

VOLUME 1

PARENTAL ILLNESS REPRESENTATIONS IN PATHOLOGICAL DEMAND
AVOIDANCE SYNDROME: PARENTAL COPING, PARENTING STRESS, PARENTAL
WELLBEING AND THE CHILD-PARENT RELATIONSHIP

by

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Thesis Overview

This thesis, submitted in partial fulfilment of a Doctorate in Clinical Psychology, comprises two volumes. Volume One includes the research component and Volume Two consists of the clinical component.

The research component includes a literature review examining the efficacy of interventions for parents of children with autism spectrum disorder (ASD) in relation to parent outcomes, an empirical paper, which presents findings of a quantitative study exploring parental illness perceptions, coping styles, wellbeing, parenting stress and perceptions of the child-parent relationship in parents of a child with pathological demand avoidance syndrome (PDA) and an executive summary of the literature review and the quantitative study.

The clinical component includes four clinical practice reports. The models report details two psychological formulations; one from a cognitive behavioural perspective and one from a systemic perspective, for a 20 year old gentleman who was experiencing anxiety and depression, and had undergone surgery to remove part of his bowel, which left him with a stoma. The service evaluation report details an investigation of the extent to which a local respiratory service was addressing the needs of COPD patients with co-morbid anxiety and/or depression. The single case experimental design presents an evaluation of a behavioural intervention for a 25 year old woman with a moderate learning disability, who presented with skin picking behaviours. The final report details a case study of a 14 year old girl, who was under investigation for Crohn's disease and referred to a paediatric psychology service for help with anxiety. The volume concludes with an abstract, which reflects on the experience of providing consultation within a looked after and adopted child's psychology service.

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LITERATURE REVIEW

PARENTS OF CHILDREN WITH AUTISM: EFFECTIVENESS OF PARENT INTERVENTIONS ON PARENTAL STRESS, WELLBEING AND SELF-EFFICACY

Abstract

Introduction: There is extensive evidence demonstrating that parents of a child with autism spectrum disorder (ASD) experience high levels of parental stress, poor emotional wellbeing and poor parental self-efficacy. Despite this, relatively few ASD intervention studies address or evaluate parent outcomes. The aim of the review is to examine the efficacy of interventions for parents of children with ASD in relation to parent outcomes.

Method: 22 papers were identified for inclusion in the current review. Papers were reviewed to assess their quality and consider their findings.

Results: Interventions varied greatly in relation to content, approach, contact hours, mode of delivery and the quality of the research, making it difficult to make direct comparisons between interventions. In this review, psychoeducation was the intervention most commonly associated with outcomes of reduced parental stress, and parenting programmes were the intervention most commonly associated with improvements in parenting confidence/self-efficacy. There is some evidence to suggest that parent interventions have better outcomes in parents who experience poor mental health prior to their participation in the intervention.

Conclusions: The extent of the variation between interventions makes it difficult to draw any reliable conclusions about the effectiveness of parent interventions in relation to parent outcomes. A meta-analysis is recommended, to allow for the control of delivery mode, contact hours and other potentially confounding factors, which would make for a more

reliable analysis. However, findings do indicate that the type of intervention offered to parents should be based on a thorough assessment, focussed on identifying parents' individual needs.

1. INTRODUCTION

Autism Spectrum Disorder (ASD) is a neurodevelopmental disorder, which is characterised by a 'triad of impairments' (Wing & Gould, 1979). The triad of impairments includes impairment in social interaction, communication, and social imagination. For social interaction, there can be difficulties initiating and maintaining social relationships, appearing indifferent to other people, and difficulties in understanding social rules and cues. In terms of communication difficulties, these can present for individuals in both verbal and in non-verbal communication. For the final element of the triad, social imagination, deficits can present through restrictions in imaginative play (e.g., rigid and repetitive play) as well as a reduced capacity to consider and envisage the consequences of one's own actions. In addition to this triad of impairments, sensory issues (i.e., hyper- or hyposensitivity to sound, light, smell, textures and touch), repetitive behaviour patterns, and difficulty tolerating change in routine are often characteristic of children and adults with ASD (Wing & Gould, 1979). According to the Fifth Edition of the Diagnostic and Statistical Manual of mental disorders (DSM-V, 2013), symptoms must relate to persistent deficit in social communication and social interaction across multiple contexts, and restricted, repetitive patterns of behaviour, interests, or activities must be evident. The DSM-V specifies that symptoms must be present in the early developmental period, symptoms must cause clinically significant impairment in social functioning (or other important areas of current functioning) and these difficulties cannot be better explained by intellectual disability or global developmental delay.

The centre for disease control and prevention (CDC, 2014) reported that approximately 1% of the world population has ASD. The CDC found a 119.4% increase in the prevalence of

autism in children in the USA between 2000 and 2010, which, if accurate, makes autism the fastest growing developmental disability. In line with CDC estimates, Brugha et al. (2012) estimated the UK prevalence of ASD at 1%, with the National Autistic Society estimating that there are approximately 700,000 individuals living with ASD in the UK. It is widely cited that ASD is more prevalent in males than females, with a ratio of 4-5:1 (Fombonne, Quirke & Hagen, 2011). Knapp, Renee and Beecham (2007) estimated that there were 100,000 children with autism in the UK and the National Autistic Society estimate that autism affects 2.8 million people in the UK, when the families of individuals with autism are also considered.

Corcoran, Berry and Hill (2015) conducted a systematic review and meta-synthesis of 14 papers investigating the lived experience of parents of children with ASD and found six major themes within the literature, three of which relate to the way that life changes for parents and families when an individual in the family has autism. One of these three themes relates to the emotional stress and strain that parents of children with ASD experience, with subthemes concerning the stress associated with caring for a child with ASD, negative emotions associated with diagnosis, letting go of some of the hopes parents had for their child with ASD, and worrying about what will happen to their child when they are no longer around. Another major theme relates to the negative effects that caring for a child with ASD can have on the family as a whole and the parents as a couple and the third major theme relates to the requirement of parents to make adaptations and accommodate the needs of the child with ASD, which involves extra planning, recognising and avoiding triggers, being hypervigilant to potential triggers, adjusting schedules and allowing extra time.

Montes and Halterman (2006) conducted a population based study and found that mothers of a child with autism were highly stressed and were more likely to report poor or fair mental health than mothers in the general population. Similarly, Dabrowska and Pisula (2010) found

that parents of children with autism experience higher levels of stress than parents of children with Down syndrome and parents of typically developing children. In addition, parents of children with disabilities who are stressed are more likely to respond to their children in ways that exacerbate problematic behaviours (Hastings, 2002) and problematic behaviours in turn are associated with increased parenting stress in this population (Tomanik, Harris and Hawkins, 2004).

Ingersoll and Hambrick (2011) showed that increased parental stress levels are in part a consequence of the severe and persistent nature of the characteristics of ASD. In addition, parenting couples of children with ASD have reported poorer relationship satisfaction than parenting couples in the general population (Brobst, Clopton, & Hendrick, 2009), which may be in part a consequence of increased stress levels.

Sharpley, Bitsika and Efremidis (1997) conducted a study that involved 219 parents of children with autism and found that 80% of parents reported sometimes feeling “stretched beyond their limits”. The authors indicated that the three most stressful factors in parenting a child with autism were (a) worries about the permanency of the condition; (b) limited acceptance of behaviours associated with autism by society, and often by other family members; and (c) the lack of social support received by parents. A number of studies have suggested there is a strong relationship between elevated mental health problems (e.g. stress, depression) and poor parental self-efficacy in parents of individuals with ASD (Kuhn and Carter, 2006; Weiss et al., 2013). Higher levels of child need have also been associated with lower parental self-efficacy (Benson, 2014). In addition, the demands placed on parents to manage the ongoing stressors associated with ASD (e.g. physical, emotional and behavioural problems) are suspected to impact negatively on parental self-efficacy (Weiss, MacMullin & Lunskey, 2014).

Despite extensive literature evidencing the impact on parents of parenting a child with ASD, little is known about what interventions are most effective in improving parental emotional wellbeing. A wide range of interventions have been developed for children and young people with ASD, including behavioural interventions, social skills groups, parent mediated interventions, and parent interventions but, to date, the main focus of these interventions has related to improvements in child outcomes only, such as children's communication and behaviour problems (Warren et al., 2011).

Schultz, Schmidt and Stichter (2011) reviewed the effectiveness of parent education programmes for parents of children with ASD, in which thirty studies were reviewed (published between 1987 and 2007), but only four included measures of parental emotional wellbeing, all of which concerned parental stress. All four studies reported a reduction in parental stress following the parent education programme. The remaining 26 papers included in the review employed measures of parent skills/knowledge and/or child skills/behaviour, to evaluate the efficacy of the interventions. This review demonstrates that parental wellbeing is not routinely evaluated in relation to ASD interventions.

A literature review by Kuhaneck, Madonna, Novak & Pearson (2015) explored parent and family outcomes (such as parent self-efficacy, family coping and resiliency, family participation in daily life and routines) of ASD interventions targeting a range of participants in 34 published studies from 2006 to April 2013. Parents, caregivers and children with ASD were the participants in all but one study, which included professionals (such as occupational therapists) as participants. The focus of this review was on the effectiveness of interventions within the scope of occupational therapy practice. This review used broad search terms, which returned a total of 4,457 references (see appendix A for search terms). Some of the search terms, such as 'stress reduction', may have resulted in bias as this is likely to return papers

that found a positive reduction in stress, as opposed to those that found no changes in stress or an increase in stress. Authors report that articles were reviewed on the basis of predetermined inclusion and exclusion criteria to determine final inclusion in the review, but the inclusion and exclusion criteria are not reported, which makes replication of this review problematic. Kuhaneck et al. (2015) did not attempt to uncover unpublished literature in their review; which increases the likelihood that the review may be prone to publication bias. Furthermore, the quality of the papers that were reviewed varied greatly, with the overall risk of bias rated as 'high risk'. In terms of the results from the review, these were mixed and inconclusive and suggestive of a body of literature in its infancy. The results highlighted limited evidence to suggest that interventions were effective in improving broad family and parental outcomes or those specific to emotional wellbeing. More positively, Kuhaneck et al. (2015) found some evidence that centre-based interventions can improve parental confidence and self-efficacy, though it is not clear whether this is to a significant degree or not.

Since April 2013, there has been a disproportionately high number of papers published investigating parent outcomes of ASD interventions. Despite the increase in research in this area, there is currently no guidance on what interventions are associated with improved outcomes (e.g. stress, self-efficacy, quality of life) in parents of children with ASD. An updated overview and synthesis of existing literature examining the effects of interventions in ASD on parent outcomes is required to help improve the quality of information available to health care professionals and service providers. Due to the inconclusive results of the literature review undertaken by Kuhaneck, Madonna, Novak and Pearson (2015), the current review will adopt a narrower focus; specifically examining the evidence and the quality of evidence relating specifically to the effectiveness of parent interventions on parent outcomes. Thus, the focus of this review will differ from that of Kuhaneck, Madonna, Novak and

Pearson's, in that the emphasis will be on parent interventions, as opposed to ASD interventions targeting children, parents/caregivers and professionals more broadly.

2. METHOD

2.1 Electronic database search

Five databases (PsychARTICLES, PsychINFO, MEDLINE, EMBASE and Health Management Information Consortium) were searched using terms synonymous with parent interventions, ASD and parent/carer wellbeing. Searches were limited to peer reviewed journals published between January 2000 and December 2015. Table 1 shows the search terms used and the construct that each maps on to.

Table 1. ‘Keyword’ searches in relation to the effects of parent interventions in an ASD population on parent outcomes.

Search Number	‘Keyword’ Searched	Construct
1	Autis* spectrum disorder* ASD Autis*	Autism spectrum disorder
2	Parent* intervention* Parent* group* Parent mediated intervention* Parent* program* Parent training	Intervention involving parents
3	Parent* wellbeing Parent* stress Parent* outcome* Parent* self-efficacy Parent* mental health	Parent outcomes

The original search returned a total of 167 papers, including duplicates (see Figure 1). In an attempt to uncover so-called unpublished literature, contact was made with the National Autistic Society and Barnardo’s, as these organisations have known involvement with the ASD population, as well as being involved in the development, commissioning and evaluation/research of programmes relating to ASD interventions. The National Autistic Society and Barnardo’s are both UK organisations. One of the two organisations replied to the

request for research evaluations that had not been published, but the additional 2 papers that were generated did not meet the eligibility criteria for the review, as they did not report parent outcomes.

Screening criteria (i.e. full text journal article, published in English) were applied to the references returned in the electronic database search. Titles, and where necessary, abstracts and full papers were read and the following inclusion and exclusion criteria were applied to potentially relevant papers:

Inclusion criteria:

- Evaluation of the effects of ASD interventions where parents/primary caregivers are the sole recipients of the intervention.
- Papers report parental outcome data.
- Papers report primarily quantitative data; at least half the data collected and reported upon is quantitative data.

Exclusion criteria:

- Parents were not the sole recipients of the intervention.
- Multi-component programmes (which might include other support) to limit the number of confounding variables when comparing findings across different studies.
- Interventions directly involving the child with ASD (child interventions or parent and child interventions).
- Papers that reported mostly qualitative data (to allow comparisons to be made between studies in relation to methodological quality).

2.2 Searching references

To ensure all relevant papers were identified, the reference list of each of the papers that met the inclusion and exclusion criteria were hand searched for additional papers that met the eligibility criteria. In addition, forward searches were carried out using Web of Science, to identify relevant studies that referenced the papers that met eligibility criteria. The backward and forward searching yielded three additional papers. From the original searches, and the forward and backward searching, a total of 17 papers were identified that satisfied the criteria for inclusion in the review.

Due to the large number of papers published (N=12) since Kuhaneck, Madonna, Novak and Pearson's review investigating parent outcomes of parents interventions in ASD, a second electronic database search was conducted using the exact search terms identified in their review paper (Appendix A) and the same databases identified for the original search. The second search was limited to peer reviewed journals published between April 2013 and December 2015; this yielded an additional five papers that satisfied the current review's eligibility criteria. Combining papers from both the original search and the replication search, a total of 22 papers were included for review.

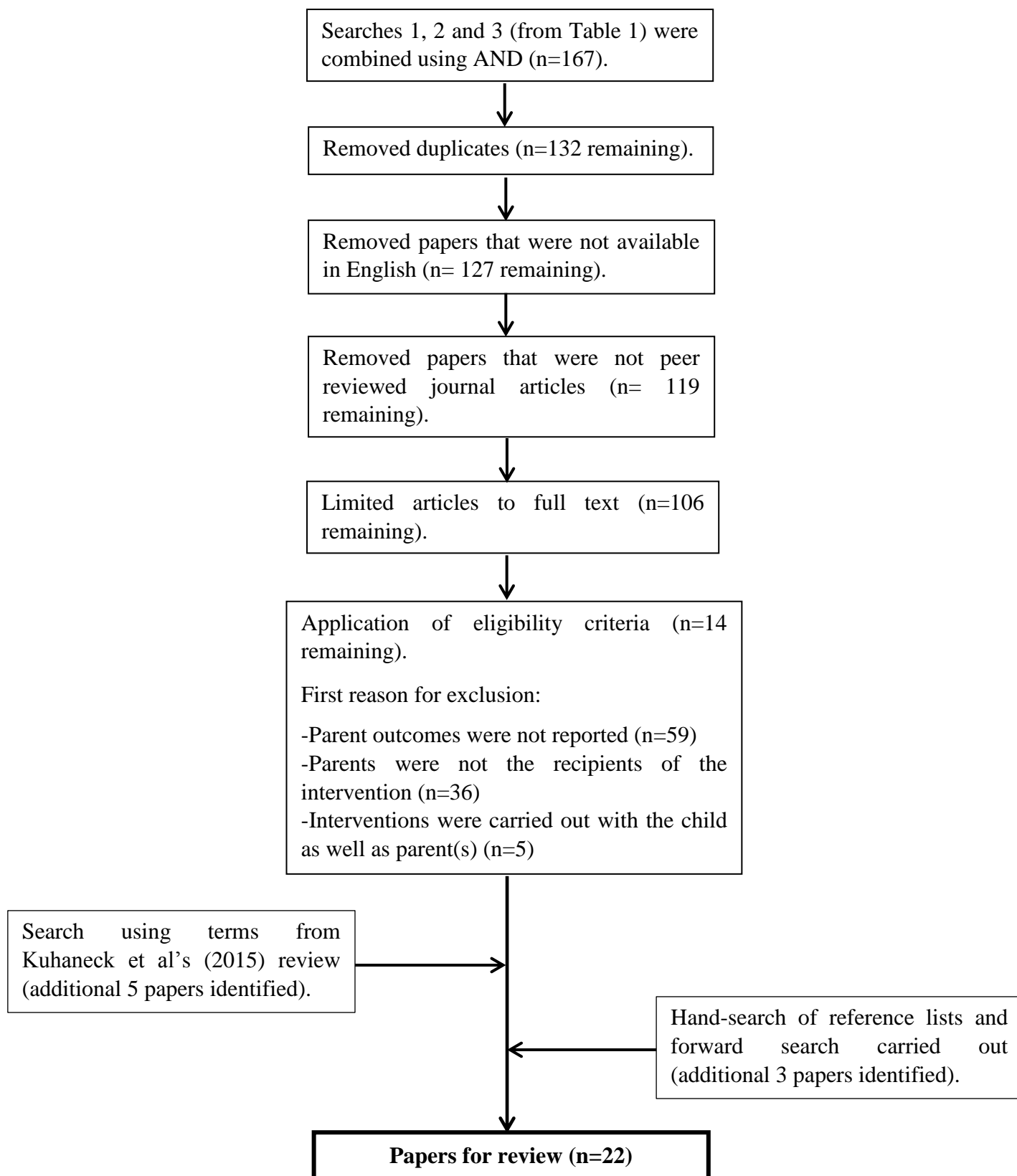


Figure 1. Search strategy employed and articles obtained

2.3 Data extraction

Data were extracted from each of the papers using a tool developed for the purpose of this review (Appendix B). The data extraction tool was adapted from guidelines for undertaking reviews in health care, developed by the NHS Centre for Reviews and Dissemination (University of York, 2009). Extracted data included information relating to sample size, design, outcome measures, key findings and limitations.

3. RESULTS

22 papers were identified for inclusion in this review. Six of the papers included in this review were also included in Kuhaneck, Madonna, Novak and Pearson's (2015) review (Bendixen et al, 2011; Keen, Couzens, Muspratt & Rodger, 2010; Solomon, Ono, Timmer & Goodlin-Jones, 2008; Tonge et al, 2006; Whittingham, Sofronoff, Sheffield & Sanders, 2009; Wong & Kwan, 2010). Kuhaneck, Madonna, Novak and Pearson's (2015) review included papers that investigated the effects of ASD interventions targeted towards parents, caregivers, children with ASD and professionals on parent and family outcomes, whereas the current review includes studies that evaluate interventions solely targeting parents and caregivers, which report on parent outcomes. The studies that met the eligibility criteria varied in theoretical approach, delivery of the intervention, outcome measures, key findings and quality of methodology. Key characteristics are summarised in Table 2.

Table 2. Data extraction for the 22 studies included in the review (in alphabetical order).

Author (Year) Country N recruited Sample	Intervention	Details
Al-Khalaf, Dempsey & Dally (2014) <i>Jordan</i> N = 10 mother father dyads Participants were mothers of preschool aged children with a diagnosis of ASD and their marital partner (the father in all cases). All children were being educated in special education centres.	<p>Psychoeducational intervention (4 x 4 hour group sessions over 4 week period), which only included mothers. Each group consisted of 5 mothers.</p> <p>The program provided parents with information about ASD:</p> <p>Session 1) triad of impairments associated with ASD.</p> <p>Session 2) social & communication difficulties.</p> <p>Session 3) repetitive behaviour & sensory issues.</p> <p>Session 4) strategies to help children during play time & ways to manage sleeping & eating problems.</p>	<p>Outcome measures:-Coping Strategy Indicator (wording changed from 6 months to 6 weeks)</p> <p>-Parent Stress Index - Short Form</p> <p>*All outcome measures were translated into Arabic by the first author and accuracy was checked by a doctoral student who was a native speaker of Arabic.</p> <p>Design: Within subject, repeated measures design.</p> <p>Assessment points: pre-intervention & 1 week post intervention (PI). Data for fathers was only collected pre-intervention.</p> <p>Key findings:-Statistically significant reduction in parenting stress (PI) in mothers.</p> <p>-Statistically significant increase in parental coping skills (PI) in mothers.</p> <p>Limitations: 1) No control group 2) Small sample size 3) No long term follow-up 4) Participants were recruited through two private centres in Jordan. 5) No assessment to confirm ASD diagnosis. 6) Participant demographics not reported. 7) No treatment fidelity checks were reported.</p>
Bendixen et al. (2011) USA N = 38 Mothers and fathers of 19 children, aged between 3 and 8 years, who had a diagnosis of 'autistic disorder' according to DSM-IV criteria. Children	<p>12 week in home training program designed to promote social interaction & parent-child reciprocity (length & number of sessions were not specified). Fathers were trained to use 4 strategies (based on social interaction theory) & then trained the mothers:</p> <p>1) Following the child's lead in play.</p> <p>2) Imitating the child's vocalisations & movements in an exaggerated, lively</p>	<p>Outcome measures:-Parenting Stress Index – Short Form</p> <p>-Family Adaptability & Cohesion Evaluation Scales II (FACES-II)</p> <p>Design: Within subject, repeated measures design.</p> <p>Assessment points: pre intervention & following last in-home session.</p> <p>Key findings:-Post intervention there was a statistically significant reduction in mothers' parenting stress.</p> <p>-Post intervention fathers' parenting stress reduced, but no statistically significant differences were found. Though PSI scores demonstrated that fathers were experiencing clinically significant stress levels prior to the intervention & each</p>

also had to score above the cut-off in each subscale of the Autism Diagnostic Interview Revised & Autism Diagnostic Observation Schedule. Children were excluded if they had physical or sensory impairments.	<p>& animated manner.</p> <p>3) Providing clear signals (i.e. facial expressions, verbal requests, labelling).</p> <p>4) Commenting on the child's actions, responses or verbalisations.</p>	<p>of these score fell below the clinically significant range post intervention.</p> <p>- There were no statistically significant changes in Faces-II scores post intervention.</p> <p>Limitations: 1) No control group 2) No long term follow-up 3) Homogeneous sample – 28 of the 38 parents who participated were Caucasian and 18 out of 19 children were male. 4) Small sample size 5) No treatment fidelity checks were reported.</p>
<p>Blackledge & Hayes (2006)</p> <p><i>USA (California)</i></p> <p>N = 20</p> <p>Parents/guardians (15 females, 5 males) of children diagnosed with autism. Ages of children are not reported.</p> <p>No assessment of autism was carried out.</p>	<p>2 day (14 hour) acceptance and commitment training group workshop.</p> <p>The workshop involved instruction, group participation and experiential exercises drawn largely from Hayes et al. (1999).</p> <p>Fidelity checks were carried out by trained raters. The time period between each workshop is not clear.</p>	<p>Outcome measures:-Global Severity Index from the Brief Symptom Inventory (BSI)</p> <p>-Beck Depression Inventory –II (BDI-II)</p> <p>-General Health Questionnaire-12 (GHQ-12)</p> <p>-Parental locus of control scale</p> <p>Design: Within subject, repeated measures design.</p> <p>Assessment points: 3 weeks before the intervention, 1 week before the intervention, 1 week after each workshop & 3 months after each workshop. By completion of the study, 11% of assessments had not been returned.</p> <p>Key findings:-Statistically significant pre to follow-up improvements were found for the BDI-II, BSI & GHQ-12, with improvement occurring primarily in participants in the clinical range or just below.</p> <p>Limitations: 1) No control group 2) Small sample size 3) No assessment to verify diagnosis of autism 4) Four participants did not receive the entire intervention, as workshops were cut short due to medical conflicts during the last 2 hours of each day.</p>
<p>Chiang (2014)</p> <p><i>USA (New York)</i></p>	<p>10 week parent education group (each session was 2 hours long).</p>	<p>Outcome measures:-Parenting Stress Index (PSI)</p> <p>-Confidence Degree Questions for Families (CDQ)</p> <p>-World Health Organisation Quality of Life-Brief</p>

<p>N = 11</p> <p>9 families of Chinese American children (all boys) with an ASD diagnosis, who were between the ages of 3 and 11 years. This included 9 mothers and 2 fathers.</p> <p>No ASD assessment was carried out.</p>	<p>Each session consisted of 4 segments:</p> <ol style="list-style-type: none"> 1) Lectured topic based on parents' interests. 2) Group discussions & role plays, associated with weekly topics. 3) Parents shared personal experiences of parenting a child with an ASD. 4) Parents shared information they thought would be useful. 	<p>Design: Within groups, repeated measures. Assessment points: pre and post intervention (no further details reported).</p> <p>Key findings:-Statistically significant reduction in parenting stress post-intervention. -Statistically significant improvement in parental confidence -Statistically significant improvement in quality of life in the domains of physical health & environment.</p> <p>Limitations: 1) No control group 2) Small sample size 3) Homogeneous sample (all participants were Chinese American, as too was the facilitator of the program) 4) No long term follow-up 5) No assessment to verify diagnosis of ASD 6) No treatment fidelity checks</p>
<p>Farmer & Reupert (2013)</p> <p><i>Australia</i></p> <p>N=98</p> <p>98 parents or family members of children diagnosed with autism. A total of 79 children were represented. 66% of the children were aged 2-6 years and 34% were older than 6 years.</p> <p>102 started the program</p>	<p>6 week parent education group (each session was 2 hours long). The program consisted of:</p> <p>Week 1-what is autism? Week 2-sensory processing in autism. Week 3-communication in autism. Week 4-social understanding in autism. Week 5-a parents' perspective Week 6-understanding & behavioural strategies (from physical receptive environment)</p> <p>Group numbers ranged from 5 to 16 participants.</p>	<p>Outcome measures:-Self-constructed questionnaire, consisting of 15 questions based on program aims.</p> <p>Design: Within groups, repeated measures. Assessment points: pre and post intervention (no further details reported).</p> <p>Key findings:-Statistically significant reduction in parental anxiety scores. -Statistically significant increase in parental knowledge of ASD. -Statistically significant increase in confidence scores.</p> <p>Limitations: 1) No control group 2) No long term follow-up 3) No reliability/validity data for the self-constructed questionnaire; no standardised measure used. Response bias associated with Likert scales. 4) No assessment to verify diagnosis of autism.</p>

