A multiple single-case design evaluation of a parent-mediated CBT intervention for children with ASD and anger management difficulties: feasibility, acceptability and an initial estimate of efficacy

Sohini Shah

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Thesis declaration form

I confirm that the work presented in this thesis is my own. Where information has been derived from other sources, I confirm that this has been indicated in the thesis.

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Overview

Part 1 of this thesis reviews the research literature on psychological interventions for young people with ASD and aggressive behaviour problems (ABPs). Eleven studies in this area were identified which used a controlled research design and included an outcome measure of aggression or irritability. They included parent training programmes, early intensive interaction, cognitive behavioural, behavioural and therapeutic horse-riding interventions. A meta-analysis of all these studies combined (N=602) revealed a moderate treatment effect size suggesting that psychological interventions can reduce the amount of ABPs show by children with ASD. An evaluation of the methodological quality of these studies indicated that further research is needed to strengthen this conclusion.

Part 2 reports on the results of an initial evaluation to understand the acceptability and feasibility of a parent-mediated CBT intervention for young people with ASD. It used a mixed-methods and small-N design consisting of a series of multiple systematic case studies (N=7). Baseline and follow-up data were collected. The results provided in depth data for each participant and preliminary evidence that this intervention was acceptable to most families and led to positive changes in anger outbursts for some young people. However, the outcome was variable across participants and so barriers to progress and ways to improve efficacy for a greater proportion of individuals are discussed.

Part 3 is a critical appraisal of the research. It includes further discussion of the results and a reflection on the use of a mixed-methods small-N design. It also expands on the limitations of the study and describes some personal reflections on the process of conducting this study.

Table of Contents

Part 3: Critical Appraisal

List of Figures

Part 1: Literature Review

Part 2: Empirical Paper

List of Tables

Appendices

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Part 1: Literature Review

Reducing aggressive behaviour problems shown by children with Autism using psychological interventions: A review of the research and meta-analysis

Abstract

Aggressive behaviour is a common co-occuring problem for children with an Autism Spectrum Disorder (ASD). Such problems left untreated can negatively affect important aspects of development and have a lasting impact as the child grows older. There is a shortage of controlled research studies on non-pharmacological interventions for these children. This review was based on a systematic search and identified 11 controlled studies of psychological interventions that have been published in this area. These included parent training programmes, early intensive interaction, cognitive behavioural and behavioural interventions and therapeutic horse-riding. A meta-analysis was conducted on these studies which collectively included a total of 602 participants with ASD. The overall treatment effect was moderate (Cohen's d = 0.47, 95% confidence intervals) and statistically significant. However, the effect sizes varied significantly across the included studies. Reasons for this are discussed in the review. The characteristics of the studies and an appraisal of the methodological quality of the identified studies is reported. A number of recommendations are made for the designs and reporting of future research in this area.

1. Introduction

Autism Spectrum Disorder (ASD) is a lifelong neurodevelopmental condition characterized by impairments in social interaction and communication together with the presence of restricted and repetitive behaviours. Recent estimates of prevalence indicate that approximately one per cent of the UK population meet diagnostic criteria for ASD (Baird et al., 2006).

1.1. Prevalence of aggressive behaviour problems in children with ASD

Young people with ASD have been reported to show aggressive behaviour problems (ABPs) such as hitting, kicking, punching, pushing, hair-pulling, spitting, throwing objects, self-injury, tantrums, and property destruction (Matson & Jang, 2014; Nebel-Schwalm & Worley, 2014). The prevalence of such difficulties are high in children with ASD. It has been estimated that 56% of children (aged 4-17 years) with ASD show aggression towards their caregivers (Kanne & Mazurek, 2011). This sample included 1380 children with a wide range of IQ scores from 13 to 167 (mean = 84.7, SD = 25.6) suggesting that such difficulties are not limited to individuals with learning difficulties.

1.2. Impact of ABPs shown by children with ASD

Studies of individuals with developmental disabilities show that severe aggression can lead to increased inpatient admission, repeated crisis referrals (Shoham-Vardi et al., 1996), use of psychotropic medication (Tsakanikos et al., 2007) and reduced overall quality of life (Gardner and Moffatt, 1990). Behaviour problems (of which ABPS is a common type) in children with ASD has repeatedly shown links with higher levels of

caregiver stress which in turn serve to maintain and escalate behaviour problems shown by the children (Zaidman-Zait et al., 2014).

Crucially, the negative effects of ABPs can affect important social, educational and cognitive stages of development and continue to have long-lasting consequences as the child grows older. For these reasons, there is a pressing need to develop an evidence base of effective interventions for children with ASD showing ABPs.

1.3. Theoretical models informing treatment approaches to ABP in ASD

This section outlines three theoretical models that inform treatment to ABP in ASD.

1.3.1. Behavioural Theories

Applied Behaviour analysis is based on operant conditioning theories (Skinner, 1938), and has been the predominant treatment approach in ASD to reduce behaviour problems such as ABPs (Bregman, Zager & Gerdtz, 2005). Behavioural interventions begin with a functional analysis which is used to establish the antecedents and consequences of the ABP. Following this, operant principles are used to systematically reinforce desirable behaviours and reduce undesirable behaviours. Behavioural interventions can focus on modifying antecedents and/or consequences. There has been a trend favouring the modification of antecedent conditions such as altering the individual's environment, making adjustments to task difficulty, increasing choice and introducing regular physical exercise (Duker & Rasing, 1989; Gabler-Halle, Halle & Chung, 1993; Munk & Repp, 1994). These focus on the prevention of ABPs as opposed to responding reactively by modifying consequences. Furthermore, there has been an increasing recognition of the social and

communication impairments of ASD that would make individuals

more likely to resort to ABPs to express anger, anxiety and

frustration. In recent years, positive and broader behaviour

approaches based on skill acquisition, functional communication

training and increasing adaptive behaviours are being used in

conjunction or in place of antecedent and consequence interventions

as a way of reducing or eliminating ABPs shown by individuals with

ASD (Bregman et al., 2005).

1.3.2. Cognitive Theories

Cognitive theories emphasise the role of perceptions and thoughts

when understanding aggression. Social information processing

theories have proposed that aggressive behaviour results from an

individual making a hostile attribution bias whereby they ascribe

negative intent to another, and react aggressively in retaliation (Baron

and Richardson, 1994). This is particularly relevant for children with

ASD who are known to have impairments in understanding another

person's mental state (Baron Cohen, 1989; Yirmiya, Erel, Shaked, &

Solomonica-Levi (1998). Consequently, they are prone to misread

social situations and misunderstand a non-threatening situation as

threatening. Furthermore, social skills deficits may render children

with ASD particularly vulnerable to using aggression to solve social

problems when they find it difficult to generate alternative social

solutions. Lastly, theory of mind deficits affect one's ability to

empathise with others. Thus, individuals with ASD may find it difficult

consider the impact of their aggression on a victim which is likely to

decrease their motivation to inhibit aggressive tendencies.

1.3.3. ASD characteristics leading to a stress-vulnerability model of ABPs

Increased levels of stress could increase the vulnerability for individuals with ASD to display aggressive behaviour. A number of features of ASD render individuals susceptible to experiencing higher levels of stress. For example, people with ASD are more likely to accumulate stress from everyday changes and transitions and sensory sensitivities (Ben-Sasson et al., 2008). They must understand and navigate a social world that they are unaccustomed to and face a higher probability of victimization and bullying (Chen & Schwartz, 2012). To support this theory, research has shown that children with ASD do show cortisol levels similar to those with chronic stress (Corbett, Mendoza, Wegelin, Carmean & Levine, 2007). Elevated levels of underlying stress could increases the probability that a seemingly trivial event can push them beyond a critical point and result in aggressive behaviour which serves to release the built up emotional energy. This theory would suggest that decreasing the overall levels of stress experienced by individuals with ASD could reduce the frequency of ABPs.

1.4. Reviews on interventions for ABPs in children with ASD

Randomized control trials of interventions for this population are largely of pharmacological treatments. Medications trialled in this population include aripiprazole, zipraprazole, risperidone, imipramine, valproate, clomipramine, atomoxetine, fluvoxamine, dextromethorphan and busipirone. Antipsychotic drugs have been used to treat children as young as three years old (Masi, Cosenza, Mucci & Brovedani (2001). It

is unclear whether these medications are acting through a specific mechanism to reduce ABPs or are simply sedating individuals. Nonetheless, there is a need to establish alternative evidence-based treatments for this population. Psychological treatments are less invasive and cause fewer side effects however, a very limited number of controlled trials are conducted on such interventions. The studies on psychological treatments in this area are largely based on behavioural methods and mostly employed single-case designs which have limited generalizability.

The published research on treatments for aggression in people with ASD was recently reviewed by Matson & Jang (2014). This review found 11 pharmacologically-based interventions and 14 psychologically-based interventions. However, none of the psychological studies identified had a control group and almost all studies were single case designs investigating behavioural methods. It is however important to note that the review was undertaken using relatively narrow search criteria (six keywords in total) which may have led to the exclusion of published papers that used alternative key words. This review aims to use a wider search criteria in order to capture a greater number of studies relevant to the treatment of ABPs shown by children with ASD. This review will also refine papers to those using a control group and will use a meta-analytic procedure to estimate overall effect sizes.

1.5. Aims of this review

To the author's knowledge, there have been no published meta-analyses on controlled studies of psychological interventions for ABPs

for children with ASD. This review is based on a systematic search to investigate the following questions:

1. What are the characteristics of the studies that used a controlled design and investigated psychological treatments to reduce ABPs in children with ASD?

2. What is the quality of published studies in this area?

3. Can psychological interventions reduce ABPs displayed by children with ASD?

4. What recommendations can be made for future research in this area?

2. Methods

2.1. Criteria for considering studies for this review

Studies were included if they met the following inclusion criteria:

- Investigated a psychological intervention which was defined as any non-pharmacological intervention that aimed to impact on psychological variables or behaviour change for therapeutic gain.

- Included an outcome measure of aggression or irritability. It did not have to be the primary outcome measure. Measures included standardized outcome measures as well as frequency outcome data.

- The sample consisted of young people aged 18 years or younger and had a diagnosis of an ASD. Studies that employed a mixed group of participants with ASD and developmental delay or ADHD (without ASD) were not included. Studies that included children with ASD and comorbid ADHD were included. Studies that included participants with 'suspected ASD' were not included.

- Study employed a controlled design (i.e. had a treatment and comparison group)

- Published between 01/01/1999 and 28/11/2015.

Studies were excluded if they met any of the following exclusion criteria:

- The psychological intervention was combined with psychopharmacological treatment

- The participants had a co-occurring physical disability

- Studies that did not report the required information to calculate an effect size and the authors did not respond to attempts to contact them about this information.

2.2. Search methods for identification of studies

2.2.1. Electronic search

A systematic search was performed across three electronic databases (PubMed, PsychINFO, Web of Sciences) in November 2015. The following search terms were used as either key terms or key words: (ASD OR autis* OR asperger* OR "pervasive developmental disorder" OR neurodevelopmental) AND (anger OR irritability OR aggressi* OR "behaviour problems" OR "behavior problems" OR external ?ing OR exclu* OR disruptive OR "emotion regulation" OR "emotional regulation") AND (intervention OR therapy OR treatment OR RCT OR "randomised control trial" or "randomized control trial" OR trial OR treat* OR CBT or cognitive OR behavioural OR behavioral OR workshop OR training) AND (child* OR adolescen* OR youth OR young OR pupil). The search was refined for studies that had been published since 01/01/1999.

2.2.2. Other searches

The reference list of included studies were also hand-searched for relevant papers.

Figure 1: Flow chart of included and excluded studies
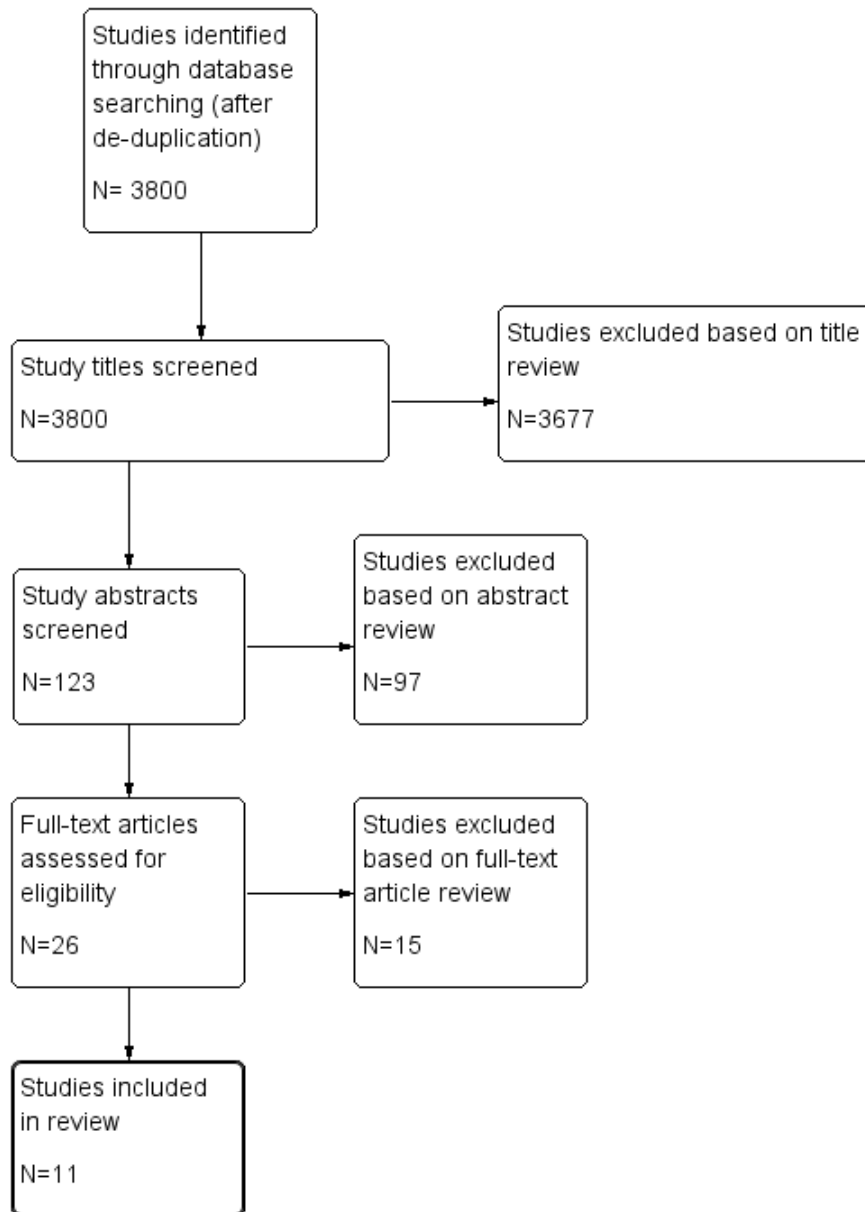
2.3. Data collection and analysis

2.3.1. Study selection

A total of 3800 studies were identified from the systematic searches. Of these, 3677 were excluded from checking the title on the basis that they did not meet inclusion criteria 1 (they were not investigating psychological interventions).

The abstracts of 123 studies were checked and 97 studies were excluded. A further 15 studies were excluded after checking the remaining papers in full. From the 123 papers, unsuitable studies were excluded for the following reasons: 9 were not investigating a psychological treatment; five investigated psychological treatment but combined with medication; 76 had no comparator group; 11 included participants where not all had an ASD according to DSM-IV or DSM-5 criteria; 11 did not have an outcome measure of aggression/irritability and one paper was excluded as it provided insufficient data which could not be resolved by contacting the authors. This resulted in 11 papers that were included in the review. A flow diagram of study selection is shown in Figure 1.

Ideally, this process would have been carried out by two independent researchers to assess the reliability with which inclusion/exclusion criteria were applied. However, there were not resources available to support this.

2.3.2. Assessment of risk of bias in included studies

All studies were evaluated for methodological quality using the Cochrane Collaboration's tool for assessing risk of bias (Higgins & Green, 2011). Treatment fidelity was added as an additional source of performance bias. Risk of bias was evaluated as per the following domains:

a) Selection bias (random sequence generation, allocation concealment)

b) Performance bias (blinding of participants and personnel, level of treatment fidelity)

c) Attrition Bias (incomplete outcome data)

d) Detection bias (blinding of outcome assessment)

e) Reporting bias (selective reporting)

f) Other bias (other potential threats to internal validity)

A judgement on the level of risk was categorised as 'low', 'high' or 'unclear' according to the Cochrane criteria for making such judgements (Higgins & Green, 2011). The 'unclear' category indicated either insufficient information available or uncertainty over the potential for bias.

Blinding of participants in psychological research is rare and this area was judged to be 'probably not done' and 'high risk' if such methods were not specifically detailed in the report.

2.3.3. *Measures of treatment effect & dealing with missing data*

All outcome measures of the included studies reported continuous data. Where available, the means, standard deviation and sample sizes were extracted. Authors of papers were contacted by email to seek required information if it was not available from the published paper.

In Bearrs et al. *(*2015), the standard deviations (SD) were not reported. The SD of the control and treatment group post-intervention scores were estimated from data of 95% confidence intervals (CI) of the group means. The following formula denoted in the Cochrane Handbook (Higgins & Green, 2011) was used:

Standard Deviation = $\sqrt{N}$ x (upper limit CI – lower limit CI) / 3.92

In Sofronoff et al., (2004) information on group sample size was not reported and could not be resolved from contacting the authors. A sample size of each group (N = 17) was estimated using the total sample (N = 51) divided by the number of groups (3) using an assumption that an equal number of participants were allocated to each group. Furthermore, in this paper there were two variants of an intervention (group and individual). The effect sizes of these subgroups were combined to get a value for one 'treatment group'.

2.3.4. *Data analysis*

The data analysis was conducted on Review Manager software, version 5.3 (The Cochrane Collaboration, 2014). The standardized mean difference (SMD) was calculated using scores of the treatment and comparison group at post-intervention. The SMD was chosen over the weighted mean difference because the papers being reviewed used varying measures to assess aggression/irritability. A random effects model was used to conduct the meta-analysis on the basis of the varying study characteristics found in this literature (e.g., treatments studied, sample sizes, age of sample and use of different outcome measures). Inconsistency between the effect size results of the studies was measured using the I^2 statistic which indicates the proportion variability in effect estimates that is due to heterogeneity

rather than sampling error (chance). Thresholds for the interpretation of the I^2 statistic were taken from the Cochrane Handbook (Higgins & Green, 2011) which advises that 30-60% may indicate moderate heterogeneity, 50%-90% may indicate substantial heterogeneity and levels above 75% can indicate considerable heterogeneity.

Meta-analyses were conducted using all the studies together and also for individual subsets of studies which were grouped by intervention type. A formal statistical analysis investigating the difference in effect sizes across treatment types was not possible due to the small number of studies in each group. However, pooled effect sizes and 95% confidence intervals for each type of intervention were calculated. Analysis was conducted on subgroups of studies where there were more than one paper of a treatment type (i.e. parent training, CBT and horse riding interventions).

3. Results

3.1. Study characteristics

The study characteristics are displayed in Table 1.

3.1.1. Sample population

There were a total of 602 children across the 11 included studies. The smallest study in this review had 11 participants (Scarpa & Reyes, 2011) and the largest had 180 (Bears et al., 2015). Mean age was reported in eight of the studies. For Garcia-Gomez et al., (2014); Smith, Groen & Wynn (2000); Scarpa & Reyes (2011), the mean age was estimated using the midpoint of the age range provided. The estimated mean age of children across the studies was 7 years and 3 months. The age ranged from 18 months to 16 years old. The age

range, without including one study (Smith et al., 2000) was 5-16 years. The percentage of male participants in ten of the studies was 86.2% (N=519). This gender statistic does not include participants from Sofronoff, Attwood, Hinton & Levin (2007; N=51) because gender ratio data was not available.

In all of the studies, authors described the participants as having a confirmed formal diagnosis of ASD. Five studies reported children with 'Autism' or an 'ASD'. One paper used a sample of children who had diagnoses of an Autistic Disorder or Asperger Disorder or PDDNOS, one with Autistic Disorder or Asperger Disorder, one with only PDDNOS. These are all terms which would be classified as ASD according to the DSM-5 (American Psychological Association, 2013).

In terms of cognitive ability, all but one study (Tellegen & Sanders, 2014), reported some type of data on the estimated Intelligence quotient (IQ) of the sample. In the ten studies where data was available, IQ levels ranged from 44 ('extremely low') to 135 ('very superior'). Two studies reported children with a wide range of ability ranging from extremely low to very superior (Solomon, Ono, Timmer, Goodlin-Jones (2008); Sofronoff et al., (2007)). But most studies (N=7) reported that all or the majority of their sample had a mean IQ above 70 (i.e. above 'borderline' range). Of these seven studies, two were conducted on samples of children diagnosed with Asperger Syndrome (Sofronoff, et al., 2007; Scarpa & Reyes, 2011). Two studies reported their IQ levels ranged between the extremely low range and the borderline range (Smith et al., (2000); Mohammadzaheri, Koegel, Rezaee & Rafiee (2015)).

