if you answered all items as best you can even if you are not absolutely certain or the item seems daft! Please give your answers on the basis of the child's behaviour over the last six months or this school year.

Child's Name .... Male/Female

Date of Birth..

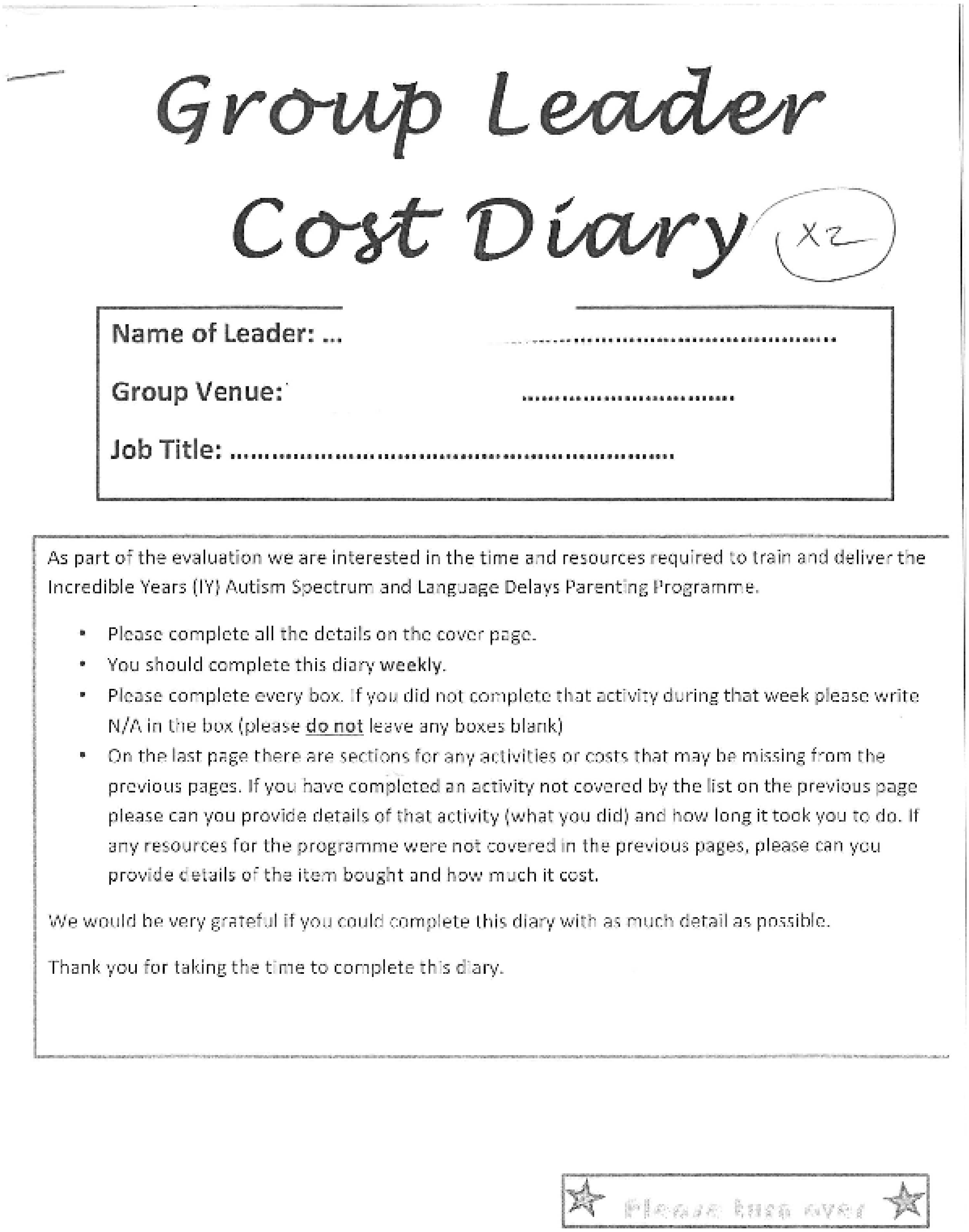
	Not True	Somewhat True	Certainly True
Considerate of other people's feelings	☐	☐	☐
Restless, overactive, cannot stay still for long	☐	☐	☐
Often complains of headaches, stomach-aches or sickness	☐	☐	☐
Shares readily with other children (treats, toys, pencils etc.)	☐	☐	☐
Often has temper tantrums or hot tempers	☐	☐	☐
Rather solitary, tends to play alone	☐	☐	☐
Generally obedient, usually does what adults request	☐	☐	☐
Many worries, often seems worried	☐	☐	☐
Helpful if someone is hurt, upset or feeling ill	☐	☐	☐
Constantly fidgeting or squirming	☐	☐	☐
Has at least one good friend	☐	☐	☐
Often fights with other children or bullies them	☐	☐	☐
Often unhappy, down-hearted or tearful	☐	☐	☐
Generally liked by other children	☐	☐	☐
Easily distracted, concentration wanders	☐	☐	☐
Nervous or clingy in new situations, easily loses confidence	☐	☐	☐
Kind to younger children	☐	☐	☐
Often argumentative with adults	☐	☐	☐
Picked on or bullied by other children	☐	☐	☐
Often volunteers to help others (parents, teachers, other children)	☐	☐	☐
Can stop and think things out before acting	☐	☐	☐
Can be spiteful to others	☐	☐	☐
Gets on better with adults than with other children	☐	☐	☐
Many fears, easily scared	☐	☐	☐
Sees tasks through to the end, good attention span	☐	☐	☐

Signature .... Date ....