<p>& 98 attended 4 weeks or more.</p> <p>No autism assessment was carried out.</p>		<p>5) Limited demographic data collected; ethnicity and socioeconomic background not recorded.</p> <p>6) No treatment fidelity checks</p>
<p>Feinberg et al. (2014)</p> <p><i>USA (Boston)</i></p> <p>N = 122</p> <p>Mothers of 122 children (mean age 34 months) who had recently (no report of how recently) received a diagnosis of ASD. Diagnosis of ASD was made by a qualified, licenced healthcare provider, most often (92%) using the Autism Diagnostic Observation Schedule (ADOS).</p>	<p>Problem solving education, a manualised cognitive behavioural intervention (6 individual sessions, lasting 30-45 minutes each). It is not clear how frequently sessions took place.</p> <p>During each session, mothers work with a trained interventionist to identify a single, measurable problem. They then set goals, brainstorm and evaluate solutions, choose a solution and create an action plan.</p>	<p>Outcome measures:-Parenting Stress Index – Short Form</p> <ul style="list-style-type: none"> -Quick Inventory of Depressive Symptomatology -Brief Coping Orientation to Problems -Parent/primary caregiver form of the Communication Scale of the Adaptive Behaviour Assessment System <p>Design: Randomised clinical trial</p> <p>Assessment points: Baseline & 3 months after the first intervention session (intervention group) or baseline & 3 months after baseline assessment (usual care group).</p> <p>Key findings:-At 3 month follow-up, mothers in the intervention condition were significantly less likely than those receiving usual care to have clinically significant levels of parenting stress.</p> <ul style="list-style-type: none"> -The reduction in mean depressive symptoms was statistically significant in the intervention condition, but the reduction in clinically significant depressive symptoms did not reach significance. <p>Limitations: 1) No long term follow-up.</p>
<p>Ferraioli & Harris (2013)</p> <p><i>USA (New Jersey)</i></p> <p>N=15</p> <p>10 mothers and 5 fathers of children under the</p>	<p>8 week mindfulness group compared with an 8 week skills-based parent training group (both 2 hours long).</p> <p><i>Mindfulness-based parent training sessions</i> 1) introduction to mindfulness, 2) non-judgmental acceptance, 3) identifying thoughts, 4)</p>	<p>Outcome measures:-Parenting Stress Index – Short Form</p> <ul style="list-style-type: none"> -General Health Questionnaire -Mindful Attention Awareness Scale -Applied Behaviour Analysis Questionnaire (developed for purpose of the study). <p>Design: Randomised comparative efficacy study.</p> <p>Assessment points: Pre-treatment, immediately post-treatment & 3 months after the completion of the treatment.</p>

age of 18, diagnosed with an ASD by a professional external to the study, according to DSM-IV criteria (mean age not reported).	accepting/distancing from thoughts, 5) staying present, 6) staying present, 7) pleasant events, 8) being effective. <i>Skills based parenting training sessions</i> 1) introduction – psychoeducation & behavioural principles, 2) reinforcement, 3) direct instruction, 4) naturalistic teaching – incidental teaching & pivotal response training, 5) social skills & play, 6) functional assessment, 7) decreasing behaviour, 8) punishment procedures.	Key findings: -Statistically significant improvement in parenting stress & global health outcomes at post intervention in the mindfulness group, though improvements in parental stress were not sustained at follow-up. -No statistically significant difference in parental stress or global health outcomes post intervention or at follow-up for the skills group. Limitations: 1) Small sample size. 2) 40% attrition in mindfulness group. 3) Analysis (t-tests) underpowered given the sample size. 4) Homogeneous sample; socioeconomic background was primarily upper-middle class, all participants had graduated college & 60% had advanced degrees. 5) No assessment to verify diagnosis of autism.
Giarelli, Souders & Pinto-Martin, Bloch & Levy (2005) <i>USA (Philadelphia)</i> N=31 31 parents of children 5 years of age or younger, diagnosed with ASD within the previous 7 days.	Brief individual nursing intervention (counselling, instruction & assistance – lasting 3 hours in total). This consisted of a 1 hour telephone call, scheduled within one week of receiving the diagnosis and a 2 hour home visit 3 months after the diagnosis with a paediatric nurse practitioner. The intervention was designed to assist parents in the implementation of the treatment plan, assist in problem solving & manage the social and emotional stresses associated with an ASD diagnosis.	Outcome measures: -Impact of Events Scale -Perceived Stress Scale Design: Randomised controlled trial. Assessment points: 1 week post diagnosis and 4 months post diagnosis. Key findings: -Differences between the intervention group & the TAU group on parental distress were not statistically significant. Limitations: 1) Small sample size. 2) Homogeneous sample; 82% were white, all participants resided in suburban Philadelphia, 94% had some college education, 71% reported having at least middle to upper earnings. 3) Does not report the assessment method or criteria used to diagnose ASD.
Hodgetts, Savage & McConnell (2013)	Stepping stones triple p (individual, 10 session format, with each session	Outcome measures: -Depression-Anxiety-Stress Scale -Parenting self-efficacy questionnaire -Supports & Services Questionnaire (developed for the

<p><i>Canada</i></p> <p>N = 15</p> <p>10 families (10 mothers and 5 fathers) of children aged 5-12 years of age, diagnosed with autism, confirmed with the ADOS (obtained through clinical records).</p>	<p>lasting approximately 1 hour).</p> <p>Session structure:</p> <ol style="list-style-type: none"> 1) Introduction & intake interview. 2) Observation of parent-child interaction & feedback. 3) Promoting children's development – parenting strategies. 4) Managing misbehaviour – parenting strategies. 5-7) Practice sessions. 8) Planned activities training – planning for high risk situations. 9) Planned activities training – planning for high risk situations. 10) Closure – family survival tips discussed. 	<p>purpose of this study)</p>
		<p>Design: Multiple case study design.</p> <p>Assessment points: Pre & post intervention (no further information reported).</p>
		<p>Key findings:-Statistically significant improvement in parental self-efficacy scores post intervention.</p> <p>-Post-intervention data was only obtained on the DASS for 3 participants. Scores improved for 2 of the 3 participants across scales. No statistical analysis was conducted with the DASS data.</p>
		<p>Limitations: 1) Small sample size (only 6 families completed the intervention).</p> <p>2) Missing data.</p> <p>3) No control group.</p> <p>4) No long term follow-up.</p> <p>5) Some participants were receiving multi-disciplinary input alongside the intervention.</p> <p>6) No statistical analysis for wellbeing data.</p>
<p>Kasari, Gulsrud, Paparella, Hellemann, Berry (2015)</p> <p><i>USA (Los Angeles)</i></p> <p>N=86</p> <p>86 parent-child dyads. All of the children were between the ages of 22 and 36 months, with a diagnosis of ASD, confirmed by independent testers with</p>	<p>Psychoeducational intervention (PEI) compared with JASPER parent mediated model (both interventions were carried out individually – 1 hour per week)</p> <p>The PEI included information on autism, details of specific behavioural impairments, principles of managing behaviour, strategies for teaching new skills, improving social interaction and communication, service availability, managing parental stress,</p>	<p>Outcome measures:-Parenting Stress Index</p> <p>-Mullen Scales of Early Learning</p> <p>-Reynell Developmental Language Scales</p>
		<p>Design: Randomised comparative efficacy study</p> <p>Assessment points: Pre-treatment, post-treatment & 6 month follow-up.</p>
		<p>Key findings:-Statistically significant reduction in child-related parenting stress in the psychoeducation condition compared to the JASPER condition.</p>
		<p>Limitations: 1) Narrow age range of children with ASD.</p> <p>2) All participants had received a 30 hour early intervention program in addition to PEI or JASPER, so conclusions cannot be drawn about the effects of PEI and JASPER alone.</p> <p>2) The paper does not report its' limitations.</p>

the Autism Diagnostic Interview-Revised.	<p>and sibling, family and community responses to autism.</p> <p>In the JASPER intervention, parents were taught to recognise the child's current developmental level of play and use of social-communication gestures. Capitalising on the child's current level of play and interests, caregivers provided opportunities for the child to initiate in a toy/activity and to establish jointly engaged play routines. Parents were taught a number of strategies to keep their child engaged and improve their frequency of social-communication gestures, spoken words and play acts.</p>	
<p>Keen, Couzens, Muspratt & Rodger (2010)</p> <p><i>Australia</i></p> <p>N=76</p> <p>39 families (total of 76 mothers & fathers) who had a child aged between 2 and 4 years with a diagnosis of ASD, based on DSM-IV criteria, received within</p>	<p>Parent focussed intervention providing parent education & parenting strategies.</p> <p>Families were assigned to a professionally supported intervention which consisted of a 2-day parent group workshop and a series of 10 home-based consultations with a facilitator, or a self-directed video intervention, covering the same information: parent education on autism, social interaction,</p>	<p>Outcome measures:-Parenting Stress Index</p> <ul style="list-style-type: none"> - Parenting Sense of Competence -Scales of Independent Behaviour Revised-early development form <p>Design: Comparative efficacy study</p> <p>Assessment points: Pre-intervention and 3 months after the completion of the intervention.</p> <p>Key findings:-Child-related parental stress scores reduced in the professionally supported condition relative to the self-directed condition (does not report whether the reduction was statistically significant).</p> <ul style="list-style-type: none"> -No overall effect on parenting satisfaction. -The professionally supported intervention was associated with increased self-efficacy when parents were scoring low in self-efficacy

6 months prior to participation in the study.	communication, play, sensory issues, behaviour, strategies to improve social interaction and communication, embedding strategies within daily routine, using a balanced approach, and selecting a child-focussed early intervention program.	prior to the intervention (no further detail reported). Limitations: 1) Children included in the study could be receiving up to 20 hours per week of services, which could confound the results. 2) Lack of random assignment. 3) Majority of the sample were well educated parents.
Minjarez, Mercier, Williams, Hardan (2012) USA N=24 24 parents (2 parents from 7 families, 1 parent from 10 families) of children between 2 and 6.11 years of age with a diagnosis of an ASD, who have documented evidence of language delays. Autism spectrum diagnosis was confirmed during a 2 hour clinical evaluation with a licensed psychologist, based on DSM-IV criteria.	10 week pivotal response group training program (PRT - 90 minute sessions). The specific content of the PRT included: 1) Presenting clear opportunities to respond while maintaining the child's attention. 2) Varying tasks (tasks the child has mastered and tasks the child has not yet mastered). 3) Incorporating child choice/shared control. 4) Providing immediate and contingent reinforcement in relation to target language behaviours. 5) Providing natural reinforcers. 6) Providing reinforcers following expressive verbal attempts & correct responses.	Outcome measures: -Parenting Stress Index– Short Form -Family Empowerment Scale
		Design: Within group, repeated measures. Assessment points: Pre and post intervention (no further information provided).
		Key findings: -Parents reported significantly lower levels of parenting stress post-intervention. -Parents reported statistically higher levels of empowerment post intervention.
		Limitations: 1) No control group. 2) No long term follow-up. 3) Small sample size. 4) Other treatments and amount of PRT used in the home was not controlled for. 5) Missing data (only 17 complete sets of data were collected and included in analysis).

<p>Ruiz-Robledillo & Moya-Albiol (2015)</p> <p><i>Spain</i></p> <p>N=17</p> <p>17 parents (10 women, 7 men) of people diagnosed with ASD. Parents were the primary caregiver and lived at home with the individual with ASD.</p>	<p>Cognitive behavioural intervention program (8 x 2 hour sessions once per fortnight plus homework tasks in between sessions – not clear whether intervention was delivered individually or in a group):</p> <ol style="list-style-type: none"> 1) Introduction, psychoeducation (ASD, stress), introduction to CBT. 2) Cognitive restructuring, leisure activities, diaphragmatic breathing techniques. 3) Importance of self-esteem, importance of leisure activities I, progressive muscle relaxation. 4) Importance of leisure activities II, acceptance vs. resignation progressive muscle relaxation II. 5) Communication styles – aggressive, passive & assertive. 6) Problem solving technique. Concept of locus of control. 7) Review & elaboration of self-care plan. 8) Review of self-care plan implementation, identification of barriers to program implementation, benefits of the program. 	<p>Outcome measures:-Profile of Mood States -Spanish version of the Somatic Symptoms Scale -Beck Depression Inventory -Burden Interview</p> <p>Design: Within group, repeated measures. Assessment points: Burden was evaluated pre & post intervention. Somatic symptoms & depression were evaluated before, immediately after the intervention and at 1 month follow-up. Mood state was evaluated pre-treatment, mid-treatment (session 4), post-intervention and at 1 month follow-up.</p> <p>Key findings:-Statistically significant reduction in burden post intervention. -Statistically significant reduction in depressive symptoms at follow-up. -Statistically significant improvements in mood states over time.</p> <p>Limitations: 1) No control group. 2) No long-term follow-up. 3) Small sample size. 4) No assessment to confirm diagnosis of ASD. 5) Age of offspring with ASD was not reported. 6) Homogeneous sample; majority of participants were married with a high level of education.</p>
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<p>Ruiz-Robledillo et al. (2015) <i>Spain</i> N=13 13 participants; 6 parents of adolescents with a diagnosis of ASD & 7 parents of age matched typically developing adolescents.</p>	<p>Mindfulness based group program over 9 sessions (each session lasting approximately 2 hours).</p> <p>Session content was as follows:</p> <ol style="list-style-type: none"> 1) Introduction to the contents of the program, principles underlying automatic pilot were explained & a short meditation was conducted. 2) Body scan, barriers to mindfulness & principles of the cognitive model of mindfulness. 3) Breathing meditation, differentiating between thoughts and facts & the importance of recalling pleasant events. 4) Stress, reactivity and parenting. Long sitting meditation. 5) Acceptance & tolerance. Nature sound meditation. 6) Emotional debt & spaciousness. Walking meditation. 7) Metta & mountain meditation. Discussion on compassion, self-compassion & self-care. 8) Review with short metta & body scan meditations. 9) Practice of metta meditation and meditation without object. <p>Homework was given after each</p>	<p>-State-Trait Anxiety Inventory (Spanish version) -Abbreviated version of the Profile of Mood States -State-Trait Anger Expression Inventory (Spanish version) -Somatic Symptoms Scale (Spanish revised version) - General Health Questionnaire -Beck Depression Inventory -Zarit Burden Inventory</p> <p>-Saliva was collected from participants to measure cortisol levels.</p> <p>Design: Between groups, repeated measures. Assessment points: Pre-treatment (session 1), mid-treatment (session 5) & post-treatment (session 9).</p> <p>Key findings:-Statistically significant reduction in mood disturbances and afternoon cortisol levels during the sessions in all participants, with reductions being greater in caregivers. -Cortisol levels did not change significantly from pre to post-intervention. -All participants showed significantly fewer depressive and somatic symptoms at the end of the program, with an improvement in their self-perceived general health</p> <p>Limitations: 1) Small sample size 2) Lack of waiting list control. 3) No long term follow-up. 4) No assessment to confirm diagnosis of ASD. 5) Main method of recruitment was through an association of relatives of people with ASD. This sample may not be representative of the general population.</p>
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	session.	
Singh et al. (2014) <i>USA</i> N=3 3 mothers and their adolescent sons diagnosed with Asperger syndrome. Each of the mothers had a history of working with behaviour analysts.	Mindfulness-based positive behaviour support (MBPBS) training. Participants were taught the basic Samantha meditation during a 2 hour individual pre-training session. Following this, participants were instructed to develop a personal meditation practice, beginning with a few minutes each day and increasing it to 30 minutes practice per day. The intervention phase consisted of 1 day individual training per week for 8 weeks. This included training on the 'four immeasurables' (loving-kindness, compassion, joy and equanimity), the 'three poisons' (attachment, anger and ignorance), Shenpa and compassionate abiding and soles of the feet meditation. Throughout the training, participants were taught how to use PBS within the context of mindfulness. The MBPBS training phase immediately followed the intervention	Outcome measures:- Perceived Stress Scale-10 -iPhone application that enabled recording of child's behaviour in real time.
		Design: Multiple baseline design across participants. Assessment points (for perceived stress): Pre-baseline, first day of baseline, last day of MBPBS training & last day of MBPBS practice.
		Key findings:- Statistically significant reductions in mothers' stress levels were correlated with the intervention.
		Limitations: 1) Small sample size. 2) No control group. 3) Participants were stressed mothers, which may not be representative of all mothers of children with ASD. 4) The intervention is not a stand-alone intervention; mothers had a history of working with behaviour analysts and the intervention built on existing knowledge of behavioural interventions. 5) Data on each child's behaviour was collected via an iPhone application, by mothers who were not blind to the purpose of the study. 6) No assessment to confirm the diagnosis of Asperger syndrome.

	phase and lasted until week 48 of the study. During this phase, mothers were requested to continue with all meditation practices they had learned and any questions were answered fully by trainers.	
Sofronoff & Farbotko (2002) <i>Australia</i> N=89 45 mothers and 44 fathers of children (aged between 6 and 12 years) recently (it is not evident how recently) diagnosed with Asperger syndrome, by a paediatrician and a psychologist, using DSM-IV criteria.	Parent management training (delivered in a 1 day workshop or 6 weekly 1 hour sessions). The following topics were covered during both intervention groups: 1) Psychoeducation relating to Asperger syndrome. 2) Comic strip conversations. 3) Social stories. 4) Management of behaviour problems. 5) Management of rigid behaviours, routines & special interests. 6) Anxiety management (ways to manage the child's anxiety).	Outcome measures:- Parental self-efficacy in the management of Asperger syndrome questionnaire (developed specifically for this study) -Eyberg Child Behaviour Inventory
		Design: Between groups, repeated measures. Assessment points: Pre-intervention, 4 weeks in to the 6 individualised sessions, 1 month after the workshop and 3 month follow-up.
		Key findings:- Statistically significant increase in mothers' self-efficacy in both intervention conditions. Changes were maintained at 3 month follow-up in the 6 session condition. -No changes in fathers' self-efficacy.
		Limitations: 1) Lack of randomisation. 2) Lack of follow-up data for waitlist control group. 3) No treatment fidelity checks. 4) Participant demographic data was not reported. 5) It is not clear how participants were allocated to the 1 day workshop or the 6 session intervention. 6) The parental self-efficacy questionnaire was developed for the purpose of this study and consequently psychometric properties have not been reported.
Solomon, Ono, Timmer & Goodlin-Jones (2008) <i>USA</i>	Parent-child interaction therapy (PCIT) (delivered individually, in two phases, spanning over a mean of 12.7 sessions).	Outcome measures:- Parenting Stress Index-Short Form -Eyberg Child Behaviour Inventory -Behaviour Assessment System for Children Parent Rating Scales

<p>N=19</p> <p>Parent(s) of 19 male subjects, aged 5-12 years, who met criteria for autistic disorder, Asperger syndrome or pervasive developmental disorder not otherwise specified according to the DSM-IV-TR; ASD or autism according to ADOS-G; and autistic disorder according to the Autism Diagnostic Interview-Revised.</p> <p>Children were excluded if they had a full scale IQ <70. Children had to demonstrate clinically significant externalising behaviour for inclusion.</p>	<p>The first phase of PCIT is child directed interaction. During this phase, parents are taught to be attuned to their child by giving positive attention and praise, by ignoring negative behaviour, and by not criticising, disciplining, making requests, giving commands or asking questions. Parents were coached until they reached 'mastery', which occurred within 8 sessions for all participants.</p> <p>The second phase of PCIT is parent directed interaction. During this phase, parents were coached to give clear, concise, direct, age appropriate, and simple commands, and to consistently reinforce compliant behaviour. Parents were also taught to use the time-out chair when children did not comply with parent requests. Parents completed this part of the intervention within 6 sessions.</p>	<p>*Shared positive affect was rated by two trained coders, using the Shared Positive Affect Coding Adapted.</p> <p>Design: Waiting-list control group design. Assessment points: Pre-intervention, midpoint and post-intervention.</p> <p>Key findings:-Statistically significant increase in shared positive affect between pre-intervention and midpoint and between pre- and post-intervention. -Statistically significantly increase in parent positive affect between pre-intervention and midpoint and between pre- and post-intervention. -No changes in parenting stress post-intervention.</p> <p>Limitations: 1) Small sample size. 2) No formal measure of treatment fidelity. 3) No long-term follow-up. 4) Demographic data is not reported for parents (only children).</p>
<p>Solomon, Van Ergen, Mahoney, Huber & Zimmerman (2014)</p> <p>USA</p>	<p>Play and Language for Autistic Youngsters (PLAY) Project home consultation program (3 hour monthly home visit for 12 months). The primary caregiver was targeted for instruction, but all caregivers were</p>	<p>Outcome measures:-Maternal Behaviour Rating Scale -Child Behaviour Rating Scale -Mullen Scales of Early Learning -MacArthur Communicative Development Inventories -Functional Emotional Assessment Scale -Parenting Stress Index</p>

<p>N=128</p> <p>128 families of children aged 2 years 8 months to 5 years 11 months, with a clinical diagnosis of ASD according to DSM-IV criteria and meeting criteria for autism or ASD on the ADOS and Social Communication Questionnaire.</p>	<p>welcome to attend.</p> <p>Parents/caregivers were trained to identify their child's subtle cues, respond contingently to the child's intentions and effectively engage in reciprocal exchanges. This is done through coaching, modelling and video feedback.</p> <p>Parents were encouraged to engage their child in 20-minute play sessions throughout the day, for a total of 2 hours per day.</p>	<p>-Center for Epidemiologic Studies Depression Scale</p> <p>Design: Randomised controlled trial.</p> <p>Assessment points: Prior to randomisation and 12 months after the intervention began (around conclusion of the intervention).</p> <p>Key findings:-Statistically significant improvement in interaction quality in the PLAY condition compared to the usual community services condition.</p> <p>-Parenting stress levels decreased over time in both groups, but changes were not statistically significant.</p> <p>-Reduction in depressive symptomatology in the PLAY group was only statistically significant when established cut-offs were applied.</p> <p>Limitations: 1) No long-term follow-up.</p> <p>2) Families were expressly allowed to obtain other interventions during participation in this study, which may have confounded results.</p> <p>3) The amount of community services received was not consistently documented by parent report, but the intervention group appeared to receive more community services than the control group.</p> <p>4) Parents were better educated and had a higher socioeconomic status than average.</p>
<p>Suzuki et al. (2014)</p> <p><i>Japan</i></p> <p>N=72</p> <p>72 mothers of pre-school (aged 2-6.5 years) children with high functioning pervasive developmental</p>	<p>Brief group psychoeducational program (4 sessions, each lasting 120 mins & held fortnightly). The number of participants in each group ranged between 3 and 5.</p> <p>During the first 60 minutes of each session, therapists presented information on 1) the characteristics of pervasive developmental disorders, 2) how to treat their children, taking</p>	<p>Outcome measures:-General Health Questionnaire-28 (GHQ-28)</p> <p>-Aberrant Behaviour Checklist</p> <p>-Zarit Caregiver Burden Interview</p> <p>-Medical Outcomes Study 36-item Short Form Health Survey</p> <p>Design: Randomised controlled trial.</p> <p>Assessment points: Baseline, post-intervention and 14 weeks post intervention.</p> <p>Key findings:-There is no evidence to suggest that the intervention alleviated maternal distress, child behaviour or caregiver burden.</p> <p>-Statistically significantly greater improvement in GHQ-28</p>

<p>disorders. To be included in the study, the child needed to have a diagnosis of autistic disorder, Asperger's disorder, or pervasive developmental disorder not otherwise specified, made by a child psychiatrist or developmental paediatrician, according to the DSM-IV, in addition to having an IQ over 70.</p>	<p>into account individual characteristics of each child, 3) information about social support resources & preparation for elementary school, 4) physical & mental health of the mother. During the latter 60 minutes, supportive group therapy was provided focussing on problem solving skills.</p>	<p>scores in the treatment as usual (TAU) group at 14 week follow-up than the intervention + TAU group.</p> <p>Limitations: 1) No treatment fidelity checks. 2) Other interventions were not controlled for, with three quarters of the participants' children in co-curricular therapy involved in other parent training programs. 3) No assessment to confirm diagnosis.</p>
<p>Tellegen & Sanders (2014) <i>Australia</i> N=64 64 parents/caregivers of a 2-9 year old child with an ASD diagnosis from a paediatrician or a child psychiatrist. Diagnoses were verified via a</p>	<p>Primary care stepping stones triple p (low intensity version of stepping stones triple p – delivered via 4 individual sessions, each designed to be 15-30 minutes long).</p> <p>The sessions were designed to address, one or two specific child problems, with the most common problems being aggression and non-compliance.</p> <p>More in-depth information about the</p>	<p>Outcome measures:-Family Background Questionnaire -Eyberg Child Behaviour Inventory -Parenting Scale -Depression, Anxiety & Stress Scale -Parenting Tasks Checklist -Parental Stress Scale -Parent Problem Checklist -Relationship Quality Index</p> <p>Design: Randomised controlled trial Assessment points: Pre-intervention, post-intervention (at approximately 8 weeks for intervention group and after 8 weeks for care as usual group) and 6 month follow-up.</p>

<p>semi-structured interview based on DSM-IV criteria.</p> <p>To be eligible, parents had to be concerned about social, emotional, behavioural or developmental problems in their child & could not be receiving parenting assistance.</p>	<p>content of the intervention is not provided.</p>	<p>Key findings:-Statistically significant short term effects on dysfunctional parenting styles, parenting confidence, parenting stress, parental conflict & relationship happiness</p> <p>-Statistically significant effects on dysfunctional parenting styles, parenting confidence and parental stress were maintained at follow-up.</p> <p>-No significant effects on parental anxiety or depression.</p> <p>Limitations: 1) Homogeneous sample; most children were Australian/White and from 2 parent families, with most families earning above the median Australian income.</p> <p>2) No assessment to verify ASD diagnoses.</p>
<p>Tonge et al. (2006)</p> <p><i>Australia</i></p> <p>N=105</p> <p>105 parents of children aged between 2½ and 5 years of age who obtained diagnosis of autism within the previous month. Each diagnosis was confirmed using a screen observation or video record of the interview by an independent clinician.</p>	<p>Parent education and behaviour management (PEBM) intervention (based on early intervention and cognitive behavioural techniques). Ten 90 minute group sessions alternated with ten 60 minute individual family sessions over a 20 week period.</p> <p>Intervention sessions included:</p> <ul style="list-style-type: none"> - Autism education. - Features of communication, social, play & behavioural impairments. - Principles of managing behaviour and change. - Teaching new skills. - Improving social interaction & 	<p>Outcome measures:-General Health Questionnaire-28 (GHQ-28)</p> <ul style="list-style-type: none"> -Parenting Stress Thermometer -McMaster Family Assessment Device (FAD) -Developmental Behaviour Checklist -Psychoeducational Profile-Revised <p>Design: Randomised controlled trial</p> <p>Assessment points: Pre-treatment, on completion of the intervention (20 weeks after the initial assessment for the non-intervention control group) & 6 months after the completion of the intervention.</p> <p>Key findings:-No statistically significant difference in overall mental health between PEBM and PEC groups.</p> <ul style="list-style-type: none"> -Statistically significant improvement in GHQ-28 scores at 6 month follow-up, when GHQ-28 total scores were above 18.29 pre-intervention in PEBM & PEC conditions. -Statistically significant improvement in parenting stress thermometer ratings and FAD scores at follow-up for those in PEBM & PEC groups when compared to non-intervention controls.