Table 1 Characteristics of the included studies in the review and meta-analysis

Author (year)	Sample Size	Age range; Mean; Gender Ratio	IQ level	ASD Diagnosis/ subtype distribution	Type of Intervention	Rater & Outcome Measure	Research Design (comparison group)
Tellegen & Sanders (2014)	64	Age range = 2-9 years; Mean = 5 years 8 months; 85.9% Male	Not reported	Children with a diagnosis of an ASD from a Paediatrician or Child Psychiatrist. Diagnosis was verified using a semi-structured interview based on DSM-IV. (16=ASD, 20 =Autism, 12 = Asperger Syndrome, 16=PDDNOS) according to DSM-IV.	Brief Parenting Programme (individual) Based on the Stepping Stones Triple P Programme (a programme specifically designed for parents of children with disabilities). Sessions target one or two specific child problems. (Most common problems targeted were aggression and non-compliance).	Parent report- Eyberg Child Behaviour Inventory- Problem subscale (how frequent behaviour problems occur)	Randomised Control Trial Experimental vs (care as usual group)
Sofronoff, Leslie & Brown (2004)	51	Age range = 6-12 years; Mean= 9 years 4 months; Gender not reported	Assumed > 70	Children diagnosed with Asperger Syndrome by a consultant paediatrician.	Parent Training (individual and group modalities were combined for this meta-analysis) Components included Psychoeducation about ASD and common difficulties, comic strip conversations, social stories, management of behaviour such as interrupting temper tantrums, anger, noncompliance and bedtime problems, management of rigid behaviours, management of anxiety.	Parent rated- Eyberg Child Behaviour Inventory - Problem subscale.	Randomised Control Trial Experimental vs (Waiting list control group)

Table 1 Characteristics of the included studies in the review and meta-analysis

Author (year)	Sample Size	Age range; Mean; Gender Ratio	IQ level	ASD Diagnosis/ subtype distribution	Type of Intervention	Rater & Outcome Measure	Research Design (comparison group)
Bearrs et al. (2015)	180	Age range = 3-6 years; Mean= 4 years 8 months; 87.8% Male	74% of children had an IQ > 70	Parents of children with ASD (DSM-4) based on clinical assessment supported by an ADOS-R completed by clinicians trained to reliability.	Parent Training (individual) 11 core sessions and up to 2 additional sessions, a home visit and up to 6 parent-child coaching sessions over 16 weeks. Sessions included specific strategies to manage disruptive behaviour including functional analysis training to understand the function of their child's behaviour, learning strategies to prevent it and reinforcement techniques for appropriate behaviour.	Parent report- Aberrant Behaviour Checklist– Irritability subscale.	Randomized Control Trial Experimental vs (control group Parent education- no behaviour management strategies)

Table 1 Characteristics of the included studies in the review and meta-analysis

Author (year)	Sample Size	Age range; Mean; Gender Ratio	IQ level	ASD Diagnosis/ subtype distribution	Type of Intervention	Rater & Outcome Measure	Research Design (comparison group)
Solomon, Ono, Timmer, Goodlin-Jones (2008)	19	Age range = 5-12 years; Mean= 8 years 2 months; 100% Male	IQ range = 79-135	Children meeting criteria for Autistic Disorder, Asperger Syndrome or PDDNOS (DSM-IV)	Parent-Child Interaction Therapy (individual) Average=12.7 sessions Treatment involved 2 phases: 1. Child directed attention (parents are coached by therapist to give child positive attention and praise, ignore negative behaviour and not use criticism). 2. Parent directed attention (parents are coached to give clear, consistent commands and reinforce compliance). Modifications were made for ASD. If child was playing in isolation or being inappropriately controlling parents were encouraged to be more directive even in first phase. If child talked excessively about special interests, this was prohibited.	Parent report-Behaviour Assessment System for Children-Aggressiveness subscale	Randomized Control Trial Experimental vs (waiting list control group)

Table 1 Characteristics of the included studies in the review and meta-analysis

Author (year)	Sample Size	Age range; Mean; Gender Ratio	IQ level	ASD Diagnosis/ subtype distribution	Type of Intervention	Rater & Outcome Measure	Research Design (comparison group)
Garcia-Gomez et al., (2014)	16	Age range = 7-14 years; Mean = not available; 81.3% Male	IQ= 50 and above. (2 subjects <50 and the rest >90)	Children 'diagnosed with ASD', attending mainstream schools in Spain No report of check.	Horse Riding (group) 24 sessions over 3 months. 45 minutes each. Twice weekly in groups of 4. Each session had three phases 1. Preparing equipment and horse 2. Mounting and riding 3. Dismount, bring in horse and tidy equipment and say goodbye to horses.	Teacher report-Behaviour Assessment System for Children-Aggressiveness subscale	Quasi-Experimental Design (no randomisation) Experimental vs (delayed-treatment group)
Gabriels et al. (2012)	42	Age range = 6-16 years; Mean= 8 years 8 months; 85.7% Male	Nonverbal IQ= 44-139 (mean = 95.2)	Children with 'Autistic or Asperger's disorder' (DSM-IV) No report of check.	Therapeutic horseback riding (group) 10 weekly lessons, 60 minutes, taught in small groups of 3-4 participants who each had an allocated volunteer. Sessions had a 2 part focus (horsemanship and therapeutic riding skills). Individual goals were set for each domain. Routine of activities included preparing, mounting, learning horse-riding skills or doing activities, dismount, groom horse, tidy equipment. The lesson plans included activities that addressed physical, psychological, cognitive and social skills as well as horsemanship skills.	Parent report-Aberrant Behaviour Checklist-Irritability subscale.	Quasi-Experimental Design. randomization process) Experimental vs (Waiting list control)

Table 1 Characteristics of the included studies in the review and meta-analysis

Author (year)	Sample Size	Age range; Mean; Gender Ratio	IQ level	ASD Diagnosis/ subtype distribution	Type of Intervention	Rater & Outcome Measure	Research Design (comparison group)
Gabriels et al. (2015)	116	Age range = 6-16 years; Mean=10years 2 months; 87.1% Male	Mean IQ = 86.4, SD= 24	Children with an ASD diagnosis (confirmed by meeting cut-off score, 15 on Social Communication Questionnaire and cut off on ADOS or ADO2 assessment).	Therapeutic Horseback riding (group) 10 weekly sessions, 45 minutes long where each participant had at least one volunteer. Sessions had a 2 part focus (horsemanship - how to lead and care for the horse and therapeutic riding skills - mounting, halting, steering, turning and trotting). Each session followed the same routine of activities.	Parent report- Aberrant Behaviour Checklist- Irritability subscale	Randomised Control Trial. Experimental vs (control group - Barn activity (without horses))
Smith, Groen & Wynn (2000)	28	Age range = 18-42 months; Mean = not available; 82.1% Male	IQ range = 35 - 75.	Diagnosed with Pervasive developmental disorder not otherwise specified (PDDNOS). No report of check.	Early intensive treatment- Individual Treatment followed the Lovaas et al 1981 manual. An intensive treatment involving interaction between the child, parent/trainer. It is based on behavioural principles and applied behavioural analysis aimed to encourage and develop socially significant behaviours. The treatment was adapted to be less intensive than the manual by having 24 rather than 40 hours per week and treatment was phased out after 18 months for children that were developing slowly rather than continuing for 10 years. Up to 3 years treatment. 24.5 hours a week for one year and then reducing over the next 1-2 years	Parent report Child Behaviour Checklist - aggression subscale.	Randomised Control Trial Experimental vs (parental training control)

Table 1 Characteristics of the included studies in the review and meta-analysis

Author (year)	Sample Size	Age range; Mean; Gender Ratio	IQ level	ASD Diagnosis/ subtype distribution	Type of Intervention	Rater & Outcome Measure	Research Design (comparison group)
Mohamma dzaheri, Koegel, Rezaee & Rafiee (2015)	30	Age range = 6-11 years; Mean= 9 years 3 months; 60% Male	IQ range = 50-70	Children diagnosed with Autism (DSM-IV)	Two treatment conditions: Pivotal Response Treatment (PRT) vs Applied Behaviour Analysis (ABA) used during a language intervention. Aim was to investigate whether PRT would result in lower levels of disruptive behaviour. PRT involved contingent consequences for the target behaviour (speech utterance attempt) that were chosen by the child based on preference/interest (e.g., child preferred treats and activities used as rewards) ABA condition involved contingent consequences for the target behaviour (speech utterance that was longer than previous attempt) that were teacher chosen pre-printed picture cards. Twice weekly 1 hour sessions over 3 months.	Independent rater- Frequency of disruptive behaviour	Randomised Control Trial. Two treatment conditions

Table 1 Characteristics of the included studies in the review and meta-analysis

Author (year)	Sample Size	Age range; Mean; Gender Ratio	IQ level	ASD Diagnosis/ subtype distribution	Type of Intervention	Rater & Outcome Measure	Research Design (comparison group)
Scarpa & Reyes (2011)	11	Age range = 5-7years; Mean = not available; 81.8% Male	Assumed IQ > 70 (described as 'high functioning')	Children with 'high functioning ASD' (confirmed by meeting cut-offs on a parental report (Social Communication Questionnaire) and observation on the Autism Diagnostic Observation Schedule.	Cognitive Behaviour Therapy- group

Group programme for children and separate concurrent parent sessions. 9 sessions (One hour each).

The sessions were structured and covered emotional regulation skills via affective education and relaxation, physical, social and cognitive strategies to 'fix' intense emotions. | Parent report- Emotion Regulation Checklist- emotion regulation subscale | Pilot Randomised Control Trial

Experimental vs (delayed treatment control group) |

Table 1 Characteristics of the included studies in the review and meta-analysis

Author (year)	Sample Size	Age range; Mean; Gender Ratio	IQ level	ASD Diagnosis/ subtype distribution	Type of Intervention	Rater & Outcome Measure	Research Design (comparison group)
Sofronoff, Attwood, Hinton & Levin (2007)	45	Age range= 10-14 years; Mean=10years 9 months; 95.6% Male	IQ range (WISC III) = 95-132 Mean IQ of Intervention group = 105.24 (SD= 22.3). Mean IQ of Control group = 108.7 (SD= 21.6)	Children diagnosed with Asperger Syndrome by a consultant paediatrician.	Cognitive Behaviour Therapy programme- group Six sessions (2 hours each) The programme consisted of a structured programme including the following main elements: 1. Affective education based on cognitive behavioural model (emotions, thoughts, behaviour & physiology) 2. Toolbox idea introduced with relaxation, social, physical and cognitive tools to manage emotions. 3. Idea of varying degrees of emotion using a 'thermometer' 4. Exploration of how social stories can be used for emotion management.	Parent report- Frequency of anger outbursts	Randomised Control Trial Experimental vs (Waiting list control group)

3.1.2. Research design

Nine studies were randomized control trials and two adopted a quasi-experimental approach (where participants were not randomly allocated to the experimental or control group (Gabriels et al., 2012 & Garcia-Gomez et al., 2014)). The majority of studies (N = 4) employed a waiting-list control group (Sofronoff et al., 2004; Gabriels et al., 2012, Solomon et al., 2008; Sofronoff et al., 2007). Others employed an active control group condition (N = 3, Gabriels et al., 2015; Bearrs et al., 2015; Tellegen & Sanders, 2014), where the control group received an intervention that controlled for an essential part of the intervention, some studies employed a delayed treatment control (N = 2, Scarpa & Reyes, 2011; Garcia-Gomez et al., 2014) and one study compared two treatment arms (Mohammadzaheri et al., 2015).

3.1.3. Intervention types

Four studies examined the effect of parent training (Tellegen & Sanders, 2014; Sofronoff et al., 2004; Bearrs et al., 2015; Solomon et al., 2008). All these interventions involved parents attending training sessions and learning strategies to prevent and manage ABPs shown by their child. Two of these interventions incorporated live coaching of the use of behavioural methods to change their child's behaviour (Bearrs et al., 2015; Solomon et al., 2008). The interventions varied in intensity from four sessions (Tellegen & Sanders, 2014) to an intervention spread over 16 weeks involving 11 core sessions, a home visit, telephone coaching and parent-child coaching sessions (Bearrs et al., 2015). Three studies investigated horse riding

interventions (Gabriels et al., 2012; Garcia-Gomez, 2014; Gabriels et al., 2015). In these studies, children participated in therapeutic horseback riding in small groups. The number of sessions varied from 10-24 across the studies. Children were assigned volunteers and each session followed the same structure which included being taught and practicing horsemanship skills (preparing the horse, grooming, mounting and riding). Two studies investigated cognitive behavioural therapies (Scarpa & Reyes, 2011 & Sofronoff et al., 2007). These interventions involved psychoeducation about emotions using a cognitive behavioural framework and also taught children cognitive, physical, social and relaxation strategies to manage emotions. One study examined behavioural methods (Mohammadzaheri et al., 2015). This study compared the application of two types of behaviour intervention (variants of contingent management strategies) aimed at reducing disruptive behaviours during a language intervention.

One study examined the effect of early intensive treatment (Smith et al., 2000). The intervention followed the Intensive Behavioural Treatment Manual (Lovaas & Smith, 1988) and was adapted to be less intensive. Across all the studies, the length of intervention varied from four sessions to thirty hours of sessions per week over two to three years. Detailed information about each intervention can be found in Table 1.

3.1.4. Outcome measures assessing ABP

The measures of ABPs across the papers included nine validated questionnaires and two frequency counts of ABP. The questionnaires included Eyberg Child Behaviour Inventory - Problem subscale (N=2,

Tellegen & Sanders, 2014; Sofronoff et al., 2004), Aberrant

Behaviour Checklist – Irritability subscale (N=3, Bearrs et al., 2015,

Gabriels et al., 2012; Gabriels et al., 2015), Behaviour Assessment

System for Children - Aggressiveness subscale (N=2, Solomon et al.,

2008) (parent report) & Garcia-Gomez et al., (2014; teacher report),

Child Behaviour Checklist - aggression subscale (N=1, Smith et al.,

2000), Emotion Regulation Checklist - emotion regulation subscale

(N=1, Scarpa & Reyes, 2011). The frequency measures included one

parent report of anger outbursts (Sofronoff et al., 2007) and one

report from an independent rater on frequency of disruptive behaviour

(Mohammadzaheri et al. (2015).

3.2. Risk of bias

A summary graph of the assessment of the risk of bias in all the studies and
per domain can be found in Figures 2 and 3. A table describing risk of bias
and support for judgement can be found in Appendix 1.

3.2.1. Selection bias

The risk of selection bias was evaluated as unclear in most of the

studies (N=8) due to insufficient information provided on the process

of randomisation. The two studies that used a quasi-experimental

design were rated as having a high risk of selection bias as they did

not employ a randomization process (Garcia-Gomez et al., 2014;

Gabriels et al., 2012). Only one study (Smith et al., 2000) adequately

reported its methods and was evaluated as having a low risk of

selection bias.

Figure 2. Graph showing risk of bias in each domain for each study

Note: '+' denotes low risk, '-' denotes high risk and '?' denotes unclear risk

3.2.2. Performance bias

3.2.2.1. Treatment fidelity

Risk of performance bias from a lack of treatment fidelity was rated as high in four papers where there was no formal measure of treatment fidelity. One of these studies did not use a standardized intervention protocol at all. Lack of treatment fidelity was evaluated as low in six studies as it was either formally assessed using a therapist-rated checklist or assessed by an independent evaluator (who assessed a random sample of recordings). All reported ratings were above 80%. The risk of performance bias from treatment fidelity was unclear in one study as an appropriate adherence check method was reported but an adherence figure was not reported.

3.2.2.2. Blinding of participants

The risk of performance bias from a lack of blinding of participants was rated as high in almost all studies (N=10). There was no evidence that studies had used initiatives to blind participants to the treatment they were allocated and it was therefore judged that they were vulnerable to expectation effects.

3.2.3. Attrition bias

Risk of bias from attrition was assessed to be low in five papers. This risk was assessed to be unclear in six papers due to insufficient reporting of rates of attrition and exclusion to permit judgement.

Figure 3. Graph showing risk of bias per domain
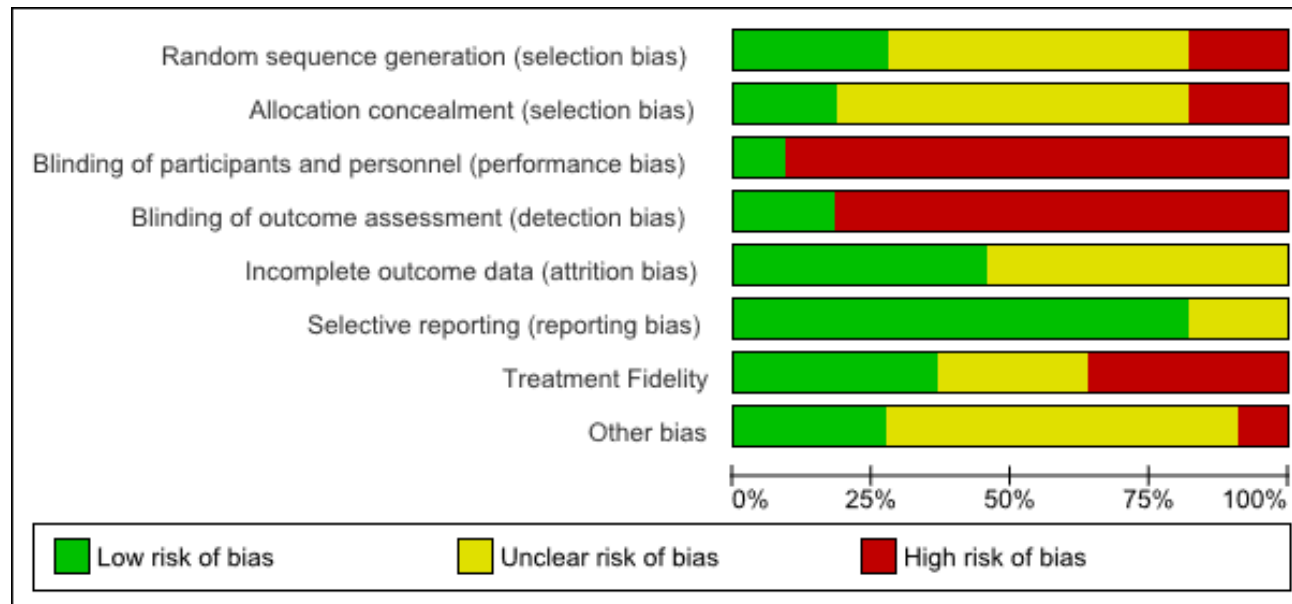
3.2.4. Detection bias

The risk of detection bias from a failure to blind the outcome assessment was evaluated to be high in nine papers as the outcome measure was completed by a parent who was aware of the treatment allocation and thus the outcomes were judged to be vulnerable to expectation effects. It was assessed to be low in two papers.

3.2.5. Other risks

Other risks identified include no long-term follow up data in five papers (Solomon et al., 2008; Scarpa & Reyes, 2011; Mohammadzaheri et al., 2015; Gabriels et al., 2012; Garcia-Gomez et al., 2014). Baseline imbalance in Tellegen & Sanders (2014) where there were more child health problems in the treatment group compared to the control group, could have resulted in underestimation of effect. Mohammadzaheri et al., (2015) used observational frequency data of the number of instances of disruptive behaviour but only collected two time points (pre and post intervention). Furthermore this study did not include a measure of change beyond the last session and beyond the clinic setting.

3.3. Efficacy of psychological treatments overall

A random-effects meta-analysis incorporating all 11 studies, revealed a statistically significant overall moderate effect of psychological interventions for reducing the ABPs of children with ASD, [N = 602, Z = 2.07, P= 0.04, pooled effect size = - 0.51 [95% CI = -1.00, -0.03]]. This outcome data can be seen in Figure 4. This outcome demonstrated high level of heterogeneity (I^2 = 85%). When the two studies using non-validated questionnaire

outcome measures were excluded (Mohammadzaheri et al., 2015; Sofronoff et al., 2007), the heterogeneity level was moderate (I^2 = 49%). The nine papers included, revealed a pooled moderate effect size of -0.38 [CI = -0.66, -0.10] that was statistically significant [N = 455, Z = 2.66, P = 0.008]. This outcome data can be seen in Figure 5.

3.4. Efficacy of treatments by subtype

3.4.1. Efficacy of Parent Training Interventions

A random-effects meta-analysis incorporating the four papers studying parent training interventions (N = 314) revealed non-significant pooled effect size of -0.44 [CI = -0.93, -0.06, Z = 1.75 (P = 0.08)]. The outcome demonstrated a high level of heterogeneity (I^2 = 71%). This outcome data can be seen in Figure 6.

3.4.2. Efficacy of Horse Riding Interventions

A random-effects meta-analysis incorporating the three papers studying horse riding interventions (N = 174) revealed a non-significant pooled effect size of -0.36 [95% CI = -0.93, 0.22, Z = 1.21, P = 0.23]. The outcome demonstrated a moderate level of heterogeneity (I^2 = 60%). This outcome data can be seen in Figure 7.

3.4.3. Efficacy of CBT Interventions

A random-effects meta-analysis incorporating the two papers studying CBT interventions (N = 56) revealed a non-significant pooled effect size of -0.13 [95% CI = -0.65, 0.40, Z = 0.47, P = 0.64]. The outcome demonstrated no heterogeneity (I^2 = 0%). This outcome data can be seen in Figure 8.

Figure 4. Figure showing results from meta-analysis including all 11 studies

Study or Subgroup	Experimental Mean	SD	Total	Control Mean	SD	Total	Weight	Std. Mean Difference IV, Random, 95% CI
Garcia-Gomez 2014	0	0	0	0	0	0		Not estimable
Mohammadzaheri 2015	1.6	0.2	15	8.5	0.56	15	1.1%	-15.97 [-20.36, -11.57]
Scarpa 2011	23.2	1.99	5	23.42	3.91	6	7.1%	-0.06 [-1.25, 1.12]
Garcia-Gomez 2014	3.333	4.926	8	1.25	1.5	8	8.1%	0.54 [-0.46, 1.54]
Solomon 2008	59.7	4.95	10	62.22	9.77	9	8.7%	-0.32 [-1.22, 0.59]
Smith 2000	56.11	9.1	15	59.67	10.4	13	9.6%	-0.36 [-1.10, 0.39]
Gabriels 2012	12.9	8.5	26	20	8.5	16	10.2%	-0.82 [-1.47, -0.17]
Sofronoff 2004	10.5	5.44	34	17.53	5.65	17	10.2%	-1.26 [-1.89, -0.62]
Sofronoff 2007	3.7	39.24	24	7.9	5.1	21	10.5%	-0.14 [-0.73, 0.44]
Tellegen 2014	14.04	8.82	35	15.46	6.08	29	11.0%	-0.18 [-0.68, 0.31]
Gabriels 2015	9.5	7.98	58	13.6	10.8	58	11.6%	-0.43 [-0.80, -0.06]
Bearss 2015	16.6	14.24	89	18.5	14.4	91	11.9%	-0.13 [-0.42, 0.16]
Total (95% CI)			**319**			**283**	**100.0%**	**-0.51 [-1.00, -0.03]**

Heterogeneity: Tau² = 0.49; Chi² = 64.85, df = 10 (P < 0.00001); I² = 85%
Test for overall effect: Z = 2.07 (P = 0.04)

Figure 5. Figure showing results from a meta-analysis run with the exclusion of studies that used non-validated outcome measures (Mohammadzehri2015 & Sofronoff, Attwood, Hinton & Levin, 2007). This analysis includes 9 studies.

Study or Subgroup	Experimental Mean	SD	Total	Control Mean	SD	Total	Weight	Std. Mean Difference IV, Random, 95% CI
Scarpa 2011	23.2	1.99	5	23.42	3.91	6	4.5%	-0.06 [-1.25, 1.12]
Garcia-Gomez 2014	3.333	4.926	8	1.25	1.5	8	5.9%	0.54 [-0.46, 1.54]
Solomon 2008	59.7	4.95	10	62.22	9.77	9	6.9%	-0.32 [-1.22, 0.59]
Smith 2000	56.11	9.1	15	59.67	10.4	13	9.0%	-0.36 [-1.10, 0.39]
Gabriels 2012	12.9	8.5	26	20	8.5	16	10.7%	-0.82 [-1.47, -0.17]
Sofronoff 2004	10.5	5.44	34	17.53	5.65	17	11.0%	-1.26 [-1.89, -0.62]
Tellegen 2014	14.04	8.82	35	15.46	6.08	29	14.2%	-0.18 [-0.68, 0.31]
Gabriels 2015	9.5	7.98	58	13.6	10.8	58	17.7%	-0.43 [-0.80, -0.06]
Bearss 2015	16.6	14.24	89	18.5	14.4	91	20.0%	-0.13 [-0.42, 0.16]
Total (95% CI)			**280**			**247**	**100.0%**	**-0.38 [-0.66, -0.10]**

Heterogeneity: Tau² = 0.08; Chi² = 15.82, df = 8 (P = 0.04); I² = 49%
Test for overall effect: Z = 2.66 (P = 0.008)

Figure 6. Figure showing results from meta-analysis on studies using Parent Training interventions

Study or Subgroup	Experimental			Control			Weight	Std. Mean Difference IV, Random, 95% CI	Std. Mean Difference IV, Random, 95% CI
	Mean	SD	Total	Mean	SD	Total			
Solomon 2008	59.7	4.95	10	62.22	9.77	9	16.5%	-0.32 [-1.22, 0.59]	
Sofronoff 2004	10.5	5.44	34	17.53	5.65	17	23.1%	-1.26 [-1.89, -0.62]	
Tellegen 2014	14.04	8.82	35	15.46	6.08	29	27.3%	-0.18 [-0.68, 0.31]	
Bearss 2015	16.6	14.24	89	18.5	14.4	91	33.2%	-0.13 [-0.42, 0.16]	
Total (95% CI)			**168**			**146**	**100.0%**	**-0.44 [-0.93, 0.06]**	

Heterogeneity: Tau² = 0.17; Chi² = 10.20, df = 3 (P = 0.02); I² = 71%
Test for overall effect: Z = 1.74 (P = 0.08)

Figure 7. Figure showing results from meta-analysis on studies using Horse Riding interventions

Study or Subgroup	Intervention Mean	SD	Total	Control Mean	SD	Total	Weight	Std. Mean Difference IV, Random, 95% CI	Std. Mean Difference IV, Random, 95% CI
Garcia-Gomez 2014	3.333	4.926	8	1.25	1.5	8	20.9%	0.54 [-0.46, 1.54]	
Gabriels 2012	12.9	8.5	26	20	8.5	16	33.0%	-0.82 [-1.47, -0.17]	
Gabriels 2015	9.5	7.98	58	13.6	10.8	58	46.1%	-0.43 [-0.80, -0.06]	
Total (95% CI)			92			82	100.0%	-0.36 [-0.93, 0.22]	

Heterogeneity: Tau² = 0.15; Chi² = 4.98, df = 2 (P = 0.08); I² = 60%
Test for overall effect: Z = 1.21 (P = 0.23)

Favours intervention   Favours control

Figure 8. Figure showing results from meta-analysis on studies using CBT interventions

	Intervention			Control				Std. Mean Difference	Std. Mean Difference
Study or Subgroup	Mean	SD	Total	Mean	SD	Total	Weight	IV, Random, 95% CI	IV, Random, 95% CI
Scarpa 2011	23.2	1.99	5	23.42	3.91	6	19.6%	-0.06 [-1.25, 1.12]	
Sofronoff 2007	3.7	39.24	24	7.9	5.1	21	80.4%	-0.14 [-0.73, 0.44]	
Total (95% CI)			29			27	100.0%	-0.13 [-0.65, 0.40]	

Heterogeneity: Tau² = 0.00; Chi² = 0.01, df = 1 (P = 0.91); I² = 0%
Test for overall effect: Z = 0.47 (P = 0.64)

-1 -0.5 0 0.5 1
Favours intervention   Favours control

4. Discussion

The present study is a literature review of psychological interventions for children with ASD that measured irritability or aggression as an outcome. It employed a review process based on a systematic search and used meta-analysis to identify and summarise the characteristics of and efficacy demonstrated by controlled studies conducted in this area. Whilst ABPs are a common problem in this population, no meta-analysis of psychological interventions for such problems has been reported yet.