Parent/Playgroup leader/Nursery teacher/Other (please specify:)

Thank you very much for your help

Appendix J



Group Leader Cost Diary

x2

Name of Leader: ... .

Group Venue: ....

Job Title: ....

As part of the evaluation we are interested in the time and resources required to train and deliver the Incredible Years (IY) Autism Spectrum and Language Delays Parenting Programme.

- Please complete all the details on the cover page.
- You should complete this diary weekly.
- Please complete every box. If you did not complete that activity during that week please write N/A in the box (please do not leave any boxes blank)
- On the last page there are sections for any activities or costs that may be missing from the previous pages. If you have completed an activity not covered by the list on the previous page please can you provide details of that activity (what you did) and how long it took you to do. If any resources for the programme were not covered in the previous pages, please can you provide details of the item bought and how much it cost.

We would be very grateful if you could complete this diary with as much detail as possible.

Thank you for taking the time to complete this diary.

☆ Please turn over ☆

Appendix K

Concept 1

Parenting interventions OR

Parent* OR
Parent* programmes OR
Parent* training

Concept 2

Autism OR

Autis*

Autistic spectrum disorder* OR ASD

OR Autis* disorder*

Concept 3

Economic evidence OR

Cost* OR
Cost effective* OR Cost effectiveness analysis OR
Cost benefit analysis OR
Cost utility analysis OR
Cost consequence analysis OR
Cost minimisation analysis OR
Unit cost* OR unit-cost* OR
Economic* OR health economic* OR economic evaluation*

Additional filters

- **Time range-10yrs (since 2009)**
- **English language only**

1a) PsycINFO

1. < Parenting interventions OR Parent* OR parent* programmes OR Parent* training>
   AND
2. <Autism OR Autis* OR Autistic spectrum disorder* OR ASD OR Autis* disorder*>

   AND

3. < Economic evidence OR Cost* OR Cost effective* OR Cost effectiveness analysis OR Cost benefit analysis OR Cost utility analysis OR Cost consequence analysis OR Cost minimisation analysis OR Unit cost* OR unit-cost* OR Economic* OR health economic* OR economic evaluation*>

1b) ASSIA

1. < Parenting interventions OR Parent* OR parent* programmes OR Parent* training>
   AND

2. <Autism OR Autis* OR Autistic spectrum disorder* OR ASD OR Autis* disorder*>

   AND

3. < Economic evidence OR Cost* OR Cost effective* OR Cost effectiveness analysis OR Cost benefit analysis OR Cost utility analysis OR Cost consequence analysis OR Cost minimisation analysis OR Unit cost* OR unit-cost* OR Economic* OR health economic* OR economic evaluation*>

1c) CINAHL

1. < Parenting interventions OR Parent* OR parent* programmes OR Parent* training>
   AND
2. <Autism OR Autis* OR Autistic spectrum disorder* OR ASD OR Autis* disorder*>

   AND

3. < Economic evidence OR Cost* OR Cost effective* OR Cost effectiveness analysis OR Cost benefit analysis OR Cost utility analysis OR Cost consequence analysis OR Cost minimisation analysis OR Unit cost* OR unit-cost* OR Economic* OR health economic* OR economic evaluation*>

1d) Pubmed (NCBI)

1. < Parenting interventions OR Parent* OR parent* programmes OR Parent* training>
   AND
2. <Autism OR Autis* OR Autistic spectrum disorder* OR ASD OR Autis* disorder*>

   AND

3. < Economic evidence OR Cost* OR Cost effective* OR Cost effectiveness analysis OR Cost benefit analysis OR Cost utility analysis OR Cost consequence analysis OR Cost minimisation analysis OR Unit cost* OR unit-cost* OR Economic* OR health economic* OR economic evaluation*>

1f) Medline (EBSCO)

1. < Parenting interventions OR Parent* OR parent* programmes OR Parent* training>
   AND
2. <Autism OR Autis* OR Autistic spectrum disorder* OR ASD OR Autis* disorder*>

   AND

3. < Economic evidence OR Cost* OR Cost effective* OR Cost effectiveness analysis OR Cost benefit analysis OR Cost utility analysis OR Cost consequence analysis OR Cost minimisation analysis OR Unit cost* OR unit-cost* OR Economic* OR health economic* OR economic evaluation*>