	<p>communication.</p> <ul style="list-style-type: none"> - Services available. - Managing parental stress, grief & mental health problems. - Sibling, family & community responses to autism. <p>*A parent education and counselling intervention (PEC) formed another treatment group, to control for the non-specific aspects of the PEBM intervention, such as therapist attention, understanding & support. The educational material was the same as for the PEBM group, however, no skills training or homework tasks were set.</p>	<p>Limitations: 1) There were pre-treatment differences in mental health between groups.</p> <p>2) Differences in regional services are not controlled for.</p>
<p>Whittingham, Sofronoff, Sheffield & Sanders (2009)</p> <p><i>Australia</i></p> <p>N=59</p> <p>59 families with a child aged between 2 and 9 years, with an ASD diagnosis from a paediatrician.</p> <p>A semi-structured</p>	<p>Stepping stones triple p group program (9 week program). Group format was used for the sessions that involved teaching parenting strategies & an individual format was used for sessions involving observation, practise & feedback.</p> <p>Session structure:</p> <ol style="list-style-type: none"> 1) Basic principles of positive parenting introduced. 2) Observation of parent-child interaction & feedback. 	<p>Outcome measures:-Eyberg Child Behaviour</p> <ul style="list-style-type: none"> -Parenting Scale -Being a Parent Scale <p>Design: Randomised controlled trial.</p> <p>Assessment points: Pre-intervention, post-intervention and 6 month follow-up for intervention group.</p> <p>Key findings:-Statistically significant improvements in parenting styles, with changes in over-reactivity & verbosity being maintained at 6 month follow-up.</p> <ul style="list-style-type: none"> -Statistically significant improvements in parental satisfaction & conflict about parenting at 6 month follow-up. -Statistically significant improvements in parental self-efficacy

<p>diagnostic interview, developed for the purpose of this study, was carried out to verify an ASD diagnosis.</p> <p>Each family was asked to nominate a primary participating parent to complete the intervention & outcome measures. The second parent was welcome to attend sessions.</p>	<p>3) Promoting children's development – parenting strategies. 4) Managing misbehaviour – parenting strategies. 5 & 6) Practice session. 7) Planned activities training – planning for high risk situations. Parents are also introduced to comic strip conversations & social stories (these strategies are not currently in stepping stones triple p). 8) Planned activities skills are practised & feedback given. 9) Closure – family survival tips discussed.</p>	<p>at alpha level of .05, but not significant at a more stringent alpha level of .025 (due to number of analyses – 2 MANOVAs).</p> <p>Limitations: 1) No follow-up data for control group. 2) Limited treatment fidelity checks (through supervision). 3) Homogeneous sample - most parents were married and living together, with at least one parent who was employed. 4) The majority of children were verbal.</p>
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3.1 Descriptive summary

Sample. The combined sample across the 22 papers was made up of 1,115 parents including guardians (Blackledge & Hayes, 2006) and family members other than parents (Farmer & Reupert, 2013). The individual sample sizes ranged between 3 participants (Singh et al., 2014) and 128 (Solomon, Van Ergen, Mahoney, Huber & Zimmerman, 2014), with a mean sample size of 51 participants ($SD=40.3$). Based on the studies that reported the number of mothers and fathers that participated, the majority of participants were mothers (69%).

The mean age of children with ASD ranged from 2.6 years to 16.7 years and the overall mean age of children across studies was 6.3 years (mean age was not reported in six studies). As is typical of ASD, the majority of children were male, with a mean of 84% of children being male across the studies that reported child gender ($n=13$). All but 7 children included in the studies had a diagnosis of an autism spectrum disorder. The inclusion of 7 typically developing children (Rui-Robledillo et al., 2015) was for the purpose of a control group. The age of parents/caregivers ranged between 34-52 years; overall mean age across studies was 39 years, although ten studies did not provide age data for parents/caregivers.

The identified studies were carried out in six countries, with a large number of studies being conducted in the USA ($n=11$) and Australia ($n=6$). As all of the studies recruited parents/caregivers of children with a diagnosis of ASD, recruitment typically took place through a health provider. One study sought out parents who were ‘stressed’ (Singh et al., 2014) and this study also targeted parents who were experiencing difficulties with their child’s behaviour.

Six of the twenty-two studies recruited homogenous samples, where the majority of participants were better educated and/or of a higher socio-economic status than the reported

average (Giarelli, Souders, Pinto-Martin, Bloch & Levy, 2005; Keen, Couzens, Muspratt & Rodger, 2010; Ferraioli & Harris, 2012; Tellegen & Sanders, 2014; Solomon, Van Ergen, Mahoney, Huber & Zimmerman, 2014; Ruiz-Robledillo & Moya-Albiol, 2015;). In one study, participant demographic data was not reported (Sofronoff & Farbotko, 2002), and in two studies limited demographic data were reported, with either no data relating to ethnicity or socioeconomic background (Farmer & Reupert, 2013) or no parent data reported; only child demographic data provided (Solomon, Ono, Timmer & Goodlin-Jones, 2008). Three studies recruited only mothers (Feinberg et al., 2014; Singh et al., 2014; Suzuki et al., 2014) and all but one study recruited only parents/caregivers of children/young people with ASD (Ruiz-Robledillo et al., 2015), where a proportion of participants were parents/caregivers of typically developing children and served as a control group.

Methodology. Seven studies employed a within-groups, repeated measures design, six studies were randomised controlled trials, three studies employed a between-groups repeated measures design (two of which included a waiting-list control), three studies were comparative efficacy studies (two of which involved randomisation), one study was a randomised clinical trial, and the remaining studies (n=2) employed a multiple case study design and a multiple baseline across participants design. Fourteen studies collected assessment data before and immediately after the intervention and two studies collected data before and within one week of completion of the intervention. Nine studies provided follow-up data at various time points: 1 month (n=1), 3 months (n=3), 14 weeks (n=1), and 6 months (n=4).

Interventions. The identified studies included a variety of interventions, which differed in length, mode of delivery and theoretical approach. Only one manualised intervention was included in the current review, namely Stepping Stones Triple P (Sanders,

Mazzucchelli, & Studman, 2003). For the purpose of this review, interventions have been grouped in to five categories based on the nature of the interventions: psychoeducation (n=7), parenting programmes (n=5), parent-mediated interventions (n=5), mindfulness interventions (n=3) and cognitive-behavioural interventions (n=2). Some studies compare two interventions, and so they appear in two different categories. Two interventions did not fit in to any one of the five derived categories: acceptance and commitment training and a brief nursing intervention.

Table 3. Intervention categories and interventions included in each category.

Author (Year)	Intervention	Intervention Category
Al-Khalaf, Dempsey & Dally (2014)	Psychoeducation	Psychoeducation
Chiang (2014)	Education program	Psychoeducation
Farmer & Reupert (2013)	Parent education program, incl. behaviour management strategies	Psychoeducation
Kasari, Gulsrud, Paparella, Hellemann & Berry (2015)	Psychoeducation vs. developmental behavioural intervention	Psychoeducation
Keen, Couzens, Muspratt & Rodger (2010)	Education & parenting strategies	Psychoeducation
Suzuki et al. (2014)	Psychoeducation group	Psychoeducation
Tonge et al. (2006)	Education & behaviour management vs. education & counselling	Psychoeducation
Ferraioli & Harris (2013)	Mindfulness vs. skills based parenting program	Parenting programme
Hodgetts, Savage & McConnell (2013)	Stepping stones triple P	Parenting programme
Sofronoff & Farbotko (2002)	Parent management training	Parenting programme
Tellegen & Sanders (2014)	Stepping stones triple P	Parenting programme
Whittingham et al. (2009)	Stepping stones triple P	Parenting programme
Bendixen et al. (2011)	Father-based in home intervention	Parent-mediated
Kasari, Gulsrud, Paparella, Hellemann & Berry (2015)	Psychoeducation vs. developmental behavioural intervention	Parent-mediated
Minjarez, Mercier, Williams, Hardan (2012)	Pivotal response training	Parent-mediated
Solomon et al. (2014)	PLAY project - home consultation - developmental relationship approach.	Parent-mediated
Solomon, Timmer & Goodlin-Jones (2008)	Parent-child interaction therapy	Parent-mediated
Ferraioli & Harris (2013)	Mindfulness vs. skills based parenting program	Mindfulness
Ruiz-Robledillo et al. (2015)	Mindfulness based group	Mindfulness
Singh et al. (2014)	Mindfulness based positive behaviour support training	Mindfulness
Feinburg et al. (2013)	CBT & problem solving education	CBT
Ruiz-Robledillo & Moya (2015)	Cognitive behavioural intervention	CBT
Blackledge & Hayes (2006)	Acceptance & commitment training	N/A
Giarelli et al. (2005)	Counselling, instruction & assistance with care plan	N/A

Quality Assessment. Each of the studies were reviewed using a quality framework (Kmet, Lee and Cook, 2004 (Appendix C)) to assess quality and credibility. Kmet, Lee and Cook's (2004) quality framework is a checklist, consisting of 13 items relating to the quality of quantitative research and each item is scored based on the degree to which it is met (yes=2, partial=1 or no=0). Items that are not applicable to the study design being assessed are rated as N/A and excluded from the summary score calculation. Summary scores are between 0 and 1, and higher scores are indicative of higher quality. Kmet, Lee and Cook's (2004) quality framework was selected because it allows for simultaneous evaluation of study quality, across disparate sources of evidence. Furthermore, this framework provides overall quality scores which are useful for synthesising information and making comparisons between studies in terms of their quality.

Quality ratings were used to evaluate the evidence the twenty-two studies provided in relation to the efficacy of parent interventions on parent outcomes. Five of the twenty-two papers (23%) were assessed for quality by a second rater to establish inter-rater reliability. Inter-rater agreement was 89% ($K=0.885$), which is indicative of 'very good agreement' (Altman, 1991). Items where disagreements occurred were discussed and a consensus was agreed.

Table 4. Quality Assessment Ratings

<div>Quality assessment questions</div> <div>Authors</div>	Question/objective sufficiently described?	Study design evident & appropriate?	Method of subject/ comparison group selection described & appropriate?	Subject (& comparison group if applicable) characteristics sufficiently described?	If interventional & random allocation was possible, was it described?	If interventional & blinding of investigators was possible, was it described?	If interventional & blinding of subjects was possible, was it reported?	Outcome measures well defined and robust to measurement/ misclassification bias? Means of assessment reported?	Sample size appropriate?	Analytic methods described/ justified and appropriate?	Some estimate of variance is reported for the main results?	Controlled for confounding?	Results reported in sufficient detail?	Conclusions supported by the results?	**Summary score
Al-Khalaf, Dempsey & Dally (2014)	Y	P	P	N	N/A	N	N/A	Y	N	Y	Y	N	Y	Y	0.58
Bendixen et al. (2011)	Y	Y	P	Y	N/A	N	N/A	Y	N	Y	Y	N	Y	P	0.63
Blackledge & Hayes (2006)	Y	Y	Y	Y	N/A	N	N/A	Y	N	Y	Y	P	Y	Y	0.73
Chiang (2014)	Y	P	N	Y	N/A	N/A	N/A	P	N	Y	Y	N	Y	Y	0.64
Farmer & Reupert (2013)	Y	Y	P	P	N/A	N	N/A	P	Y	P	Y	N	Y	P	0.58
Feinberg et al. (2014)	Y	Y	Y	Y	Y	Y	N	P	P	Y	Y	Y	Y	Y	0.86
Ferraioli & Harris (2013)	Y	P	Y	Y	Y	N	N	P	N	P	N	Y	Y	Y	0.61
Giarelli, Souders & Pinto-Martin, Bloch & Levy (2005)	Y	Y	Y	P	P	N	N	Y	N	Y	N	P	P	P	0.54
Hodgetts, Savage & McConnell (2013)	Y	Y	P	Y	N/A	N/A	N/A	Y	N	P	N	N	Y	Y	0.58
Kasari, Gulsrud, Paparella, Hellemann, Berry (2015)	Y	Y	Y	Y	Y	Y	N	P	P	Y	Y	Y	Y	P	0.82
Keen, Couzens, Muspratt & Rodger (2010)	Y	Y	Y	Y	N	Y	N	P	P	Y	Y	N	Y	Y	0.71
Minjarez, Mercier, Williams, Hardan (2012)	Y	Y	Y	P	N/A	N	N/A	Y	N	Y	Y	N	Y	Y	0.71
Ruiz-Robledillo & Moya-Albiol (2015)	Y	Y	Y	Y	N/A	N	N/A	Y	N	Y	Y	N	Y	Y	0.75
Ruiz-Robledillo et al. (2015)	Y	Y	P	Y	N/A	N	N/A	Y	N	Y	Y	N	Y	Y	0.71
Singh et al. (2014)	Y	Y	Y	Y	N/A	N	N/A	P	N	Y	N	N	Y	Y	0.63
Sofronoff & Farbotko (2002)	Y	N	P	P	N	N	N	P	N	Y	Y	N	Y	Y	0.46
Solomon, Ono, Timmer & Goodlin-Jones (2008)	Y	Y	Y	P	Y	Y	N	Y	N	Y	Y	Y	Y	Y	0.82
Solomon, Van Ergen, Mahoney, Huber & Zimmerman (2014)	Y	Y	Y	Y	Y	Y	N	Y	Y	Y	Y	P	Y	Y	0.89
Suzuki et al. (2014)	Y	Y	Y	Y	Y	N	N	Y	Y	P	Y	P	Y	Y	0.79
Tellegen & Sanders (2014)	Y	Y	P	Y	Y	N	N	P	Y	Y	Y	Y	Y	Y	0.79
Tonge et al. (2006)	Y	Y	Y	Y	Y	Y	N	P	Y	Y	Y	Y	Y	P	0.86
Whittingham, Sofronoff, Sheffield & Sanders (2009)	Y	Y	Y	Y	Y	N	N	Y	Y	Y	Y	Y	Y	Y	0.86

* Y = Yes, N = No, P = Partial, N/A = Not applicable

**Please see Appendix C for summary score algorithm.

Quality ratings in Table 3 were assigned as ‘strong’ (green, n=6 studies), ‘moderate’ (amber, n=11 studies) or ‘weak’ (red, n=5 studies), based on summary scores. Kmet, Lee and Cook (2004) do not provide an overall cut-off or threshold score for quality, so for the current review relative quality codings were developed such that those studies with a summary score equal to or below the 25th percentile were assigned a weak quality rating, those with a summary score between the 25th and the 75th percentile were assigned a moderate quality rating and those with a summary score equal to or above the 75th percentile were assigned a strong quality rating.

The quality assessment showed that areas of weakness within the current literature related to the lack of interventional and random allocation, lack of blinding (of participants and investigators), small samples, and lack of controlling for confounding variables, which is in line with the risks of bias identified in Kuhaneck, Madonna, Novak and Pearson’s (2015) review.

Table 5. Mean methodological quality ratings per category.

Intervention Category	Mean Quality Rating
Psychoeducation	0.71
Parenting programme	0.66
Parent-mediated intervention	0.77
Mindfulness	0.65
CBT	0.81

3.2 Interventions

Interventions ranged between 3 hours and 36 hours in length, with a mean intervention length of 12.9 hours (excluding four studies that did not clearly specify the length of sessions/the entire intervention: Bendixen et al., 2011; Keen, Couzens, Muspratt & Rodger,

2010; Solomon, Ono, Timmer & Goodlin-Jones, 2008; Whittingham, Sofronoff, Sheffield & Sanders, 2009).

i. Psychoeducation

The most commonly studied intervention was psychoeducation (n=7), but the content, format and length of psychoeducation differed in each study. In two of the seven studies psychoeducation was evaluated in comparison to another active intervention (Kasari, Gulsrud, Paparella, Hellemann & Berry, 2015; Tonge et al., 2006). The evidence for the effectiveness of psychoeducation programmes was mixed, which is likely to be reflective of the variety of psychoeducational interventions and the quality of studies evaluating psychoeducation programmes. Three studies investigating the effects of psychoeducation found a statistically significant reduction in overall parenting stress following the intervention (Al-Khalaf, Dally & Dempsy, 2014; Chiang, 2014; Tonge et al., 2006), though one study (Tonge et al., 2006) used a parenting stress thermometer (a visual analogue rating of general level of stress (rated from 0 to 4) to measure parental stress) and psychometric properties for this measure are not provided. Two additional studies found a reduction specifically relating to child-related parenting stress (Kasari, Gulsrud, Paparella, Hellemann & Berry, 2015; Keen, Couzens, Muspratt & Rodger, 2010). The remaining two studies evaluating psychoeducation interventions (Suzuki et al., 2014 and Farmer & Reupert, 2013) did not measure parenting stress. The three studies that found that psychoeducation was associated with a statistically significant reduction in overall parenting stress ranged from weak to strong quality ratings.

Two studies investigating the effects of psychoeducation reported statistically significant improvements in parental confidence (Chiang, 2014; Farmer & Reupert, 2013). Farmer & Reupert (2013) used a self-constructed questionnaire, part of which specifically focussed on

confidence relating to parenting tasks. Chiang (2014) used the confidence degree questions for families (CDQ, Okuno et al., 2011) to measure parental confidence, but did not report psychometric properties for this measure or domains that the CDQ measures. Three of the seven psychoeducation studies measured mental health; two of these used the General Health Questionnaire-28 (GHQ-28; Goldberg, 1978). Suzuki et al. (2014) found a no changes in GHQ-28 scores following a brief psychoeducational group. Tonge et al. (2006) found no significant change in GHQ-28 scores in either of the treatment groups; following psychoeducation and behaviour management (PEBM) or psychoeducation and counselling (PEC), but found statistically significant improvements in mental health in PEBM and PEC groups at 6 month follow-up, above pre-treatment scores of 18.49 and 18.29 respectively. This shows that PEBM and PEC are more effective with parents with poorer mental health. One study measuring anxiety found a statistically significant reduction in parental anxiety scores post-intervention (Farmer & Reupert, 2013), though anxiety was measured using the previously mentioned self-constructed questionnaire.

One study found a statistically significant increase in coping skills in mothers following a psychoeducational intervention (Al-Khalaf, Dally & Dempsy, 2014), while other studies found statistically significant improvements in parental quality of life, in the domains of physical health and environment (Chiang, 2014), and general family functioning (Tonge et al., 2006). Only one out of the seven studies evaluating psychoeducation measured maternal distress and caregiver burden, and no significant effect was found in relation to these outcomes.

Overall, the findings of the studies included in the current review indicate that psychoeducation is associated with a reduction in parenting stress (both overall parenting stress and child-related parenting stress). The evidence suggests that more intensive and

lengthier psychoeducational interventions have a positive impact on parental mental health in parents who have poor mental health prior to starting the intervention. The mean quality rating for the studies evaluating psychoeducation is 0.71, which lies within the moderate range. Together this information suggests that psychoeducation is a beneficial intervention for parents of children with ASD.

ii. Parenting programmes

Five studies evaluated parenting programmes and three out of the five evaluated the efficacy of the Stepping Stones Triple P Positive Parenting Program, a manualised parenting program for parents of children with disabilities (Sanders et al., 2003). Although Stepping Stones Triple P is a manualised programme, the format, content and length of the intervention differed across studies, with Hodgetts, Savage and McConnell (2013) employing an individual, 10 session format, Tellegen and Sanders (2014) employing a low intensity, individual 4-session format and Whittingham, Sofronoff and Sheffield (2009) employing a 9-session group and individual format (4 sessions were individual and 5 sessions were group sessions). All three Stepping Stones Triple P studies found a statistically significant improvement in parental self-efficacy/parenting confidence post-intervention (Hodgetts, Savage & McConnell, 2013) and/or at follow-up (Tellegen & Sanders, 2014; Whittingham, Sofronoff and Sheffield, 2009). Each of these studies used a different outcome measure to evaluate parental self-efficacy and only two out of the three obtained follow-up data.

Statistically significant improvements in parental stress and ‘relationship happiness’ were found following engagement in the individual four session format, with statistically significant effects on parental stress being maintained at 6-month follow-up (Tellegen & Sanders, 2014). Statistically significant improvements in parenting styles and parental conflict

were found following the four session (Tellegen & Sanders, 2014) and the nine session format (Whittingham, Sofronoff & Shefflied, 2009). Effects on conflict about parenting were maintained at 6 month follow-up (Whittingham, Sofronoff & Shefflied, 2009), as were effects on dysfunctional parenting styles (Tellegen & Sanders, 2014; Whittingham, Sofronoff & Shefflied, 2009). Data for the 10 session format of Stepping Stones Triple P (Hodgetts, Savage & McConnell, 2013) was limited, due to the small sample and high levels of attrition (40%). Sofronoff and Farbotko (2002) found mothers' self-efficacy increased to a statistically significant degree following parent management training, but there were no significant changes in fathers' self-efficacy following the intervention. Parental self-efficacy was the only parental outcome that was measured in this study and it was assessed using the 'parental self-efficacy in the management of Asperger syndrome' questionnaire, which was developed for the purpose of the study, but lacks published reliability and validity data. Ferraioli & Harris (2013) compared a skills based parenting program with mindfulness and found no statistically significant changes in parenting stress or global health outcomes post intervention or at follow-up for the skills group.

Overall, quality ratings for studies investigating the effects of parenting programmes ranged from weak to strong, with a mean quality of 0.66. Taking the quality of the evidence into account, the two strongest studies within the 'parenting program' category investigated the effects of Stepping Stones Triple P (Tellegen & Sanders, 2014; Whittingham, Sofronoff & Shefflied, 2009) and findings of these studies were in agreement, suggesting that Stepping Stones Triple P is effective in improving parental self-efficacy, parenting styles and parental conflict.

iii. Parent-mediated interventions

Five studies examined the efficacy of parent-mediated interventions, with the content, length and format of each intervention differing. One commonality between the studies that evaluated parent-mediated interventions was that parents were trained to address their child's deficits, namely in language, play or social interaction. All five studies evaluated parenting stress pre and post-intervention, with two out of the four studies finding a statistically significant reduction in parenting stress post-intervention (Bendixen et al., 2011; Minjarez, Mercier, Williams, Hardan, 2012), though this only applied to mothers and not fathers in the father-based in home intervention (Bendixen et al., 2011). Other findings of parent-mediated intervention studies include a statistically significant improvement in interaction quality (Solomon, Van Ergen, Mahoney, Huber & Zimmerman, 2014), a significant increase in shared positive affect and parent positive affect (Solomon, Ono, Timmer & Goodlin-Jones, 2008), a significant improvement in parenting styles (Bendixen et al., 2011) and significantly higher levels of parent empowerment (Minjarez, Mercier, Williams, Hardan, 2012) post-intervention. Only one of the five studies investigating the effects of parent-mediated interventions considered their effect on mental health (Solomon, Van Ergen, Mahoney, Huber & Zimmerman, 2014), with findings demonstrating a statistically significant reduction in depressive symptomatology when established cut-offs were applied. Kasari, Gulsrud, Paparella, Hellemann & Berry (2015) compared the effects of a joint attention, symbolic play, engagement and regulation (JASPER) intervention with psychoeducation with parents of toddlers with ASD. The parent-mediated intervention (JASPER) was associated with significant changes in child outcomes, but no significant changes were found in parenting stress.

The majority of findings in this category seemed to relate to child outcomes, with relatively few effects on parent outcomes. Three of the studies that evaluated the efficacy of parent-mediated interventions received a ‘strong’ quality rating. The mean quality rating for parent-mediated interventions was 0.77.

iv. Mindfulness

Three studies focussed on the effects of mindfulness and, again, the content and intensity of the mindfulness intervention differed in each study. One of the studies incorporated positive behaviour support principles in the context of mindfulness (Singh et al., 2014), while another compared mindfulness with a skills-based parenting program (Ferraioli & Harris, 2013). Findings were relatively consistent across the three studies that evaluated mindfulness interventions. One study found a statistically significant reduction in parenting stress following the intervention (Ferraioli & Harris, 2013) though these changes were not maintained at 3-month follow-up and another found a statistically significant reduction in general stress following mindfulness training (Singh et al., 2014). General stress continued to reduce over the ‘practice phase’ which followed the intervention phase and lasted between 32 and 37 weeks (Singh et al., 2014). During this phase, participants were encouraged to continue with all mindfulness practices they had learned and to mindfully utilise positive behaviour support techniques. The sample size in the mindfulness-based positive behaviour support program was small ($n=3$), but statistical significance was obtained due to a large effect size (partial eta-squared=0.94). Ruiz-Robledillo et al. (2015) did not measure self-perceived parental stress, but they found a statistically significant reduction in cortisol levels during mindfulness sessions in caregivers of people with ASD. However, there were no significant changes in cortisol levels from pre to post-intervention. Ruiz-Robledillo et al.

(2015) found a statistically significant reduction in mood disturbances, depressive symptoms and somatic symptoms at the end of the intervention.

Overall, the evidence indicates that mindfulness is associated with reductions in general stress levels, but it is not clear whether these effects are maintained over time. All three studies that evaluated mindfulness interventions received a moderate quality rating, with a mean quality rating of 0.65.

v. Cognitive behavioural interventions

Two studies evaluated the efficacy of CBT interventions, though the format and content of these interventions differed, with Feinberg et al. (2014) investigating the efficacy of a brief individual (six-sessions, each session 30 minutes) intervention and Ruiz-Robledillo and Moya (2015) investigating the efficacy of an eight-session intervention (each session lasted 2 hours, but it is not clear whether this intervention was delivered individually or in a group). Parents who participated in the brief intervention reported a statistically significant reduction in depressive symptoms and a clinically significant reduction in parenting stress when compared to treatment as usual. Ruiz-Robledillo and Moya (2015) also found statistically significant improvements in depressive symptoms, mood states and caregiver burden over time, but did not measure parenting stress. The studies that evaluated the effects of CBT were of relatively high quality and received quality ratings of moderate and strong, with a mean quality rating of 0.81. The quality of the evidence, along with the consistency in findings relating to depressive symptoms indicate that CBT is associated with improvements in depressive symptomatology.

vi. Other interventions

Other interventions included in this review are acceptance and commitment training (Blackledge & Hayes, 2006) and a brief nursing intervention (counselling and advice)

(Giarelli, Souders & Pinto-Martin, Bloch & Levy, 2005). No significant changes were found following the nursing intervention and the quality of the study was rated as weak. Statistically significant improvements were found in depressive symptoms, general emotional wellbeing and psychological distress following acceptance and commitment training and this study received a moderate quality rating.

Across all interventions there is some evidence (Solomon, Van Ergen, Mahoney, Huber & Zimmerman, 2014; Tonge et al., 2006) across intervention categories to suggest that interventions have significant effects in parents who experience poor mental health prior to the intervention. Studies discovered significant intervention effects on overall mental health and wellbeing (Tonge et al., 2006) and depressive symptomatology (Solomon, Van Ergen, Mahoney, Huber & Zimmerman, 2014) only when cut-offs were applied.