This study identified 11 papers which included a total of 602 children with ASD. The number of studies of psychological treatments in this area is limited and previous reviews have reported very few or no control studies on psychology interventions however, this review employed a wide search criteria and a thorough screening process is are likely to have resulted in the identification of more papers than has been reported before. For example, a recent review on treating aggression in persons with ASD did not identify any controlled studies on psychological interventions (Matson & Jang, 2014). A separate review of CBT for ASD and disruptive behaviours cited only two studies; one controlled study on CBT and one pre-post small-N design of a mindfulness intervention involving three participants (Singh et al., 2006).

4.1. Characteristics of studies

The review revealed five types of interventions that have been studied in this area using a controlled design. They included parent training (N=4), horse riding therapy (N=3), CBT (N=2), behavioural methods (N=1) and early intensive interaction (N=1). Notably, the review process identified a number of other psychological interventions studied and many (72 out of 123) were excluded from this review due to having no comparator group. Other

psychological interventions studied (but not included in this review) include a vast number of behavioural studies and a few mindfulness-based interventions, TEACHH methods and interventions based on social stories. Future studies could also aim to investigate such treatments using larger controlled studies.

The literature on ABPs in non-ASD populations indicates that multi-systemic therapy (MST) has shown efficacy to help young people showing ABPs (Henggeler, 2011). In comparison, ASD literature appears to lack research on interventions that target multiple systems connected to the young person. Given that, like for TDCs, the ABPs of children with ASD are likely to be determined by multiple factors, there may be benefits to conducting research on an MST for children with ASD. For example, the intervention could intervene at an individual level (cognitive and behavioural aspects), environment level (such as structure, routine and environment), parent level (psychoeducation and reducing parental stress), school level (issues related to education and learning) and social issues (increasing awareness of ASD with peers at school, initiatives to facilitate friendship building and target bullying). This type of research and treatment is likely to be costly but may reduce long-term costs of untreated or ineffectively treated ABP for individuals with ASD.

The gender distribution of participants in the studies included in this review was 86.2% male which is similar to a recent meta-analysis of CBT for anxiety in children with ASD (N=511, 83.6% male, Ung, Selles, Small & Storch, 2015). This distribution is common in ASD literature. However, the samples are likely to be unrepresentative of the actual gender ratio of children living with ASD. A growing body of research suggests that girls with ASD show differences in symptomology and are being underdiagnosed (Van

Wijngaarden-Cremers, Van Eeten, Groen, Van Deurzen, Oosterling & Van der Gaag (2014). Thus, they are likely to be underrepresented in research. As awareness and research into the female ASD phenotype is increasing, along with the development of female-sensitive diagnostic instruments, future studies will hopefully have a more representative gender distribution in their samples.

The diagnosis of participants in the studies varied both within and between studies. Studies ranged from including participants with a mix ASD subtypes to some studies only including children with one subtype (e.g., only PDDNOS or only Asperger syndrome). Having such heterogeneous populations makes it difficult to compare studies and generalise findings. There was also a wide range and mix of cognitive abilities of the samples between and within some of the studies, although not all. It is likely that children with different levels of cognitive functioning will respond differently to different types of treatment. Not all papers reported information on IQ level and it will be important for future studies to fully report the mean IQ and range of their sample in order to understand which type of ASD populations the results can generalise to. Furthermore, it will be helpful for future studies to use homogenous groups with regard to IQ levels or moreover incorporate IQ as a moderator in their analysis to understand the effect of this on response to treatment.

In terms of sample size, most studies used relatively small samples compared to psychopharmacological trials. Only two used samples greater than 100 participants. This is likely to be because carrying out psychological RCT with large sample sizes requires the availability of trained clinicians and clinic space and can easily become time consuming and costly.

4.2. Quality of the evidence

The methodological quality of studies was evaluated using the critical appraisal guidance outlined in the Cochrane handbook (Higgins & Green, 2011). A strength of the literature on psychological interventions for children with ASD and ABPs is the low risk of reporting bias. This reduces the risk of a 'file drawer effect' that can lead to interventions erroneously being portrayed as more effective than they are. Therefore the results of this review are unlikely to reflect inflated effect sizes stemming from reporting biases. However, no studies were rated as having a low risk of bias in all domains. There were repeated problems with the reporting of random sequence generation and allocation concealment leading to unclear understanding of selection bias in most studies. Studies have shown that research papers where concealment is judged to be inadequate or unclearly reported show on average 18% more beneficial effect sizes (Pildal, 2007). Thus there is a risk that a number of the effect sizes in this review could be inflated.

There was a high risk of performance bias in most studies. Given the nature of psychological interventions, performance bias stemming from lack of blinding participants is difficult to avoid. However, future studies should aim to use manualised protocols and treatment adherence checks. Detection bias was also a problem for many of the included studies. The majority of studies used parent-rated outcome measures and parents were likely to be aware of the allocation group. This makes the studies vulnerable to bias from expectation effects. Lack of outcome rater blinding has been linked with an increased average effect size of 9% (Pildal et al., 2007). However, this inflation of effect size is reduced when objective measures are used (Wood et al., 2008) and studies in this review used measures that collected

objective data on child behaviour which buffer some of the risk of overestimation of effect size from detection bias.

One study (Mohammadzaheri et al., 2015) did use an independent rater of ABP that was blind to allocation group, which significantly lowers the risk of performance bias. Interestingly, this study showed an unusually large effect size. However, ratings were only collected at two time points and also only during sessions in the clinic which reduces the reliability of this measure and generalizability of the findings beyond the clinic session. Blind independent raters observing children in natural environment instead of parent-rating would reduce performance bias. However, there are benefits to using parent-rated outcome measures in this area as parents are most likely to be able to observe small but relevant changes in their child's behaviour. An alternative way of reducing risk of bias and still use parent-rated outcomes in such studies could be to blind parents to the allocation group (e.g., by having an active control group where parents cannot easily guess which the treatment group is).

4.3. Treatment efficacy

A meta-analysis was conducted on all 11 studies to understand whether there is evidence that psychological treatments show efficacy to reduce ABPs shown by children with ASD.

The controlled studies on psychological treatments showed a significant and moderate effect size for reducing ABPs in children with ASD. However, a number of potential biases were introduced to these studies through the design (e.g., performance and detection biases) and a number of unreported methodological details make the evaluation of other important biases (e.g., selection bias and attrition biases) unclear. Thus, the results

from the meta-analysis must be interpreted with caution and further research is needed to strengthen the conclusion drawn.

The meta-analysis showed considerable variability in effect size between the studies. There are a number of possible sources of this variability. One source of the variability in overall effect size could be the variability between the types of psychological treatments included in the analysis. Due to the small number of studies available for each type of treatment, a formal analysis comparing effect sizes across treatment types was not possible. However, pooled effect sizes were calculated across the subtypes of interventions (i.e. CBT, parent training and horse riding types). The confidence intervals of each of these effect sizes obtained crossed zero and thus were non-significant and this is likely to reflect the limited power available in each subgroup analysis.

Another source of the variability between the effect sizes of studies could reflect the different outcome measures used across the studies. However, standardized mean differences were used to account for this. Furthermore, when only studies reporting scaled standardized questionnaire measures of ABP were included and studies using frequency data were excluded from the meta-analysis (Mohammadzaheri et al., 2015; Sofronoff et al., 2007; 75 participants), there was less but still substantial variability between the effect sizes. The overall significant and moderate pooled effect size remained unchanged. This suggests that psychological treatments can have a moderate effect size to reduce ABP in children with ASD but there is substantial variability between the effect size across different studies that is not accounted by the mix of standardized questionnaire with unstandardized frequency outcome measures.

Other sources of variability include the outcome raters (parent, teacher or independent rater) and varying characteristics of the samples (e.g., cognitive ability and age). Once there is a greater number of studies published in this area, future reviewers should conduct moderator analysis to reveal which variables moderate treatment response. This will help to understand the variability in effect sizes observed in this review.

In conclusion, there are a handful of controlled studies of psychological interventions aimed at reducing ABP in children with ASD. Overall these treatments have a pooled moderate effect size although there is significant heterogeneity between the studies. The pooled effect sizes obtained for each type of intervention (CBT, parent training and horse riding) were not significant which is likely to reflect a lack of statistical power. Additional studies, using similar interventions and outcome measures, conducted with larger sample sizes, greater methodological rigour and detailed reporting of methodology are needed to replicate findings and fully understand the efficacy of psychological treatments for children with ASD and ABPs.

4.4. Limitations of the review

Limitations of this review should be considered. Firstly, this review focused on studies that used a control group. Whilst it increases the internal validity of the findings and provides an overview of the controlled studies conducted in this area, it does not reflect the breadth of psychological treatments in this area as it excludes a significant number of small-N and uncontrolled research studies that have been conducted in this area.

Secondly, due to small amount of controlled research designs conducted in this area, this review employed relatively wide inclusion criteria

(in terms of population age and outcome measures used to measure ABP). One benefit of this is that is generated 11 studies and described the range of characteristics of the existing studies in this area. However, it did result in a heterogeneous group of studies which had drawbacks for the meta-analysis. Had stringent inclusion criteria been set, there would be very few studies included. Measures were used to account for the influence of the heterogeneity (such as using standardized mean differences and a random-effects model) but the results of the meta-analysis should still be interpreted a consideration of the heterogeneity between the studies.

Thirdly, the results of this review are limited by incomplete data available for the included studies. Methodological details, participant characteristics and outcome data were not fully reported for all studies and therefore results were either reported with missing data or estimates were used (as reported in the review).

Fourth, due to resource limitations this review could not assess inter-rater reliability of study selection of risk of bias tool.

4.5. Implications for clinical practice

The results of the meta-analysis suggest that non-pharmacological interventions are warranted and should be considered by service providers to reduce the amount of ABPs shown by children with ASD as an alternative to psychopharmacological interventions. There is currently no evidence that one specific type of psychological intervention is indicated above others. Thus, services should not restrict the type of psychological treatment options available and instead aim to offer a range of psychological treatment options to service users.

The results of this meta-analysis also indicate that children receiving psychological treatments will not consistently show the same amount of change in symptoms (as reflected in the heterogeneity of the magnitude of effect sizes across studies). It is possible that the effect of different types of treatment will vary according to the characteristics of the treatment, characteristics of the child, and/or presenting problem and more research is needed to fully understand the role of such moderators.

4.6. Implications for future research

Implications for future research studies looking at psychological treatments to reduce ABPs for children with ASD have been noted throughout the discussion and the main points are summarised below. Improving research designs and reporting style would improve the quality of evidence available and increase confidence in the effect estimates provided in studies. It would be desirable for future research to address the following:

- More randomised controlled trials on behavioural treatments, cognitive behaviour therapy, horse riding and intensive interaction treatments for ABPs in children with ASD to replicate current findings

- Moderator analysis to understand the effect of participant characteristics (e.g., diagnostic subtype, IQ, age, gender, severity of problem) on the efficacy of treatment

- Research focusing on the long-term treatment effects and cost-benefit analysis of psychological treatments in this area

- The use of procedures for blinding outcome raters and/or having independent raters to decrease detection bias as well as the measurement of children's behaviour in natural environments to increase generalizability of findings

- The development of more controlled research designs investigating psychological treatments that have been shown to be efficacious in small-N designs

- Larger samples sizes that include a representative proportion of girls with ASD

- Careful reporting of outcome data to include group size, mean and standard deviations for each measure

- A CONSORT (Consolidated Standards of Reporting Trials) flow diagram of the trial

- Thorough reporting of participant demographics including gender, age range and mean, parental education level and income and cognitive ability estimates.

- Thorough reporting of methodological details to include details of randomization process (random sequence generation and allocation concealment procedures) as well as attrition rates, reasons and how this was managed to allow readers to judge potential bias adequately.

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Part 2: Empirical Paper

A multiple single-case design evaluation of a parent-mediated CBT intervention for children with ASD and anger management difficulties: feasibility, acceptability and an initial estimate of efficacy

Abstract

Children with Autism Spectrum Disorder (ASD) suffer from higher levels of emotional problems such as anger and aggression. The aim of this study was to investigate the acceptability, feasibility and obtain an initial estimate of efficacy for a cognitive behavioural intervention for young people (8-14 years old) with ASD and anger management problems. The study employed a mixed-methods, small-N design which consisted of a series of systematic case studies. Participants (N=7) acted as their own controls and baseline and follow-up data was collected. The quantitative and qualitative results indicated preliminary evidence that this intervention was acceptable to most families and led to reductions in anger outbursts for some young people as well as positive changes in parental stress and improved communication between parents and children. However, the outcome varied across participants. Obstacles to progress highlighted through post-intervention interviews with parents and therapist checklists related to the young person's motivation to change, their ability to generalize skills outside sessions and underlying anxiety. These are discussed and suggestions for improvements to the intervention are made. Overall, the findings support further investigation of a refined version of the intervention through a larger study.

1. Introduction

1.1. Background to Autism Spectrum Disorder

Autism Spectrum Disorder (ASD) is a neurodevelopmental condition that affects an individual's social and communication skills, thinking style and behaviour. Symptoms of Autism were first described simultaneously but separately by Leo Kanner and Hans Asperger in the 1940's, (Kanner, 1943; Asperger, 1944). Since then, there has been a growing body of research, interest and awareness into the characteristics, causes, prevalence, impact and treatments for this condition. The current Diagnostic and Statistical Manual, Fifth Edition, (DSM-5, American Psychological Association, 2013) describes a single diagnostic umbrella term, 'Autism Spectrum Disorder' which subsumes the separate diagnostic categories that were previously described in the DSM-IV, (APA, 2000).

ASD affects approximately 1% of the population, (Baird, Simonoff, Pickles, Chandler, Loucas, Meldrum & Charman, 2006) and the estimated prevalence is steadily increasing. For affected individuals, ASD symptoms are present from early development. Core features of ASD include clinically significant and persistent impairments in social communication and social interaction as well as rigid and repetitive patterns of behaviour or interests (RRBIs) and differences in sensory processing (American Psychological Association, 2013). The cognitive abilities of people with ASD can range from gifted to severely challenged (Charman, Pickles, Simonoff, Chandler, Loucas & Bairds, 2011). The exact cause of the condition is not fully understood but there is evidence that ASD is arises from a complex interplay between genetic and environmental risk factors (c.f. Mandy & Lai, 2016).

1.2. Associated emotional and behavioural difficulties: Anger and aggression

In addition to the core deficits, research indicates that many individuals with ASD show abnormalities in emotional regulation (Mazefsky, 2015) and show elevated levels of emotional and behavioural problems including anger and aggression. Aggression has been defined as an overt behaviour that can result in harm to self or others (Connor, 2002). Theoretical distinctions have been made between subtypes of aggression (Dodge, 1991; Vitiello & Stoff, 1997). Proactive aggression is controlled and instrumentally driven whereas reactive aggression is an emotionally-driven and impulsive. The latter occurs in response to frustration, a perceived threat or provocation and often takes the form of 'angry temper outbursts' (Matthys & Lochman, 2010, p.14). Over half of children with ASD have shown aggression towards a caregiver (Kanne & Mazurek, 2011). Over 25% of a sample of individuals with ASD were reported by parents to have problematic levels of temper tantrums and problems with being explosive and easily angered (Lecavalier, 2006). A more comprehensive summary of the high prevalence rate of aggression in children with ASD is described by Kanne & Mazuerk (2011).

Untreated anger and aggression can have a longstanding and negative impact on the child and their family and can disrupt the child's psychosocial development through a number of ways. In school, anger and aggression can interfere with learning and can lead to temporary and permanent exclusions (Barnard, Prior & Potter, 2000). Socially, poor control of anger can interfere with relationships. It can alienate peers and/or elicit bullying and victimization by others (Reiffe, Camodeca, Pouw, Lange & Stockman, 2012). Aggression can result in physical injury to the child and

others. It can lead to the prescription of sedative medications in children as young as three years old (Masi, Cosenza, Mucci & Brovedani (2001) and in severe cases, psychiatric admission (Shoham-Vardi et al. 1996) or involvement with the criminal justice system (Siponmaa, Kristiansoon, Johnson, Nyden & Gillberg, 2001). At the family level, anger and aggression can cause high levels of distress at home and severely affect family functioning. Studies have shown that the severity of the child's problem behaviour, as opposed to autism symptoms, is predictive of parenting distress and family functioning (Manning, Wainwright & Bennet, 2010). Furthermore, there is evidence of a bi-directional relationship between parenting stress and child behaviour problems (including aggression) (Zaidman-Zait et al, 2014). Thus, the need to establish evidence-based and effective treatments for anger and aggression in this population is crucial for both the young person (YP) and systems around them. Furthermore, interventions which involve carers and reduce their stress levels may be an important feature of effective treatments.

1.3. Current Interventions

Pharmacological treatments are commonly used to treat aggression shown by children with ASD and are currently recommended in the National Institute of Clinical Excellence guidelines (NICE, 2013) as the second line of treatment for those who show behaviour that challenges. Antipsychotics have been shown to reduce levels of aggression in this population (Marcus et al., 2009; Owen et al. 2009). However, these same studies report a high risk of side effects including weight gain, appetite increase, anxiety, sedation and fatigue and in some cases vomiting (Robb, 2010; Marcus et al., 2009; Owen et al. 2009). NICE guidelines (NICE, 2013) recommend regulatory controls for administration such as clearly defined and monitored target behaviours and adverse reactions. However, it is

unclear what extent these are being followed in routine clinical practice. Whilst practice in UK has not been systematically studied, one American study reported that the average duration of treatment was 5.3 years in a sample of 104 individuals with intellectual disabilities showing behaviour that challenges (Marshall, 2004). The long-term effects and dangers of taking such medications for long periods of time is unknown. Research that has investigated long term effects tends to report on periods up to six months (Malone, Maislin, Choudhury, Gifford & Delaney, 2002; Troost et al. 2005) and have found the onset of dyskinesias in some children when the antipsychotic was removed (Malone, Maislin, Choudhury, Gifford & Delaney, 2002). For these reasons, there is a pressing need to find effective short-term psychological interventions for this population to reduce the need to resort to the use of long-term antipsychotic medication.

The most widely investigated psychological interventions for aggression shown by children with ASD are based on behaviour theory (Matson & Jang, 2014). Behaviour theory and its application to individuals with ASD is outlined in the literature review above. Few controlled research designs have investigated the impact of behavioural interventions applied to this area (see systematic review). However, a number of small-N research designs have reported the success of behavioural interventions to reduce unwanted aggressive behaviours in the context of ASD and comorbid intellectual disability (Matson, 2009).

There is a need to identify alternative effective psychological treatments for children with ASD who show emotional regulation difficulties such as anger and aggression. Firstly, the behavioural interventions evolved from individuals with intellectual disability and most research studies using such methods involve children with ASD and comorbid intellectual disability and/or likely impairments

in verbal ability. Many behavioural interventions focus on communication training as an alternative means to communicate and access needs and it is unclear that such methods would be appropriate for aggression shown by those with above average verbal abilities. Consequently, there is scope to explore alternative interventions for children with ASD with average and above cognitive and verbal abilities. Secondly, behavioural methods rely on intensive external input from carers or staff and once intervention and contingencies are withdrawn, problem behaviours are likely to recur. The development of alternative evidence-based treatments for children that encourages self-regulation of behaviour could empower children to learn skills to regulate their own emotions and behaviour across settings and without requiring continuous input from others.

1.4. Cognitive Behavioural Interventions

In the non-disability field, Cognitive Behaviour Therapy (CBT) has grown to be the most common approach for anger management problems (Beck & Fernandez, 1998) and has been shown to be effective for both adults and children. CBT is a time-limited intervention which teaches the individual a set of skills to regulate their own emotions and behaviour. A meta-analysis of 40 studies revealed a medium effect size for CBT targeting anger-related problems in YP without ASD (Cohen's d=0.67; Sukhodolsky, Kassinove & Gorman, 2004).

Central to the CBT model is the idea that anger is produced by the appraisal and meaning given to events rather than objective properties of the events. Anger has been conceptualised as a human emotion that is a product of cognitive processing, physiological arousal and behavioural reactions, all of which are linked through reciprocal connectedness (Novaco, 1975). CBT involves learning new ways of responding to cues that would have previously

evoked anger and aggression. Core components of CBT programmes designed to reduce anger include education about anger within a cognitive behavioural model, self-monitoring of anger episodes and situational triggers, teaching of behavioural techniques (e.g., relaxation), cognitive restructuring techniques and in some programmes other skills training such as problem-solving and assertive communication as well as an emphasis on practicing these skills (Taylor & Novaco, 2005; Deffenbacher, Dahlen, Lynch, Morris, and Gowensmith, 2000). A meta-analysis of 40 studies on CBT revealed that practices of CBT varied on a scale from "less behavioural" (affective education and problem solving) to "more behavioural" (eclectic treatments and skills development) (Sukhodolsky, Kassinove & Gorman, 2004). This study also identified a number of therapeutic techniques used in CBT including instruction, discussion, modelling, role-play, feedback, emotion identification, relaxation, self-instruction, exposure, homework and reinforcement.

1.5. CBT interventions applied in the ASD population

A growing body of research is being published on the application of modified CBT interventions to individuals with ASD and these studies have shown promising results (Sofronoff et al., 2005; Chalfant, Rapee & Carroll, 2007; Keehn, Lincoln, Brown, & Chavira, 2013; Wood et al., 2009). The primary modifications made to CBT when delivered to children with ASD include greater use of concrete and visual methods (e.g., emotion statements, pictures, visual worksheets, social stories, and role-play), incorporating the young person's special interests and involving a parent or carer (Moree & Davis, 2010). A recent meta-analysis included 14 such studies and reported significant and moderate treatment effects for reducing anxiety symptoms using modified CBT in this population (Ung, Selles, Small & Storch, 2015).

At present, CBT research for the ASD population has focused on anxiety management. However, these findings demonstrate the potential effectiveness of using CBT with this population for anger management and associated aggression. There are a number of mechanisms through which CBT could be a particularly effective treatment for children with ASD and such difficulties. Firstly, individuals with ASD have deficits in theory of mind skills (Baron-Cohen, 1995). They find it difficult to understand the thoughts and feelings of other people and themselves and may often make incorrect assumptions about other's intentions, leading them to feel angry. Furthermore, recent research on mechanisms underlying maladaptive behaviour in young people with ASD has identified that in comparison to their typically developing peers, they are less likely to use cognitive reappraisal as an emotion regulation strategy (Samson, Hardan, Lee, Phillips & Gross, 2015). CBT specifically focuses on directly teaching and practicing skills to identify thoughts and consider alternative ways of thinking as an emotional regulation strategy. In light of the aforementioned reasons, this may be an effective treatment for excessive anger in children with ASD. Secondly, individuals with ASD show weaker executive functioning skills (Hughes, Russell & Robbins, 1994), and have difficulty overriding a pre-potent response (Russell, Mauthner, Sharpe & Tidswell, 1991). This makes them vulnerable to acting quickly and impulsively 'without thinking'. CBT provides a structured way of identifying common triggers, concrete ways of identifying anger and a set of concrete skills in order for them to use to regulate their emotions. Thirdly, individuals with ASD show significantly more difficulties in identifying, processing and describing their own emotions (Hill, Bertoz & Frith, 2004). Such difficulties may lead them to physical expressions of anger to express mood and release emotional energy. CBT includes an affective education component helps children identify, label and understand their emotions. Additionally, CBT is a structured, concrete and goal focused form of

71

therapy which can be argued to involve 'logical thinking' and is thus likely to suit individuals with ASD more than more open-ended, abstract forms of psychotherapy.

There has been one randomized control study evaluating a group CBT intervention specifically for anger-related difficulties for ASD with Asperger's Syndrome (Sofronoff, Attwood, Hinton & Levin, 2007). This intervention was highly structured and delivered in a group format. The authors reported significant reductions in anger episodes in the participants following the intervention and these were maintained at six week follow-up. A non-randomised study has been conducted on a group CBT programme for young children (5-7 year olds) with ASD targeting anxiety and anger (Scarpa and Reyes, 2011). This study found that following the programme, children showed reductions in the frequency of anger outbursts although no significant improvement on an emotional regulation outcome measure.

Both these interventions have investigated the effect of CBT delivered in a group format. Children with ASD are a population with great phenotypic heterogeneity and varying needs. Individual treatment can offer greater flexibility and treatment can be customised to meet each child's level of cognitive and communications skills. They may result in improved outcomes. Secondly, some children may show aggressive and disruptive behaviour that excludes them from safely participating in a group programme. These children may respond better through individual CBT where they receive 1:1 attention from a clinician who can adapt flexibly to their individual needs. Thirdly, although in the above studies the programme content was shared with parents in a separate session, they were not involved in sessions with their child. A number of factors suggest that parental involvement in the session may improve treatment outcome. Firstly, parental involvement has been highlighted as an important component for this population. It aids generalization of CBT

skills (Reaven &Blakely-Smith, 2013) and has resulted in significantly better outcomes (Sofronoff, Attwood & Hinton, 2005). Secondly, research has shown a bi-directional relationship between parental stress and child behaviour problems (Zaidman-Zait et al, 2014). Thirdly, clinicians in this field report observations that many parents of children with ASD have difficulties communicating emotion (Attwood & Scarpa, 2013). These authors suggest that parental involvement may 'encourage solutions to problems experienced by other family members that have a positive influence on the emotional atmosphere at home, and consequently the emotional equilibrium of the child or adolescent with ASD' (Attwood & Scarpa, 2013, p. 41).

For these reasons, it is important to investigate the potential efficacy of an individually administered CBT intervention involving the parent-child system in sessions. To the author's knowledge there has been no research on such a programme for YP with ASD showing anger and aggression.

1.6. Aims and objectives of this study

This study aims to investigate a CBT intervention which was developed at a UK National Clinic for High Functioning ASD in line with the principles outlined above for anger-related problems in children diagnosed with ASD. RCTs are the 'gold standard' method of carrying out scientific research on the efficacy of an intervention. However, they are both costly and time-consuming. It has been recommended that they 'should only be used when there is prior evidence that the experimental treatment is beneficial' (Barker & Pistrang, 2002, p. 157). Furthermore, the Medical Research Council framework for evaluating complex interventions describes a phase of running exploratory or pilot trials before conducing an RCT (Campbell, Murray, Darbyshire, Emery, Farmer, Grifiths...Kinmonth, 2007).

Given that there is little research in this area, this study employs a small-N research design to investigate the feasibility, acceptability and an initial estimate of efficacy of this intervention in order to inform the feasibility of a larger scale randomised trial. The main objectives of this study are to investigate the following:

(1) the acceptability of this anger management intervention to families

(2) the feasibility of the intervention and the measures used to evaluate it

(3) whether the intervention is associated with a reduction in the frequency of anger outbursts in children with ASD

(4) whether the intervention is associated with a reduction in levels of parenting stress

2. Methods

2.1. Design

In order to evaluate the feasibility, acceptability and obtain an initial estimate of efficacy of the intervention, this research employed a mixed-method, small-N design consisting of a series of multiple systematic case studies.

2.1.1 Background to the single case experimental design

Single case experimental designs (SCED) originate from the single case study approach but have undergone significant advancements in methodology and analysis (c.f. Smith, 2012). The Medical Research Council guidance describes 'N-of-1' designs as a valid type of experimental design for evaluating complex interventions (Craig, Dieppe, Macintyre, Michie, Nazareth, & Petticrew, 2008). Furthermore, SCEDs have been argued to play a pivotal role in the initial stages of intervention development. They can investigate feasibility and acceptability, contribute to the development of treatment manuals, pilot test measures and collect preliminary evidence that

the treatment can be beneficial before investing in a larger scale evaluation (Barlow, Nock & Hersen, 2009, p. 28-29). They can also function to build evidence of effectiveness and inform trial design (Craig, Dieppe, Macintyre, Michie, Nazareth & Petticrew, 2008, p. 4). There are a number of advantages for the use of a SCED for the aims of this research. Firstly, SCEDs hold the advantage of staying sensitive to individual variation between and within individuals which Randomized Control Trials (RCTs) are unable to do as they estimate the average effect of a large group. Secondly, they do not face the practical problems of collecting large numbers of participants and are comparatively less costly than RCTs. Thirdly, SCEDs are not limited by ethical issues of withholding treatment to a control group.

2.1.2. Increasing internal validity

This study employed a replicated single case AB design where repeated outcome measures were used and participants acted as their own controls. In such designs, a stable behavioural baseline (A) followed by a change in behaviour that coincides with the start of an intervention period (B) can provide support for the causal inference that the treatment programme is responsible for behavioural change (Perone & Hursh, 2013). The influence of other factors must be ruled out in order to increase the validity of such claims. Various design features were adopted to address threats to validity as recommended by Kazdin (1982) and Barker, Pistrang & Elliot (2002). These are outlined below:

Baseline period: Prior to starting the treatment programme, each child's parent completed a baseline assessment phase where they recorded the daily frequency of anger outbursts (FAO). They completed the measure daily for a week at two time points (4 weeks and then 2 weeks before the first session), resulting in 14 baseline data points.

This phase was designed to inform the potential impact of threats to internal validity (e.g., time based symptom fluctuation, repeated exposure to the measurement, spontaneous improvement). Data from this phase aimed to provide a predictive function by estimating the pattern of behaviour (by trend of the data points) before participants commenced the treatment programme.

Frequency and type of outcome measures: Multiple assessments of change over time were administered (see Figure 1). Outcome measures were chosen that were quantitative and objective (i.e. frequency of anger outbursts). Both standardized questionnaires and individuals outcome measures were used.

Monitoring extraneous variables: Information on changes in medication and participation in other therapeutic interventions was collected to rule out as many alternative explanations as possible.

Multiple cases: Multiple cases were recruited to bolster the validity of improvement seen in any individual case and reduce attribution to extraneous events (Kazdin, 1992).

Qualitative data: A qualitative approach was employed to supplement the findings yielded from the quantitative measures, to explore processes which may be responsible for any change and to rule out alternative explanations for any change that occurred. Qualitative methods have the potential to be more sensitive to establishing effects, collect information about events in therapy deemed important to the change process and systematically search for evidence of alternative explanations (non-therapy processes deemed to bring about change) (Elliott, 2002).

2.2. Participants

Seven young people diagnosed with ASD and aged between 9 and 13 years old and their parent(s) completed the study. One family dropped out before the first session due to child illness. All children included in the study had received a diagnosis of AN ASD from a UK National Clinic for High Functioning Autism. Diagnosis was established through multidisciplinary clinical consensus with the aid of gold standard standardized diagnostic tools: The Autism Diagnostic Observation Schedule 2 (ADOS; Lord et al., 2000) and the developmental, dimensional and diagnostic interview (3Di; Skuse et al., 2004). Families were eligible for the study if they met the following inclusion criteria: (1) child was aged 8-14 years and received a clinical consensus primary diagnosis of ASD, (2) child was identified by clinicians at the clinic as presenting with significant anger management problems (3) child scored 'definitely present' for more than three items on a parent-report conduct disorder questionnaire and where 'loses temper' was one of them (Appendix 2), (4) families were verbally fluent in English, (5) family were able/committed to attend ten sessions of therapy. Participants were excluded from the study if they met any of the following exclusion criteria; (1) taking psychotropic medication (unless they intended to stay on a stable dose of medication throughout the study), (2) taking part in another form of psychological therapy during the study, (3) subjection to a child protection plan. Participants were not excluded on the basis of secondary comorbidities such as ADHD, anxiety or depression.

2.3. Measures

2.3.1. Primary outcome measure

2.3.1.1. Frequency of Anger Outbursts (FAO). This is a parental monitoring measure of the daily frequency of anger outbursts

displayed by their child. Anger outbursts were operationalised as instances where the young person 'is unable to maintain emotional control and behaved or spoke inappropriately in anger' (as in Sofronoff, Attwood, Hinton & Levin, 2007). Parents were asked to complete this measure daily for two weeks before commencing the programme to obtain a baseline measure. They were then asked to complete the daily measure for a week, every alternate week for the duration of the programme and also at six week follow-up after the programme finished (see Figure 1). This measure can be found in Appendix 3.

2.3.2. Secondary Outcome Measures

2.3.2.1. *Strengths and Difficulties Questionnaire (SDQ,* Goodman, 2001). This is a parent-report questionnaire for 4-17 year olds consisting of a list of 25 positive and negative attributes. Parents are asked to use a 3-point Likert scale to indicate how far each attribute applies to their child. The questionnaire yields a total difficulties score, an impact score and five sub scores: emotional problems; conduct problems; hyperactivity; peer problems; prosocial skills. The questionnaire has demonstrated good reliability, as measured by internal consistency (Cronbach's Alpha, $\alpha = 0.82$) in a British sample of 5-15 years olds (Goodman, 2001). It also demonstrated validity as a dimensional measure of child mental health where the odds of disorder were demonstrated to increase at a constant rate across the range (odds ratios between 1.14 and 1.28 per one-point increase in SDQ score, Goodman & Goodman, 2009). In terms of criterion validity, the SDQ was highly correlated with the Child Behaviour Checklist and was similarly able to detect internalizing and externalizing problems (Goodman & Scott, 1999). Parents completed

78

this measure at the first, fifth and final session and at six week follow-up (see Figure 1).

2.3.2.2. *The Parenting Stress Index– Short Form* (PSI-SF, Abidin, 1995). This is a 36-item questionnaire for parents which yields a total stress score and three subscales: parental distress, parent-child dysfunction and difficult child. The questionnaire has demonstrated good to excellent reliability for 184 parents of children with ASD as measured by internal consistency where Cronbach's alpha was between 0.88 and 0.95 for

all scales and subscales (Dardas & Ahmad, 2014). In this study, parents completed the PSI-SF at the first, fifth and final session and at six-week follow-up, Figure 1)*.*

2.3.2.3. *Goal Based Outcomes* (GBO). This is an idiographic measure of change designed for young people in a Child and Adolescent Health setting (www.corc.uk.net). The GBO form uses a 0-10 point scale to capture the progress made towards client-identified goal where 0 represents the goal is not at all met, 5 is half way to reaching the goal and 10 denotes the goal has been reached. The GBO has been demonstrated to capture higher order and underlying factors that cannot be captured by normed outcome measures (e.g., confidence, resilience and coping) (Jacob, Edbrooke-Childs, Holley, Law & Wolpert, 2015). Families were encouraged to identify up to three goals at the start of the programme (both child and parent were encouraged to identify a personal goal as well as a joint goal). The progress towards each goal was re-rated by families at the first, fifth and final session and at six-week follow-up (see Figure 1).

2.3.2.4. *Post-Intervention Interview.* Parents took part in a telephone interview 4-8 weeks after they finished the programme. The interview

was semi-structured and aimed to capture parental views about the usefulness and acceptability of the programme and information about change (Appendix 4). It included questions from the Change Interview (Elliott, Slatick & Urman, 2001), about any changes they had noticed since the programme started and attributions of any changes, what they found helpful, missing and disappointing about the programme. It also included questions around the feasibility of the outcome measures used.

2.3.2.5. *Treatment fidelity checklist.* Therapists completed a treatment fidelity checklist for each family (Appendix 10). Therapists marked the number of core components of the programme which were administered in the allocated sessions. They also documented factors which interfered with the delivery of the intervention and any adaptations that were made to the manual.

2.4. Procedure

This study was granted ethical approval by Egbaston NHS Research Ethics Committee (15/WM/0140, see Appendix 6). The research took place in a UK National Clinic for High Functioning Autism. The study flow is outlined in Figure 2. Written informed consent was obtained from parents and assent from children in the first session. Parents and young people completed the goal based outcomes with their therapist in sessions. The post-intervention interview was conducted by a specially-trained Assistant Psychologist who was not involved in any other part of the treatment programme or research. The interview was audio-recorded and then transcribed verbatim.

```
┌─────────────────────────────────────────────────────────────┐
│  Families were routinely seen for an assessment of ASD at     │
│  the clinic                                                   │
└─────────────────────────────────────────────────────────────┘
                              ↓
┌─────────────────────────────────────────────────────────────┐
│  Clinicians identified children presenting with anger         │
│  outbursts, briefly explained the intervention programme      │
│  and gained consent to be contacted by the researcher         │
└─────────────────────────────────────────────────────────────┘
                              ↓
┌─────────────────────────────────────────────────────────────┐
│  Researcher contacted family, screened for eligibility,       │
│  explained the study. Researcher sent Participant             │
│  Information Sheet and example consent form to eligible        │
│  families                                                     │
└─────────────────────────────────────────────────────────────┘
                              ↓
┌─────────────────────────────────────────────────────────────┐
│  Families agreeing to participate were given appointment      │
│  times and baseline measures were explained over the phone    │
│  and posted out                                               │
└─────────────────────────────────────────────────────────────┘
                              ↓
```

Baseline

┌───┐
│ **4 weeks before first session**   │
│ Parents completed baseline measures (FAO) for a week   │
└───┘

┌───┐
│ **2 weeks before first session**   │
│ Parents completed baseline measures (FAO) for a week   │
└───┘

Intervention

┌───┐
│ INTERVENTION PROGRAMME   │
│   │
│ Session 1 (complete consent forms, GBO & return FAO)   │
│   │
│ Session 2 (return SDQ, PSI-SF)   │
│   │
│ Session 3 (return FAO)   │
│   │
│ Session 4   │
│   │
│ Session 5 (return FAO, SDQ, PSI-SF, complete GBO)   │
│   │
│ Session 6   │
│   │
│ Session 7 (return FAO)   │
│   │
│ Session 8   ⎫   │
│   ⎬ Final session   │
│ (Session 9) ⎪ (Return FAO, SDQ, PSI-SF, complete   │
│   ⎪   GBO).   │
│ (Session 10) ⎭   │
└───┘
                              ↓

Follow-up

┌───┐
│ **4 weeks post programme**   │
│ Families were sent follow-up questionnaires by post (FAO,   │
│ GBP, PSI-SF, SDQ)   │
└───┘
                              ↓
┌──────────────────────────────────────┐   ┌──────────────────────┐
│ **4-8 weeks post programme**   │ → │ Families were   │
│ Parents completed follow-up phone   │   │ reminded to send   │
│ interview   │   │ back follow-up   │
│   │   │ questionnaires   │
└──────────────────────────────────────┘   └──────────────────────┘

Figure 1. Participant flow through study including frequency of outcome measures returned by participants. FAO=frequency of anger outbursts, GBO= goal based outcomes, PSI= parenting stress index & SDQ=strength and difficulties questionnaire

2.5. Intervention programme

2.5.1. Development and details of the intervention

The programme was developed by the clinical team at a UK National Clinic for High Functioning Autism. It is based on CBT principles and draws on strategies outlined in CBT manuals for anger and aggression (Sukhodolsky & Scahill, 2012; Attwood, 2004). The intervention was individually administered to each family and revolved around bespoke CBT case conceptualisation. The programme was guided by an intervention manual (Appendix 10) and was split into six phases. Table 1 outlines the aim and content of each phase of the programme.

To increase accessibility for children with ASD the intervention had a number of adaptations from standard CBT for children. (1) There was extensive use of visual, rather than verbal, information; (2) a greater emphasis on emotion education and recognition of varying levels of anger; (3) a greater emphasis on behavioural over cognitive strategies; (4) simplification of cognitive activities where necessary (e.g., instead of producing alternative thoughts, replacing a distressing angry thought with a pre-prepared standard thought or affirmation); (4) planning scheduled breaks to maintain attention; (5) incorporating the child's interests to facilitate engagement, attention and understanding of programme material; (7) involving the parent(s) in the sessions to facilitate communication about solutions to the problem between the child and parent and to support the implementation of skills outside of therapy sessions.

Table 1. The main aims and content of each phase of the intervention programme

| Phase	Aim	Content
1	Engagement, Assessment and Motivation to change	Introductions, exploring hobbies/strengths, normalizing anger, generate examples of anger in the family, pros/cons of reactions to anger, goals for therapy, explain the programme
2	Affective Education & Socialisation to CBT model	Exploring anger using CBT model: effects on thinking, the body and behaviour. Introduce different intensities of emotion (emotion thermometer). Identify unique early warning signs of anger.
3	Introducing alternative ways of responding	Using comic strip conversations, explore alternative ways of responding: What would make me feel better?
4	Behavioural Techniques	Explore behavioural techniques to manage anger (physical/relaxation/special interests/social strategies)
5	Cognitive Techniques	Introduce cognitive restructuring. Link the alternative way of thinking with consequences using comic strip type conversations.
6	Generalising the learning and relapse prevention	Reviewing practice outside sessions and troubleshooting. Increasing accessibility of effective techniques for child.

2.5.2. Implementation of the intervention

The intervention programme was delivered by two therapists at the clinic. Five families were seen individually by the author (a trainee in the Clinical Psychology Doctorate programme). In two of these cases, this was joint work with the Principal Clinical Psychologist (PCP) at the clinic who has had over seven years of experience working with ASD. Two families were seen individually by the PCP. Both therapists completed treatment fidelity checklists (Appendix 5) for each family to indicate whether or not the core components of the manual were administered. The programme consisted of weekly eight sessions lasting 1-1.5 hours, with two additional 'spill over' sessions offered to families where the main elements of the protocol were not accomplished within eight sessions. Each session involved meeting with the child and parent(s) together as well as the child individually.

3. Results

3.1. Participant flow information

Figure 2 shows participant flow through the study. At least one parent attended all sessions of the programme for all seven families. Six out of seven young people attended all sessions. One young person (P5) did not attend the final four sessions of the programme, and the session content was completed with the mother.
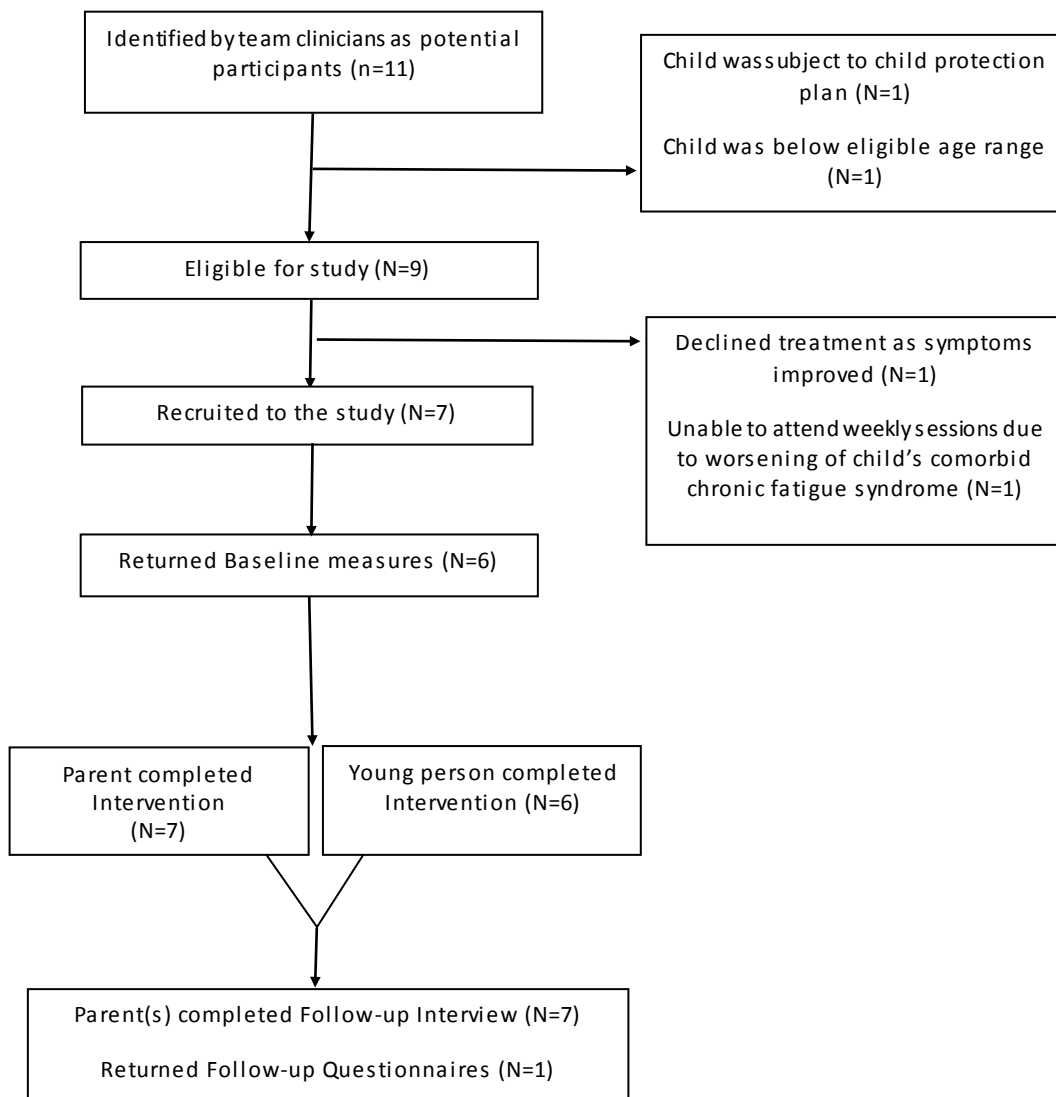
Figure 2 Participant flow through the study

3.2. Patient demographic data

The characteristic of participants is summarised in Table 2.