3.3 Outcome measures

The reliability and validity of outcome measures are important when considering the quality of research. The reported reliability and validity of the measures used in each of the studies varied greatly, with some studies using pre-validated questionnaires and other studies using questionnaires developed for the respective studies. All but two of the studies measured parental outcomes via self-report data, in the form of questionnaires. Ruiz-Robledillo et al. (2015) collected saliva from each participant to measure cortisol levels and in Solomon, Ono, Timmer and Goodlin-Jones' (2008) study, shared positive affect was rated by two trained coders.

The most commonly assessed parental outcome was parental stress (n=14). Studies evaluated a number of other parental outcomes, including parental depression (n=7), overall mental health/wellbeing (n=5), parenting self-efficacy/confidence (n=5), parental anxiety

(n=4) and carer burden (n=3). The most commonly used outcome measure was the Parenting Stress Index (n=10) (Abidin, 1995), which is designed to evaluate the magnitude of stress in the parent-child system. The Parenting Stress Index is a robust measure that maintains its validity within a variety of different cultures. Some studies used just one outcome measure, while others used up to eight outcome measures.

4. DISCUSSION

4.1 Summary of findings

This review assessed the effects of parent-focussed interventions on parent outcomes in the ASD population. The search strategy resulted in 22 studies, which included a range of interventions, categorised here as: psychoeducation, parenting programs, parent-mediated interventions, mindfulness and cognitive behavioural interventions. Comparisons between studies and between interventions were complicated due to the differences in content, delivery and length. At the time of the current review, there were not enough papers in each category to carry out a meta-analysis. A meta-analysis would have allowed for the control of delivery mode, length of the intervention and other potentially confounding variables. When there are more papers/higher quality papers, this will make for a more reliable analysis. The results of this review suggest a body of literature that is in its early stages, but when compared to the results of Kuhaneck, Madonna, Novak and Pearson's (2015) review, it is evident that the literature in this area is growing.

The most commonly evaluated intervention type was psychoeducation. Based on the findings of this review, psychoeducation is the intervention most commonly associated with a reduction in parental stress (71% of psychoeducation interventions included in this review reported a significant reduction in parenting stress), with the mean quality of the papers in this category lying within the moderate range, although the quality of the evidence varied (two

studies received a strong quality rating, one received a moderate quality rating and two received a weak quality rating). Parenting programs were most commonly associated with improvements in parenting confidence/self-efficacy (75% of parenting programs included in this review) and the quality of the studies that found improvements in self-efficacy received a mean quality rating of moderate. The evidence included in this review suggests that longer, more intensive parenting interventions do not have better outcomes than brief (4 sessions) parenting interventions.

The intervention category with the strongest mean quality rating (0.81) is CBT, although only two studies evaluated the effects of CBT. The second strongest evidence is for parent-mediated interventions (mean quality rating of 0.76) and the weakest evidence is for mindfulness interventions (mean quality rating of 0.65).

The only interventions evaluated in single studies were acceptance and commitment training and a brief counselling and advice intervention. The study investigating the effects of acceptance and commitment training was conducted in 2006 and no further research has been carried out to investigate the effects of this type of intervention in an ASD population since 2006. This study received a moderate quality rating and results were promising; there was evidence of improvements in psychological wellbeing, across a number of measures (Beck Depression Inventory, General Health Questionnaire-12 and the Global Severity Index of the Brief Symptom Inventory). Due to the promising results, further research is recommended in relation to acceptance and commitment training.

Stress was the most commonly evaluated parental outcome, with fifteen of the twenty-two studies evaluating parental stress (parenting stress (n=12) and general stress (n=3)) and ten of the fifteen studies finding a statistically significant reduction in parental stress

(parenting stress (n=10), general stress (n=0)) either post-intervention or at follow-up. This finding is in line with the evidence base, which demonstrates extensive literature relating to parenting stress in an ASD population, relative to other parent outcomes. Given the extensive evidence demonstrating the elevated stress levels in parents of children with ASD, it is recommended that future research evaluates parenting stress as standard practice.

Stepping Stones Triple P was the only manualised intervention included in this review and it was evaluated in three separate studies. However, the format of the program differed in each study and the quality of the studies ranged from weak to strong. Despite differences in the format of interventions and the quality of the studies, there was some agreement in relation to the findings, with evidence demonstrating that Stepping Stones Triple P is associated with improvements in parental self-efficacy, parenting styles and parental conflict in parents of children with ASD. Given the association between parental self-efficacy and parental stress (Kuhn & Carter, 2006), improvements in self-efficacy may lead to a reduction in stress over time.

Of the nine studies that collected follow-up data, four maintained the majority of the changes that were found post-intervention. Of the other five studies that collected follow-up data, one reported further improvement between baseline and follow-up, one study reported that improvements seen post-intervention were maintained at follow-up for one of two outcomes, two studies did not clearly report changes between post-intervention and follow-up and one study reported no changes between baseline and follow-up. Overall, the maintenance of changes suggests that changes are relatively stable over time, but there is little evidence to suggest that further improvements are made post-intervention.

It is important to note that parent/parent-mediated interventions may have been designed to complement rather than replace existing interventions. In a number of studies, either parents, their child(ren) with ASD or both were receiving treatment as usual alongside the intervention under study, which makes it difficult to identify effects specific to the intervention under study without a control group (9 studies did not include a control group).

There is some evidence to suggest that parent interventions are more effective for those who are poorer in mental health prior to intervention. This has implications for screening potential participants and also for exploring pre-intervention mental health as a confounding variable in research.

There is a growing body of research with aims relating to reducing parental stress/distress, improving parental mental health, improving parental confidence/self-efficacy and improving parental quality of life. In order to improve the quality of the research in this area, future research should include large samples, interventional and random allocation, blinding (of participants and investigators), control for confounding, the use of pre-validated measures and long-term follow-ups.

4.2 Limitations

Limitations of the papers

The majority of studies (n=17) were carried out in the USA and Australia and a number of studies included in this literature review used samples that were better educated, or had higher socio-economic status than average, which may impact on the external validity of the results. It may be the case that parents who have reached a higher level of education, or those from a higher socio-economic background are more likely to engage in parent/parent-mediated interventions than the general population.

A number of studies did not include follow-up data and the studies that included a follow-up have a relatively short follow-up period, which provides only a limited understanding of the longer term effects of parent interventions in ASD. The current review included studies with small samples, with the smallest sample including only three participants.

Most of the studies included in the current review used pre-validated questionnaires and more than one measure to evaluate parent outcomes. However, the quality of the papers varied considerably, which impacts on the conclusions that can be drawn.

Limitations of the review

A major limitation of this review, or reviews of this type is that there is so much variation between interventions that it is difficult to draw any reliable conclusions about what interventions are effective.

The current review is limited to papers that are published in English in peer reviewed journals, which may mean that other relevant papers have been excluded. However, known ASD organisations were contacted in an attempt to uncover some unpublished literature. The current review excluded interventions where children with ASD or their teachers were the participants. Consequently, the current review may underestimate the effects of ASD interventions on parent outcomes.

The current review identified 17 papers that were not included in Kuhaneck, Madonna, Novak & Pearson's (2015) review, that were published within the time frame used in Kuhaneck, Madonna, Novak & Pearson's (2015) review. Utilising the search terms identified in Kuhaneck, Madonna, Novak & Pearson's (2015) review in the current review

returned an additional five papers, making it more likely that all relevant papers have been identified.

4.3 Recommendations

Given the ambiguity of the current evidence and the current economic climate, caution is warranted when considering financial costs to service providers of delivering parent interventions. The information provided in this review in relation to specific parent outcomes may help to inform decisions regarding the type of intervention that might be most beneficial to each parent/family, based on need. For example, psychoeducation may be the most appropriate intervention for a parent who is stressed.

Further high quality research is required in this area, in order to determine the efficacy of parent/parent-mediated interventions for parents of children with ASD. To enhance understanding, researchers may wish to consider measuring a number of variables, such as stress, self-efficacy, depression and parental relationship conflict. Researchers investigating the effects of parent interventions may also want to consider alternatives to parent self-report to measure parent outcomes, such as clinical interviews or monitoring cortisol levels. Clinicians and researchers alike may wish to measure parent outcomes as routine practice, even when interventions are carried out with children/young people and teachers. Furthermore, future research should consider investigating the effects of parent interventions with parents who are experiencing elevated/clinically significant levels of stress/depression/mental health problems, to characterise which parents are most likely to benefit from intervention. Given the promising results associated with acceptance and commitment training in Blackledge and Hayes' (2006) study, further research including larger samples is recommended in this area.

4.4 Summary

There is an extensive body of literature demonstrating the high levels of stress, poor parenting self-efficacy and increased rate of mental health problems experienced by parents of children with an ASD. The importance of the relationship between parent outcomes and child outcomes makes it imperative that health services work towards improving parent outcomes in this population. Due to the variation in findings and in the quality of the research, it is recommended that future research develops the evidence base by improving the quality of research, in order to address the gaps in current understanding relating to the efficacy of interventions for parents of a child with ASD. Furthermore, a meta-analysis or meta-synthesis is required to control for potentially confounding factors (such as contact hours, mode of delivery, etc), which will allow for direct comparisons between interventions and intervention types.

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EMPIRICAL PAPER

CHILDHOOD PATHOLOGICAL DEMAND AVOIDANCE: PARENTAL ILLNESS PERCEPTIONS, COPING, WELLBEING AND THE CHILD-PARENT RELATIONSHIP

Abstract

Background: Leventhal's self-regulation model (SRM) theorises that people make sense of health stressors by developing an organised pattern of implicit common sense beliefs, which predict medical, psychological and behavioural outcomes. This study examined the applicability of Leventhal's SRM to parents' understanding of childhood Pathological Demand Avoidance syndrome (PDA).

Method: A cross-sectional web-based design was used to explore the relationships between parental illness perceptions, coping, parenting stress, wellbeing and the perceived quality of the child-parent relationship in parents of children with a diagnosis of PDA (diagnostic group, n=71) and parents who suspected their child has PDA (non-diagnostic group, n=118). Participants were recruited through the PDA Society website, Facebook group and Twitter page.

Results: Parental illness perceptions of personal control and illness coherence differed significantly between groups (diagnostic > non-diagnostic). Across both groups, parents perceived PDA as a chronic condition, with considerable negative consequences. Furthermore, both groups of parents reported high levels of parenting stress and poor wellbeing. Parental perceptions of greater consequences, lower levels of personal control and a poorer understanding of PDA were associated with higher levels of parenting stress, poorer wellbeing and higher levels of child-parent conflict. Coping was not found to mediate the relationship between parental illness perceptions and outcomes.

Conclusions: Parents of children with PDA and suspected PDA are identified as an 'at risk' group. Findings indicate that the SRM is applicable to parents of children with PDA, which has important implications for assessment and intervention. Suggestions for future research and limitations of the study are discussed.

Keywords: illness perceptions, PDA, pathological demand avoidance, coping, parenting stress, wellbeing, child-parent relationship.

1. INTRODUCTION

1.1 Pathological Demand Avoidance Syndrome

Pathological Demand Avoidance (PDA) is a term increasingly used by practitioners in the UK (O’Nions, Viding, Greven, Ronald & Happe, 2014). The term PDA, initially coined by Professor Elizabeth Newson in the 1980s, is used to describe a profile of obsessive resistance to everyday demands and requests, using socially manipulative and ‘outrageous’ behaviour to avoid demands (Newson, Le Marechal & David, 2003). In the 1980s, Newson and her team recognised that children who were referred to their specialist clinics for diagnostic assessments (Child Development Research Unit, Nottingham University), shared some traits of autism spectrum disorder (ASD), but they were not ‘typically autistic’. These children also presented differently to children with atypical autism or pervasive developmental disorder not otherwise specified (PDD-NOS) (Diagnostic and Statistical Manual-IV, DSM-IV), though there were a number of commonalities. Newson and her team noticed that these children were similar in a number of significant ways, which subsequently lead to the proposal of PDA.

There is a growing but limited literature in relation to PDA and although PDA is not currently formalised in published diagnostic manuals (i.e. DSM-V, American Psychiatric Association, 2013; ICD-10, World Health Organisation, 1992), it is becoming increasingly recognised (O’Nions et al. 2015). Clinicians and researchers are using the criteria defined by Newson (2003) for a diagnosis of PDA, but there is some uncertainty as to whether PDA is distinct from autism spectrum disorder (ASD) or a manifestation of this (Christie, 2007). Wing and Gould (2002) argued that PDA is not a distinct syndrome, as behavioural features that are consistent with PDA are found in individuals with an ASD. In 2003, Newson, Le Marechal and David made a proposal to recognise PDA as a separate entity within pervasive

developmental disorders, instead of coming under ‘pervasive developmental disorder not otherwise specified’ (DSM-IV). More recently, O’Nions et al. (2016) found that all but one case recognised as having a high number of features consistent with PDA, as identified by the Diagnostic Interview for Social and Communication Disorders (DISCO, Wing, Leekam, Libby, Gould & Larcombe, 2002), met diagnostic criteria for an ASD, which might suggest that PDA is not distinct from ASD. Anecdotally, the diagnosis of PDA is still very controversial amongst professionals in the UK, with the ability or willingness to diagnose PDA varying between professionals and between regions. The reluctance among some professionals to diagnose PDA may relate to a lack of knowledge or understanding of the disorder as well as the absence of PDA as a distinct category within diagnostic classification systems. PDA is described as a life-long developmental disorder, but due to the relative newness of the criteria and diagnosis, it has predominantly been diagnosed in children. Additionally, Gillberg, Gillberg, Thompson, Biskupsto and Billstedt (2014) suggest that criteria for PDA are unlikely to still be met in late adolescence and early adult life. For the current study, the focus is on PDA in childhood and the remainder of the text will refer to childhood PDA. The diagnostic criteria for PDA, defined by Newson (2003), are as follows:

- Passive early history in the first year.
- Continues to resist and avoid ordinary demands of life, with strategies of avoidance being essentially socially manipulative.
- Surface sociability, but an apparent lack of social identity.
- Lability of mood, led by need to control.
- Comfortable in role play and pretend.
- Language delay.
- Obsessive behaviour.

- Neurological involvement (i.e. poor communication).

The avoidance of demands, which is often described as obsessive, is conceptualised as being driven by anxiety and a need for control. Newson suggested that extreme outbursts and challenging behaviour that are typical of PDA are best understood as panic attacks. The strategies employed to avoid demands for those with PDA are seen as being socially manipulative (Newson & Le Marechal, 1998), demonstrating that these children have more advanced social interaction and communication skills than children with typical autism. Children with PDA can demonstrate empathy, social awareness and understanding, but these skills can appear superficial, with no sense of responsibility, pride or shame (Newson & Marechal, 1998). It is common for children with PDA to identify with adults rather than children and behave like adults, telling others the rules (Newson, Le Marechal & David, 2003).

Another characteristic of children with PDA is sudden and excessive ‘mood swings’ that may have no obvious trigger. Additionally, these children differ from other children on the autism spectrum, in that they have a good imagination and are very comfortable in role-play; mimicry of others can be taken to an extreme extent, such that they sometimes lose touch with reality (Newson, Le Marechal & David, 2003).

The early language delay in children with PDA seems to be due to passivity and there is often a remarkable degree of catch-up. Similarly to other children on the autism spectrum, children with PDA may have difficulty understanding sarcasm and metaphors (Newson & Le Marechal, 1998).

Obsessive behaviour in PDA may encompass behaviours such as taking on another persona in an obsessive manner, being fixated on a certain individual or doing a number of

things in an ‘obsessive way’. Furthermore, Newson hypothesised that there is neurological involvement in PDA due to some degree of poor co-ordination and called for further investigation involving neurological assessment due to limited evidence.

O’Nions, Viding, Greven, Ronald and Happe, (2013) explored the behavioural profile of children with PDA (n=29), ASD (n=35), and conduct problems and callous-unemotional traits (CP/CU; n=28) (mean age of 11 years). Results highlighted that children with PDA displayed severe impairments across multiple domains. Comparisons between behaviour in PDA, ASD and CP/CU revealed levels of peer problems and autistic-like traits in PDA comparable to ASD. Results also revealed levels of anti-social traits, manipulative behaviour and lack of pro-social behaviour in PDA comparable to those in CP/CU. Findings showed that children with PDA had significantly higher scores than children with ASD for conduct problems and lack of pro-social behaviour. Children with PDA also scored significantly higher than children with CP/CU for peer problems. Additionally, the PDA group had significantly higher levels of parent reported emotional symptoms (anxiety/internalising problems) and behavioural problems than either the ASD or the CP/CU group. The differentiation between children with PDA and children with ASD and CP/CU in relation to emotional symptoms may support the hypothesis that avoidance behaviours seen in PDA are driven by or associated with anxiety.

1.2 Parental Wellbeing

Extensive research demonstrates that parents of children with ASD experience higher levels of stress than parents of children with other disabilities (such as Down’s syndrome, intellectual disabilities and cerebral palsy) and parents of typically developing children (Hayes & Watson, 2013; Dabrowska & Pisula, 2010; Griffith, Hastings, Nash & Hill, 2010; Estes, Munson, Dawson, Koehler, Zhou & Abbott, 2009; Davis & Carter, 2008). Parents of

children with a developmental disability, particularly parents of children with ASD, are at a higher risk of experiencing mental health problems than parents of typically developing children (Estes et al., 2013). Two thirds of mothers of children with a diagnosis of pervasive developmental disorder (PDD) reported significantly elevated stress levels, with behaviour being a significant predictor of maternal distress, accounting for 32% of the variance in maternal distress (Tomanik, Harris and Hawkins, 2004). Children's non-compliance in PDD was one factor that was found to be significantly related to high stress levels. Given that non-compliance is a major factor in PDA, and there is overlap in behavioural profiles of children with PDA and PDD/ASD, it is hypothesised that there may be some overlap between the experiences of parents of a child with PDA and experiences of parents of a child with ASD. Lecavalier, Leone and Wiltz (2006) found strong associations between behaviour problems and parent stress and, in particular, conduct problems were found to be a significant predictor of stress in parents of children with ASD. Furthermore, parent stress was found to exacerbate behaviour problems. Overall, the literature presents a consistent profile of results that shows parents of children with behaviour problems, developmental disabilities and particularly ASD are at an increased risk of experiencing elevated levels of parenting stress and mental health problems.

It is evident that parents are affected differently by the challenges that their children bring, however, there is little evidence demonstrating what factors, other than child behaviour, contribute to parental stress and emotional wellbeing, given that child behaviour only accounts for a proportion of the variance in parenting stress (Tomanik, Harris & Hawkins, 2006).

1.3 The Self-Regulation Model

Leventhal, Meyer and Nerenz (1980) and Leventhal, Nerenz and Steele (1984) outlined and developed the common sense model of self-regulation, which theorised that people make sense of an illness or condition they are diagnosed with by developing an organised pattern of implicit common sense beliefs about the health threat. The self-regulation model (SRM, Leventhal, Nerenz & Steele, 1984, 1997) offers a theory to explain why individual responses to the same health threat may differ, which helps to predict how individuals may perceive, behave and adjust to health-related stressors. According to the SRM, when threats to health occur, people generate two types of representations: cognitive representations (interpretations of the nature of the threat) and emotional representations (e.g. fear and anger). The SRM proposes that cognitive representations comprise of five components:

- i) Identity – the symptoms an individual classifies as being part of the illness/disorder.
- ii) Consequences – anticipated effects and outcome of the illness/disorder.
- iii) Timeline – the length of time the individual believes the illness/disorder will last.
- iv) Control/cure – perceived control over the disorder or symptoms of the disorder/availability of a cure
- v) Cause – ideas about what caused the illness or symptoms.

Each representation produces its own attempt at regulation; firstly of the threat itself and secondly of the consequent emotions. Hence, the model hypothesises that individuals' 'common sense beliefs' about a condition influence the coping strategies that they develop and employ, and subsequently influence a range of medical, psychological, and behavioural outcome variables (Hagger & Orbell, 2003; Scharloo Kaptein, & Weinman, 1999). Nerenz

and Leventhal (1983) acknowledged that illness representations are not necessarily bio-medically valid, but it is these views that influence coping and outcomes. Figure 1 illustrates the SRM developed by Leventhal, Nerenz and Steele (1984).

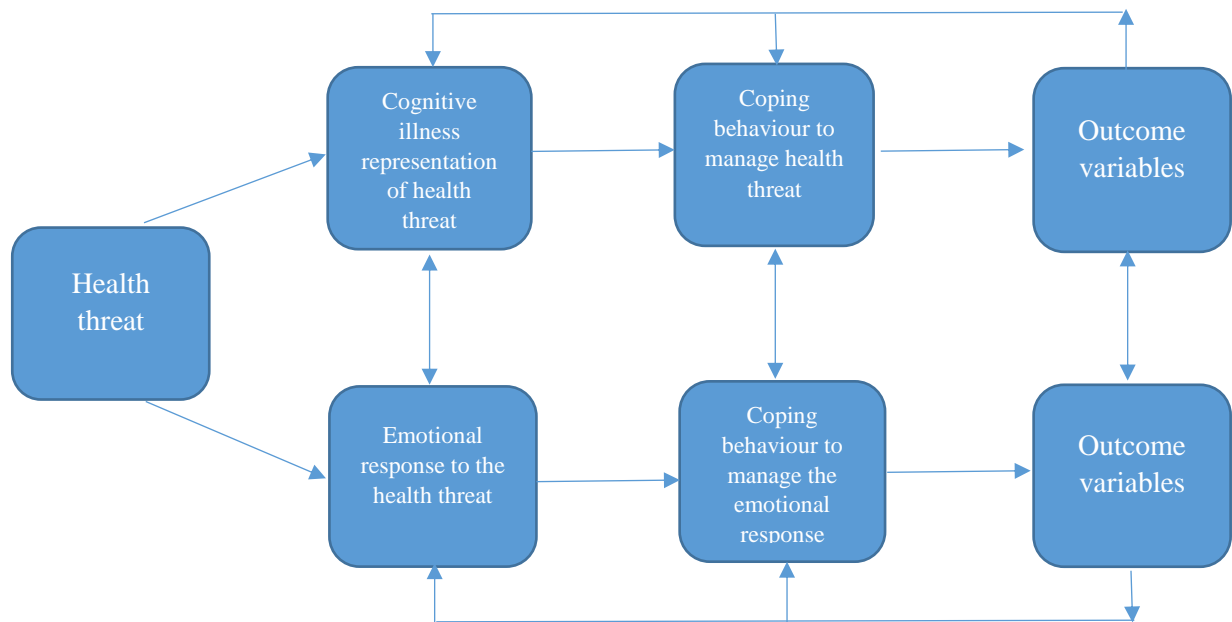


Figure 1. The self-regulation model.

The majority of research relating to illness representations has focussed on chronic physical health problems. Hagger and Orbell (2003) conducted a meta-analytic review of 45 empirical studies adopting the SRM, within the context of physical health problems (e.g., diabetes, psoriasis). The analysis showed that perceptions of a strong illness identity were significantly and positively associated with coping strategies of avoidance/denial and emotion expression. Perceived controllability of the health threat was found to be significantly associated with cognitive reappraisal, expressing emotions and problem-focussed coping strategies. Furthermore, representations that the illness was controllable/curable were significantly and positively related to psychological wellbeing and social functioning, and significantly and negatively related to psychological distress and health state.

More recently, research has examined illness perceptions within the context of mental health. Baines & Wittkowski (2013) carried out a review of the literature, examining 13 studies that explored illness perceptions within the context of a range of mental health conditions (e.g., depression, psychosis, eating disorders). Mental health problems were consistently viewed by those who had experienced them as chronic, with serious negative consequences. Illness perceptions were found to be associated with a range of outcomes, including levels of anxiety and depression, help seeking behaviours, coping and treatment choices.

The use of the SRM has also been applied to parents', family members' and spouses' understanding of a health condition, as well as individuals' understanding of their own health condition (e.g. Rodríguez-Orozco, Kanán-Cedeño, Guillén Martínez & Campos Garibay, 2011; Klok, Brand, Bomhof-Roordink, Duiverman & Kaptein, 2011). Recently, research has explored interactions associated with parental illness representations in child and adolescent mental health (Shanley & Reid, 2014). Shanley & Reid (2014) found significant associations between parental illness representations and child problem severity, parental adjustment, and treatment acceptability.

1.4 Applicability to ASD

To date, no study has explored the applicability of the SRM in the context of PDA, though the SRM has been applied to ASD. Al Anbar, Dardennes, Prado-Netto, Kaye & Contejean (2010) explored the role of parental illness perceptions in treatment choices among 89 parents of a child with ASD. Authors found that parents had a coherent model of ASD and 65% of parents referred to genetics as the most important cause of ASD. Significant associations were found between parental illness representations of ASD and treatment choices. Results showed that parental illness representations related to how parents coped

with having a child with ASD, such as speaking to friends, seeking information, and choosing treatments.

In 2011, Dardennes et al. investigated the relationship between causal beliefs and treatment choices in parents of children with a diagnosis of ASD. Again, the most commonly cited causal beliefs were genetics and brain abnormalities. Dardennes et al. (2011) also discovered a link between causal beliefs and treatment choices (e.g., psychoeducation, medication), and the authors suggested that identifying/modifying parental illness beliefs may be important in facilitating appropriate interventions, reducing distress and enhancing wellbeing.

Gatzoyia et al. (2014) found that parental perceptions of greater consequences and greater chronicity relating to ASD were associated with increased psychological distress and increased severity of depressive symptoms. Furthermore, 34% of parents reported clinically significant depressive symptoms and 55% reported clinically significant levels of psychological distress.

1.5 Coping styles in parents of children with ASD

The literature on coping among parents of children with ASD suggests that coping styles can impact the extent to which parents are affected psychologically by their child's ASD. Lai, Goh, Oei and Sung (2015) found that parents of children with ASD reported higher levels of parenting stress and a higher number of depressive symptoms, as well as more frequent use of active avoidance coping than parents of typically developing children. In line with this finding, Benson (2010) found that avoidant coping was significantly associated with higher levels of depression and anger in mothers of children with autism, and cognitive reframing was associated with greater levels of maternal wellbeing.

Evidence suggests that emotion-oriented coping is associated with increased parent and family stress in parents of children with ASD (Dabrowska & Pisula, 2010; Lyons, Leon,

Roecker Phelps & Dunleavy, 2009) and task-oriented coping is associated with lower physical incapacity related stress (Lyons, Leon, Roecker Phelps & Dunleavy, 2009). Evidence also indicates that illness representations are associated with coping (Baines & Wittkowski, 2013). Specifically, perceptions that mental health problems are chronic and controllable, with negative consequences were found to be associated with more active coping strategies and help-seeking behaviours across a range of mental health problems. Outside of the context of ASD, some studies have found that coping mediates the relationship between illness representations and psychological wellbeing (e.g. Savage, 2011; Gould, Brown & Bramwell, 2010), while others have found direct effects of illness representations, unmediated by coping (e.g., Kaptein et al., 2006).