Table 2. Characteristics of participants

Gender	Age at start of programme	Estimated Cognitive Ability	Co-morbid diagnoses	Ethnicity
F	11 years 1 month	Overall IQ [a] =Average (132) *VCI= very superior (140)* *PRI= very superior (140)* *WMI= average (110)* *PSI= average (97)*	Auditory processing disorder	White British
M	13 years 7 months	Overall IQ=Borderline (74) *VCI= average (71)* *PRI= average (100)* *WMI= profound (59)* *PSI= low average (83)*	Generalized anxiety disorder	White/ Black Caribbean
M	9 years 3 months	Overall IQ=Average (99) *VCI= average (95)* *PRI= average (104)* *WMI= average (94)* *PSI= average (103)*	Attention deficit hyperactivity disorder	White British
F	9 years 4 months	Overall IQ=Average (98) *VCI= average (106)* *PRI= average (96)* *WMI= average (97)* *PSI= low average (88)*	None	White British
M	13 year 1 month	Cognitive assessment could not be completed due to difficulties with engagement	None	Black Caribbean
M	13 years 4 months	Overall IQ=Average (98) *VCI= low average (85)* *PRI= average (108)* *WMI= average (97)* *PSI= average (106)SI= average (66)*	None	White British
M	13 years 1 month	Overall IQ [a] =Average (103) *VCI= average (112)* *PRI= average (104)* *WMI= average (110)* *PSI= borderline (75)*	Neuro-muscular difficulties, hypermobility, dyspraxia, chronic tic disorder	White British

VCI= Verbal Comprehension Index, PRI= Perceptual Reasoning Index, WMI= Working Memory Index, PSI= Working Memory Index. Composite scores are reported in brackets.
[a] PRI and PSI scores were significantly discrepant but overall IQ was calculated to provide an indication of overall ability for research purposes

3.3. Treatment Fidelity

Treatment fidelity checklists indicated that at least 83.54% of items were administered for all families (range=64-100, SD=15.12). An average of 72% of items (range= 43-100, SD=20.86) were covered in the allocated session and the remainder could not be covered in the allocated session. This was due to additional factors such as the child's poor engagement in the programme, families not completing homework activity, the child's difficulty with

inattention/hyperactivity, needing more breaks, families arriving late to sessions and difficulty moving off topics that the child was fixated on and/or distressed about. For four young people, therapists outlined that engagement and motivation needed a longer period of time than was allocated in the treatment manual. Obstacles noted included the child not seeing anger as problematic (P5), being particularly reluctant to discuss 'anger outbursts' in sessions (P3), holding a fixed idea that others were at fault and they should change (P3), feeling a strong sense of injustice about events and that their anger/aggression was justified (P2) and holding a negative belief about the efficacy of treatment (P5 and P6). For P6, reinforcers helped to motivate the child to try the strategies at home. Of note is that, for P5, the manual had to be adjusted significantly (longer engagement phase and the final four sessions were delivered to the mother only) as the young person did not want to continue the intervention. For all participants, therapists noted that the YP required reminders by parents to use the strategies at home.

3.4. Anger Outbursts: Graphical Display

The data for the daily frequency of anger outbursts were visually analysed by constructing graphical displays using Excel™ and following the guidelines on visual analysis for single case data by Morley (2015). These graphs for each participant are shown in Figure 3. Data from P6 is not displayed due to missing data.

On visual inspection, for P1, P2 and P5, there appear to be higher levels of overall anger outbursts during the baseline phase compared to the treatment phase. P4 and P7 appear to show a reduction in anger outbursts during the treatment phase. However, the baseline phase shows that there was a lower level of anger outbursts before treatment compared to the start of treatment. P3

shows a marked decrease in anger outbursts during the baseline phase and a variable pattern during the treatment phase.

3.5. Anger Outbursts: Statistical Analysis

In addition to visual display, a Tau-U statistical analysis was performed on the frequency data to test for discontinuities in trend between the baseline and treatment phase for each participant. This is a non-parametric technique developed by Parker, Vannest, Davis, & Sauber (2011) and described in Morley (2015) as a technique to statistically analyse data in small-N designs. It was developed to be used when there is a trend in the slope of baseline data and is a test of 'dominance' which can be used to understand whether the slope of the treatment phase differs significantly from the slope of the baseline phase. Vannest and colleagues argue that Tau-U controls positive baseline trend better than regression-based approaches (Allison & Gorman, 1993). Tau-U is comprised of four indices which are reported in this paper: (a) Trend in A, which can be interpreted as the trend in the baseline phase, a negative value indicates a downwards slope (b) Trend in B, which can be interpreted as the trend in the treatment phase, a negative value indicates a downwards slope (c) A versus B, which can be interpreted as the tendency for scores in the treatment phase to be consistently different (lower or higher) to scores in the baseline phase, and (d) Tau-U which can be interpreted as the trend in the treatment phase, controlling for the trend in the baseline phase. This is designed to describe any symptom improvements during treatment that are above and beyond change that would be expected based on data from the baseline phase. Tau values and their respective significance values were calculated using an online calculator (Vannest, Parker, & Gonen, 2011). The data for P6 was not included in this analysis due to missing data.

3.5.1. Checking for baseline trend

As shown in table 3, only P3 showed a significant decline in anger outbursts over the baseline period (Tau= -.65, p=.001).

3.5.2. Baseline trend versus treatment trend

As shown in table 3, for all participants there was a negative, but non-significant, trend towards symptom reduction during the treatment phase. As shown in table 4, the difference in trend of anger outbursts between baseline and treatment phase was significant for P1 (Tau-U=-.68, p=.006) and P5 (Tau-U=-.45, p=.027). This indicates that for P1 and P5 the treatment phase was associated with a statistically significant reduction in anger outbursts compared to what would naturally occur if they were not attending the treatment programme. There was no significant difference between the baseline and treatment phase trends for P2 (Tau-U=-.09, p=.649), P3 (Tau-U=.02, p=.904), P4 (Tau-U=.24, p=.21) and P7 (Tau-U=-.01, p=.979). This indicates that for P2, P3, P4 and P7, there was no significant reduction in anger outbursts over and above the change that would naturally occur if they were not attending the treatment programme.
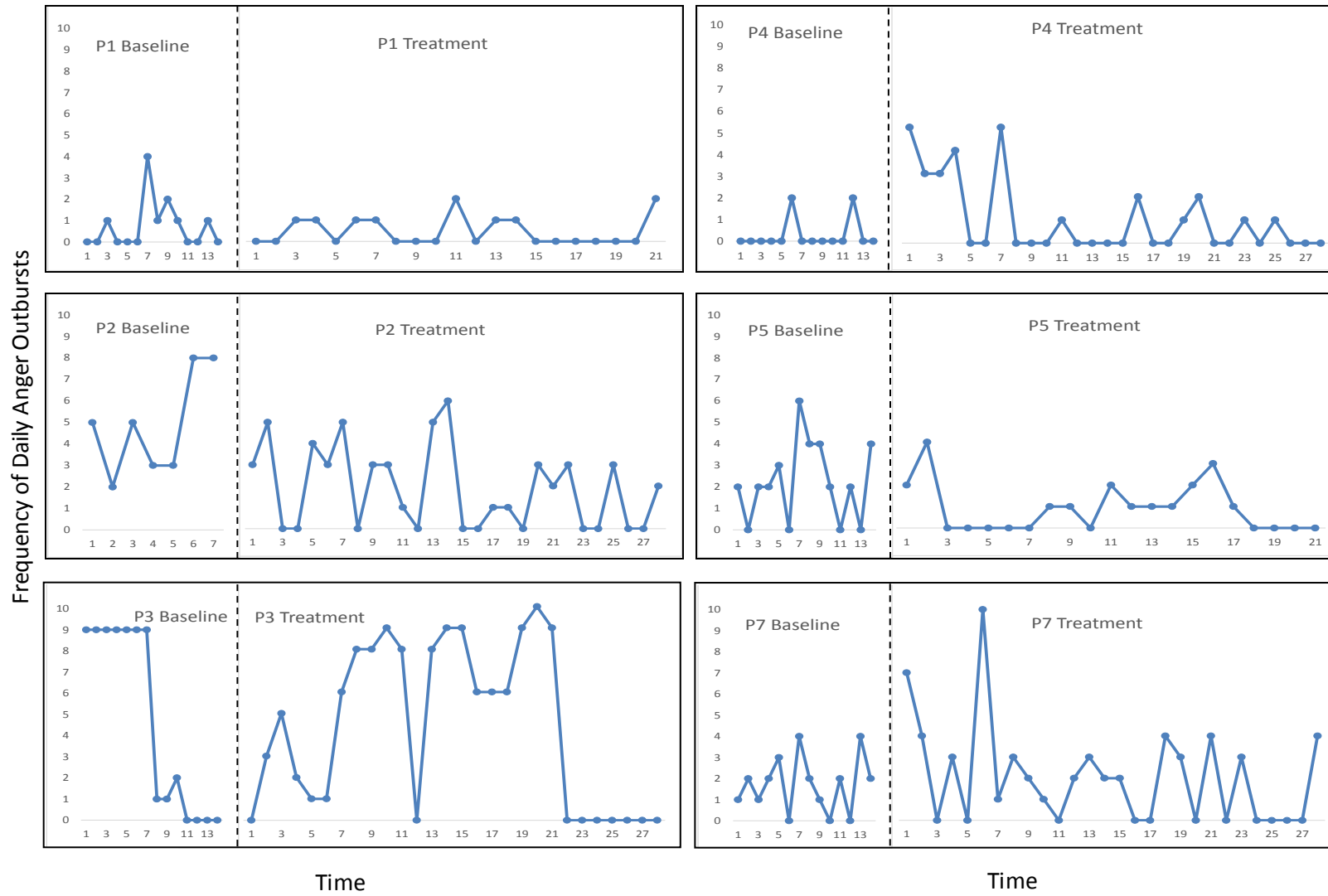
Figure 3 The frequency of parent-reported anger outbursts over the baseline phase and treatment phase per participant. Each data point marks the daily frequency of anger outbursts.

Table 3. The calculation of trend in the baseline phase (Trend$_A$) and the trend in the treatment phase (Trend$_B$). A negative Tau value indicates a downwards slope.

Participant number	Trend $_A$ (baseline phase)					Trend $_B$ (treatment phase)				
	S	Pairs	Tau	P-value	CI (90%)	S	Pairs	Tau	P-value	CI (90%)
1	8	21	.38	.230	-0.14 <> 0.90	-69	378	-.18	.173	-0.40<>0.04
2	7	91	.08	.702	-0.25 <>0.41	-10	210	-.05	.763	-0.31<>0.21
3	-59	91	-.65	**.001****	-0.98 <>-0.32	-31	378	-.08	.540	-0.30<>0.14
4	6	91	.07	.743	-0.26 <>0.40	-87	378	-.23	.086	-0.45<>-0.01
5	6	91	.07	.743	-0.26<>0.40	-11	210	-.05	.740	-0.31<>0.21
7	4	91	.04	.827	-0.29<>0.37	-72	378	-.19	.155	-0.41<>0.03

CI= confidence interval; ** $p < 0.01$

Table 4. The calculation of the A vs B comparison which indicates the tendency for scores in the treatment phase to be consistently different (lower or higher) to scores in the baseline phase, and the calculation of Tau-U which indicates the trend in the treatment phase, controlling for the trend in the baseline phase.

Participant number	A vs. B					Tau-U (A vs B − Trend in A)				
	S	Pairs	Tau	P-value	CI (90%)	S	Pairs	Tau	P-value	CI (90%)
1	-126	196	-.64	**.009**	-1.05<>-0.24	-134	196	-.68	**.006**	-1.09<>-0.28
2	-20	294	-.07	.736	-0.40<>0.26	-27	294	-.09	.649	-0.42<>0.24
3	-50	392	-.13	.505	-0.44<>0.19	9	392	.02	.904	-0.29<>0.34
4	100	392	.26	.182	-0.06<>0.57	94	392	.24	.21	-0.08<>0.55
5	-125	294	-.43	**.035***	0.76<>-0.09	-131	294	-.45	**.027***	-0.78<>-0.11
7	2	392	.01	.9787	-0.31<>0.32	-2	392	-.01	.979	-0.32<>0.31

CI= confidence interval; ** p < 0.01 * p < 0.05

3.6. Reliable Change on the SDQ and PSI-SF

Tables 5 and 6 show the pre and post scores for each participant for the SDQ and PSI-SF. The reliable change index (RCI, Jacobson and Truax, 1991) was used to calculate whether the difference between participants' pre and post treatment scores on the PSI-SF and SDQ showed reliable change beyond what would be expected from measurement error. The reliable change criterion for the SDQ and PSI-SF and their respective subscales can be found in Appendix 11. For an individual to have made a reliable change, their change score must be larger than the RCI value. Graphical displays were generated for each subscale using the Leeds Reliable Change Index Calculator (Morley & Dowzer, 2014). Figures 4a and 4b (SDQ) and Figure 5 (PSI-SF) depict the level of change that occurred for each participant from pre to post-treatment across each subscale of the questionnaires. The middle section within the red lines portrays no reliable change, the top left segment, beyond the red line depicts a reliable improvement, and the bottom right segment depicts reliable deterioration. Clinically significant change was also considered. For the SDQ, cut-off values were obtained from the SDQ manual which represent the threshold above which 10% of a UK population score. No normative values are available for the PSI-SF so it was not possible to derive a meaningful cut off point and conceptualise clinically significant change on this measure.

3.6.1.   SDQ

As shown in Figures 4a and 4b, no participants showed any reliable or clinically significant improvements on any subscales of the SDQ (including the total difficulties, behavioural difficulties, emotional symptoms, hyperactivity, peer problems, prosocial behaviour and total impact).

### 3.6.2.	PSI-SF

Of the six families who completed the PSI-SF, three reported reliable improvements (P1, P2, and P6) and one showed a reliable deterioration (P3) on the total stress scale. Similarly, three families reported reliable improvements (P1, P7, and P6) and one showed a reliable deterioration (P3) on the difficult child scale. One parent showed a reliable improvement on the parental distress scale (P6) and one family made a reliable deterioration (P7). On the parent-child dysfunctional interaction subscale, one family made a reliable improvement (P3). Level of change for each participants on the PSI-SF is shown in Figure 5.

3.7. Goal based outcomes

Each family rated their progress towards at least one personal goal that they set at the first session. Progress was rated again at the mid-point and end of the intervention. Five out of seven families rated that they had made progress (an increase of at least 2 points) on one of their goals. P3, P4 and P5 rated one of their goals as above eight at the end of the intervention. Data from the GBO is depicted in Figure 6.

Table 5 Summary of the pre-treatment and post-treatment scores and level of reliable change for each participant across the subscales of the SDQ

Measure	P1	P2	P3	P4	P6	P7
SDQ- Total Distress						
Pre-treatment	26	27	26	30	12	30
Post-treatment	27	31	30	27	10	25
RCI	*NC*	*NC*	*NC*	*NC*	*NC*	*NC*
SDQ - Total Impact						
Pre-treatment	7	9	10	8	4	7
Post-treatment	8	10	10	7	5	9
RCI	*NC*	*NC*	*NC*	*NC*	*NC*	*NC*
SDQ- Emotional Difficulties						
Pre-treatment	5	7	7	9	4	6
Post-treatment	7	9	8	9	2	9
RCI	*NC*	*NC*	*NC*	*NC*	*NC*	*NC*
SDQ- Behavioural Difficulties						
Pre-treatment	6	5	7	6	2	9
Post-treatment	7	5	9	4	2	7
RCI	*NC*	*NC*	*NC*	*NC*	*NC*	*NC*
SDQ- Hyperactivity						
Pre-treatment	9	7	10	8	6	10
Post-treatment	8	8	10	7	6	9
RCI	*NC*	*NC*	*NC*	*NC*	*NC*	*NC*
SDQ- Peer Problems						
Pre-treatment	6	8	2	7	0	5
Post-treatment	5	9	3	7	0	4
RCI	*NC*	*NC*	*NC*	*NC*	*NC*	*NC*
SDQ- Prosocial Behaviour						
Pre-treatment	4	4	3	5	5	3
Post-treatment	4	4	3	7	5	2
RCI	*NC*	*NC*	*NC*	*NC*	*NC*	*NC*

Note: RCI = Reliable Change Index; NC = No Change

Table 6 Summary of the pre-treatment and post-treatment scores and level of reliable change for each participant across the subscales of the PSI-SF

Measure	P1	P2	P3	P4	P6	P7
PSI- Total Stress						
Pre-treatment	125	130	92	108	121	109
Post-treatment	111	121	105	105	104	116
RCI	*RC***	*RC***	*D*	*NC*	*RC***	*NC*
PSI- Parent Distress						
Pre-treatment	37	42	26	29	31	28
Post-treatment	33	41	29	30	23	42
RCI	*NC*	*NC*	*NC*	*NC*	*RC***	*D*
PSI- Parent-Child Dysfunctional Interaction						
Pre-treatment	36	43	29	30	44	30
Post-treatment	34	37	21	28	42	24
RCI	*NC*	*NC*	*RC***	*NC*	*NC*	*NC*
PSI- Difficult Child						
Pre-treatment	52	45	38	49	46	51
Post-treatment	44	43	55	47	39	40
RCI	*RC***	*NC*	*D*	*NC*	*RC***	*RC***

Note: RCI = Reliable Change Index; RC** = Reliable Change; NC = No Change;
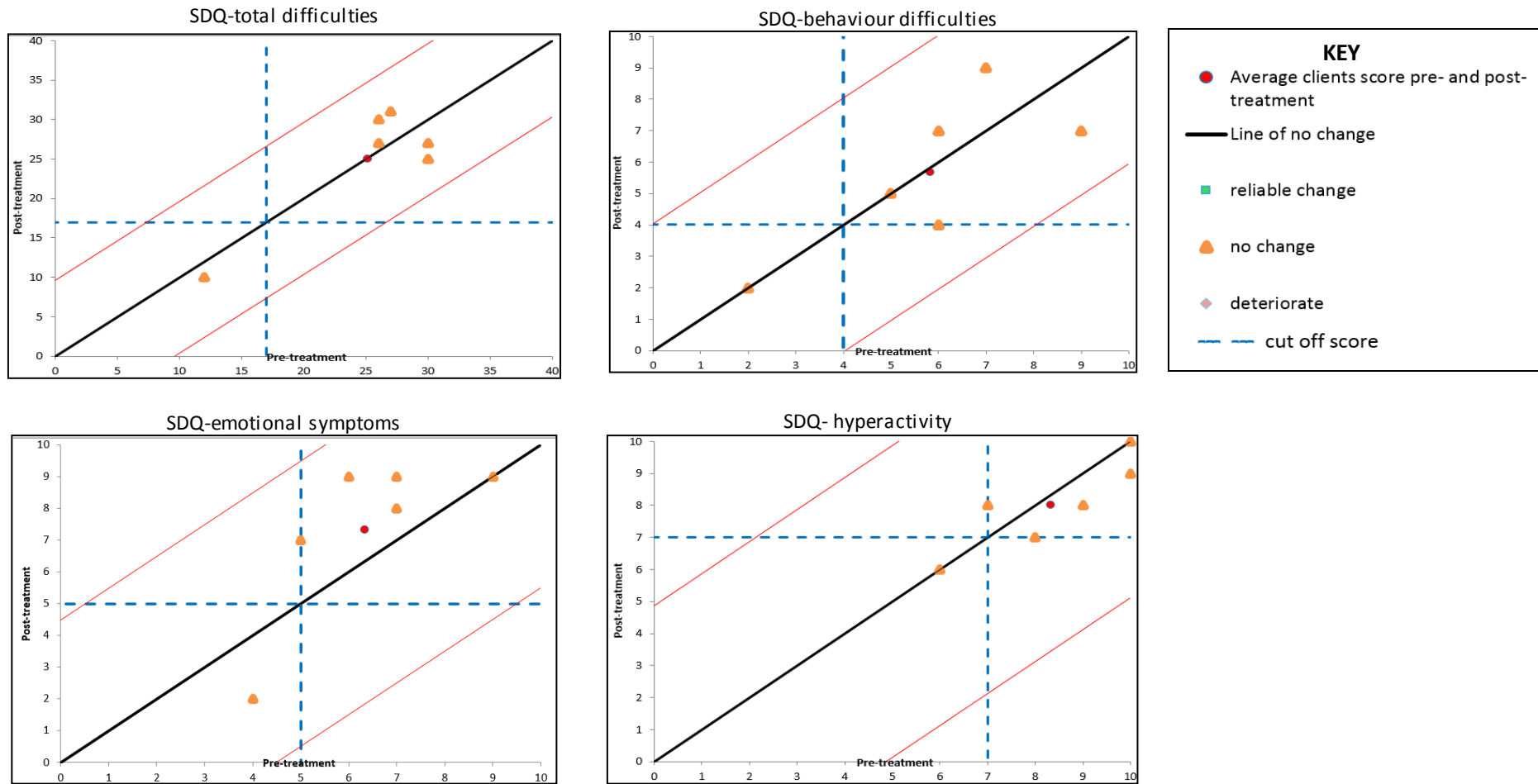D = Reliable Deterioration

Figure 4a The pre-treatment and post-treatment scores for each participant for the subscales of the Strength and Difficulties Questionnaire (SDQ)
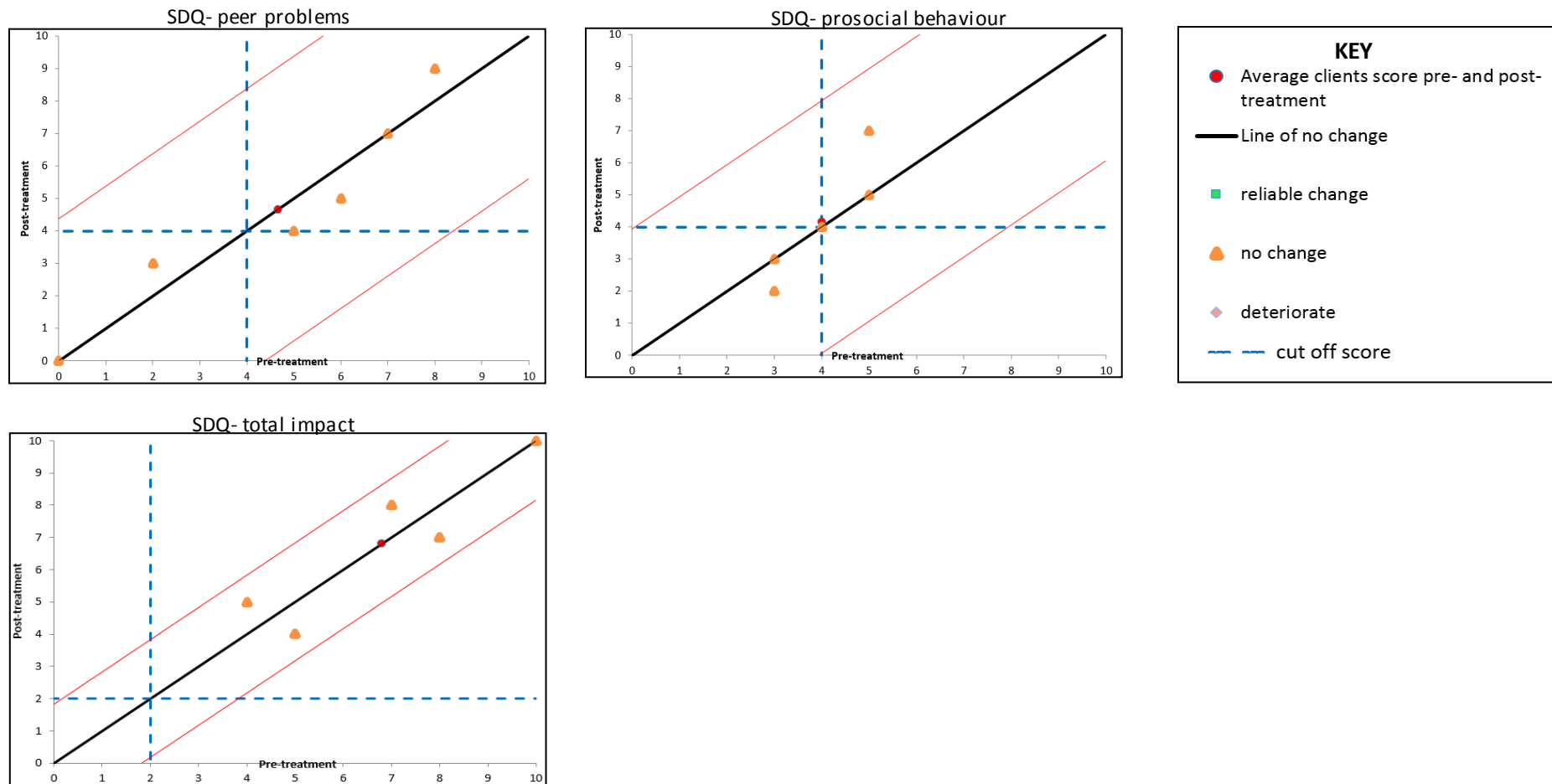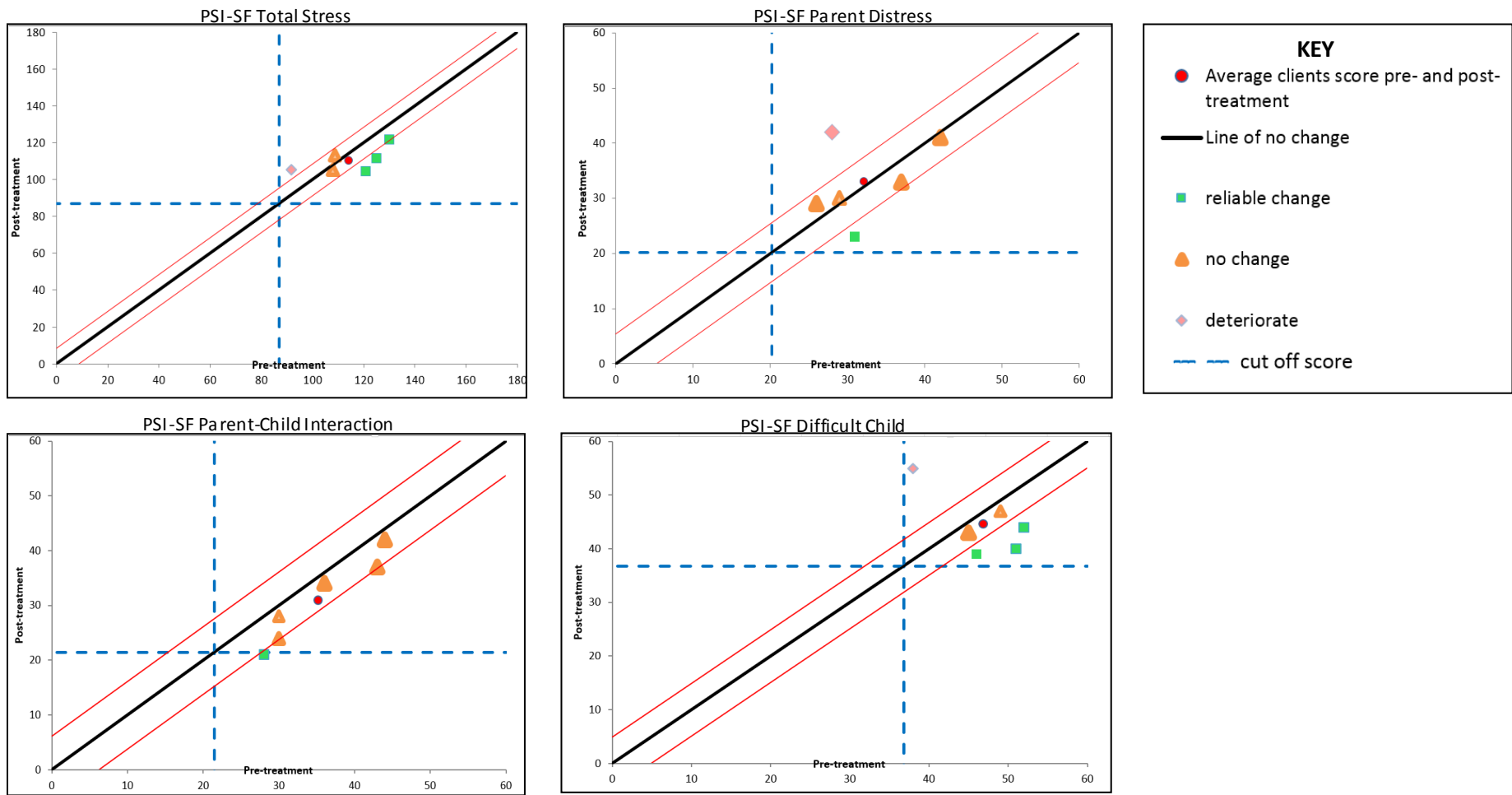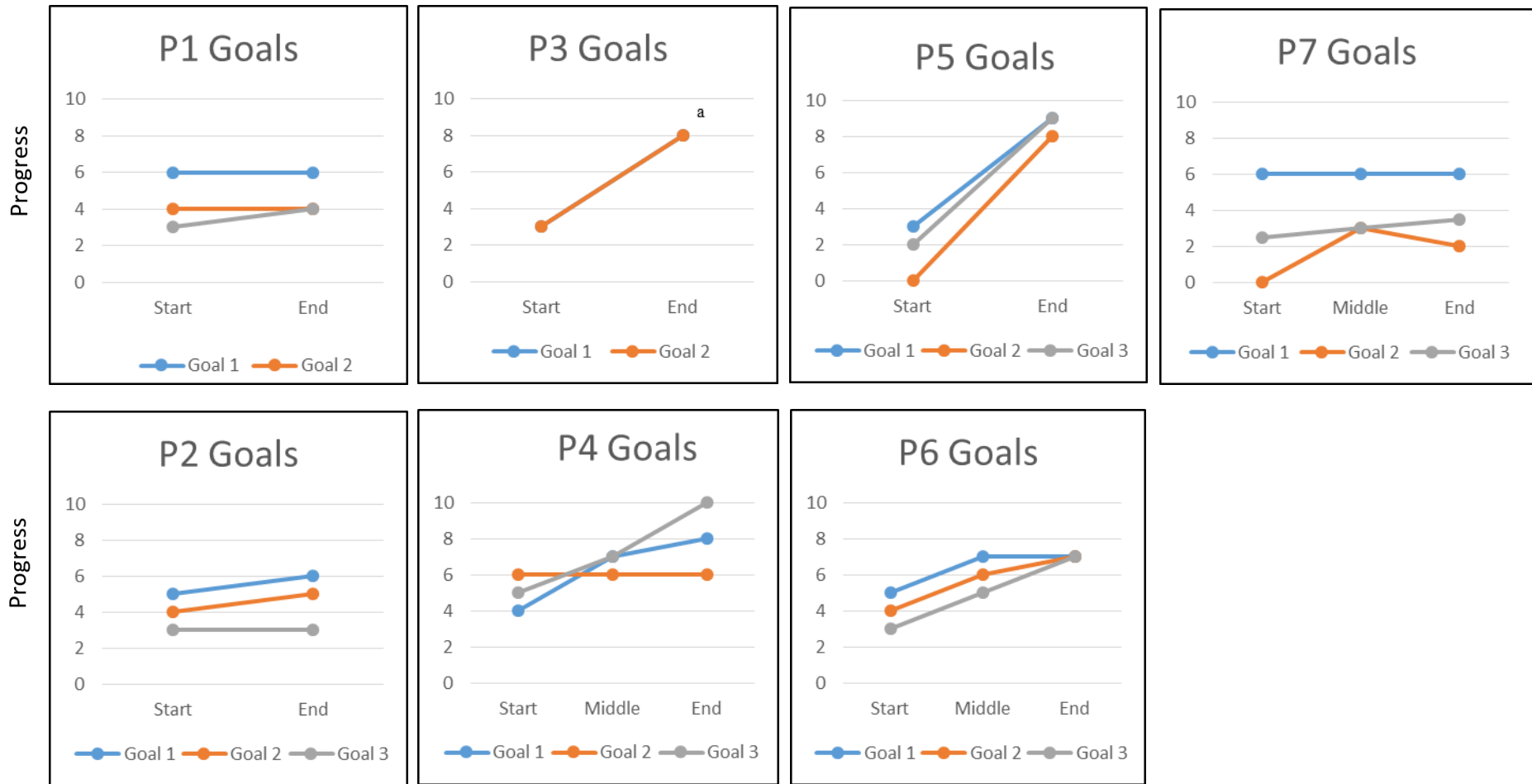
Figure 4b The pre-treatment and post-treatment scores for each participant for the subscales of the Strength and Difficulties Questionnaire (SDQ)
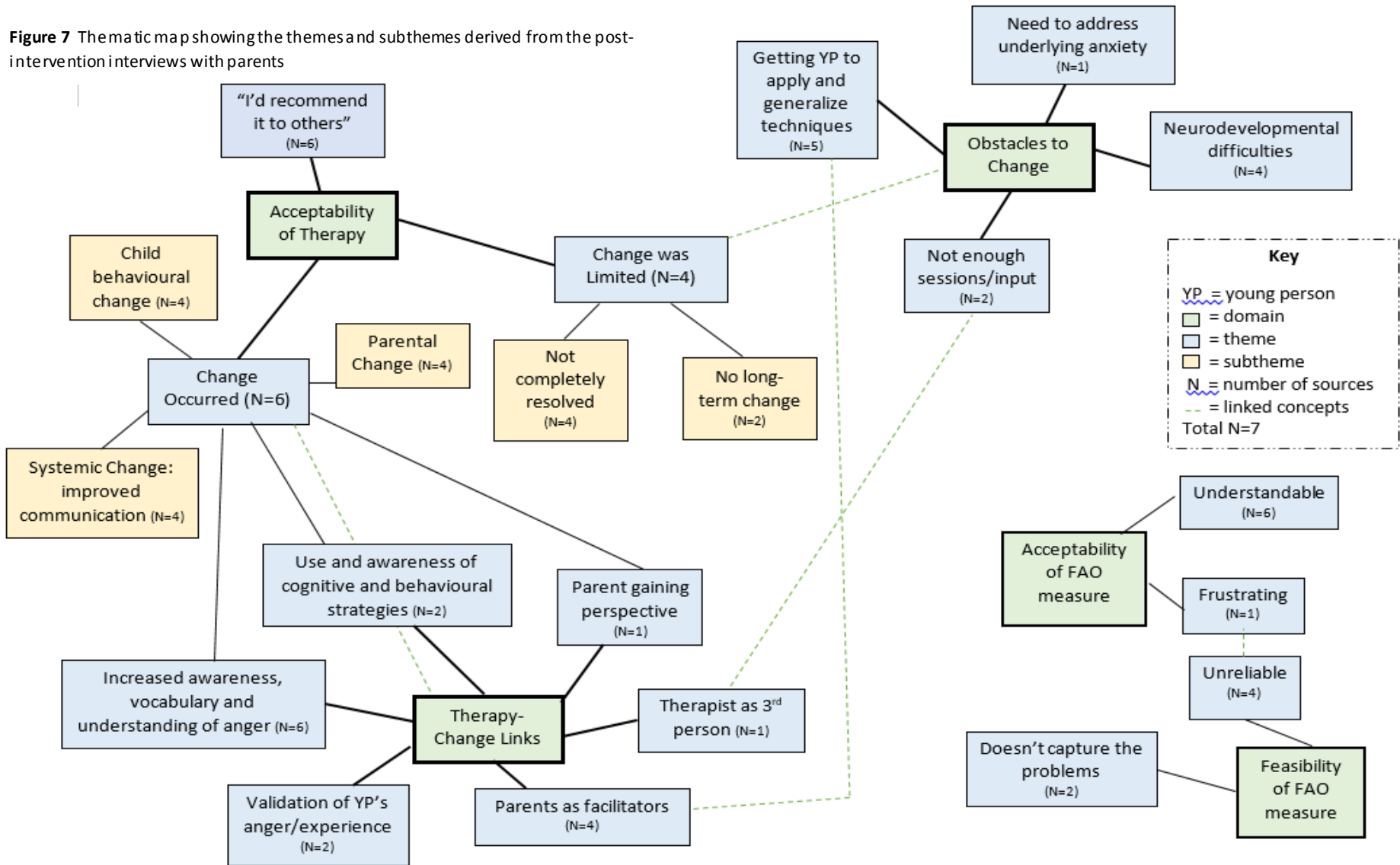
Figure 5. The pre-treatment and post-treatment scores for each participant for the four subscales of the Parenting Stress Index- Short Form (PSI-SF)

Figure 6 Graphs depicting each family's progress towards their personal goals as measured by the Goal Based Outcome (GBO) measure.
[a] P3's Goals 1 and 2 readings are the same

Figure 7 Thematic map showing the themes and subthemes derived from the post-intervention interviews with parents

Need to address underlying anxiety (N=1)

Getting YP to apply and generalize techniques (N=5)

Obstacles to Change

Neurodevelopmental difficulties (N=4)

"I'd recommend it to others" (N=6)

Acceptability of Therapy

Child behavioural change (N=4)

Change was Limited (N=4)

Not enough sessions/input (N=2)

Parental Change (N=4)

Not completely resolved (N=4)

No long-term change (N=2)

Change Occurred (N=6)

Systemic Change: improved communication (N=4)

Key

YP = young person

☐ = domain

☐ = theme

☐ = subtheme

N = number of sources

--- = linked concepts

Total N=7

Understandable (N=6)

Acceptability of FAO measure

Frustrating (N=1)

Use and awareness of cognitive and behavioural strategies (N=2)

Parent gaining perspective (N=1)

Unreliable (N=4)

Increased awareness, vocabulary and understanding of anger (N=6)

Therapy-Change Links

Therapist as 3rd person (N=1)

Doesn't capture the problems (N=2)

Feasibility of FAO measure

Validation of YP's anger/experience (N=2)

Parents as facilitators (N=4)

100

3.8 Post-intervention interviews: Thematic analysis

All interviews were transcribed verbatim. The data was analysed using thematic analysis and followed the six phases outlined in Braun and Clarke (2006). The transcripts were re-read and initial ideas were noted. Next, the data was coded to identify extracts that were perceived to be related to the research questions about change, acceptability and feasibility of the intervention. Other extracts that appeared important were also coded. The computer software programme (Qualitative Data Analysis Miner version 4.1.31) was used to code the data. (See Appendix 12 for an annotated example). The author identified codes that appeared conceptually related and sorted these into potential themes and sub-themes. An initial stage of this process can be seen in Appendix 13. Themes and subthemes were represented in an initial thematic map and were reviewed and refined in relation to each other and the original data set until a final thematic map was constructed (Figure 7). Quotations from transcripts were selected to illustrate examples of each theme. A note regarding the author's background and theoretical orientation can be found in Appendix 14 to help readers interpret potential bias in interpretation (Elliot, Fischer & Rennie, 1999). A credibility check of the themes generated was completed by a Clinical Psychologist (who works in the field of Autism and uses a multi-systemic/biopsychosocial framework) who checked the original transcript data with the themes.

The following section describes the domains, themes and subthemes derived from the thematic analysis as shown in Figure 7.

3.8.1. Acceptability of Intervention

3.8.1.1. Good enough to recommend

As a measure of acceptability, parents were asked if they would recommend the programme to a friend if they were experiencing similar problems. All but one parent said that they would, and three

had already recommended it to someone, which suggests that the programme was acceptable to most parents. P3's parents said their recommendation would depend on the child because they would not recommend it for children with ADHD. Reasons for this are further described in the 'obstacles to change' section.

3.8.1.2. Change occurred

During the interviews, all families (N=6) reported some positive change since starting the programme. Three main levels of change were identified: change at the level of a) the child, b) the parent and c) at a systemic level in terms of improved communication and interactions between parents and child.

3.8.1.2.1. Change at the level of the child

As well as improvements in awareness, understanding and vocabulary of anger (further described in section 3.1.3.2), many parents noted some improvements in anger outbursts (N=4). These varied in terms of whether an improvement in frequency, intensity or duration was reported.

"I'd say tantrums don't go on as long, there's probably less of them." - P6

3.8.1.2.2. Systemic change: communication and interactions

The majority (N=5) of families talked about some type of improvement of the interactions and communication between family members.

P7 described her child beginning to communicate his feelings and seeing evidence that expressing himself resulted in positive change occurring. P6 described an improvement in communication between

family members as an alternative to members of the family 'losing their tempers'. P2 talked about their child being more able to talk to their parents about incidents that made him feel angry.

"He gets distressed but it's easier to calm him down and it's easier to talk with him, talk it through. And he seems to be finding it easier to come and talk it through rather than getting so angry." – P2

P4 also mentioned an improved ability for her and her daughter to 'talk things through' in the programme, although this did not continue once the programme ended. P5 described a different type of change in the interactions whereby the parent was better able to recognise when to stop communicating with her son at times when this was distressing him.

"From coming to that programme, I've learnt about watching the signs before he gets upset, before he gets frustrated and before he gets angry and it's the signs I've learnt about within your therapy, that make, you know a change to my son because rather than I'm gonna carry on talk to him while he gets upset and carry on with it when I realise that okay, I see that he's getting annoyed, I see the sign now I need to stop talk and that's the changes that um, I learned from you know, from your surgery." - P5

3.8.1.2.3.   *Change at the level of the parent*

Some parents described changes they noticed in themselves after starting the programme including increased levels of confidence, coping and feeling calmer. P4 talked about a short-term improvement in her perceptions of coping that she linked to the improvements she was seeing in her daughter's behaviour during the programme. P2

described changes in her confidence to deal with her child's anger outbursts.

"I think it sort of helped in terms of teaching me maybe more of a way of what - how to sort of go about things with him and discuss things and situations if they arise. Um…so it kind of in a way has given me a bit of _confidence_." – P2

P5 reported reductions in her own levels of frustration when dealing with the problem and described feeling calmer since starting the programme.

"Yes, there is um, change with me because um_, I used to get frustrated_ as well, with _dealing with all these issues. But I realised that I'm a bit calmer_ and I get to understand how to deal with this problem and _the fact that I understand his problem make me a bit very calm you know? I'm okay with myself now, yeah." - P5

3.8.1.3.   Change was limited

Although most parents mentioned some positive changes, some parents mentioned that the change did not last after they finished the programme and some mentioned that the problem was not totally resolved.

3.8.1.3.1.   No long term change

P3 reported that the small changes they noticed were not a result of the programme.

"Part of it to a degree is growing up over the course of the few months we were going to the appointments, and we did talk about it but I wouldn't say the positives were so significant that it would not have happened anyway if that makes sense…because of his inattention in the sessions, we don't really feel we achieved much of it." – P3

A couple of parents reported the changes were short-lived and not sustained after the programme ended.

"*We've not had sort of a like a major transformation of everything suddenly being easier, um we did experience, um, some improvement that tended to be fairly temporary…. She did seem to occasionally be able to use the techniques and manage her anger better. Um, but to be honest it hasn't really translated into a permanent change.*" - P1

3.8.1.3.2.   *Not completely resolved*

Some families also mentioned that whilst there are positive changes, the problems were not 'completely' resolved.

"*yeah I have noticed, he is, he is calmer, all round I'd say he is a bit calmer, however, there is still issues to do with um, his anger. It's not completely resolved…he's calmer in terms of, he hasn't been hitting himself and he hasn't been getting so so distressed like he was before.*" – P2

3.8.2. Obstacles to change

During the interviews, the parents mentioned some difficulties, obstacles and processes which made it difficult for the programme to be effective in reducing the number of anger outbursts. These included the child's ability and motivation to generalize and apply the techniques outside of the sessions, ASD traits such as inflexible thinking and theory of mind and inattention in sessions. A couple of families also said that the number of sessions were not enough and

their child needed on-going or longer input and one family commented that their child's anxieties needed to be addressed.

3.8.2.1. *YP applying and generalizing techniques*

Many parents reported that it was difficult for their child to independently apply the skills and techniques of the programme in everyday life. For example, P4 reported that her daughter showed a clear awareness of the theory and the strategies she could adopt in various situations but did not apply them in practice.

"She will just by rote say perhaps I could do this and she will go through the, she will talk through the strategies suggested like the stress ball or you know perhaps I could think of a happy place or something. But when push comes to shove, she doesn't use them…But she knows they are there. She knows they're in the bag, she knows they're available to there. It was again when [therapist] was around and we were talking about what she could do. Yes she knows, we wrote the things that could help her. But actually putting them into practice, she doesn't." - P4

Some parents reported constantly prompting their child to use the ideas outside of sessions.

"He still needed an awful lot of parental guidance to be able to do it, he wasn't so great at initiating the ideas by himself, but he was at least willing to try a different approach with that parental guidance" - P7

However, for some parents, taking on this 'co-therapist' role with their child outside of sessions was problematic. For example, P3 explained

that bringing up strategies from the 'toolbox' as a way of managing his emotions appeared to make their son feel more angry.

"we try and use the tool box, but he is not keen on that one to be honest with you…um he relates it to being different that's why, he is fully aware that he is different from other regular kids if you like, so by bringing up stuff like that to do with the hospital makes him angry because he is different." - P3

P2 described not knowing how to help her daughter apply the techniques herself.

"(young person) had great difficulty engaging with the techniques...I had absolutely no idea how I could get her to engage with any techniques, or what the solution to that would be." – P1

3.8.2.2. Neurodevelopmental difficulties as an obstacle

Many parents spoke about their child's autistic characteristics as an obstacle to progress. For example, a tendency to show fixed ways of thinking, difficulties with theory of mind and a tendency to want to follow their own agenda. P1 discussed the difficulty of trying to get her son to 'shift' his thinking and consider another perspective as he could get very 'fixed on an idea'.

"it's part of the Asperger's with his literal thinking, very black and white, and it's hard to sort of get the idea shift, you know the- how he views something and his perception, he's very, very [inaudible] that that is the way it is and that is really quite hard to shift- to try and help him shift that way of thinking. That skill like um, a big um… sort of thing that I'm trying to deal with, with him is when he's got a- fixated

on an idea, he finds it hard to sort of take on board what you might be saying." - P2

P3 and P2 both described a tendency for their child to show resistance to external direction as an obstacle to change and engagement with the programme/techniques. For example, P3 described their son as following his own agenda in sessions and did not engage with the material of the session.

"And we found that [young person] controlled the session, that's the best way of putting it... He talked about what he wanted to do rather than doing what was asked of him." – P3

Similarly P2 described being 'directed from the outside' as being a problem for her child.

"I think that is generally speaking, a problem for her, that she doesn't engage with things, you know, even when she's got the best, um, she's so sort of um, difficult to direct from outside and I think that's part of her disability." - P1

P6 described his son's difficulties with theory of mind as an obstacle to thinking about how he comes across to others. P3 had comorbid ADHD and his parents reported that his difficulties sustaining attention in the sessions was a major obstacle to making progress on the programme.

"I don't think it helped [young person] massively because of his inattention, it was difficult for him to stay focused on any topic…because of his inattention in the sessions, we don't really feel we achieved much of it… now that he has got his ADHD medication,

and that's helping him quite a lot, it would probably be more helpful now that he is medicated." - P3

3.8.2.3. *Not enough sessions/input*

P1 and P4 both explained the short-term nature of the programme as a short-coming in addressing the problem. Both reported that on-going input for their child would be more helpful.

"it's a relatively small amount of sessions. Um, so no, there's only so much they can do, it can't be ongoing and I think, I think that's something you know that is a bigger issue, a wider issue that isn't reflective of the actual therapy itself but in terms of not having more of an ongoing thing, I think it's quite hard to sort of put things in place long term when it's only been a relatively short period that you've sort of started to look at it.. to put things in place I think it takes time to sort of establish- for them to be habits you know." - P2

P4 described the need for her daughter to have on-going input. She described how she was considering employing another person to continue aspects of the programme on an on-going basis.

3.8.2.4. *Underlying anxiety*

P7 commented that the programme was helpful to enable the child to express his emotions and as a result it was apparent that anxiety was underlying his anger outbursts. She described the need to address the underlying problem of anxiety.

"the anger is the result of the anxieties and the things that are going wrong, um, and having strategies to deal with the anger is great, but until you can deal with the anxiety and the cause of the anxiety, all you are really doing is firefighting at the end of the day. As I say it's very valuable then to actually understand it [child's anxieties], I think

it's actually really really important part of the process I think it can be very difficult when you've got a child that doesn't really know how to express their emotions to do that and to have that kind of expert opinion in there who can tease out what the child is trying to say is invaluable"-P7

3.8.3. Therapy-change links

Themes in this domain are focused on specific ideas and processes that parents mention as being helpful and suggest the processes by which change may have occurred for some families. Parents linked changes to therapy processes a number of times.

3.8.3.1. *Validation of young person's anger/experience*

Two parents (P1 and P4) spoke about the importance of having someone listen, acknowledge and validate their child's feelings and experiences and that this was an important part of the programme to help their child.

"… I think in the therapy as well he really started to feel heard a bit more…I think the acknowledgement for (patient) that you know was made in the therapy makes a difference to him…that's what calms him down I think, if you fight against it then it gets progressively worse." - P2

3.8.3.2. *Increased awareness, vocabulary and understanding of anger*
Parents mentioned that they or their child benefitted from learning a language to talk about anger (N=2) and an awareness of recognising early signs of anger (N=4).