1.6 Child-parent relationship

In exploring child-parent relationships, Hoppes & Harris (1990) found that mothers of children with autism described their children as significantly less responsive and reported less emotional closeness and reciprocity in their relationship with their child than mothers of children with Down's syndrome. Findings showed that a mother's perception of her child's attachment was related to her feelings of gratification. Authors suggest that the lack of reciprocity and reinforcement for mothers may help to explain why mothers of children with autism experience higher levels of stress than mothers of children with Down's syndrome and other disabilities. These findings demonstrate the importance of exploring the nature and quality of the parent-child relationship in PDA.

1.7 Hypotheses

There is a wealth of literature evidencing that illness representations are associated with psychological wellbeing (Hagger & Orbell, 2003) and with coping (Baines & Wittkowski, 2013). Coping styles have been found to be associated with psychological

adjustment and psychological wellbeing (Lai, Goh, Oei & Sung, 2015; Benson, 2010; Lyons, Leon, Roecker Phelps & Dunleavy, 2009; Roesch & Weiner, 2001). Building on the theoretical model of the SRM, the extensive literature demonstrating that parents of children with ASD experience elevated levels of stress and mental health problems and the overlap in behavioural profiles between ASD and PDA, it seems pertinent to investigate the associations between parental illness representations of PDA, coping and parenting stress, parental wellbeing and the child-parent relationship. Hypotheses are presented in Table 1.

Table 1. Hypotheses

Hypothesis 1 Illness perceptions → outcome	There will be a relationship between parental illness perceptions of PDA and self-reported parenting stress, parental wellbeing, conflict in the child-parent relationship and closeness in the child-parent relationship.
Hypothesis 2 Illness perceptions → coping	There will be a relationship between parental illness perceptions of PDA and coping strategies employed by parents to manage their child's PDA.
Hypothesis 3 Coping → outcome	There will be a relationship between parental coping strategies and self-reported parental wellbeing, parenting stress, child-parent conflict and child-parent closeness.
Hypothesis 4 Illness perceptions → coping → outcome	Coping will mediate the relationship between parental illness perceptions of PDA and outcomes of: (a) parental wellbeing, (b) parenting stress, (c) child-parent conflict, and (d) child-parent closeness.

2. METHOD

2.1 Design

This cross-sectional web-based survey was hosted by Lime Survey. Ethical approval for this study was granted by the University of Birmingham's Science, Technology, Engineering and Mathematics Ethical Review Committee (ERN-14-0474; approved 5th February 2015) (Appendix D & Appendix E).

2.2 Procedure

Two sources of support for parents of children with PDA were identified: charities known as the PDA Society and the National Autistic Society (NAS). Both the PDA Society and the NAS were contacted via their websites; the PDA Society confirmed their willingness to advertise the research on their website and via their social media groups (i.e., Facebook and Twitter) and NAS did not respond. Initially, only parents/main carers of children with a confirmed (self-reported) diagnosis of PDA were invited to participate. Parents who were interested in finding out more about the study, based on a short advert (Appendix F), could click through to a participant information sheet (Appendix G) that provided further details about the study. For those parents/carers willing to participate, a consent form was completed (Appendix H).

32 complete questionnaires were returned during the first week of the survey's launch and only 7 additional complete questionnaires were returned over the following 4 weeks, which raised concerns regarding response rates. In addition, a number of parents, who suspected their child has PDA, e-mailed the researcher querying whether they could take part. Consequently, the researcher decided to expand the inclusion criteria to include parents who suspected their child has PDA, but do not have a confirmed diagnosis of PDA. The research

was re-advertised, via the same sources, inviting both parents of children with a diagnosis of PDA and parents who suspect their child has PDA to participate.

Entry into a prize draw was offered to compensate participants for their time. Parents wanting to be entered into the prize draw (£50, £20 and £10 of Amazon vouchers) had to supply an email address within the survey.

2.3 Participants

189 parents/caregivers participated; 71 parents/caregivers of a child with a diagnosis of PDA and 118 parents/caregivers who suspected their child had PDA. 93.4% of participants were mothers, 3.3% were fathers, and 3.3% were primary caregivers/guardians, which included family members who were special guardians; overall 96% of the participant sample were female. The parental relationship status of participants was: married (58.7%), co-habiting (13.0%), separated (13.0%), divorced (11.3%), and 'other' (4.0%). Ethnicity data fell into the following categories: white British (84.6%), white other (6.0%), mixed ethnic background (6.0%), black Caribbean (0.6%) and other (2.8%).

Ages of children ranged from 4.8 to 18.04 years; 68.1% of children were boys and 31.9% were girls. For the diagnostic group (n=71), time since diagnosis of PDA ranged from 0.16 to 9.85 years. Mean child age, duration of diagnosis, number of comorbid diagnoses, parent age and number of siblings are detailed in Table 2.

Table 2. Sample statistics

	Mean (standard deviation) Diagnostic Group	Mean (standard deviation) Non-diagnostic group
Child's age (in years)	10.35 (3.62)	9.85 (3.53)
Length of time since diagnosis of PDA (in years)	1.98 (2.22)	
Number of diagnoses (other than PDA)	<i>2.15 (1.54)</i>	<i>1.66 (1.55)</i>
Parent's age (in years)	37.59 (15.59)	38.04 (11.44)
Number of siblings	1.34 (1.11)	1.44 (1.07)

*Median (interquartile range) are presented in italics

2.4 Measures & Materials

The survey comprised the following seven questionnaires (Appendix I):

Demographic data questionnaire: Consisted of 17 items designed to capture demographic data.

Extreme Demand Avoidance Questionnaire (EDA-Q: O’Nions, Christie, Gould, Viding and Happe, 2013): A 26-item questionnaire used to indicate whether or not a child’s profile is consistent with PDA. Parents indicate on a four-point scale (0 = ‘not true’ to 3 = ‘very true’) how true each statement is for their child; higher scores are indicative of more symptoms of PDA. Scores of 50 and over identify individuals with an elevated risk profile consistent with PDA in 5-11 year olds; scores of 45 and over identify individuals with an elevated risk profile consistent with PDA in 12-17 year olds. Published Cronbach’s alpha is good at 0.87 (O’Nions, Christie, Gould, Viding, Happé, 2013).

Illness Perception Questionnaire – Revised (IPQ-R: Moss-Morris, Weinman, Petrie, Horne, Cameron and Buick, 2002): A 56-item questionnaire that measures the five cognitive components of illness perceptions in Leventhal’s self-regulation model. The wording of the IPQ-R was altered (as authors permit) to make it specific to PDA (e.g., ‘my

child's PDA will improve in time'). For the identity subscale, items were altered to reflect the characteristics associated with PDA. Twelve further items were added in order to capture parents' beliefs about different treatments for PDA. The majority of items use a 5-point Likert type scale (1 = 'strongly disagree' to 5 = 'strongly agree'), where parents indicate the degree to which they agree with each statement. High scores on the identity, timeline, consequences and cyclical domains represent strongly held beliefs about the number of symptoms attributed to PDA, the chronicity of PDA, the negative consequences associated with PDA and the cyclical nature of PDA. High scores on the personal control, treatment control and coherence domains represent positive beliefs about the controllability (personal and treatment control) of PDA and a good personal understanding or comprehension of PDA. This measure has been shown to have good internal reliability across all subscales, with Cronbach's alpha ranging from 0.79 for the timeline cyclical dimension to 0.89 for the timeline acute/chronic dimension (Moss-Morris, Weinman, Petrie, Horne, Cameron and Buick, 2002).

Brief COPE (Carver, 1997): A 28-item, validated self-report measure of coping styles, consisting of 14 theoretically derived subscales. Each item is scored on a four-point scale with higher scores indicating more use of a particular strategy/behaviour in an attempt to cope with their child's PDA. Carver (1997) reported Cronbach's alpha values ranging from 0.50 to 0.90 across the 14 subscales. Cooper, Katona and Livingston (2008) collapsed the 14 subscales into three subscales: emotion-focussed coping (e.g., 'getting emotional support', 'trying to make it seem more positive'), problem-focussed coping (e.g., 'getting help and advice from other people', 'concentrating efforts on doing something about the situation') and dysfunctional coping (e.g., 'giving up trying to deal with it', 'using alcohol or other drugs to feel better) and found good reliability, demonstrated by Cronbach's alpha values ranging from 0.72 to 0.84. The three factor structure was used to analyse data in the current study.

Parental Stress Scale (PSS: Berry and Jones, 1995): An 18-item measure of parenting stress. Responses are based on a five-point Likert scale, ranging from strongly disagree to strongly agree and higher total scores indicate greater levels of parenting stress, with total scores ranging from 18-90. Berry and Jones (1995) found satisfactory levels of internal reliability (Cronbach's alpha of 0.83), test-retest reliability (0.81) and convergent validity, suggesting that the Parental Stress Scale serves as an adequate replacement for the widely used Parenting Stress Index (Abidin, 1986).

General Wellbeing Index (GWBI: McKenna & Hunt, 1992): This is a 22-item standardised quality of life measure. The GWBI is the British adaptation of the American Psychological General Wellbeing Index (PGWBI, Dupuy, 1984), which has not been validated with a British population. The GWBI is very similar to the PGWBI, differing only in terms of wording of some items. Responses are based on five options, the wording of which differs for each item. Scores range from 22-132 and higher scores are indicative of greater wellbeing over the past month. McKenna and Hunt (1992) demonstrated high levels of internal consistency (ranging between 0.92 and 0.96). Despite the PGWBI having six subscales with reportedly adequate levels of internal reliability, subscales were not recommended for the GWBI due to high inter-correlations (Hunt & McKenna, 1993).

Child-Parent Relationship Scale-Short Form (CPRS: Pianta, 1992): A 15-item self-report measure, completed by parents to assess their perceptions of their relationship with their child. Responses are rated on a five-point Likert scale and the ratings can be summed into groups of items corresponding to conflict (8 items, e.g., 'my child easily becomes angry at me') and closeness (7 items, e.g., 'I share an affectionate, warm relationship with my child') subscales, with higher scores indicating more conflict and more closeness in the child-parent relationship. Reported Cronbach's alpha values are 0.78 and 0.66 respectively (Pianta, 1992).

2.5 Variables

The independent variables were parental illness perceptions. Dependent variables were measures of parental coping, parental wellbeing, parenting stress and child-parent relationship.

2.6 Data analysis

All scales and subscales included in this study were analysed using SPSS to examine their internal reliability and establish the normality of data. Most subscales achieved an acceptable level of internal reliability ($\alpha \geq .07$) (George & Mallery, 2003). Table 3 shows alpha levels and the final number of items in each scale (some items were removed to increase reliability).

Table 3. Cronbach's alpha values

Subscale	Cronbach's alpha	Number of items
Extreme Demand Avoidance Questionnaire		
EDA-Q Total	.69	26
Illness Perceptions Questionnaire-Revised		
IPQ Timeline	.69 (item 18 removed)	5
IPQ Consequences	.73 (item 9 removed)	5
IPQ Personal control	.74	6
IPQ Treatment control	.70 (item 20 removed)	4
IPQ Illness coherence	.87	5
IPQ Timeline cyclical	.72	4
IPQ Emotional representations	.80	6
Brief COPE		
Emotion-focussed strategies	.76 (items 20 & 24 (acceptance subscale) removed)	8
Problem-focussed strategies	.86	6
Dysfunctional coping strategies	.67	12
Parental Stress Scale		
PSS Total	.86	18
General Wellbeing Index		
Anxiety	.83	5
Depressed mood	.79	3
Positive wellbeing	.81	4
Self-control	.81	3
General Health	.84	3
Vitality	.84	4
Total wellbeing	.94	22
Child-Parent Relationship Scale-Short Form		
Conflict	.65	8
Closeness	.70	7

Somewhat unsurprisingly given the nature of the sample, the majority of data were skewed (all scales and subscales other than overall wellbeing), indicating high levels of demand avoidant behaviour, strongly held beliefs about PDA, frequent use of unhelpful coping strategies and poor quality child-parent relationships. Where data were normally distributed, parametric statistical tests were used for inferential analyses of the data, and where data were not normally distributed, non-parametric tests were used. A significance level of $p < 0.05$ was adopted, unless otherwise stated.

For the purpose of the analyses, parents of children with a diagnosis of PDA will form one group (diagnostic group) and parents who suspect their child has PDA, but they do not

have a formal diagnosis, will form the second group (non-diagnostic group). To test hypotheses 1-4, only the diagnostic group will be used.

Analysis strategy for testing hypotheses. The majority of data were not normally distributed and correcting sample distributions for normality is not an appropriate correction to these data. When testing hypotheses where variables were not normally distributed, traditional asymptotic probability estimates will be supplemented by bootstrap parameter estimates and 95% confidence intervals. The nonparametric bootstrap method is independent of population distribution and offers robust confidence intervals in small sample situations (Moore & McCabe, 2005).

3. RESULTS

PDA and comorbidities. 177 out of the total 189 EDA-Q scores were above the suggested cut-offs, thus identifying 94% of children at elevated risk of having a profile consistent with PDA. There was no difference in EDA-Q scores between groups, with the median score being 61 for both groups (interquartile range was 11 for the diagnostic group and 9.25 for the non-diagnostic group). There was no significant difference in children's ages between groups. 86% of children in the diagnostic group had comorbid diagnoses and 68% of children in the non-diagnosis group had comorbid diagnoses, as reported by parents. In comparing the mean number of diagnoses across both groups, the diagnostic group had a significantly higher number of comorbid diagnoses. The most common co-morbid diagnoses were autism, anxiety and ADHD, with 44.4%, 37.5% and 21.7% of the total sample of parents reporting these comorbid diagnoses respectively.

3.1 Between group comparisons

Illness Perceptions

To explore differences between groups in relation to illness perceptions, Mann-Whitney U tests were conducted. Results showed a statistically significant difference in subscale scores between groups on only two subscales: personal control and illness coherence. Table 4 reports scores for each IPQ-R subscale, by group.

Table 4. IPQ-R subscales for diagnostic and non-diagnostic groups

IPQ-R subscale	Diagnostic group		Non-diagnostic group		p value
	Median ¹	Interquartile range	Median ¹	Interquartile range	
Timeline acute/chronic (e.g., his/her PDA will pass quickly)	4.50	0.67	4.16	0.58	.083
Consequences (e.g. his/her PDA is a serious condition)	4.11	0.36	4.01	0.39	.108
Personal control (e.g., the course of his/her PDA depends on me)	3.54	0.61	3.24	0.72	.008
Treatment control (e.g. his/her treatment can control his/her PDA)	3.06	0.71	2.97	0.63	.348
Illness coherence (e.g., I don't understand his/her PDA)	3.72	0.84	3.31	0.85	.003
Timeline cyclical (e.g. his/her symptoms come and go in cycles)	3.92	0.87	3.80	0.78	.171
Emotional representations (e.g. his/her PDA makes me feel anxious)	3.81	0.72	3.92	0.76	.352

¹ Adjusted median score (sum of scale items divided by number of items); all scores range from 1-5 with higher scores indicating greater endorsement of a subscale construct.

Results show that parents have strongly held beliefs about the chronicity of PDA, the negative consequences and negative emotional consequences related to PDA, and the cyclical nature of PDA. Parents in the diagnostic group reported a significantly greater understanding/comprehension of PDA (higher illness coherence) than parents in the non-diagnostic group. Furthermore, parents in the diagnostic group reported significantly greater perceptions of personal control in relation to PDA than parents in the non-diagnostic group.

Identity

There was no statistically significant difference in the total number of symptoms endorsed by parents (i.e., symptoms attributed to PDA) between groups. The median number of symptoms attributed to PDA was 9 (interquartile range = 3), with a maximum of 12 possible symptoms to endorse. Not all parents completed the identity subscale, with the number of participants completing individual items ranging from 149 (for sudden and excessive mood swings) to 179 (for avoidance of every day demands). This may suggest that parents do not have a clear idea about the symptoms/characteristics that make up PDA. Figure 2 illustrates differences between groups in relation to the individual symptoms parents attribute to PDA.

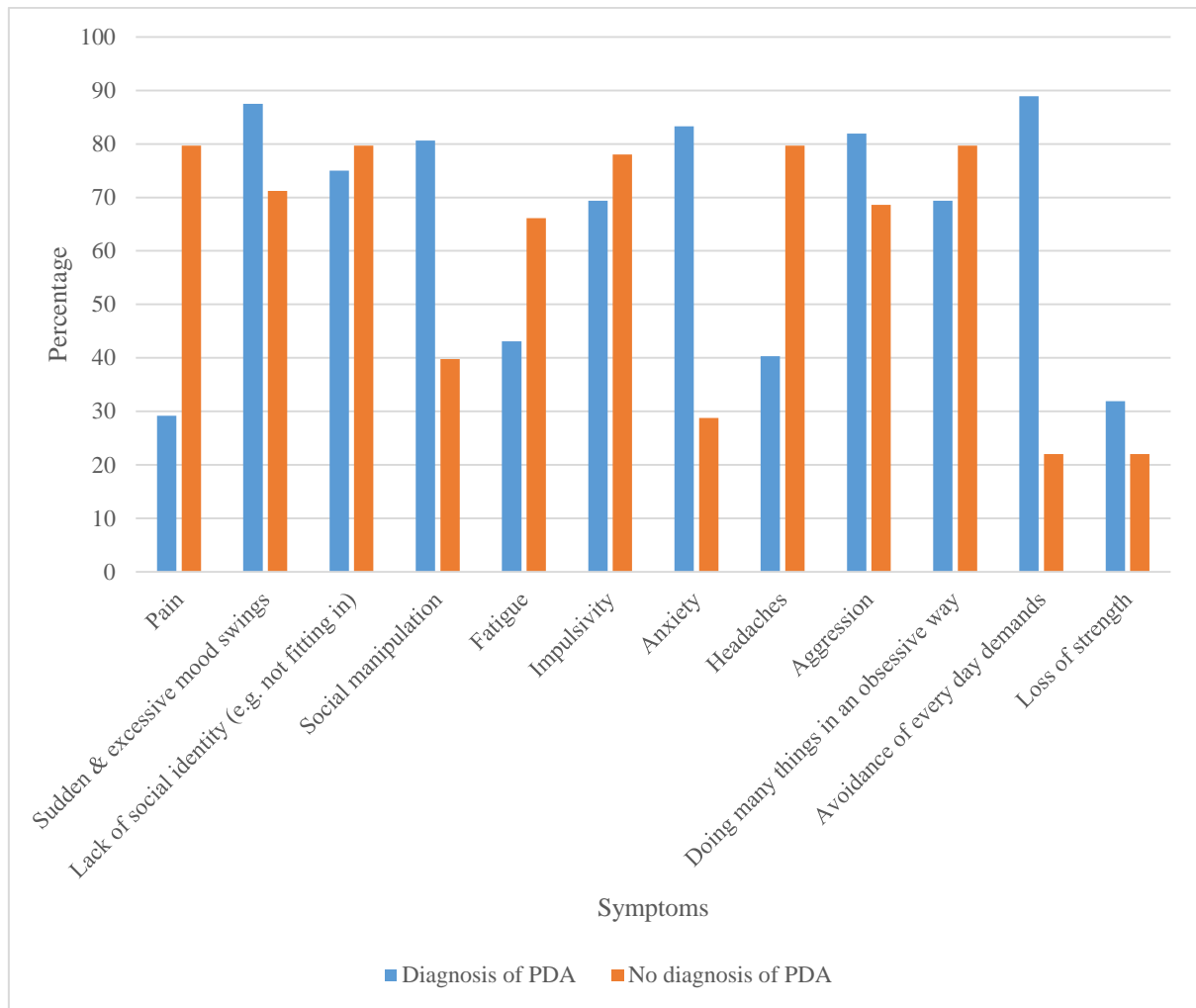


Figure 2. Frequency of symptoms attributed to PDA, by group

The three symptoms most commonly attributed to PDA by parents in the diagnostic group were ‘avoidance of everyday demands’, ‘sudden & excessive mood swings’, and ‘anxiety’, all of which are commonly associated with PDA. The four symptoms most commonly attributed to PDA by parents in the non-diagnostic group were ‘pain’, ‘lack of social identity’, ‘headaches’, and ‘doing many things in an obsessive way’. Interestingly, two of the three least commonly endorsed items in the non-diagnostic group (‘avoidance of every day demands’ and ‘anxiety’) are commonly associated with PDA, with the avoidance of ordinary demands of life being one of the defining components of PDA (Newson, 2003). These data suggest that parents in the non-diagnostic group do not have a clear understanding

of PDA or the symptoms that make up PDA. Parents in the diagnostic group seem to have a better understanding of the symptoms that are attributable to PDA.

Cause

Comparisons were made between the diagnostic and non-diagnostic groups' causal attributions for PDA (Table 5). Statistically significant differences were found for 6 out of the 23 causes listed (a more conservative p value of $p < 0.01$ was adopted, due to the large number of comparisons).

Table 5. Perceived causes of PDA, by group

	Diagnostic group	Non-diagnostic group	
Cause	Mean (standard deviation)	Mean (standard deviation)	p-value
Hereditary	3.94 (1.00)	2.79 (1.47)	<.001
Chance or bad luck	2.87 (1.41)	2.25 (1.24)	.002
Stress	2.35 (1.39)	2.73 (1.45)	.080
Child's personality	2.20 (1.37)	2.65 (1.28)	.022
Child's emotional state	2.11 (1.32)	2.44 (1.46)	.122
Child's mental attitude	1.92 (1.22)	2.17 (1.27)	.178
Child's own behaviour	1.77 (1.02)	2.17 (1.24)	.024
Growing up	1.77 (1.12)	1.74 (1.03)	.816
Parent's emotional state	1.76 (1.04)	2.04 (1.07)	.078
Pollution	1.72 (0.92)	2.01 (1.20)	.810
A germ or virus	1.55 (0.86)	2.08 (1.27)	.002
Diet or eating habits	1.72 (0.93)	1.88 (1.06)	.284
Poor medical care	1.70 (0.93)	1.79 (1.05)	.581
Parent behaviour	1.70 (1.03)	1.88 (1.00)	.245
Parent personality	1.66 (0.97)	1.85 (1.02)	.219
Changes in child's body	1.65 (1.07)	1.78 (0.97)	.386
Family problems or worries	1.63 (0.95)	2.47 (1.38)	<.001
Parent thinking negatively	1.59 (0.92)	1.66 (0.87)	.603
Overwork	1.55 (0.91)	1.87 (1.10)	.038
Lack of parental control	1.52 (0.92)	1.76 (0.95)	.089
Family member drinking	1.37 (.078)	1.39 (0.80)	.842
Accident or injury	1.37 (.066)	1.74 (1.05)	.008
Family member smoking	1.35 (0.76)	1.91 (1.21)	.001

Scores range from 1-5 with higher scores indicating greater agreement that the item was a cause of their child's PDA.

Across the total sample, the most commonly cited cause of PDA was genetics (hereditary, runs in the family), with 84 out of 189 parents (44%) rating it as the most important cause of PDA. 15 parents (8%) named the second most important cause of PDA as problems in pregnancy or during the birth (e.g., stress in pregnancy, low birth weight, birth trauma) and 9 parents (5%) cited their child's personality as the third most important cause of their child's PDA. Findings suggest that the diagnostic group have a clearer conceptualisation of PDA than the non-diagnostic group (who endorsed a wider range of causes), which is in line with the lower levels of illness coherence found in the non-diagnostic group.

To explore the inter-relationships between the illness perception scales across the sample, they were correlated with each other. These data are shown in Table 6.

Table 6. Inter-correlations between IPQ-R dimensions

IPQ-R Subscales	1.	2.	3.	4.	5.	6.	7.	8.
1.Identity	-							
2.Timeline Acute	.08	-						
3.Consequences	.26**	.51**	-					
4.Personal Control	-.08	-.08	-.14	-				
5.Treatment Control	-.07	-.20**	-.20**	.50**	-			
6.Illness Coherence	-.02	.30**	.23**	.23**	-.02	-		
7.Timeline Cyclical	.16*	-.03	.13	-.06	.03	-.04	-	
8.Emotional Representations	.25**	.14	.38**	-.21**	-.19**	-.10	.08	-

** . Correlation is significant at the 0.01 level (2-tailed).

* . Correlation is significant at the 0.05 level (2-tailed).

Table 6 shows a number of significant inter-correlations between subscales of the IPQ-R. The strongest inter-correlation exists between the timeline acute subscale and the consequences subscale ($r=.51$), with parents' beliefs about the chronicity of PDA increasing as perceptions of the negative consequences generated by PDA increased. Other predictable relationships were found, for example, an increased number of symptoms attributed to PDA (illness identity) was associated with greater perceived consequences and more negative

emotional consequences; and a negative relationship between increased perceived consequences and reduced perceptions of ability to treat PDA. Interestingly, perceptions of personal control in relation to PDA were moderately associated with perceptions of treatment control, which might be explained by the fact that the main ‘treatment’ for PDA currently is parental management of behaviour.

Parental Coping

To explore differences between groups in relation to each of the three coping styles (Carver, Katona & Livingston, 2008), Mann-Whitney U was used. Results showed that emotion-focussed coping did not differ significantly between groups. However, parents in the non-diagnostic group used significantly more problem-focussed strategies and dysfunctional coping strategies than parents in the diagnostic group.

Table 7. Coping subscales, by group

	Diagnostic group		Non-diagnostic group		p value
	Median	Interquartile range	Median	Interquartile range	
Emotion-focussed coping	2.13	0.88	2.25	1.16	.22
Problem-focussed coping	1.75	0.58	2.00	0.83	<.0001
Dysfunctional coping	3.50	1.17	4.00	1.67	<.0001

Scores range from 1-4 with higher scores indicating greater use of coping strategy.

Parenting Stress

Mann-Whitney U was used to investigate differences between groups in relation to parenting stress. Findings showed a statistically significant difference between groups, with parents of children with a diagnosis of PDA reporting significantly higher levels of parenting stress than parents in the non-diagnostic group.

Table 8. Parenting stress, by group

	PDA diagnosis		No PDA diagnosis		p value
	Median	Interquartile range	Median	Interquartile range	
Parental Stress Scale	57	15	53	13	.03

Scores range from 18-90 with higher scores indicating higher levels of parenting stress.

The PSS total median score of 57 (IR: 15) for the diagnosis group is 1.5 standard deviations higher than the mean of 43.2 (SD: 9.1) found by Berry and Jones (1995) in parents of children with behavioural problems. The non-clinical sample mean was 37.1 (SD: 8.1). The median parental stress scale score of 53 (IR: 13) for the non-diagnostic group is still over 1 standard deviation higher than the mean found the clinical sample (Berry and Jones, 1995). These data suggest that parents in both groups are experiencing higher levels of stress than comparative samples of parents and non-clinical samples of parents.