"he's got more <u>vocabulary</u> to talk about it… he's been able to talk about his emotions more…we realised that (patient) didn't have much of a vocabulary to discuss it and now he does" - P6

"He is <u>aware of his anger</u>…He can <u>tell</u> us when he is getting angry. And he can <u>understand</u> a bit more about <u>why</u> he is angry." - P3

3.8.3.3. Use and awareness of cognitive and behavioural strategies

Two parents described the use or increased awareness of cognitive and behavioural strategies to use when the young person was feeling angry. P1 described the continued use of a cognitive strategy (statement to aid perspective-taking). P4 described the success of a behavioural strategy (listening to CDs and DVDs) when frustrated on public transport. P4 also described the young person's ability to describe cognitive (think of a happy place) and behavioural strategies (stress ball) that she could use when feeling angry.

"she was more aware of um techniques she could try and employ to to sort of help with her frustration…[YP] will talk through the strategies suggested like the stress ball or you know perhaps I could think of a happy place or something." - P4

3.8.3.4. Parents as facilitators of young people employing behavioural and cognitive techniques

Many parents eluded to their role as a coach to the child, by helping and prompting their child to engage with techniques learnt in the programme.

<u>"the sort of things that um I was able to remind (patient) that I can tell that you're beginning to feel really angry, what's the right way to deal with it</u>?" - P4

3.8.3.5. Therapist as a 3rd person

In addition to the techniques, P4 talked about the therapist as a 3rd person available to the YP who could help them talk about and address the problem. The parent thought a 3rd person helped her daughter talk more openly without a fear of upsetting.

"I think actually it was valuable to her to have that weekly ..get err, come away, talk things through…I just felt that <u>she really she benefited from that sort of almost a third party and talk things through without feeling as if she was upsetting anybody else.</u>" - P4

3.8.3.6. *Parent gaining perspective*

One parent described the programme as a way of 'taking a step back' and gaining a wider perspective which she twice described as a process that helped her to help her son.

"it's been <u>useful</u> in terms like <u>taking a step back</u>, and seeing it from a you know, a <u>wider perspective</u> and being able to maybe- <u>that's helped me to help him, for me to be able to view it.</u>"- P2

3.8.4. Acceptability and feasibility of measures

3.8.4.1. *Forms were understandable*

Parents were asked about how easy it was to understand and complete the outcome measures. All families reported the FAO was very easy to complete. P5 highlighted that the FAO was significantly easier to understand than the standardized questionnaires which she didn't understand. P7 commented that they liked that the standardized measures were completed less frequently.

3.8.4.2. *Forms were frustrating*

One parent found completing the FAO frustrating which he thought affected the accuracy of the data.

"going back to the forms, they are a little frustrating, because they are difficult to fill and you get frustrated, you get a skewed opinion of the day" – P3

3.8.4.3. Forms were completed unreliably

However, many (N=4) said it was difficult to accurately complete the FAO. For example, P3 and P1 both described not remembering or having no time to record each incident because they were busy trying to manage their child's behaviour.

"No, it's not that it was <u>difficult it was just, whether you always remember each one as well</u>, I think in a way <u>it isn't easy when you're going through it, I think the last thing you think of is the tally chart</u>. But I'd go back you know and sort of recap over the week how many times that I can remember you know, I've got fairly decent memory at times but <u>I wouldn't say it was 100% accurate at the time</u>." – P2

3.8.4.4. Forms don't capture problems

P1 explained the FAO did not capture the number of outbursts her child was having at school where many of the incidents occurred. P7 felt that the results on the FAO didn't really capture or reflect what was 'going on at home'.

4. Discussion

This study employed a replicated case series (N=7) and mixed-methods design to explore a short-term, parent-mediated and manualised CBT intervention for children with ASD presenting with anger outbursts. This section will begin by discussing findings relevant to understanding the acceptability of the intervention, then go on to considering the acceptability and feasibility of the measures used. The findings relevant to understanding the efficacy of the intervention will then be discussed before moving on to

clinical and research implications. Finally the strengths and limitations of the study will be considered before ending with a final conclusion.

4.1. Acceptability of intervention

Assessing acceptability has been discussed as a critical early step in program development (Ayala & Elder, 2011). Attendance rates are often used as a proxy measure of acceptability (as in Martinsen, Kristin, Kendall, Stark & Neumer, 2014). Attendance rates were high for parents (parents attended all of the sessions offered) and all but one set of parents said they would recommend the intervention to others. Three YP had difficulty engaging with aspects of the programme and one did not attend the final four sessions. Taken together, this data suggests the intervention programme was acceptable to the majority of families although less so to a subset of YP. Of note is that one set of parents said they would not recommend it for children with ADHD due to inattention being a significant barrier to progress in sessions. Wider research has shown that young people with comorbid ADHD receiving CBT, have significantly poorer outcomes than their non-comorbid counterparts (Halldorsdottir et al., 2015). Although generalizations cannot be made upon one participant, this raises an important point given the frequency of children with ASD and concurrent ADHD. It will be important to understand the suitability and acceptability of the intervention for children with co-occurring ADHD.

Acceptability was also informed by the degree to which parents found the intervention helpful. All parents reported at least one positive change. However, a subset of parents were disappointed by some aspects of the programme outcome. This included the YP's inability to independently apply some of the CBT strategies and for two parents, the temporary nature of the improvement which they believed stopped after the intervention finished. A

114

couple of the families thought that longer term input from a third party was required. Thus, it seems that the intervention was on the whole acceptable to most parents and there is also scope for improvement. Proposed modifications to the intervention to increase acceptability and efficacy are discussed in sections 4.3 and 4.4.

4.2. Acceptability and feasibility of measures

In addition to capturing the acceptability of interventions, researchers are encouraged to assess the acceptability of the measures used to assess outcomes. One strength of the current study is that a variety of measures were incorporated including parent-rated frequency measure, standardized questionnaires, idiographic measures of change and also data from an open-ended interview. The FAO was chosen to provide objective frequency data to protect against expectation effects and increase reliability of results (Wood et al., 2008). Furthermore, it holds comparative value as it was used in previous research of a CBT anger management interventions for ASD (Sofronoff et al., 2007). On one hand, qualitative data indicated that the FAO measure was easy to understand for all parents. For one parent, it was reportedly much easier than the standardised questionnaires with which she had great difficulty. On the other hand, qualitative data revealed that the FAO was challenging to complete at the time of (or shortly after) their child's anger outburst. Half of the parents completed it in retrospect and reported low confidence that it was accurate and reliable. Qualitative data from interviews also implied that the FAO was not sensitive enough to capture improvements in the intensity, duration or severity/type of behaviour exhibited in anger outbursts. It was also deemed to be unreflective of the problem occurrences at school. Whilst it may be that the YP's behaviour did not actually improve, it should be considered that shortcomings of the

measure could account for the lack of improvement reflected by the FAO for the majority of children. In future studies, the sensitivity of the FAO could be improved by including a rating scale for more dimensions of behaviour change (e.g., intensity, duration and severity) and by including a measure of the problem occurrence at school. Additionally, it will be essential for future studies to capture child-report data on their behaviour and it would be useful for researchers to collect direct-observation data to complement parent and child report measures of child behaviour.

4.3. Initial estimate of efficacy

An initial estimate of the efficacy of this intervention can be obtained from a number of sources. These include systematic data of the FAO, pre and post-intervention data from the SDQ, PSI-SF and GBO and finally qualitative data from an interview with parents that took place at least six weeks post intervention.

Looking holistically across all these measures, one could conclude that the intervention is likely to have been efficacious for families who report (statistically significant, reliable or qualitative) improvements on at least three of these measures. Using this criteria, the data from three out of the seven families (P1, P5 and P7) showed evidence of efficacy. This represents over a third of the sample and is comparable to the proportion of participants that have been reported to show improvements from efficacious interventions investigated through randomised control trials (Gordon et al., 2015; Wong, 2008).

The conclusion on the estimate of efficacy is complicated by the discrepancy between the quantitative and qualitative data. Whilst the FAO suggests that only two YP showed significant reductions in anger outbursts, the qualitative analysis revealed that most parents reported some type of

improvement in anger outbursts which they attributed to the intervention. In addition to levels of anger outbursts, many parents reported other positive behaviour changes in their child such as an increased awareness of anger, increased vocabulary to talk about anger and an increased awareness of strategies to use to manage anger, changes in the parent and changes in interactions between parent and child. Given that the FAO was the primary outcome measure, it is unclear how much weight to give the qualitative findings when estimating efficacy. These findings also raise an interesting point about how improvement is defined and the type and level of change required to conclude evidence of efficacy. For example, one could argue that an autistic child's improvement in awareness and communication of anger is a substantial improvement. In this case, the initial estimate of efficacy of this intervention would be higher than 40%. The estimates of efficacy drawn from these findings will vary depending on how one judges the type and level of change needed to qualify as an improvement.

Nevertheless, it is apparent that families benefited in different ways and each to a different extent. There is a need to better understand how the intervention can be modified to benefit a greater proportion of participants. A strength of this in-depth, mixed method research design is the ability to gather rich data about individual participants as opposed to quantitative results reflecting the averages of large groups. Information from the qualitative data of interviews and treatment fidelity checklists that were deemed relevant to understanding barriers to progress and ways to improve the efficacy of the intervention are discussed below.

Firstly, the qualitative analysis revealed that some children developed an awareness of strategies to manage anger but did not apply them at home. This finding is consistent with other literature which has found that children with ASD have difficulty generalizing taught skills to novel

scenarios. A recent Cochrane review on interventions based on theory of mind for ASD reported that generalization of skills beyond specific items and settings appeared to be a repeated problem for children with ASD (Fletcher-Watson, McConnel, Manold & McConachie, 2014). In the current study, parents were encouraged to act as co-therapists to aid generalization/application of learning outside of sessions. However, for two families, parents prompting the young person in an angry moment further angered the young person. It may be that liaison with schools and conducting the sessions in the home or school environment would facilitate the application of self-regulation strategies in those settings. Additionally, clinicians could emphasise role-playing techniques and using social stories to promote generalization. Furthermore, a gradual spacing out of the length between sessions and the provision of post-intervention 'drop in' sessions may help the child to practice and apply the skills. This may also help to maintain gains.

Secondly, for three out of the seven cases, low levels of engagement and motivation were identified as obstacles to progress through treatment fidelity checklists. The reasons identified by therapists are consistent with findings from Deffenbacher (1999) and DiGuiseppe (1999) who identified similar factors which were detrimental to adults' engagement in anger management programmes (i.e. feeling forced to attend anger management programme, disagreement that anger is problematic and a belief that anger is a symptom of an external problem). In this study, motivational enhancement techniques were included in the intervention such as exploring personal goals and negative consequences of aggression. However, for some individuals with ASD, a tendency to show inflexible thinking patterns, may pose an additional challenge to moving out of a pre-contemplative stage of change in a short space of time. This intervention

could be adapted to include a 'pre-treatment phase' to assess and address readiness to change and only those YP who are ready to use CBT to change move onto the main part of the programme. However, it may be that a proportion of YP with ASD do not reach a stage where they want to learn self-regulation skills. The behaviour of these YP may be better managed through external and environmental means.

Thirdly, the qualitative and treatment fidelity checklist data indicated that there was a need to address one participant's underlying anxieties that became apparent during the intervention. Ambler, Eidels & Gregory (2015) investigated the link between anxiety and aggression in 12-18 year olds with ASD and found a strong correlation between anxiety and aggression for YP with ASD but not their neurotypical counterparts. On one hand, this link could indicate that aggression is an overt symptom of anxiety. If this is the case, researchers and clinicians should consider whether observed anger and aggression is best tackled with teaching anger-control strategies or by uncovering and addressing underlying difficulties (e.g., anxiety). On the other hand, it could be argued that the correlation observed between anger/aggression and anxiety could reflect the presence of an underlying mechanism causing both. Weiss (2014) discusses maladaptive emotional regulation processes underlying emotional problems such as anxiety, depression and anger. According to this theory, a transdiagnostic treatment that targets underlying emotional regulation impairments could work better for individuals with ASD.

4.4. Clinical implications

The results of this study suggest that a parent-mediated CBT intervention for anger management can be associated with positive benefits for YP with ASD and their parents. Thus, clinicians should consider it as a treatment

option for this population. For clinicians who plan to offer parent-mediated CBT for anger management to this population, it will be useful to consider factors such as the YP's motivation and readiness to change, underlying anxiety, difficulties linked to comorbid ADHD, ways to facilitate the generalization of skills learnt in sessions and strategies to ensure gains are maintained once the programme has ended. For example, it will be beneficial to introduce a 'pre-treatment phase' to assess and address readiness to change. To aid the generalization of skills, clinicians could consider emphasizing the use of role-play or social stories in sessions, liaising with schools and/or conducting sessions at a home/school setting. To maintain gains, clients may benefit from the gradual spacing out the length of time between sessions and/or the provision of post-intervention 'drop in' sessions.

4.5. Research implications

The preliminary positive results of this study suggest that a larger scale investigation of a refined CBT intervention programme is viable. An evaluation of the measures used in this study and recommendations for the measures used in future research of this programme are discussed in section 4.2.

Given the heterogeneity in the level of improvements seen in this study, it would be worthwhile for future research to further understand the characteristics of individuals with ASD who are most likely to benefit from CBT for anger. It is beyond the remit of this study to make any conclusions on this but factors such as levels of motivation, severity of problem at the start, levels of cognitive flexibility, levels of cognitive ability (specifically verbal ability), gender, levels of social and emotional developmental maturity, and comorbidity could be considered. Furthermore, it would be

interesting to understand the role of therapist experience on the outcome of this intervention.

4.6. Strengths and limitations

A strength of this study is that it has provided an in-depth understanding of a parent-mediated CBT intervention applied to seven families who have a child with ASD and difficulties managing anger. The mixed-methods design captured data that would not have been possible in a quantitative or qualitative design alone. It provided an exploration at an individual level which would not have been possible through a larger group comparison study. There are however, some noteworthy limitations to this study. First, this study is a case series of seven families and so the findings cannot generalise beyond the seven families nor beyond the setting and clinicians through which the intervention was delivered. However, given that this intervention has not been empirically studied before, this study was intended to be informative of the gains possible in this population and also to gather preliminary data. A second limitation of this study is the absence of a control group which meant that improvements reported cannot be ascribed to the intervention itself. The effects of extraneous variables, placebo effect or non-specific therapy factors cannot be ruled out. However, a baseline phase was included to compensate for this and participants acted as their own controls. Furthermore, a number of therapy-change links were made by parents in the follow-up interviews which provides support to the idea that, for most families, positive changes were a result of the intervention. Nevertheless, a controlled study is needed to ascertain further evidence of these preliminary findings. A third limitation is that the study relied heavily on parent report. More data from other sources such as the

YP and teachers would provide further useful information for the development of the programme.

4.7. Conclusions

This study has provided preliminary evidence that a manualised short-term parent-mediated CBT programme for anger management is acceptable to most families and can lead to positive changes for some children with ASD and their parents. The nature of this design allowed the collection of in depth individual data which indicated that the outcome varied across participants. Ultimately, it will be clinically valuable for research to investigate what factors are linked to the families who are most likely to benefit from this type of intervention programme. Qualitative information contributed to understanding how the delivery of this CBT intervention could be adjusted and refined to better meet the needs of this population. The findings, taken together, support further investigation of the efficacy of a refined version of the intervention programme in a large scale controlled study which includes quantitative and qualitative measures.

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Part 3:

Critical Appraisal

Reflections on the use of a mixed-methods and small-N design

The small-N and mixed methodology design allowed me to capture rich data at an individual level and paint a complex picture of the benefits and challenges of applying this type of intervention to children with ASD. This outcome spurred me to reflect on how much of the complexity, intricacies and differences of individual participants are missed when the averages of quantitative data of groups are compared in randomised control trials. This is something I had not considered before when using RCTs to inform my clinical practice. As a clinician, I will now be seeking out qualitative research and small-N designs as well as the results of larger scale RCT.

Before planning this research, I had taken a dichotomous approach to research design in terms of thinking of qualitative methods (QLM) and quantitative methods (QNM). I perceived QNMs as the 'superior' method for investigating clinical interventions. My bias towards QNM was probably influenced by RCTs classified as the 'gold standard' for evaluating clinical interventions and the fact that a considerably larger number of quantitative studies appear in prestigious research journals. Nonetheless, some of the research questions in this study were exploratory and had a focus on acceptability so an inductive approach using qualitative methods allowed for a better understanding of the experiences of families. Despite my initial bias, I was intrigued by the richness and relevance of the information which the QLM captured. The qualitative data seemed to bring the results 'to life' and raised crucial issues about understanding the acceptability and usefulness of the intervention. For example, it threw light on the limited extent of change for some participants, useful ideas about obstacles to change as well as different types of improvements that would not have been captured by quantitative data. The outcome illustrated a number of advantages of QLM that have been described in the literature (i.e. Barker,

Pistrang & Elliot, 2002, p 74) and these certainly outweighed the relatively longer time required for the process.

This research design also prompted me to consider the relative strengths and weaknesses of QNM and QLM and the philosophical positions from which each method is derived. QNM, rooted in a positivist approach uses measures selected by the researcher. Thus, the scope of the data captured depends on the researcher's predictions and assumptions about the type of change they expect to occur. Whilst this deductive approach provides a good method to confirm a prediction, it closes down opportunities gained from an inductive approach such as the discovery of new pieces of information and changes experienced by participants that weren't initially considered by the researcher. Additionally, whilst quantitative data can be simpler to process, it can reduce complex concepts to numbers which may not accurately reflect the construct being investigated. These points were apparent in the findings of the current study where the qualitative approach appeared to hold greater sensitivity and reflected more positive changes than were captured from the quantitative measures (e.g., changes in intensity, duration and type of the YP's anger outbursts, YP's awareness and understanding of anger, YP's language to express emotions and parental confidence and perceived coping). Thus the strengths of the QLM to avoid pre-judgments, capture novel information and provide detail and depth to results was clear. Nonetheless, this research did also exemplify how qualitative data collection and analysis is more time-consuming. It highlighted why this method is carried out on smaller samples. This tendency to use small samples brings an extra caveat to QLM in that the results are not easily generalizable to the wider population. Thus, it would be very informative if QLM were used in largescale randomized studies. Change process research (Greenberg, 1986) has been widely used to investigate the processes which bring about therapeutic change in psychotherapy. It has been argued to be 'a necessary

138

complement to randomized clinical trials and other forms of efficacy research' (Elliot, 2010 p. 1). Qualitative change process research can support causal relationships observed in RCTs and can contribute to the credibility of interventions (e.g., EMDR, Shapiro, 1996). However, given that QLM is resource intensive for a large sample, researchers and funding bodies may be reluctant to carry out such extensive pieces of research. However, QLM can be combined with QNM in creative ways. For example, QLM could be used to elucidate QNM (Barker, Pistrang & Barker (2002). Specifically, QNM could be used to identify subsets of participants that either did or did not respond well to the intervention and QLM could then be used to collect more in depth data for these participants. Alternatively an efficient method of collecting qualitative data could be through the use of brief open-ended post-intervention questionnaires and this would eliminate the requirement for lengthy interviews and transcribing processes.

Given that both QLM and QNM have their pros and cons, I would encourage researchers to consider which methods would answer their research question best and moreover, not restrict themselves to one approach but consider mixing methods in a complementary fashion to benefit from the best of both worlds.

However, in addition to the advantages of mixed-methodology, I did also discover the hidden challenges that can result from combining QLM and QNM. Methodological triangulation (Denzin, 1978) has been proposed to increase the validity of the data sources. When data sources from multiple methods (e.g., QLM and QNM) provide mutual confirmation, then one can be more confident in the validity of each source (Denzin, 1978). However, the results of the current study raised the dilemma of how researchers should respond when the qualitative and quantitative data contradict each other. This dilemma has also been raised by other authors (e.g., Bryman, 1992). If we conclude that discrepant results indicate that one data source is not valid, this raises the question of how to decide which account to 'prioritise' or which source is 'superior'. In this study, I felt inclined to favour the

qualitative data especially as a few participants had spoken about difficulties with accurately completing the quantitative primary outcome measure. However, it has been argued that the use of different methodologies means that different issues are being examined by each method and thus they cannot be directly compared (Fielding & Fielding, 1986). Additionally, some argue that different methodologies 'tap different domains of knowing' (Mathison, 1988, p.14). For example interviews have been argued to tap into more private versus public views (Morgan, 1993). Given these points, it seems more reasonable to combine QLM and QNM to elaborate results from one to the other and increase the breadth and depth of knowledge rather than as a tool of validity.

Further reflections on the outcome of the study

Given that there are so few studies investigating a cognitive behavioural approach for children with ASD and difficulties managing anger, the results of this study look promising for the potential of individually administered CBT interventions to be useful to some children with ASD. Nonetheless, a striking outcome of this research to me, was the clear evidence of variability between the outcomes of participants. Consequently, the more useful question might be which types of individual does this intervention work best for, as opposed 'to does this intervention work on for a group of heterogeneous individuals with ASD?'. This type of information would enable services and service-users to make better informed decisions about which treatments they are likely to benefit from. It would also help researchers to investigate alternative interventions for the subsets of populations that in practice are not benefitting from the current evidence-based ones. RCTs can provide such information if they obtain large enough sample sizes to investigate the moderators of change. I plan to use this type of research more to inform my clinical practice in terms of understanding which type of individual would benefit most from a particular intervention.

As mentioned above, I found the information gathered from the post-intervention interviews to be extremely informative and thought-provoking. It was particularly interesting to hear about the parents who reported temporary (and sometimes quite significant) improvements in their child's behaviour that diminished after treatment ended. Firstly, this finding made me reflect on the process by which change occurred when the temporary improvements were seen. As mentioned by two of the participants, it is possible that attending weekly therapy sessions may have provided the motivation to the child by giving 'importance' to the anger outburst (P2) or through receiving positive reinforcement (from the therapist) for their efforts to employ strategies at home (P4). It was however a positive sign that there were improvements in anger outbursts and it will be important to understand how these gains can be maintained. If the abovementioned factors were the 'active ingredient' in getting the child to use the techniques outside of sessions, then gains could be maintained by attending post-intervention 'review' sessions with the therapist (2, 3 and then 6 months after ending the programme). Secondly, this finding also speaks to the importance for research studies to collect follow-up data and for readers to carefully consider their evaluation of a study if there is no evidence of long-term effects. Thirdly, this finding made me reflect on my own clinical practice when implementing short-term interventions with young people. It is unusual to obtain long-term feedback from clients one has worked with. Consequently, I plan to be aware of not falling into a trap of resorting to the provision of short term 'quick fixes' that show initial improvements without focusing on maintaining long-term benefits. I plan to place greater emphasis on ways to increase long-term improvements in my clinical practice and will stay mindful of including wider systems and considering alternative interventions if it will increase the maintenance of gains. This seems especially relevant in the current climate, where services are under pressure to see more clients with less resources and when initiatives such as 'payment by results'

are introduced which could tempt clinicians/services to value short term over long term outcomes.

A further discussion of limitations of the study

Whilst there are many strengths of this research, it does not come without limitations. These are discussed in the empirical paper and are further reflected on here. One major limitation of this project is that it relied heavily on parental report. Individuals with ASD have difficulties with self-awareness and introspection (Stewart, Barnard, Pearson, Hasan, & O'Brien, 2006) which may decrease the reliability of the child-report measure. Many studies report discrepancies between child and parent reports of symptoms in non-ASD (De Los Reyes & Kazdin 2005) and ASD literature (White, Ollendick, Scahill, Oswald & Albano, 2009). The decision to focus on parent-report in this study was made because the main construct being measured was anger outbursts, an observable behaviour deemed likely to be more accurately and reliably documented by parents. Nevertheless, future studies could further boost validity by triangulating parent, child and clinician report of symptoms. Additionally, young people with ASD could be helped to express themselves through qualitative methods (e.g., in Calzada, Pistrang & Mandy, 2012). This additional data from young people will be valuable in further evaluating the outcome of the programme and crucially for developing the acceptability, motivation, attendance and ultimately efficacy for young people.