Parental Wellbeing

To explore differences between groups in relation to overall wellbeing, a t-test was carried out, but no significant difference was found.

Table 9. Parental wellbeing, by group

	PDA Diagnosis		No PDA Diagnosis		p value
	Mean	Standard deviation	Mean	Standard deviation	
GWBI total	57.68	16.37	61.38	14.34	.22

The GWBI total mean scores of 57.68 (SD: 16.37) for the diagnostic group and 61.38 (SD: 14.34) for the non-diagnostic group are considerably lower than the median values of 73.0 (pre-intervention) and 86 (post-intervention) in a sample of individuals undergoing treatment for stress-related exhaustion (Lundgren-Nilsson, Jonsdottir, Ahlborg and Tennant, 2013). These data suggest that parents across both groups have poor psychological wellbeing.

Parent-child relationship

The Mann Whitney-U test found statistically significant differences between groups in relation to CPRS closeness scores, with parents in the non-diagnostic group reporting a closer relationship with their children than those in the diagnostic group. No differences in conflict scores were found between the two groups.

Table 10. Child-parent relationship, by group

	PDA diagnosis		No PDA diagnosis		p value
	Median ¹	Interquartile range	Median ¹	Interquartile range	
Conflict	4.25	0.75	4.25	0.75	.36
Closeness	3.83	1.33	4.17	1.33	.03

¹ Adjusted median score (sum of scale items divided by number of items); all scores range from 1-5 with higher scores indicating more conflict on the conflict subscale and more closeness on the closeness subscale.

Levels of closeness in both groups were more than 1 standard deviation lower than the mean of 4.62 (0.35) found in a non-clinical sample and levels of conflict across both groups were more than 3 standard deviations higher than the mean of 1.92 (0.64) found in a non-clinical sample (Driscoll & Pianta, 2011).

To explore relationships between outcome variables and parent reported severity of PDA symptoms across the sample, Pearson's *r* was used.

Table 11. Inter-correlations between PDA severity (EDA-Q) and outcomes across groups.

	1.	2.	3.	4.	5.
1.EDA-Q	-				
2.Parenting Stress	.02	-			
3.Wellbeing	-.30*	-.45**	-		
4.Conflict	.48**	.48**	-.44**	-	
5.Closeness	-.28*	-.35**	-.07	-.22	-

** . Correlation is significant at the 0.01 level (2-tailed).

* . Correlation is significant at the 0.05 level (2-tailed).

Table 11 shows that higher EDA-Q scores were significantly associated with reduced overall wellbeing, reduced closeness in the child-parent relationship, and increased conflict in

the child-parent relationship. In addition, the data show that parenting stress was associated with other outcomes in expected directions: moderate relationships between increased parental stress and reduced wellbeing, increased conflict, and reduced closeness in the child-parent relationship. Increased/better wellbeing was moderately associated with reduced conflict in the child-parent relationship.

3.2 Hypothesis Testing

To test the hypotheses, all subsequent analysis focuses only on the data from the diagnostic group (n=71).

For bivariate correlation analyses (hypotheses 1-3) a series of bootstrapped Pearson's r correlation coefficients were used. Bootstrap samples comprised 1,000 re-samples with bias correction, and non-parametric 95% confidence intervals calculated for parameter estimates.

Relationships between child age, duration of diagnosis, severity of PDA (measured by EDA-Q), parental illness perceptions, coping, parenting stress, wellbeing and the quality of the child-parent relationship) were explored for the diagnostic group, using a series of bootstrapped Pearson's r correlation coefficients (Table 12).

Table 12. Inter-correlations between variables for the diagnostic group

	EDA-Q	Child's age	Duration of diagnosis
1.EDA-Q	-	-.37**	-.32*
2.Child's Age	-.37**	-	.52**
3.Duration of diagnosis	-.32*	.52**	-
4.Identity	.37**	.09	-.22
5.Timeline Acute/Chronic	.26	-.24	-.13
6.Consequences	.54**	.09	-.17
7.Personal Control	-.16	-.02	.23
8.Treatment Control	-.42**	.22	.41**
9.Coherence	-.04	-.01	.16
10.Timeline Cyclical	.27	.19	.14
11.Emotional Representations	.22	.07	-.14
12.Emotion-focussed coping	-.06	-.04	.10
13.Problem-focussed coping	.27	-.08	-.12
14.Dysfunctional coping	.27	.05	.05
15.Wellbeing	-.36**	.09	.18
16.Parenting stress	-.04	.19	-.13
17.Conflict	.53**	.10	-.18
18.Closeness	-.26	-.10	.15

*

Correlation is significant at the 0.05 level (2-tailed).

** . Correlation is significant at the 0.01 level (2-tailed).

Table 12 shows a significant negative association between reported PDA severity and duration of diagnosis, indicating that parent reports of PDA severity decrease as the length of time a child has been diagnosed with PDA increases. EDA-Q scores were also significantly related to parental perceptions of identity, consequences and treatment control in relation to PDA, with the total number of symptoms parents attribute to PDA increasing, perceptions regarding the consequences associated with PDA increasing and perceptions of treatment control reducing as parent reports of PDA increase. Parental perceptions of treatment control were also significantly and positively related to duration of PDA diagnosis. EDA-Q scores were significantly and negatively related to parental wellbeing, and significantly and positively related to child-parent conflict. Accordingly, EDA-Q scores will be controlled for in subsequent mediation analyses.

Hypotheses 1: There will be a relationship between domains of parental illness perceptions of PDA and self-reported parenting stress, parental wellbeing, conflict in the child-parent relationship and closeness in the child-parent relationship.

Parental illness perceptions of PDA were correlated with parental wellbeing (General Wellbeing Index), parenting stress (Parental Stress Scale), child-parent conflict (Child-Parent Relationship Scale) and child-parent closeness (Child-Parent Relationship Scale). Results are shown in Table 13.

Table 13. Bootstrapped Pearson's r correlation coefficients between parental illness perceptions of PDA and outcomes¹.

	PSS Total	GWBI Total	Conflict	Closeness
Identity	.17	-.22	.20	-.13
Timeline Acute/Chronic	.12	-.11	.08	-.13
Consequences	.27*	-.30*	.35**	-.20
Personal Control	-.31*	.20	-.40**	.28*
Treatment Control	-.08	.13	-.33**	.18
Coherence	-.13	.10	-.34**	.26*
Timeline Cyclical	-.08	-.15	.16	.03
Emotional Representations	.50**	-.44**	.39**	-.19

** . Correlation is significant at the 0.01 level (2-tailed).

* . Correlation is significant at the 0.05 level (2-tailed).

¹ Correlations were deemed significant if the 95% confidence intervals did not include zero.

Table 13 shows significant relationships between parental illness perceptions of PDA and parenting stress, parental wellbeing, and the child-parent relationship. Parenting stress and child-parent conflict were significantly and positively correlated with perceptions of illness consequences and emotional representations, and parental wellbeing was significantly and negatively correlated with perceptions of illness consequences and emotional representations (i.e., a greater number of perceived consequences relating to PDA and higher levels of negative emotional consequences are associated with increased parenting stress, increased child-parent conflict and reduced wellbeing). Parenting stress and child-parent

conflict were significantly and negatively correlated with personal control (i.e., perceptions of lower levels of personal control were associated with increased levels of parenting stress), while child-parent closeness was significantly and positively related to personal control. Additionally child-parent conflict and child parent closeness were significantly associated with illness coherence, with conflict reducing and closeness increasing as parents' understanding of PDA improves. Finally, child-parent conflict was found to reduce as perceptions of treatment control increased.

Overall, hypothesis 1 is supported by the data, which indicate that parental perceptions of PDA are significantly associated with parent reports of parenting stress, wellbeing, and conflict and closeness in the child-parent relationship.

Hypothesis 2: There will be a relationship between domains of parental illness perceptions of PDA and coping strategies employed by parents to deal with their child's PDA.

Parental illness perceptions of PDA were correlated with parental coping. Results are shown in Table 14.

Table 14. Bootstrapped Pearson's *r* correlation coefficients between parental illness perceptions of PDA and parental coping strategies¹.

	Emotion-focussed	Problem-focussed	Dysfunctional coping
Identity	.03	.37**	.14
Timeline Acute/Chronic	-.01	.04	.22
Consequences	.06	.25*	.22
Personal Control	.35**	.02	.02
Treatment Control	.07	-.19	.10
Illness Coherence	.05	.05	-.006
Timeline Cyclical	.04	-.03	.15
Emotional Representations	-.002	.06	.38**

** . Correlation is significant at the 0.01 level (2-tailed).

* . Correlation is significant at the 0.05 level (2-tailed).

¹ Correlations were deemed significant if the 95% confidence intervals did not include zero.

Table 14 shows a small number of significant relationships between parental illness perceptions of PDA and coping strategies employed by parents to deal with their child's PDA. Greater use of emotion-focussed coping was significantly associated with increased perceptions of personal control over PDA. Results also show that the more symptoms parents identify with PDA and the more negative consequences they perceive in relation to PDA, the more likely they are to use problem-focussed coping strategies. For dysfunctional coping, greater use of this strategy was associated with higher levels of negative emotional consequences of PDA.

Overall, hypothesis 2 is supported by the data with a small number of significant, weak to moderate strength correlations, indicating that parental perceptions of PDA were related to coping.

Hypotheses 3: There will be relationships between parental coping strategies and self-reported parental wellbeing, parenting stress, child-parent conflict and child-parent closeness.

Parental coping styles were correlated with parental outcomes. The relationships between coping and outcomes are shown in Table 15.

Table 15. Bootstrapped Pearson's r correlation coefficients between parental coping strategies, parental wellbeing and stress, and severity of PDA¹.

	Emotion-focused (confidence interval)	Problem-focused (confidence interval)	Dysfunctional coping (confidence interval)
PSS Total	-.16	-.03	.25
GWBI Total	.29*	-.16	-.14
CPRS Conflict	-.14	.04	.26
CPRS Closeness	-.07	-.12	-.06

**. Correlation is significant at the 0.01 level (2-tailed).

*. Correlation is significant at the 0.05 level (2-tailed).

¹ Correlations were deemed significant if the 95% confidence intervals did not include zero.

Table 15 shows a significant relationship between emotion-focussed coping and parental wellbeing, such that higher levels of overall wellbeing were associated with greater use of emotion-focussed coping strategies. As only emotion-focussed coping was related to outcome variables, there is only partial support for hypothesis 3.

Hypothesis 4a: Coping will mediate the relationship between parental illness perceptions of PDA and parental wellbeing

The role of coping as a mediator in the relationship between illness perceptions and wellbeing was explored using the asymptotic and bootstrap methods for examining and comparing indirect effects. Hayes (2013) outlined multiple mediator models and the parallel multiple mediator model has been employed here, which allows for simultaneous examination of the three coping styles as potential mediators of the relationship between parental illness perceptions and parental wellbeing. Only the illness perceptions that were significantly correlated with parental wellbeing and at least one of the coping styles (see Tables 14 and 15) were selected for further analysis.

Accordingly, consequences and emotional representations were selected as independent variables in the model depicted in Figure 3. As EDA-Q scores were significantly correlated with overall wellbeing scores (Table 12), EDA-Q was included as a covariate to control for the impact of PDA severity on parental wellbeing. Results are shown in Table 16.

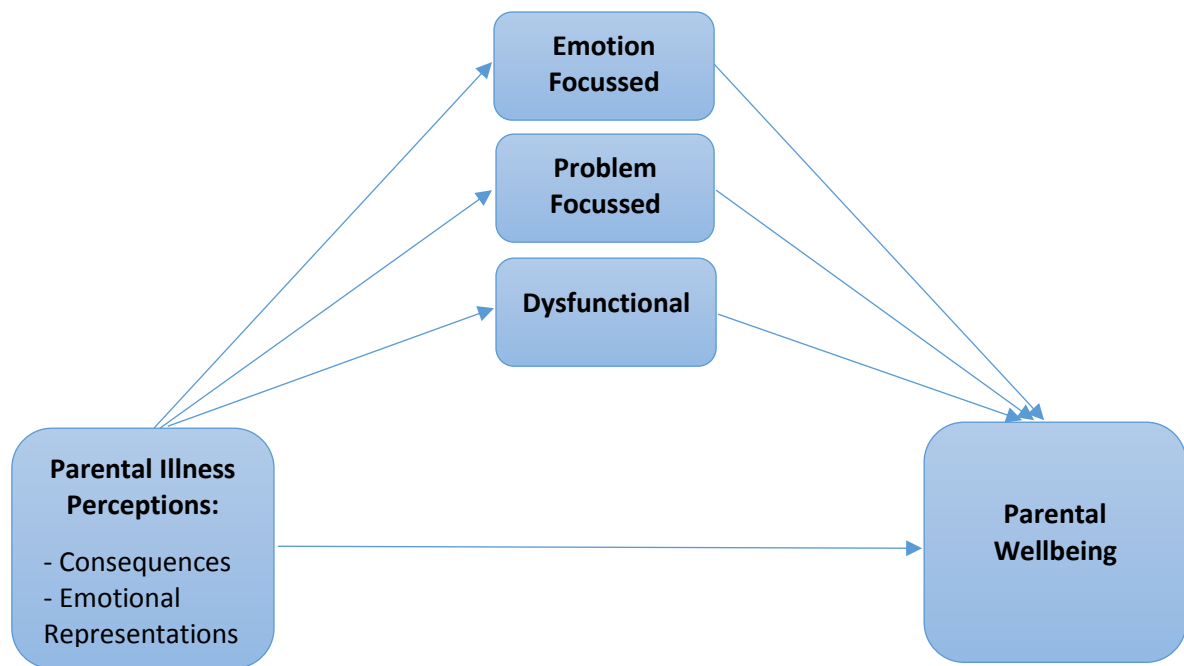


Figure 3. Hypothesised direct and indirect relationship between parental illness perceptions and overall wellbeing.

Table 16. Direct and indirect relationships between parental illness perceptions and parental wellbeing

	Asymptotic Path Estimate	Bootstrap Path Estimate	Bias	Standard Error	Lower 95% Bootstrap Confidence Interval	Upper 95% Bootstrap Confidence Interval
Illness Perceptions: Consequences (R²=0.18; p =0.02)						
Direct Effect	-1.7890			.9742	-3.7347	.1567
Emotion Focused	.8434	1.0378	.1944	.2411	-.1172	.9653
Problem Focused	-.5982	-.8358	-.2376	.2547	-.9869	.0772
Dysfunctional	-.3810	-.3958	-.0148	.1922	-.5848	.2910
Total of Indirect Effects		-.0580		.3300	-.8098	.5629
Illness Perceptions: Emotional Representations (R²=0.27.; p < 0.001)						
Direct Effect	-1.5047			.4391	-2.3817	-.6276
Emotion Focused	.5668	.5478	-0.0190	.0845	-.3037	.0918
Problem Focused	-.5942	-.5975	-.0033	.0649	-.1650	.1119
Dysfunctional	-.0807	-.0496	.0311	.1958	-.2935	.5330
Total of Indirect Effects		.0088		.2655	-.3349	.52175

As can be seen from Table 16, R-squared values show significant effects of parental perceptions of illness consequences and coping, and emotional representations and coping on the variance of overall wellbeing. The variance in parental wellbeing relating to emotional

representations is most likely to be explained by the direct effect of emotional representations on parental wellbeing. Results show that the mediated pathways were non-significant for both models presented in Table 16.

In contrast to Leventhal's self-regulation model, coping did not mediate the relationship between illness consequences and parental wellbeing or the relationship between emotional representations and parental wellbeing.

Hypothesis 4b: Coping will mediate the relationship between parental illness perceptions and parenting stress

The role of coping style as a mediator in the relationship between illness perceptions and parental stress was examined (Figure 4). Results are shown in Table 17.

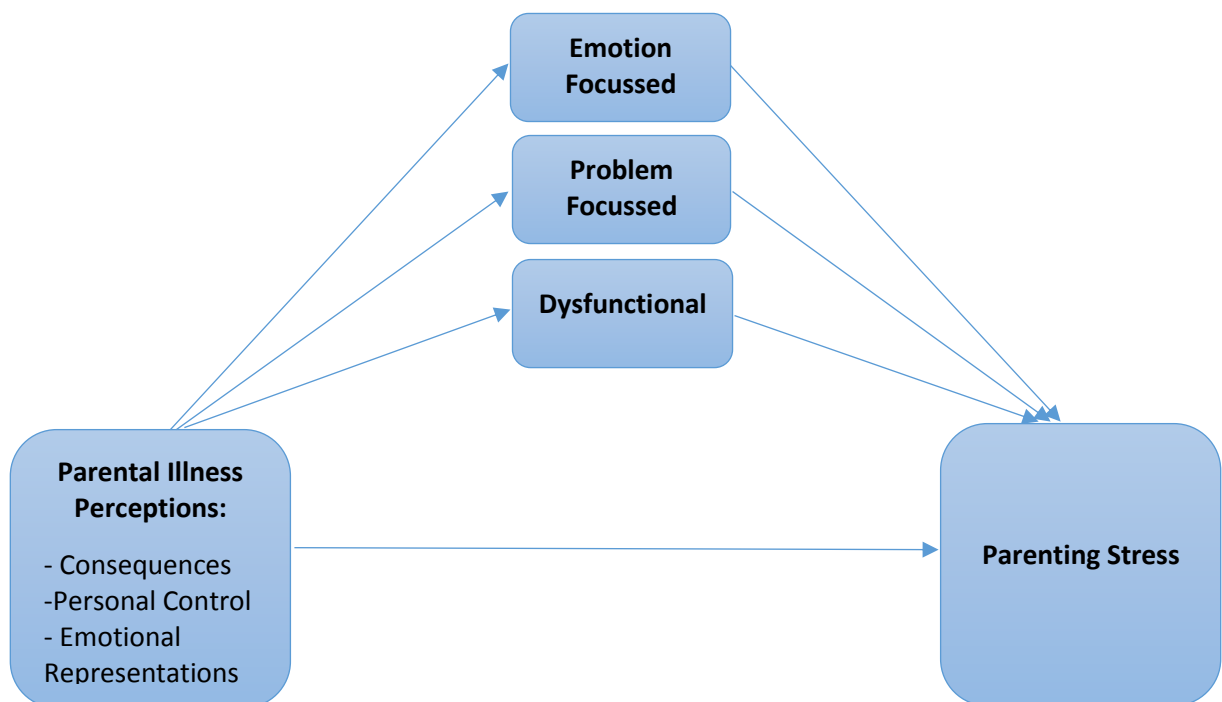


Figure 4. Hypothesised direct and indirect relationship between illness perceptions and parenting stress

Table 17. Direct and indirect relationships between parental illness perceptions and parenting stress

	Asymptotic Path Estimate	Bootstrap Path Estimate	Bias	Standard Error	Lower 95% Bootstrap Confidence Interval	Upper 95% Bootstrap Confidence Interval
Illness Perceptions: Consequences ($R^2=0.15$; $p=0.03$)						
Direct Effect	1.0796			.5178	.0458	2.1135
Emotion Focused	-.3026	-.3449	-.0423	.0905	-.3279	.0692
Problem Focused	-.0233	-.0333	-.0100	.2003	-.3664	.4393
Dysfunctional	.5070	.5699	.1529	.1684	-.0670	.6526
Total of Indirect Effects		.1006		.2669	-.3809	.6559
Illness Perceptions: Personal Control ($R^2=0.16$; $p=0.02$)						
Direct Effect	-.7637			.3249	-1.4124	-.1149
Emotion Focused	-.0900	-.1258	-.0358	.1306	-.3234	.2105
Problem Focused	.1413	.1433	.0020	.0436	-.0474	.1289
Dysfunctional	.5355	.5293	-.0062	.0931	-.2381	.1598
Total of Indirect Effects		-.0399		.1519	-.3535	.2788
Illness Perceptions: Emotional Representations ($R^2=0.2738$; $p=0.0003$)						
Direct Effect	1.0549			.2601	.5356	1.5742
Emotion Focused	-.1497	.1442	.0055	.0336	-.0352	.1231
Problem Focused	.0286	.0295	.0009	.0380	-.0639	.0856
Dysfunctional	.2366	.3363	.0997	.1019	-.0629	.3553
Total of Indirect Effects		.1061		.1154	-.1054	.3737

As can be seen from Table 17, R-squared values show there were significant effects for the models including illness consequences, personal control and parental emotional representations on the variance of parenting stress. The variance explained in parenting stress across the three models presented in Table 17 is most likely explained by the direct effects of illness consequences, personal control and emotional representations on parental wellbeing (as indicated by confidence intervals that do not include zero). Results show that the mediated pathways were non-significant, indicating that coping did not mediate the relationship between parental illness perception domains and parenting stress.

Again, in contrast to Leventhal's self-regulation model, the likely direct effects of consequences, personal control and emotional representations on parenting stress were

significant, but this interaction was not mediated by any of the three coping styles.

Hypothesis 4c: Coping will mediate the relationship between parental illness perceptions and child-parent conflict.

The role of coping style as a mediator in the relationship between illness perceptions and child-parent conflict was examined (Figure 5). Results are shown in Table 18.

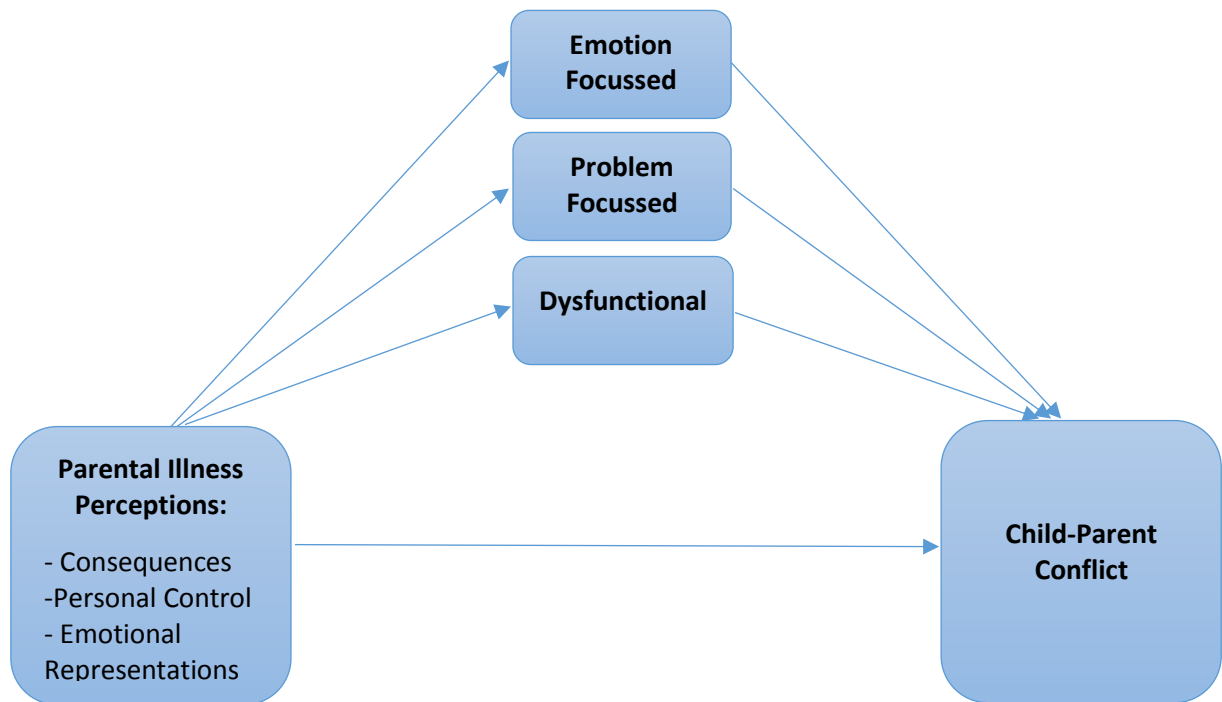


Figure 5. Hypothesised direct and indirect relationship between illness perceptions and child-parent relationship.

Table 18. Direct and indirect relationships between parental illness perceptions and child-parent conflict

	Asymptotic Path Estimate	Bootstrap Path Estimate	Bias	Standard Error	Lower 95% Bootstrap Confidence Interval	Upper 95% Bootstrap Confidence Interval
Illness Perceptions: Consequences ($R^2=0.29$; $p=0.0004$)						
Direct Effect	.4008			.2358	-.0702	.8718
Emotion Focused	-.1062	-.1307	-.0245	.0366	-.1343	.0215
Problem Focused	-.0823	-.1150	-.0327	.0603	-.2103	.0522
Dysfunctional	.1051	.1092	.0041	.0475	-.0867	.1290
Total of Indirect Effects		-.0531		.0719	-.2106	.0765
Illness Perceptions: Personal Control ($R^2=0.34$; $p<0.0001$)						
Direct Effect	-.3585			.1273	-.6127	-.1043
Emotion Focused	.0090	.0126	.0036	.0446	-.0867	.1042
Problem Focused	-.0532	-.0556	-.0024	.0149	-.0481	.0149
Dysfunctional	.0925	.0967	.0042	.0193	-.0229	.0586
Total of Indirect Effects		.0054		.0461	-.0764	.1200
Illness Perceptions: Emotional Representations ($R^2=0.33$; $p<0.0001$)						
Direct Effect	.2837			.1096	.0648	.5025
Emotion Focused	-.0535	-.0517	.0018	.0140	-.0142	.0514
Problem Focused	-.0740	-.0744	-.0004	.0128	-.0318	.0220
Dysfunctional	.0194	.0269	.0075	.0403	-.0735	.0986
Total of Indirect Effects		.0089		.0469	-.0925	.1117

As can be seen from Table 18, R-squared values show there were significant effects for the three models that included perceptions of illness consequences, personal control and emotional representations on the variance in child-parent conflict. The variance in child-parent conflict for the models relating to personal control and emotional representations is most likely explained by the direct effects of personal control and emotional representations on child-parent conflict. Results show that coping did not mediate the relationship between parental illness perceptions and perceived conflict in the child-parent relationship.

In summary, these findings do not fit with the model associations suggested by Leventhal's self-regulation framework. Although the direct effects of personal control and emotional representations on child-parent conflict were significant, the interactions were not

mediated by any of the three coping styles.

Hypothesis 4d: Coping will mediate the relationship between parental illness perceptions and child-parent closeness.

The role of coping style as a mediator in the relationship between illness perceptions and child-parent closeness was examined (Figure 6). Results are shown in Table 19.

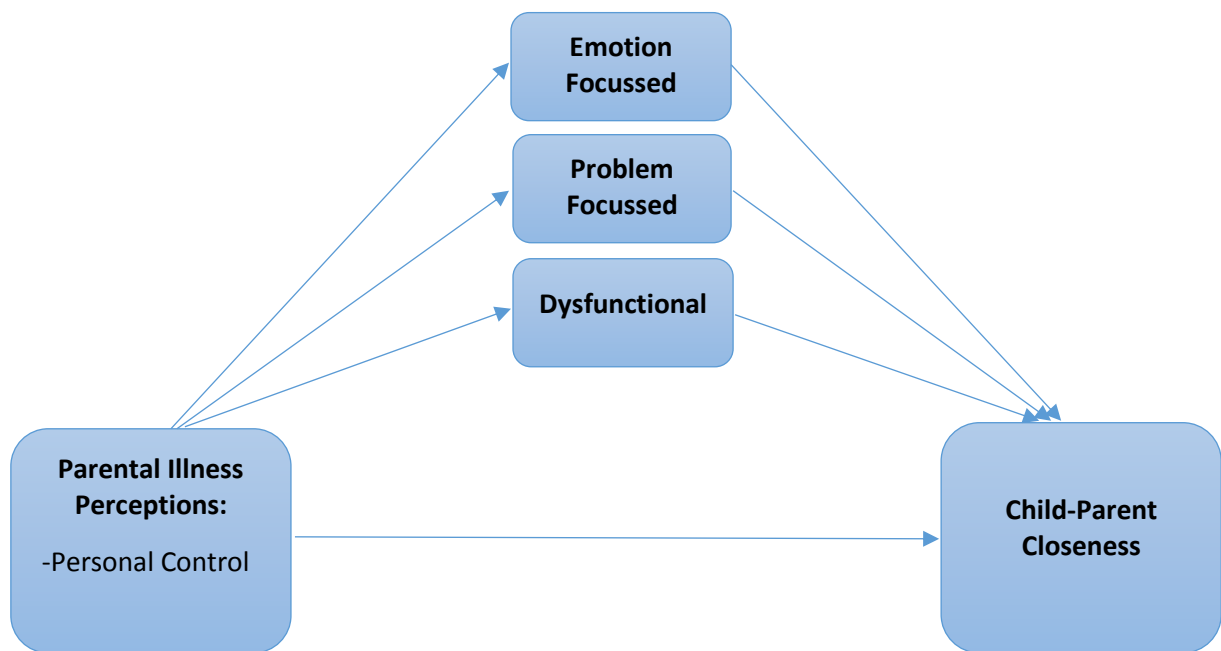


Figure 6. Hypothesised direct and indirect relationship between illness perceptions and child-parent closeness

Table 19. Direct and indirect relationships between parental illness perceptions and child-parent closeness

	Asymptotic Path Estimate	Bootstrap Path Estimate	Bias	Standard Error	Lower 95% Bootstrap Confidence Interval	Upper 95% Bootstrap Confidence Interval
Illness Perceptions: Personal Control ($R^2=0.14$; $p=0.08$)						
Direct Effect	.3995			.1714	.0571	.7419
Emotion Focused	-.0829	-.1164	-.0335	.0709	-.2129	.0861
Problem Focused	-.1168	-.1220	-.0052	.0219	-.0770	.0227
Dysfunctional	-.0600	-.0627	-.0027	.0267	-.0852	.0306
Total of Indirect Effects		-.0414		.0707	-.2058	.0888

Table 19 shows a statistically significant direct effect of personal control on child-parent closeness, however the model as a whole was not significant in explaining the variance in child-parent closeness. Results show that the mediated pathways were non-significant.

Once again, these findings do not fit with the hypothesised links in Leventhal's self-regulation model, in that coping did not mediate the relationship between perceptions of personal control and child-parent closeness.

4. DISCUSSION

The current study aimed to: (1) explore the pattern of perceptions parents hold about PDA or suspected PDA, (2) explore coping among parents with a child with PDA or suspected PDA, (3) explore levels of parental stress, wellbeing, and child-parents relationships in PDA and suspected PDA, and (4) explore the interrelationships between these variables, to determine the applicability of the SRM (Leventhal, Nerenz and Steele, 1984). This is the first study to explore these variables, and their interaction, in the context of childhood PDA

Findings show that PDA was viewed as a chronic condition with considerable negative consequences across groups. These findings are comparable with the findings of Baines & Wittkowski's (2013) review, which found that mental health problems were viewed as chronic with serious negative consequences. The illness perception domains that were least strongly held across both groups were treatment control and personal control, indicating that parents do not hold particularly positive beliefs about the controllability of PDA.

Significant differences were found between the diagnostic group and the non-diagnostic group in relation to parental illness perceptions across personal control and coherence domains, with parents of children with a diagnosis of PDA perceiving they have

more control over their child's PDA and a better, more coherent, understanding of the condition. Parents' responses on the identity and causes subscales also indicated that parents in the diagnostic group had a greater conceptualisation of PDA than parents in the non-diagnostic group, such that parents in the non-diagnostic group endorsed fewer symptoms that are recognised characteristics of PDA and a greater number of causes at a generally higher level than parents in the diagnostic group. One hypothesis to explain these findings is that parents gain information and support post-diagnosis, improving their understanding of PDA.

Across groups, findings show that dysfunctional coping was the coping style most frequently used by parents to cope with their child's PDA/PDA symptoms and problem-focussed coping was the least frequently used coping style. The non-diagnostic group used dysfunctional coping (e.g. giving up trying to deal with it and using alcohol or other drugs to feel better) and problem-focussed coping (e.g. getting help and advice from other people and concentrating efforts on doing something about the situation) significantly more than the diagnostic group, and the reasons, although speculative, might relate to additional difficulties in getting an assessment and a diagnosis for PDA.

Parents in the diagnostic group reported significantly higher stress levels than parents in the non-diagnostic group. Across both groups, parenting stress levels were high and overall wellbeing was poor, even in comparison to clinical samples (Berry and Jones, 1995; Lundgren-Nilsson, Jonsdottir, Ahlborg and Tennant, 2013) indicating that parents of a child with PDA and those who suspect their child has PDA are an 'at risk' group. Inter-correlations suggest that elevated stress levels may pose risks for mental health difficulties and the child-parent relationship.

Across both groups, parent reports indicated that child-parent closeness was lower than in a non-clinical sample and child-parent conflict was considerably higher than in a non-clinical sample (Driscoll & Pianta, 2011). There was a statistically significant difference between groups in relation to child-parent closeness, with the diagnostic group reporting less closeness in their relationship with their child. Child-parent closeness was significantly and negatively related to parenting stress, and given that parents in the diagnostic group reported less closeness in their relationship with their child, as well as higher levels of parenting stress, it is possible that child-parent closeness impacts directly on parenting stress, or responses to parenting stress impact on child-parent closeness, which may, to some extent, explain differences in parenting stress between groups.

The SRM was used to explore the relationships between variables. Findings show that emotional representations was the illness perception most strongly associated with outcomes across both groups. It is possible that there is some conceptual overlap between parental perceptions of emotional representations (i.e., the severity of the emotional responses generated by PDA) and parenting stress, overall wellbeing and the quality of the child-parent relationship, as the emotional representations subscale of the IPQ-R includes items pertaining to feeling depressed, upset, angry and anxious in relation to their child's PDA.

For the diagnostic group, parents' reports of higher levels of parenting stress, poorer overall wellbeing and conflict in the child-parent relationship were found to be significantly associated with perceptions of greater consequences and greater negative emotional consequences generated by PDA. Parental reports of higher levels of parenting stress and child-parent conflict, and lower levels of closeness were significantly associated with perceptions of reduced personal control in relation to PDA. Perceptions of a clearer understanding of PDA were significantly associated with lower levels of conflict and higher

levels of closeness in the child-parent relationship. Additionally, perceptions of less treatment control were found to be significantly associated with increased child-parent conflict. Child-parent conflict was more strongly related to parental perceptions of PDA (consequences, personal control, treatment control, illness coherence and emotional representations) than any other outcome variable.

In line with the SRM, significant relationships were found between four illness perception domains (identity, consequences, personal control, emotional representations) and coping for the diagnostic group. A moderate positive correlation was found between emotion-focussed coping and perceptions of personal control in relation to PDA, positive associations were found between problem-focussed coping and the number of symptoms parents attributed to their child's PDA (moderate correlation) and perceptions of the negative consequences related to PDA (weak correlation), and dysfunctional coping was found to be moderately and positively correlated with perceptions of the negative emotional consequences associated with PDA.

The SRM predicts that relationships will exist between coping and outcomes, but only one significant (weak) relationship was found between parental coping and wellbeing for the diagnostic group. Parental wellbeing was found to increase as the use of emotion-focussed coping increased, indicating that emotion-focussed coping is a favourable coping style. Previous research has found both beneficial effects of emotion-focussed coping (Pottie & Ingram, 2008; Savage, 2011) and detrimental effects of emotion-focussed coping (Dabrowska & Pisula, 2010; Lyons, Leon, Roecker Phelps & Dunleavy, 2009) in parents of children with ASD. The ambiguity in current literature could be associated with a lack of coherence in understanding what behaviours contribute to emotion-focussed coping. Further research may be required in order to better understand the relationship between emotion-focussed coping

and parental wellbeing. Although relationships between problem-focussed coping, dysfunctional coping and outcomes did not reach statistical significance, results suggest that problem-focussed coping and dysfunctional coping are unhelpful coping strategies. Longitudinal research may be helpful in better understanding the direction of the relationships between coping, wellbeing and the child-parent relationship.

Across both groups, moderate correlations were found between child-parent conflict and both parenting stress and overall wellbeing, so it may be that child-parent conflict impacts on parenting stress and parental wellbeing, or child-parent conflict is a response to high levels of parenting stress and poor wellbeing. Further research is required to enhance the understanding of the relationships between child-parent conflict and parental wellbeing and establish causality.

Across both groups, parental wellbeing and child-parent closeness were found to reduce and child-parent conflict was found to increase as parent reported severity of PDA increased, indicating that the severity of PDA may have clinical implications for parental and relational outcomes, highlighting the importance of considering systemic factors in clinical work with this population. Additionally, findings showed that parent reports of PDA severity decreased, as the length of time following diagnosis of PDA increased. One hypothesis to explain this latter finding is that parents learn/develop strategies to deal with their child's PDA symptoms over time, indicating that coping may change over time. Once a child receives a diagnosis of PDA, parents may become more skilled at dealing with/reducing associated behaviours and symptoms, which in turn reduces the frequency or severity of PDA symptoms. In support of this hypothesis, treatment control was the only illness perception domain found to increase with duration of diagnosis, indicating that parents' belief that PDA can be managed/controlled increases over time. When asked specifically about treatments,

parents provided details of management strategies, such as carefully wording requests and offering choices, with some explicitly stating that there are no available ‘treatments’ for PDA, which may suggest that there is some overlap between treatment control and personal control, as evidenced by the moderate correlation between these illness perception domains.

Findings showed significant relationships between parental illness perceptions and coping, between illness perceptions and all four outcomes, and between coping and parental wellbeing, demonstrating the utility of the SRM with parents of a child with PDA and parents who suspect their child has PDA. One criticism of using the SRM in relation to PDA is that the SRM refers to ‘illness’ and ‘health threats’, which has the potential to medicalise the behaviours associated with PDA. The medicalisation of such behaviours can disempower individuals and their families (Valentine, 2010) and lead to a search for a cure/treatment.

Mediation analyses did not find evidence to show that coping mediated the relationship between illness perceptions and outcomes, as the SRM predicts. The percentage of variance in outcomes explained by illness perceptions directly and through coping (ranging from 14% to 34%) indicates that there is a large proportion of variance that is left unaccounted for by the SRM. Although coping has not been found to mediate the relationship between illness perceptions and outcomes, the amount of variance in outcomes accounted for by coping and illness perceptions, along with the direct associations with coping, indicate that coping is still an important factor. Further research is required to determine what other factors contribute to the variance in outcomes.

Hagger and Orbell’s (2003) meta-analytic review did not test the hypothesis that coping mediates the relationship between illness representations and outcomes, and a more recent systematic review in the field of mental health (Baines and Wittkowski, 2013) only

addressed the direct relationships between illness perceptions, coping and outcomes. Individual studies have found mixed results, with some finding that coping does mediate the relationship between illness representations and outcomes (e.g. Ziarko, Mojs, Piasecki & Samborski, 2014; Knowles, Swan, Salzberg, Castle & Langham, 2014), some finding that coping does not mediate the relationship between illness perceptions and outcomes (e.g. Kaptein et al., 2006; Edgar & Skinner, 2003), and others finding that some coping styles mediate the relationship, while others do not (e.g. Tiggelman, van de Ven, Schayck, Kleinjan & Engels, 2014).

4.1 Clinical implications and implications for future research

As hypothesised, parents of children with a diagnosis of PDA (and those who suspect their child has PDA) report high levels of parenting stress and poor wellbeing, suggesting that they are an ‘at risk’ group. These findings are in line with the literature relating to parental stress and wellbeing in parents of children with ASD (Hayes & Watson, 2013; Estes et al., 2013). Results of this study highlight the importance of taking a systemic and relational approach to the assessment of children presenting with PDA or with suspected PDA. Results emphasise the need for interventions targeting parenting stress, parental wellbeing and the child-parent relationship in this population, particularly given the well documented implications of parental stress on child outcomes (Osborne, McHugh, Saunders & Reed, 2008; Le Cavalier, Leone & Witz, 2006; Hastings, 2002; Essex, Klein, Cho & Kalin, 2002). Statutory organisations should be offering comprehensive assessments and targeted interventions, for parents of children with PDA/suspected PDA. Findings show moderate associations between child-parent conflict, parenting stress and parental wellbeing, and weak associations between child-parent closeness and parenting stress. Longitudinal research is required to determine the direction of these associations.

The SRM, along with the findings of the current study, indicate that targeting parental illness perceptions may have a beneficial impact on parenting stress, parental wellbeing and/or the child-parent relationship. Therefore, future research should focus on the evaluation of interventions that aim to alter parental illness perceptions of consequences, personal control, illness coherence and emotional representations, with the aims of increasing perceptions of personal control, increasing parents' understanding of PDA, reducing perceptions of emotional consequences and overall consequences associated with PDA. Cognitive interventions to explore and address assumptions, interpretations and potential cognitive biases associated with PDA may help to reduce the emotions generated in response to PDA (emotional representations). Research demonstrates that illness perceptions can be modified, with positive effects (Keogh et al., 2011).

Findings also indicate that the IPQ-R is a useful tool in the context of childhood PDA, which could be used in clinical settings to assess parental illness perceptions. The IPQ-R focuses, to some degree, on disorder and deficit; with a larger number of items reflecting deficits than those reflecting resilience factors. Hence, this measure may be valuable in identifying perceptions that are associated with negative outcomes, which may aid the development of individualised interventions.

Researchers may want to consider further research exploring the relationships between coping and parental stress/wellbeing, in order to gain a greater understanding of what coping strategies are most beneficial. This understanding could be used to inform interventions aimed at improving parental wellbeing and the child-parent relationship.

Evidence suggests that psychoeducation, parenting programs and CBT are effective in improving parent outcomes: parenting stress (e.g. Al-Khalaf, Dally & Dempsy, 2014; Chiang,

2014), parenting self-efficacy (e.g. Tellegen & Sanders, 2014; Whittingham, Sofronoff & Shefflied, 2009) and depressive symptoms (e.g. Feinberg et al., 2014; Ruiz-Robledillo & Moya, 2015) in parents of children with ASD. Research involving parents of children with ASD also indicates that they experience low levels of parenting self-efficacy, therefore it may be important to evaluate levels of parenting self-efficacy and interventions that have been found to be effective in an ASD population in future research with parents of children with PDA.

4.2 Limitations

One limitation of this research is that 86% of parent participants classed themselves as ‘white British’ and 96% of participants were female. The homogeneity of the sample may limit the generalisability of the findings. Additionally, diagnoses were based solely on parent report and inclusion in the non-diagnostic group was based on parents suspecting their child has PDA.

414 parents/caregivers who started the survey, did not complete the survey. The relatively low number of complete responses may be indicative of the length of the survey, the limited time and resources available to these parents due to the day-to-day demands of caring for children with PDA/suspected PDA and/or the high stress levels evidenced in this population. It may be helpful to consider the length of surveys and the demands placed on these parents when planning future research.

As a consequence of the cross-sectional design employed in the current study, relationships between variables can be identified, but causality cannot be established. It is possible that the relationships are bi-directional, i.e. parental illness perceptions of PDA effect outcomes of parenting stress, parental wellbeing and the child-parent relationship and

outcomes of parenting stress, parental wellbeing and the child-parent relationship effect parental illness perceptions of PDA. This hypothesis is consistent with the SRM, where outcomes feedback into illness perceptions and coping strategies (Figure 1).

4.3 Summary

Overall, this study shows that parents of child with PDA/suspected PDA view PDA as a chronic condition with considerable negative consequences. These parents have been found to have elevated levels of parenting stress and low levels of psychological wellbeing, which is in line with findings for parents of children with ASD. Reports indicate that there is more conflict in the child-parent relationship than closeness for children with PDA/suspected PDA. The most commonly used coping style was dysfunctional coping, which results suggest is unhelpful. Relationships were found between illness perceptions and outcomes, though these relationships were not mediated by coping. Findings indicate the applicability of the SRM to PDA and the results have important implications for clinicians working with parents of a child with PDA/suspected PDA.

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PUBLIC DISSEMINATION DOCUMENT

The work presented in Volume 1 of this thesis details the research undertaken as part of the Clinical Psychology Doctorate at the University of Birmingham. It is comprised of two components: (1) a review of the literature exploring the effectiveness of interventions designed for parents of children with a diagnosis of autism spectrum disorder (ASD) and (2) a research paper investigating the relationships between parental beliefs about pathological demand avoidance syndrome (PDA), coping styles, wellbeing, parenting stress and the quality of the relationship between children and parents. Details of each paper are provided below:

1) Parents of children with autism: the effectiveness of parent interventions on parental stress, wellbeing and parenting confidence.

Introduction: There is wealth of evidence that shows parents of a child with ASD can experience high levels of parental stress, poor emotional wellbeing and low confidence in parenting (parental self-efficacy). Despite extensive research evidencing the impact on parents of parenting a child with ASD, there are relatively few intervention studies that have focussed specifically on parents as the main focus of research. The aim of the review was to examine the evidence for how effective interventions for parents of children with ASD are.

Method: 22 papers were included in the review. The research papers were reviewed to assess their quality and consider their findings. Parent interventions were grouped into five categories: psychoeducation, parenting programme, parent-mediated intervention, mindfulness and Cognitive Behavioural Therapy (CBT).

Results: Both within and between intervention categories, interventions varied greatly in relation to content delivered, approach, length of the intervention, how the intervention was

delivered and the quality of the research, making it difficult to make direct comparisons between interventions and intervention categories. In this review, education programmes that provided parents with information about ASD (psychoeducation) were the intervention type most commonly associated with reductions in parental stress; parenting programmes (which provided parents with management strategies) was the intervention type most commonly associated with improvements in parenting confidence/self-efficacy. There is some evidence to suggest that parent interventions have better outcomes in parents who experience poor mental health prior to the start of the intervention.

Conclusions: The studies varied widely in the nature of their interventions, which made it difficult to draw any reliable conclusions about the effectiveness of interventions for parents. When there are more papers and better quality papers, a statistical analysis of the results across all papers will allow researchers to take account of factors that might influence outcomes for parents (e.g., how interventions are delivered; amount of contact hours) which would make for more reliable conclusions. Clinicians and researchers may wish to measure parent outcomes as part of routine practice, even when interventions do not directly involve parents, because there is evidence to suggest that child interventions can improve parent outcomes (Karst et al., 2014).

2) Parental illness representations in pathological demand avoidance syndrome: parental coping, stress, wellbeing and the child-parent relationship.

Introduction: The self-regulation model (SRM; Leventhal, Nerenz & Steele, 1984) offers a theory to explain why individuals may differ in their responses to the same health condition. The SRM proposes that people make sense of a health condition they are diagnosed with by developing a pattern of beliefs (i.e., illness perceptions) about the condition, which predict

medical, psychological and behavioural outcomes. Research has found evidence to show that the SRM has been helpful in understanding people's responses to a range of mental health conditions (Baines & Wittkowski, 2013). More specifically, relationships have been found between parental illness perceptions and how parents cope with having a child with ASD (Al Anbar, Dardennes, Prado-Netto, Kaye & Contejean, 2010). In addition, parents' beliefs about the chronicity of ASD and the consequences related to ASD were found to be associated with parents' levels of psychological distress and the severity of depressive symptoms (Gatzoyia et al., 2014).

Pathological demand avoidance (PDA) is a term used to describe a pattern of obsessive resistance to everyday demands and requests, using socially manipulative and 'outrageous' behaviour to avoid demands (Newson, Le Marechal & David, 2003). Those with PDA have been found to share some traits of ASD, but the evidence base in relation to pathological demand avoidance is currently limited. As such, and using the theory of the SRM, this study explored parents' beliefs of PDA, how parents cope with their child's PDA, parents' stress and overall wellbeing, and the child-parent relationship in PDA.

Method: 189 parents were recruited through the PDA Society website, Facebook group and Twitter; 71 parents of children with a diagnosis of PDA (diagnostic group) and 118 parents who suspected their child has PDA, but do not yet have a formal diagnosis (non-diagnostic group). Parents completed an online survey, which included questionnaires asking about parental perceptions about PDA, parental coping styles, parenting stress, parental wellbeing and the child-parent relationship.

Results: Parents viewed PDA as a chronic condition with considerable negative consequences. Parents in both groups reported high levels of parenting stress and low levels

of psychological wellbeing. The findings showed that there was more conflict in the child-parent relationship than closeness for children with PDA and suspected PDA. For parents in both groups, higher levels of parenting stress, poor overall wellbeing and conflict in the child-parent relationship were found to be associated with strongly held beliefs about the negative emotional consequences associated with PDA and parental perceptions of greater consequences related to PDA. In addition, parent reports of higher levels of parenting stress and child-parent conflict, and lower levels of closeness, were found to be associated with beliefs of less personal control of PDA. Beliefs that indicated a clearer understanding of PDA were associated with lower levels of conflict and higher levels of closeness in the child-parent relationship. Emotion-focussed coping (e.g., parents getting emotional support and looking for something good in their child's PDA) was associated with positive effects on psychological wellbeing. In line with some other research (e.g., Kaptein et al, 2006), but in contrast to the SRM, coping was not found to explain the relationship between parental illness perceptions and outcomes.

Conclusions: Findings indicated that the self-regulation model is a fitting model for PDA, which can be used to predict relationships between parental illness perceptions, coping and outcomes. The results have important implications for clinicians working with parents of a child with PDA/suspected PDA. Given the high stress levels and poor emotional wellbeing reported by parents, clinicians and researchers should consider interventions targeting parental illness perceptions, particularly perceptions of the consequences associated with PDA and perceptions of the degree of personal control parents have in relation to PDA.

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APPENDICIES FOR VOLUME 1

Appendix A: Search terms used in Kuhaneck, Madonna, Novak & Pearson's (2015) Review

1. "autism" or "asperger syndrome" or "pervasive developmental disorder
2. Parent mediated interventions
3. Parent implemented instruction
4. Parent self efficacy
5. Family coping
6. Self determination
7. Resilience
8. Positive psychology
9. Family quality of life
10. Stress reduction
11. Family life
12. Marriage
13. Spousal relationships
14. Marital relations
15. Self management
16. Decision making
17. 2 OR 5 OR 6 OR 7 OR 8 OR 9 OR 10 OR 11 OR 12 OR 13 OR 14 OR 15 OR 16
18. 1 and 17 were combined used AND.

Appendix B: Data Extraction Tool

Author(s) (Year)	
Country of origin	
Sample	
Study design	
Details about the intervention	
Outcome measures used	
Assessment points	
Key findings	
Limitations	

Appendix C: Standard Quality Assessment Criteria for Assessing the Quality of Quantitative Research. Kmet, Lee & Cook (2004)

- 1 Question / objective sufficiently described?
- 2 Study design evident and appropriate?
- 3 Method of subject/comparison group selection or source of information/input variables described and appropriate?
- 4 Subject (and comparison group, if applicable) characteristics sufficiently described?
- 5 If interventional and random allocation was possible, was it described?
- 6 If interventional and blinding of investigators was possible, was it reported?
- 7 If interventional and blinding of subjects was possible, was it reported?
- 8 Outcome and (if applicable) exposure measure(s) well defined and robust to measurement / misclassification bias? Means of assessment reported?
- 9 Sample size appropriate?
- 10 Analytic methods described/justified and appropriate?
- 11 Some estimate of variance is reported for the main results?
- 12 Controlled for confounding?
- 13 Results reported in sufficient detail?
- 14 Conclusions supported by the results?

Manual for Quality Scoring of Quantitative Studies

Definitions and Instructions for Quality Assessment Scoring

How to calculate the summary score

Total sum = (number of “yes” * 2) + (number of “partials” * 1)

Total possible sum = 28 – (number of “N/A” * 2)

Summary score: total sum / total possible sum

Appendix D: Ethical approval

Dear Dr Law & Dr Madurai

Re: “Parental representations of childhood Pathological Demand Avoidance syndrome: illness representations, coping and wellbeing”
Application for Ethical Review ERN_14-0474

Thank you for your application for ethical review for the above project, which was reviewed by the Science, Technology, Engineering and Mathematics Ethical Review Committee. The study was granted conditional ethical approval on 5th February 2015.

On behalf of the Committee, I can confirm the conditions of approval for the study have now been met and this study now has full ethical approval.

I would like to remind you that any substantive changes to the nature of the study as described in the Application for Ethical Review, and/or any adverse events occurring during the study should be promptly brought to the Committee’s attention by the Principal Investigator and may necessitate further ethical review.

Please also ensure that the relevant requirements within the University’s Code of Practice for Research and the information and guidance provided on the University’s ethics webpages (available at <https://intranet.birmingham.ac.uk/finance/accounting/Research-Support-Group/Research-Ethics/Links-and-Resources.aspx>) are adhered to and referred to in any future applications for ethical review. It is now a requirement on the revised application form (<https://intranet.birmingham.ac.uk/finance/accounting/Research-Support-Group/Research-Ethics/Ethical-Review-Forms.aspx>) to confirm that this guidance has been consulted and is understood, and that it has been taken into account when completing your application for ethical review.

Please be aware that whilst Health and Safety (H&S) issues may be considered during the ethical review process, you are still required to follow the University’s guidance on H&S and to ensure that H&S risk assessments have been carried out as appropriate. For further information about this, please contact your School H&S representative or the University’s H&S Unit at healthandsafety@contacts.bham.ac.uk.

Thank you,

Gemma Williams
Deputy Research Ethics Officer
Research Support Group
Finance Office
Aston Webb, B Block
Edgbaston, Birmingham
B15 2TT

Web: www.birmingham.ac.uk/researchsupportgroup

Appendix E: Ethical approval for amendments

Dear Dr Law and Dr Madurai

Re: “Parental Illness Representations in Pathological Demand Avoidance Syndrome: Parental Coping, Stress and Wellbeing”
Application for amendment ERN_14-0474A

Thank you for the above application for amendment, which was reviewed by the Science, Technology, Engineering and Mathematics Ethical Review Committee.

On behalf of the Committee, I can confirm that this amendment now has full ethical approval.

I would like to remind you that any substantive changes to the nature of the study as now amended, and/or any adverse events occurring during the study should be promptly brought to the Committee’s attention by the Principal Investigator and may necessitate further ethical review. A revised amendment application form is now available at <https://intranet.birmingham.ac.uk/finance/accounting/Research-Support-Group/Research-Ethics/Ethical-Review-Forms.aspx> . Please ensure this form is submitted for any further amendments.

Please also ensure that the relevant requirements within the University’s Code of Practice for Research and the information and guidance provided on the University’s ethics webpages (available at <https://intranet.birmingham.ac.uk/finance/accounting/Research-Support-Group/Research-Ethics/Links-and-Resources.aspx>) are adhered to and referred to in any future applications for ethical review. It is now a requirement on the revised application form (<https://intranet.birmingham.ac.uk/finance/accounting/Research-Support-Group/Research-Ethics/Ethical-Review-Forms.aspx>) to confirm that this guidance has been consulted and is understood, and that it has been taken into account when completing your application for ethical review.

Please be aware that whilst Health and Safety (H&S) issues may be considered during the ethical review process, you are still required to follow the University’s guidance on H&S and to ensure that H&S risk assessments have been carried out as appropriate. For further information about this, please contact your School H&S representative or the University’s H&S Unit at healthandsafety@contacts.bham.ac.uk.

If you require a hard copy of this correspondence, please let me know.

Kind regards

Susan Cottam
Research Ethics Officer
Research Support Group
C Block Dome
Aston Webb Building
University of Birmingham
Edgbaston B15 2TT

Appendix F: Research Advert

UNIVERSITY OF
BIRMINGHAM
UNIVERSITY OF
BIRMINGHAM

PATHOLOGICAL DEMAND AVOIDANCE RESEARCH STUDY FOR PARENTS

Living with a child with pathological demand avoidance (PDA) symptoms can have considerable consequences. As such, this research project is interested in understanding the relationship between childhood PDA symptoms and parent/carer stress.

This study was originally launched in June 2015, but due to demand we have expanded the criteria to include parents who suspect their child has PDA, but have not yet received a diagnosis. Both parents/main carers of children (under the age of 18) with suspected PDA and parents/main carers of children with a diagnosis of PDA are now invited to take part in this research. It will involve completing a number of online questionnaires about their child with PDA symptoms and their own wellbeing. Responses will be completely anonymous and the survey should take approximately 30-40 minutes. Participants will be entered into a prize draw for £10, £20 or £50 worth of amazon vouchers.

For more information and to take part in the study, please click on one of the following links:

Click on <https://lesweb2.bham.ac.uk/surveys/index.php?r=survey/index/sid/528939/lang/en> if you suspect your child has PDA, but he/she has not yet received a diagnosis.

Click on <https://lesweb2.bham.ac.uk/surveys/index.php?r=survey/index/sid/171121/lang/en> if your child has a diagnosis of PDA.

Appendix G: Information Sheet

INFORMATION SHEET

STRESS AND COPING ASSOCIATED WITH CARING FOR A CHILD WITH PATHOLOGICAL DEMAND AVOIDANCE SYMPTOMS

My name is Lauren Good and I am currently doing a doctorate in clinical psychology at the University of Birmingham. I am conducting research into childhood pathological demand avoidance (PDA) symptoms and parental stress and coping. I would like to invite parents of children/young people between the ages of 5 and 18, with symptoms of PDA to complete a set of questionnaires about PDA symptoms, stress and coping.

What is the purpose of this research?

There has been very limited research in relation to PDA and I am interested in finding out more about what it is like for parents of children with PDA symptoms. This will be an opportunity for parents to contribute to the understanding of PDA, which may help you or others in the future.

Who can participate?

Participants must be the parent or main carer of a child or young person (up to 18 years of age), who is suspected to have PDA.

What will happen if I agree to take part?

You will complete a set of questionnaires online, the majority of which are made up of multiple choice questions. There are lots of questions because this is the first survey of its type and we want to collect as much data as possible. The survey will take a total of approximately 30-40 minutes to complete. If you would like to complete the survey in more than one sitting or you would like to take a break, click on the 'resume later' button in the bottom left corner of the page and you will be asked to enter a name and password (which you can e-mail to yourself as a reminder by entering your e-mail address). When you are ready to come back to the survey, click on the link to the survey or enter the web address and click on 'load unfinished survey'. You will be asked for the name and password that you entered previously and you will be taken into your survey, where you will find your responses.

In recognition of the time it will take to complete the survey, participants will be entered in to a prize draw for £10, £20 or £50 worth of amazon vouchers. Participants will need to provide an e-mail address that they can be contacted on in order to be entered in to the prize draw. All information will remain confidential and data will be anonymised. Information will only be viewed by me and my supervisors, Dr Gary Law and Dr Teresa Madurai, clinical psychologists at the University of Birmingham.

What will happen if I decide that I do not want to carry on with the study?

You can withdraw from the study any time up until 31st January 2016. You will be given a reference number that is unique to you once you have completed the survey and you will need to keep the reference number. If you wish to withdraw your information at a later stage, you can contact me using the details below, quoting your unique reference number and your data will be withdrawn. It will not be possible to withdraw your information without your reference number.

What will happen to the results of the study?

The results will be used for a university doctoral thesis and will also be submitted for publication in a journal. An overall summary of the research findings will be available on the websites that the research is advertised on (PDA society website, PDA society Facebook group & Twitter page). Participants will also be asked to note down the email and postal addresses of the academic supervisors at the end of the survey, so that they can seek summary results of the study at some future time.

What happens if I have any further concerns?

If you have any further concerns about your emotional wellbeing or how you are coping with the challenges that living with a child with PDA symptoms can bring, please contact your G.P.

Alternatively, if you require further information or support you may find the following links helpful:

www.pdasociety.org.uk contains a wide range of information and resources for parents/carers of children with PDA. It contains links to local support groups and details of 'family fun days' for families of children with PDA. There are also private forums, where parents and carers can share their experiences and support each other.

www.autism.org.uk/about-autism/related-conditions/pda-pathological-demand-avoidance-syndrome.aspx contains information about PDA and provides links to other resources.

www.facebook.com/groups/PDAsupport is an online parent/carer support group.

www.parentlineplus.org.uk is the leading national charity that provides help and support to anyone caring for children. They provide web-chat, e-mail support and a free, 24 hour helpline **(0808 800 2222)**.

www.samaritans.org.uk provides confidential support, 24 hours a day **(08457 90 90 90)**.

If you would like to discuss any aspect of this research please contact:

Contact details:	Lauren Good
Supervisors:	Dr Gary Law and Dr Teresa Madurai
Address:	University of Birmingham, School of Psychology, Edgbaston, Birmingham.
Post Code:	B15 2TT

Appendix H

CONSENT FORM

Title of Project: Pathological Demand Avoidance Syndrome: Parental Stress & Wellbeing

Researcher: Lauren Good

Please tick box

1. I confirm that I have understood the information sheet dated July 2014 for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily. ☐
2. I understand that my participation is voluntary and that I am free to withdraw at any time during the research interview, without giving any reason, without my own or my child's medical/social care or legal rights being affected. ☐
3. I understand that the data collected during this study will be looked at by the researcher and relevant others at the University of Birmingham to ensure that the analysis is a fair and reasonable representation of the data. ☐
4. I agree to take part in the above study. ☐

Appendix I: Questionnaires
Demographic Data Questionnaire

Does your child have a diagnosis of PDA?

What professional diagnosed your child with PDA?

Year and month of diagnosis of PDA:

What treatment is your child receiving for his/her PDA?

Diagnoses:

Relationship to child:

D.O.B of child:

Gender of child:

Medication:

Number of siblings the child has:

Country of origin:

Parental relationship status:

Who is in the household?

How many hours of respite are provided for your child each month?

Gender of the person completing the survey:

Education status of the person completing the survey:

D.O.B. of the person completing the survey:

Extreme demand avoidance questionnaire (EDA-Q)

Please choose the most appropriate answer in relation to your child with PDA.

		Not true	Somewhat true	Mostly true	Very true
1	Obsessively resists and avoids ordinary demands and requests.				
2	Complains about illness or physical incapacity when avoiding a request or demand.				
3	Is driven by the need to be in charge.				
4	Finds everyday pressures (e.g. having to go on a school trip/ visit dentist) intolerably stressful.				
5	Tells other children how they should behave, but does not feel these rules apply to him/herself.				
6	Mimics adult mannerisms and styles (e.g. uses phrases adopted from teacher/parent to tell other children off).				
7	Has difficulty complying with demands unless they are carefully presented.				
8	Takes on roles or characters (from TV/real life) and 'acts them out'.				
9	Shows little shame or embarrassment (e.g. might throw a tantrum in public and not be embarrassed).				
10	Invents fantasy worlds or games and acts them out.				
11	Good at getting round others and making them do as s/he wants.				
12	Seems unaware of the differences between him/herself and authority figures (e.g. parents, teachers, police).				
13	If pressurised to do something, s/he may have a 'meltdown' (e.g. scream, tantrum, hit or kick).				
14	Likes to be told s/he has done a good job.				
15	Mood changes very rapidly (e.g. switches from affectionate to angry in an instant).				
16	Knows what to do or say to upset specific people.				
17	Blames or targets a particular person.				
18	Denies behaviour s/he has committed, even when caught red handed.				
19	Seems as if s/he is distracted 'from within'.				
20	Makes an effort to maintain his/her reputation with peers.				

21	Uses outrageous or shocking behaviour to get out of doing something.				
22	Has bouts of extreme emotional responses to small events (e.g. crying/giggling, becoming furious).				
23	Social interaction has to be on his or her own terms.				
24	Prefers to interact with others in an adopted role, or communicate through props/toys.				
25	Attempts to negotiate better terms with adults.				
26	S/he was passive and difficult to engage as an infant.				

How to score the EDA-Q

Questions 1 - 26 (apart from questions 14 and 20)

Not true = 0

Somewhat true = 1

Mostly true = 2

Very true = 3

Questions 14 & 20

Not true = 3

Somewhat true = 2

Mostly true = 1

Very true = 0

Results

For children aged 5 to 11 a score of **50** and over...

For children aged 12 to 17 a score of **45** and over...

...identifies individuals with an elevated risk of having a profile consistent with PDA.

The EDA-Q should not be considered a diagnostic test. For diagnosis, a thorough assessment by an experienced professional is required.

Illness Perceptions Questionnaire-Revised (adapted)

Listed below are a number of symptoms that your child may or may not have experienced as part of their PDA. Please indicate whether your child has experienced any of these symptoms and whether you believe that these symptoms are related to their illness.

	My child has experienced this symptom	My child has not experienced this symptom	This symptom is related to my child's PDA	This symptom is not related to my child's PDA
Pain	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Sudden and excessive mood swings	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Lack of social identity (e.g. not fitting in)	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Social manipulation	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Fatigue	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Impulsivity	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Anxiety	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Headaches	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Aggression	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Doing many things in an obsessive way	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Avoidance of every day demands	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Loss of strength	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>

We are interested in your own personal views of how you see your child's PDA.

Please indicate how much you agree or disagree with the following statements about your child's PDA by checking the appropriate answer.

	Strongly disagree	Disagree	Neither agree nor disagree	Agree	Strongly agree
His/her PDA will last a short time	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
His/her PDA is likely to be permanent rather than temporary	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
His/her PDA will last for a long time	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
His/her PDA will pass quickly	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I expect that he/she will have PDA for the rest of his/her life	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
His/her PDA is a serious condition	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
His/her PDA has major consequences on his/her life	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
His/her PDA does not have much of an effect on his/her life	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
His/her PDA strongly affects the way others see him/her	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
His/her PDA has serious financial consequences	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
His/her PDA causes difficulties for those who are close to him/her	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
There is a lot that I can do to control his/her symptoms	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
What I do can determine whether his/her PDA symptoms get better or worse	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
The course of his/her PDA depends on me	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Nothing I do will affect his/her PDA symptoms	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I have the power to influence his/her PDA symptoms	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
My actions will have no affect on the outcome of his/her PDA symptoms	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
His/her PDA symptoms will improve in time	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
There is very little that can be done to improve his/her PDA symptoms	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
His/her treatment will be effective in curing his/her PDA symptoms	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
The negative effects of his/her PDA can be prevented (avoided) by his/her	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>

treatment					
His/her treatment can control his/her PDA symptoms	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
There is nothing which can help his/her PDA symptoms	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
The symptoms of his/her PDA are puzzling to me	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
His/her PDA is a mystery to me	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I don't understand his/her PDA	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
His/her PDA doesn't make any sense to me	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I have a clear picture or understanding of his/her PDA	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
The symptoms of his/her PDA change a great deal from day to day	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
His/her symptoms come and go in cycles	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
His/her PDA is very unpredictable	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
He/she goes through cycles in which his/her PDA gets better and worse	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I get depressed when I think about his/her PDA	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
When I think about his/her PDA I get upset	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
His/her PDA makes me feel angry	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
His/her PDA does not worry me	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
His/her PDA makes me feel anxious	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
His/her PDA makes me feel afraid	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>

*We are interested in what you consider may have been the cause of your child's PDA. As people are very different, there is no correct answer for these questions. We are most interested in your own views about the factors that caused your child's PDA rather than what

others including doctors or family may have suggested to you. Below is a list of possible causes for your child's PDA. Please indicate how much you agree or disagree that they were causes for you by checking the appropriate box.

	Strongly disagree	Disagree	Neither agree nor disagree	Agree	Strongly agree
Stress or worry	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Hereditary - it runs in the family	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
A germ or virus	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Diet or eating habits	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Chance or bad luck	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Poor medical care in my child's past	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Pollution in the environment	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
My child's own behaviour	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
My child's mental attitude, e.g. thinking about life negatively	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Family problems or worries caused my child's PDA	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Overwork	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
My child's emotional state, e.g. feeling down, lonely, anxious or empty	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Growing up	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Family member drinking alcohol	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Family member smoking	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Accident or injury	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
My child's personality	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Changes in my child's body	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
My behaviour	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Lack of parental control	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
My personality	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
My emotional state, e.g. feeling down, lonely, anxious or empty	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Me thinking about life negatively	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>

In the spaces below, please list in rank order, the three most important factors that you now believe caused your child's PDA. You may use any of the items above, or you may have ideas of our own.

- The most important cause for me
- The second most important cause for me
- The third most important cause for me

We are interested in your views on each particular treatment/intervention/strategy that is used to manage your child's PDA symptoms. Please answer the following questions for the medications/treatments/interventions/strategies you listed at the beginning of the survey.

Medication/Treatment/Intervention/Strategy 1:

Answer

Medication/Treatment/Intervention/Strategy 1:

	Strongly disagree	Disagree	Neither agree nor disagree	Agree	Strongly agree
This treatment will be effective in curing the symptoms of my child's PDA	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
This treatment can prevent the negative effects of my child's PDA	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
This treatment can control the symptoms of my child's PDA	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>

Medication/Treatment/Intervention/Strategy 2:

Answer

Medication/Treatment/Intervention/Strategy 2:

	Strongly disagree	Disagree	Neither agree nor disagree	Agree	Strongly agree
This treatment will be effective in curing the symptoms of my	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>

child's PDA

This treatment can prevent the negative effects of my child's PDA

☐ ☐ ☐ ☐ ☐

This treatment can control the symptoms of my child's PDA

☐ ☐ ☐ ☐ ☐

Medication/Treatment/Intervention/Strategy 3:

Answer

Medication/Treatment/Intervention/Strategy 3:

	Strongly disagree	Disagree	Neither agree nor disagree	Agree	Strongly agree
This treatment will be effective in curing the symptoms of my child's PDA	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
This treatment can prevent the negative effects of my child's PDA	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
This treatment can control the symptoms of my child's PDA	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>

Brief COPE (adapted)

These items deal with ways you've been coping with the stress associated with your child's PDA. There are many ways to try to deal with problems. These items ask what you've been doing to cope. Obviously, different people deal with things in different ways, but I'm interested in how you've tried to deal with your child's PDA. Each item says something about a particular way of coping. I want to know to what extent you've been doing what the item says. How much or how frequently. Don't answer on the basis of whether it seems to be working or not—just whether or not you're doing it. Use these response choices. Try to

rate each item separately in your mind from the others. Make your answers as true FOR YOU as you can.

	I haven't been doing this at all	I've been doing this a little	I've been doing this a medium amount	I've been doing this a lot
1. I've been turning to work or other activities to take my mind off things.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
2. I've been concentrating my efforts on doing something about the situation I'm in.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
3. I've been saying to myself "this isn't real".	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
4. I've been using alcohol or other drugs to make myself feel better.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
5. I've been getting emotional support from others.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
6. I've been giving up trying to deal with it.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
7. I've been taking action to try to make the situation better.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
8. I've been refusing to believe that this has happened.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
9. I've been saying things to let my unpleasant feelings escape.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
10. I've been getting help and advice from other people.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
11. I've been using alcohol or other drugs to help me get through it.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
12. I've been trying to see it in a different light, to make it seem more positive.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
13. I've been criticizing myself.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
14. I've been trying to come up with a strategy about what to do.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
15. I've been getting comfort and understanding from someone.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
16. I've been giving up the attempt to cope.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
17. I've been looking for something good in what is happening.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
18. I've been making jokes about it.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
19. I've been doing something to think about it less, such as going to movies, watching TV, reading, daydreaming, sleeping, or shopping.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
20. I've been accepting the reality.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
21. I've been expressing my negative feelings.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
22. I've been trying to find comfort in my religion or spiritual beliefs.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
23. I've been trying to get advice or help from other people about what to do.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
24. I've been learning to live with my child's PDA.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
25. I've been thinking hard about what steps to take.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>

26. I've been blaming myself. ☐ ☐ ☐ ☐
27. I've been praying or meditating. ☐ ☐ ☐ ☐
28. I've been making fun of the situation. ☐

Parental Stress Scale

The following statements describe feelings and perceptions about the experience of being a parent. Think of each of the items in terms of how your relationship with your child with PDA typically is. Please indicate the degree to which you agree or disagree with the following items by using the rating scale below.

	Strongly disagree	Disagree	Undecided	Agree	Strongly agree
I am happy in my role as a parent	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
There is little or nothing I wouldn't do for my child if it was necessary.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Caring for my child sometimes takes more time and energy than I have to give.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I sometimes worry whether I am doing enough for my child.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I feel close to my child.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I enjoy spending time with my child.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
My child is an important source of affection for me.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Having my child gives me a more certain and optimistic view for the future.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
The major source of stress in my life is my child.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Having my child leaves little time and flexibility in my life.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Having my child has been a financial burden.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
It is difficult to balance different responsibilities because of my child.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
The behaviour of my child is often embarrassing or stressful to me.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
If I had it to do over again, I might decide not to have my child.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I feel overwhelmed by the responsibility of being a parent.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Having my child has meant having too few choices and too little control over my life.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I am satisfied as a parent	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I find my child enjoyable	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>

General Wellbeing Index

This section of the survey contains questions about how you feel and how things have been going with you. For each question, check the answer which best applies to you

*How have you been feeling in general during the past month?

Choose one of the following answers

- ☐ In very good spirits
- ☐ In good spirits mostly
- ☐ I have been up and down a lot
- ☐ In low spirits mostly
- ☐ In very low spirits

*During the past month have you been bothered by any illness, pains or fears about your health?

Choose one of the following answers

- ☐ All the time
- ☐ A lot of the time
- ☐ Some of the time
- ☐ A little bit
- ☐ Not at all

*Did you feel depressed during the past month?

Choose one of the following answers

- ☐ Yes, very much so
- ☐ Yes, quite a bit
- ☐ Sometimes, enough to bother me
- ☐ A little depressed now and then
- ☐ No, not at all

*During the past month have you felt in firm control of your actions, thoughts, or feelings?

Choose one of the following answers

- ☐ Yes, definitely
- ☐ Yes, mostly
- ☐ Not too well
- ☐ No, hardly at all
- ☐ No, not at all

*Have you been bothered by your nerves during the past month?

Choose one of the following answers

- ☐ Very much so
- ☐ Quite a bit
- ☐ Sometimes
- ☐ A little
- ☐ Not at all

*During the past month how much energy or vitality did you have?

Choose one of the following answers

- ☐ Lots of energy
- ☐ Fairly energetic most of the time
- ☐ Energy varied quite a bit
- ☐ Low in energy mostly
- ☐ No energy at all

*Have you felt downhearted and sad over the past month?

Choose one of the following answers

- ☐ All of the time
- ☐ Most of the time

- ☐ From time to time
- ☐ Very occasionally
- ☐ Not at all

*During the past month how tense have you been?

Choose one of the following answers

- ☐ Extremely tense all of the time
- ☐ Very tense most of the time
- ☐ From time to time
- ☐ Rarely tense
- ☐ Not tense at all

*How happy or satisfied have you been with your personal life during the past month?

Choose one of the following answers

- ☐ Very satisfied
- ☐ Fairly satisfied
- ☐ Satisfied on the whole
- ☐ Rather dissatisfied
- ☐ Very dissatisfied

*Over the past month did you feel well enough to do the things you like to do or had to do?

Choose one of the following answers

- ☐ Yes, definitely
- ☐ Yes, or the most part
- ☐ About half the time
- ☐ No, not often
- ☐ No, not at all

*Have you felt so sad, disheartened or had so many problems that you wondered if anything was worthwhile over the past month?

Choose one of the following answers

- ☐ All the time
- ☐ Most of the time
- ☐ From time to time

- ☐ Very occasionally
- ☐ Not at all

*During the past month have you been waking up feeling fresh and rested?
Choose one of the following answers

- ☐ Every day
- ☐ Most days
- ☐ Less than half the time
- ☐ Not often
- ☐ Not at all

*Have you had any worries or fears about your health during the past month?
Choose one of the following answers

- ☐ Yes, all the time
- ☐ Most of the time
- ☐ From time to time
- ☐ Not a lot
- ☐ Not at all

*During the past month have you wondered if you were losing control over your actions, thoughts, feelings or memory?
Choose one of the following answers

- ☐ All the time
- ☐ Most of the time
- ☐ From time to time
- ☐ No, hardly at all

- ☐ Not at all

*Has your daily life been filled with things that interest you during the past month?
Choose one of the following answers

- ☐ All of the time
- ☐ Most of the time
- ☐ Some of the time
- ☐ A little
- ☐ Not at all

*During the past month how active and vigorous have you felt?
Choose one of the following answers

- ☐ Very active every day
- ☐ Mostly active
- ☐ Fairly active
- ☐ Seldom active
- ☐ Not at all active

*Have you been anxious, worried, or upset over the past month?
Choose one of the following answers

- ☐ Very much so
- ☐ Quite a lot
- ☐ Sometimes, enough to bother me
- ☐ A little bit
- ☐ Not at all

*During the past month have you felt emotionally stable and sure of yourself?

Choose one of the following answers

- ☐ All of the time
- ☐ Most of the time
- ☐ Some of the time
- ☐ Now and then
- ☐ Not at all

*How relaxed have you felt over the past month?

Choose one of the following answers

- ☐ Very relaxed all the time
- ☐ Mostly relaxed
- ☐ Relaxed about half the time
- ☐ Rarely felt relaxed
- ☐ Not at all relaxed

*During the past month how cheerful have you been?

Choose one of the following answers

- ☐ Not cheerful at all
- ☐ A little cheerful now and then
- ☐ Cheerful about half of the time
- ☐ Mostly quite cheerful
- ☐ Very cheerful all the time
-

*Have you felt tired, worn out or exhausted during the past month?

Choose one of the following answers

- ☐ All of the time
- ☐ Most of the time
- ☐ Some of the time
- ☐ Now and then
- ☐ Not at all

*Over the past month have you been under any stress or pressure?

Choose one of the following answers

- ☐ Yes, almost more than I could bear
- ☐ Yes, more than usual
- ☐ About the same as usual
- ☐ Yes, a little
- ☐ No, not at all

Please provide any additional information about your emotional wellbeing that you think is relevant or important for us to know.

CHILD-PARENT RELATIONSHIP SCALE

Please reflect on the degree to which each of the following statements currently applies to your relationship with your child with PDA. Please select the appropriate rating for each item.

Definitely does not apply	Not really	Neutral, not sure	Applies somewhat	Definitely applies
1	2	3	4	5

1. I share an affectionate, warm relationship with my child.	1	2	3	4	5
2. My child and I always seem to be struggling with each other.	1	2	3	4	5
3. If upset, my child will seek comfort from me.	1	2	3	4	5
4. My child is uncomfortable with physical affection or touch from me.	1	2	3	4	5
5. My child values his/her relationship with me.	1	2	3	4	5
6. My child appears hurt or embarrassed when I correct him/her.	1	2	3	4	5
7. My child does not want to accept help when he/she needs it.	1	2	3	4	5
8. When I praise my child, he/she beams with pride.	1	2	3	4	5
9. My child reacts strongly to separation from me.	1	2	3	4	5
10. My child spontaneously shares information about himself/herself.	1	2	3	4	5
11. My child is overly dependent on me.	1	2	3	4	5
12. My child easily becomes angry at me.	1	2	3	4	5
13. My child tries to please me.	1	2	3	4	5
14. My child feels that I treat him/her unfairly.	1	2	3	4	5
15. My child asks for my help when he/she really does not need help.	1	2	3	4	5

16. It is easy to be in tune with what my child is feeling.	1	2	3	4	5
17. My child sees me as a source of punishment and criticism.	1	2	3	4	5
18. My child expresses hurt or jealousy when I spend time with other children.	1	2	3	4	5
19. My child remains angry or is resistant after being disciplined.	1	2	3	4	5
20. When my child is misbehaving, he/she responds to my look or tone of voice.	1	2	3	4	5
21. Dealing with my child drains my energy.	1	2	3	4	5
22. I've noticed my child copying my behavior or ways of doing things.	1	2	3	4	5
23. When my child is in a bad mood, I know we're in for a long and difficult day.	1	2	3	4	5
24. My child's feelings toward me can be unpredictable or can change suddenly.	1	2	3	4	5
25. Despite my best efforts, I'm uncomfortable with how my child and I get along.	1	2	3	4	5
26. I often think about my child when at work.	1	2	3	4	5
27. My child whines or cries when he/she wants something from me.	1	2	3	4	5
28. My child is sneaky or manipulative with me.	1	2	3	4	5
29. My child openly shares his/her feelings and experiences with me.	1	2	3	4	5
30. My interactions with my child make me feel effective and confident as a parent.	1	2	3	4	5

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