A second limitation of this study is the poor rate of return for the postal follow-up questionnaires. Reviews indicate the postal response rate can be increased by repeat mailing and telephone reminders (Nakash, Hutton, Jorstad-Stein, Gates, & Lamb, 2006). Despite the implementation of these strategies, only one out of seven families returned the questionnaires by post with is a much lower rate than I had expected. Interestingly, all participants gave their time to complete the follow-up interviews and fortunately this allowed the collection of valuable follow-

142

up data. In hindsight, the return rates of the quantitative data could have been increased by inviting participants to attend an appointment held at the clinic to complete both the follow-up interview and quantitative measures.

Thirdly, it is possible that bias was introduced through demand characteristics due to my dual role in both conducting the research aspects and providing the intervention to more than half the participants. Had there been more resources available for this study (i.e. clinician time to undertake interventions with multiple families in the time period required for study completion), then it would be desirable for the researcher to have no contact with the participants except to collect baseline and outcome measures. This would increase objectivity from the researcher's point of view and it would reduce risk of bias from demand characteristics.

Lastly, there is potential for the results of this study to reflect bias stemming from researcher allegiance (Munder, Brutsch, Leonhart, Gerger, & Barth, 2013). Given that the two clinicians providing the intervention were invested in the research project, it is unclear to what extent the results would be replicated if carried out by other clinicians. However, this research was intended to explore the potential for efficacy in these conditions. The current study does benefit from the involvement of two clinicians with varying experience and expertise with ASD. Given that participants seen by both clinicians showed improvements, this provides some preliminary evidence that improvement is not dependent on significant levels of expertise. Further research is needed to establish efficacy using controlled studies, in other settings. The next stage would be to 'roll out' the intervention to test whether the results are replicated in other settings with other clinicians.

A reflection on methodological challenges

There were a couple of obstacles that presented during the course of the research. One issue that arose from this study was getting enough suitable participants recruited in the timescale needed. With the support of the internal supervisor who had experience working in the clinic for a number of years, we had considered but not anticipated any problems finding enough suitable participants. However, during the 11 month recruitment window, clinicians advised us that there were much fewer young people presenting with anger outbursts then had been anticipated. In hindsight, it may have been helpful to carry out a 'pilot recruitment stage' by sampling the number of suitable participants that came through the clinic in a sample time period. Additionally, I think this issue was complicated by the fact that neither the internal supervisor nor myself were working in the clinic at the initial recruitment time which meant we depended on other clinicians to identify suitable participants. Given that clinicians are already under pressure to carry out their workload, it may be possible that suitable participants attended the clinic but were missed in this way. More initiatives to remind or ease the process for clinicians to keep recruitment in mind and enquire about anger outbursts may have helped (e.g., asking all families who are seen at the clinic for assessment to complete a screening questionnaire). Alternatively, it is possible that rates were actually lower than previous years. It may have been helpful to start the recruitment at an earlier stage to create a bigger time window. However, this was not possible due to the time required to get NHS ethics approval and time restraints linked to the deadline of the research project. Given that clinical research is most often conducted in a working service, similar issues related to recruitment are likely to span across most clinical research studies. In future research I will give more weight to fully understanding the rates of suitable participants in a clinic and potential pitfalls in this area during the initial planning stages of a study.

144

In retrospect, I have realised that this type of intervention study was a highly ambitious, challenging and somewhat risky venture for a doctorate research project due to the uncertainty of recruitment of families, attendance and the complex logistics required. I had underestimated the practical and administrative challenges posed by carrying out this type of research in a busy NHS clinic within the time constraint of one research day a week. One example was the complex coordination that was required to ensure that families were given the correct measures and booked in at appropriate times and intervals and juggling appointments, cancellations, rescheduled appointments to fit in with the time frame of the study, limited room availability, and therapist availability, etc. There were various other similar challenges which required a high degree of resilience, perseverance, time management and organisational skills. I would urge future researchers not to underestimate these challenges and allow for and make contingency plans for the unexpected. This is probably true of most clinical research but possibly more significant in this project due to my dual role as conducting the intervention and coordinating and organising the research aspects at the same time.

Final thoughts

All in all, the experience of conducing this doctoral research project has been a journey that has been challenging, at times stressful but also incredibly rewarding, informative and thought-provoking. The growth I have experienced in my understanding of planning and carrying out a clinical piece of research as well as the knowledge I gained through the results of the project is invaluable. I hope that the findings will be as interesting and useful to fellow researchers and clinicians working in this area and ultimately for young people with ASD and their families.

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Appendix 1: Table describing risk of bias and support for judgement for each study

Bearss et al (2015) Risk of Bias Table

Bias	Authors' judgement	Support for judgement
Random sequence generation (selection bias)	Unclear risk	"The data center randomly assigned eligible children to treatment….
Allocation concealment (selection bias)	Low risk	"...randomly assigned […] in a 1:1 ratio using permuted blocks allowing for concelment of allocation prior to enrollment"
Blinding of participants and personnel (performance bias)	High risk	Not reported. Probably not done. Parents and therapists very likely to be aware of the assigned treatment condition.
Blinding of outcome assessment (detection bias)	High risk	Primary measures was parent ratings who were not blind to treatment. Secondary measure was an independent evaluator. "To protect the treatment binding, we maintained separate study binders for therapists and independent evaluators. Parents were instructed to avoid discussing the treatment during assessments with independent evaluators."
Incomplete outcome data (attrition bias)	Low risk	Attrition in experimental group was 11.2% and 8.8% for control group. In experimental group: 7 exited the study, 3 discontinued treatment but completed follow-up. In control, 2 exited study before post intervention, 2 discontinued intervention but completed measures, All participant data was included in the primary analysis.
Selective reporting (reporting bias)	Low risk	4/5 outcomes are reported in the paper. "since development of the protocol, questions have been raised about the ecological validity of the SOAP and whether it is representative of a child's behaviour (Handel et al 2013). The SOAP will be presented in a separate report".
Treatment Fidelity	Low risk	Treatment was delivered by 23 therapists across 6 sites. "Therapists undertook systematic training and achieved certificates. Weekly supervision occurred. "The training manual included verbatim scripts and instructions for therapists" Checklist used each session. Independent raters scored 10% of recordings. Treatment fidelity ratings for treatment condition were high: mean (SD)=96.7% (8.3) and 97.2% (6.4) for controls.
Other bias	Low risk	

Garcia-Gomez et al. (2014) Risk of bias table

Bias	Authors' judgement	Support for judgement
Random sequence generation (selection bias)	High risk	"Two groups were formed" not randomised. "The quasi-experimental design, with the control groups not being selected at random may have influenced the results"
Allocation concealment (selection bias)	High risk	The groups were not allocated at random.

Blinding of participants and personnel (performance bias)	High risk	No blinding of participants or researchers.
Blinding of outcome assessment (detection bias)	Low risk	Primary outcome data was completed by teachers
Incomplete outcome data (attrition bias)	Low risk	the sample consisted of 32 pupils [...] but there is only enough information about 16" Then "two groups were formed" 8 in each.
Selective reporting (reporting bias)	Low risk	All intended outcomes reported fully.
Treatment Fidelity	High risk	No measure of treatment fidelity. "the diversity of the sample, to a certain extent conditioned how the intervention sessions were carried out and thus may have altered effects of the treatment".
Other bias	Unclear risk	No long term follow-up

Mohammadzaheri, Koegel, Rezaee & Rafiee, (2015) Risk of Bias Table

Bias	Authors' judgement	Support for judgement
Random sequence generation (selection bias)	Unclear risk	"Each participant in each dyad was then randomly assigned to one of the two treatment groups"
Allocation concealment (selection bias)	Unclear risk	Not reported
Blinding of participants and personnel (performance bias)	Low risk	'Parents and teachers were informed that their children/students would receive speech and language services, but were naïve to the target behaviour and the intervention condition to which their child was assigned".
Blinding of outcome assessment (detection bias)	Low risk	Raters were blind for primary outcome (disruptive behaviour). "teachers, parents, individuals who scored the tests and evaluated the children were not aware of the purpose of the study, the intervention approach to which children were assigned, nor did they have access to the randomization list". "videotapes were randomly presented and scored by two SLPs who were unaware of the hypothesis of the study and [child's experimental condition]"
Incomplete outcome data (attrition bias)	Unclear risk	Not reported
Selective reporting (reporting bias)	Low risk	All intended outcomes reported fully.
Treatment Fidelity	Unclear risk	The use of pre-existing training manuals were used for each condition and measure of treatment fidelity was always above 80%.
Other bias	Unclear risk	1. Primary outcome measure was observation of frequency of disruptive behaviours in first session compared to last session. Only 2 data points per participant 2. No long term follow up or measure of generalisation of benefits apparent in treatment clinic to real life settings (home and school settings)

Scarpa & Reyes (2011) Risk of Bias Table

Bias	Authors' judgement	Support for judgement
Random sequence generation (selection bias)	Unclear risk	Details not reported. "children were randomly assigned to…."
Allocation concealment (selection bias)	Unclear risk	Details not reported
Blinding of participants and personnel (performance bias)	High risk	Not reported. Probably not done. Families were "assigned to experimental or delayed treatment control group... who started the intervention approximately one week after the experimental group"
Blinding of outcome assessment (detection bias)	High risk	Raters (parents) were not blinded to allocation group. 'During the intake session, parents and children met the experimenters and were informed about the study's main objectives and procedures'. primary outcome measure is child and parent report.
Incomplete outcome data (attrition bias)	Unclear risk	"One family dropped out of the study due a family emergency, leaving a final sample of 11 children". 11 children were randomised so dropout appears to be before randomisation process. No more information reported.
Selective reporting (reporting bias)	Low risk	All intended outcomes reported fully.
Treatment Fidelity	Unclear risk	No detailed report of formal/independent treatment fidelity check. "The intervention's treatment manual was followed and sessions were reviewed for treatment adherence"
Other bias	Unclear risk	1. No long term follow-up. 2. Generalizability limited to Caucasion, high-income families.

Smith, Groen & Wynn (2000) Risk of Bias Table

Bias	Authors' judgement	Support for judgement
Random sequence generation (selection bias)	Low risk	"Using a random numbers table [the statistician] assigned one member of each [matched] pair to the intensive treatment group and the other to the parent training group".
Allocation concealment (selection bias)	Low risk	random assignment was carried out by an "independent statistician"
Blinding of participants and personnel (performance bias)	High risk	Not reported. Probably not done.
Blinding of outcome assessment (detection bias)	High risk	Raters (parents) were not blinded to allocation group .
Incomplete outcome data (attrition bias)	Low risk	"There were no dropouts"
Selective reporting (reporting bias)	Unclear risk	The data of 4 participants was removed due to a 'design change'. 'children with mental retardation and not Pervasive dv disorder were removed from the study. The authors report 'the data do not alter the results of the significance testing presented". Pg 272. Otherwise All intended outcomes seem to be reported fully

Bias	Authors' judgement	Support for judgement
Treatment Fidelity	High risk	No formal/independent measure of adherance to treatment manual
Other bias	High risk	Treatment length varied between and within the 2 groups (3-9 months for parent training and 2-3 years for Intensive Early Intervention). pg 274: "the number of months of treatment was more variable than had been planned". In the intensive treatment 'contingent aversive were employed briefly for the first 4 children, they were then stopped for all children.

Sofronoff, Attwood, Hinton & Levin, 2007 Risk of Bias Table

Bias	Authors' judgement	Support for judgement
Random sequence generation (selection bias)	Unclear risk	"Participants were randomly assigned to........."
Allocation concealment (selection bias)	Unclear risk	Not reported
Blinding of participants and personnel (performance bias)	High risk	Not reported. Probably not done.
Blinding of outcome assessment (detection bias)	High risk	Raters (parents) were not blinded to allocation group.
Incomplete outcome data (attrition bias)	Unclear risk	The number of dropouts were not reported.
Selective reporting (reporting bias)	Low risk	All intended outcomes reported fully.
Treatment Fidelity	Unclear risk	All parents received a manual that contained all the information covered by the 6 components of the intervention package. However, in terms of therapists adherence to the manual in sessions, adherence checklists were completed by therapists but an adherence figure is not reported. There was no adherence check from an independent evaluator. "Each therapist followed the format of each session from a therapist's manual and completed a checklist to indicate that all components had been completed".
Other bias	Low risk	

Sofronoff 2007 Risk of Bias Table

Bias	Authors' judgement	Support for judgement
Random sequence generation (selection bias)	Unclear risk	"families were randomly assigned to either the intervention or wait-list condition as consent forms were returned.
Allocation concealment (selection bias)	Unclear risk	Not reported
Blinding of participants and personnel (performance bias)	High risk	Not reported. Probably not done.
Blinding of outcome assessment (detection bias)	High risk	Raters (parents) were not blinded to allocation group .
Incomplete outcome data (attrition bias)	Low risk	No missing outcome data. "preliminary analysis revealed that the distribution was normal and homogenous with no missing data"

Selective reporting (reporting bias)	Unclear risk	All intended outcomees reported fully
Treatment Fidelity	Low risk	"Therapists completed a checklist after each session to indicate adherence to the protocol and 25% of sessions were videotaped for protocol adherence.
Other bias	Low risk	

Solomon, Ono, Timmer, Goodlin-Jones (2008) Risk of Bias Table

Bias	Authors' judgement	Support for judgement
Random sequence generation (selection bias)	Unclear risk	Methods of randomization not reported in detail. "[participants paired for age, cognitive level and behaviour symptoms] one subject from each pair was then randomly selected to receive intervention first".
Allocation concealment (selection bias)	Unclear risk	Not reported
Blinding of participants and personnel (performance bias)	High risk	Not reported. Probably not done.
Blinding of outcome assessment (detection bias)	High risk	Raters (parents) were not blinded to allocation group .
Incomplete outcome data (attrition bias)	Unclear risk	Insufficient reporting of attrition/exclusion and numbers used in analysis to permit judgement.
Selective reporting (reporting bias)	Low risk	All intended outcomes reported fully
Treatment Fidelity	High risk	No formal measure of treatment fidelity (although "regular team coding meetings" occurred over the study where tapes of tx sessions were reviewed and discussed both behavioural and coaching issues" pg 1771) Also 3/5 therapists were officially trained and 2/5 therapists were trained in the team by working on at least 3 cases with trained therapists
Other bias	Unclear risk	No long term follow up data.

Tellegen & Sanders (2014) Risk of Bias Table

Bias	Authors' judgement	Support for judgement
Random sequence generation (selection bias)	Low risk	"Participants were randomly assigned using a computer generated random number sequence"
Allocation concealment (selection bias)	Unclear risk	Not reported
Blinding of participants and personnel (performance bias)	High risk	Not reported. Probably not done.
Blinding of outcome assessment (detection bias)	High risk	"Allocation to conditions was not concealed to the researcher. Participants were not blinded to allocation." This design is vulnerable to expectation effects as outcomes are parent report.
Incomplete outcome data (attrition bias)	Low risk	Missing outcome data balanced in numbers across groups, with similar reasons for missing data across groups. Intent to treat analysis conducted. For both groups, Post tx= 9 dropouts. Followup- 10 dropouts. "Chi-squared tests for independence revealed no significant difference in attrition between

153

		the two groups at post-intervention stage or follow up. There were no significant differences on demographic characteristics on dependent variables at preintervention between completers and non-completers. "
Selective reporting (reporting bias)	Low risk ▼	All intended outcomes reported fully
Treatment Fidelity	Low risk ▼	Manual used, supervision, protocol adherence checklists completed by therapist and independent assessor checked random sample 20% of session recordings. 97% fidelity rating.
Other bias	Unclear risk ▼	Baseline imbalance- There were significantly more child health problems in families in the experimental group (n=17) compared to the control (n=7). This could lead to underestimation of effect. "with the exception of child health problems, the groups did not differ significantly on demographic characteristics."

Appendix 2: Conduct disorder questionnaire

Please indicate whether your child shows any of the following, more than you would expect in comparison to children of the same age	Definitely not	Possibly true /uncertain	Definitely
Lose temper?			
Arguing with adults?			
Deliberately defying adults?			
Blaming others for own mistakes or bad behaviour?			
Touchy or easily annoyed by others?			
Complain unjustifiably about not being treated fairly?			
Destructive toward own property?			
Spiteful or vindictive?			
Fails to keep promises?			
Lying?			
Frequent fighting?			
Has s/he done any of the following:			
Used a deadly weapon?			
Physically cruel to a person?			
Mugging or purse snatching?			
Stealing?			
Forced another person into sexual activity?			
Has s/he done any of the following to anyone else's property:			
Destroyed property?			
Deliberate setting fires?			
Persistent stealing?			
Broken in?			
Cruel to an animal?			
Victim of bullying?			
Bullying to others?			
Has s/he seemed out of control for any of the following reasons:			
Staying out late without permission?			
Truanting, beginning under the age of 13?			
Running away?			

Appendix 3: Frequency of anger outbursts questionnaire

Tally of Anger Episodes

Please indicate the number of times that your child was 'unable to maintain emotional control and behaved or spoke inappropriately in anger' (anger episode) at home and/or when out with the family using the tally chart below:

Day	Tally of Anger Episodes	Comments
Monday		
Tuesday		
Wednesday		
Thursday		
Friday		
Saturday		
Sunday		
Total number of anger episodes this week:		

In your opinion, how accurate is the tally that you have recorded on a scale from 0 to 7?

0	1	2	3	4	5	6	7

Not at all accurate   Very accurate

To be completed by clinician:

Week Commencing _____

Session number _____

Appendix 4: Post-Intervention Interview

Great Ormond Street
Hospital for Children
NHS
NHS Trust

Joint Research and Development Office
Division of Research and Innovation

Post-Intervention Interview

Hello, my name is _____. I'm calling from the Social and Communication Disorders Clinic at GOSH. I believe that you have consented to taking part in some research about a CBT programme that you and your child have recently attended at our clinic. I am part of the research team and I am conducting the post-intervention interviews. I wanted to check that this is still a suitable time to do this with you? If no, rearrange a time.

It will take approximately 10-20 minutes and I will be asking you questions about your experience of the programme and any effects that you have noticed.

We'd like to audio-record the comments you give us so that we can analyse the results later. Before we start can I take your consent to audio-record the conversation? (take verbal consent- if not then type their answers verbatim).

*****PROMPT THE PARENT IF ANSWER IS VAGUE*****

1.   What <u>changes</u>, if any, have you noticed in your child since the programme started?*

2.   What <u>changes</u>, if any, have you noticed in yourself since the programme started?*

   *(For example, Are you <u>doing, feeling, or thinking</u> differently from the way you did before?
   And what <u>specific ideas</u>, if any, have you gotten from therapy so far?)*

3.   Thinking about your child, has anything changed for the <u>worse</u> for you since the programme started?*

4. Thinking about your child, is there anything that you <u>wanted</u> to change that hasn't since the programme started?*

5.   In general, what do you think has <u>caused</u> these various changes? In other words, what do you think might have brought them about?
   (Including things both <u>outside</u> of therapy and <u>in</u> therapy)

6.   Can you sum up what has been <u>helpful</u> about the programme so far? Please give examples. *(For example, general aspects, specific events)*

7. What kinds of things about the programme have been unhelpful, negative or disappointing for you*? (For example, general aspects. specific events)*

8. Has anything been <u>missing</u> from your treatment? (What would make/have made your therapy more effective or helpful?)*

9. Would you <u>recommend</u> this intervention programme to a friend if they were experiencing similar problems? (please circle)

Yes/No

10. Were the questionnaires and anger outburst logs easy to understand? (please circle)

Yes/No

Any comments/suggestions

_____

11. Were the questionnaires and anger outburst logs easy to complete? (please circle)

Yes/No

Any comments/suggestions

_____

12. Do you have any <u>suggestions</u> for us, regarding the research or the programme?*

13. Are there any other comments that you would like to make?

Appendix 5: Treatment fidelity checklist

CHECKLIST ACTIVITY	Attempted in allocated session?	Attempted at all?	*If no, state the reason*	Ready to move on?	Did any EXTRANEOUS VARIABLES interfere? Which?	Adaptations Made?
Session 1						
Introductions						
Getting to know the young person						
Discussions about Anger (Normalizing anger and discussing family's experiences of anger)						
Triggers and Child's responses to anger						
Hopes for Therapy						
HW- Family Storyboard						
Session 2						
Review homework						
Affective Education (CBT model)						
Intensities of emotion (thermometer)						
Recognise early warning signs of anger						
HW- Family Storyboard Enhanced						
Session 3						
Explore connections between thoughts, emotions, behaviour, physiology. Build formulation						
Explore consequences of alternative ways of responding (using comic strips)						

HW- triggers, thoughts, feelings, behaviours that make them feel better)						
Session 4						
Review Homework						
Behavioural techniques to manage anger						
Introduce toolbox						
HW- practice using behavioural techniques from toolbox. Note down difficulties to discuss next session.						
Session 5						
Review Homework						
Introduce cognitive strategies						
Link alternative ways of thinking with concrete consequences						
HW- practice using cognitive strategies						
Session 6						
Review Homework						
Continue building on behavioural and cognitive strategies						
Generalising						
HW- Others encourage use of effective technique.						
HW- make techniques accessible (cards, notes in house)						

165					
Session 7, 8 (9+10)					
Generalizing Blueprint and Relapse prevention (what makes me angry, what are the early warning signs, what makes me feel better?)					

Appendix 6: NHS Ethical Approval

NHS
Health Research Authority
NRES Committee West Midlands - Edgbaston
The Old Chapel
Royal Standard Place
Nottingham
NG1 6FS

21 May 2015

Dr William Mandy
Senior Lecturer
University College London
Floor 4 Torrington Place
1-19 Torrington Place
London
WC1E 6BT

Dear Dr Mandy,

Study title:	**The feasibility, acceptability and effectiveness of a family CBT intervention aimed at families of children with a diagnosis of Autism Spectrum Disorder and presenting with significant anger management difficulties**
REC reference:	**15/WM/0140**
IRAS project ID:	**169795**

Thank you for your letter of 20 May 2015, responding to the Committee's request for further information on the above research and submitting revised documentation.

The further information has been considered on behalf of the Committee by the Chair.

We plan to publish your research summary wording for the above study on the HRA website, together with your contact details. Publication will be no earlier than three months from the date of this favourable opinion letter. The expectation is that this information will be published for all studies that receive an ethical opinion but should you wish to provide a substitute contact point, wish to make a request to defer, or require further information, please contact the REC Manager, Mrs Helen Poole,
Under very limited circumstances (e.g. for student research which has received an unfavourable opinion), it may be possible to grant an exemption to the publication of the study.

Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised, subject to the conditions specified below.

Re-issue 17.06.15 – reinstate child PIS documents into checklist

Conditions of the favourable opinion

The favourable opinion is subject to the following conditions being met prior to the start of the study.

Management permission or approval must be obtained from each host organisation prior to the start of the study at the site concerned.

Management permission ("R&D approval") should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements.

Guidance on applying for NHS permission for research is available in the Integrated Research Application System or at http://www.rdforum.nhs.uk.

Where a NHS organisation's role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"), guidance should be sought from the R&D office on the information it requires to give permission for this activity.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of approvals from host organisations

Registration of Clinical Trials

All clinical trials (defined as the first four categories on the IRAS filter page) must be registered on a publically accessible database. This should be before the first participant is recruited but no later than 6 weeks after recruitment of the first participant.

There is no requirement to separately notify the REC but you should do so at the earliest opportunity e.g. when submitting an amendment. We will audit the registration details as part of the annual progress reporting process.

To ensure transparency in research, we strongly recommend that all research is registered but for non-clinical trials this is not currently mandatory.

If a sponsor wishes to request a deferral for study registration within the required timeframe, they should contact ███████████████████ The expectation is that all clinical trials will be registered, however, in exceptional circumstances non registration may be permissible with prior agreement from NRES. Guidance on where to register is provided on the HRA website.

It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

Ethical review of research sites

NHS sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

Re-issue 17.06.15 – reinstate child PIS documents into checklist

Approved documents

The final list of documents reviewed and approved by the Committee is as follows:

Document	Version	Date
Covering letter on headed paper [Cover letter for resubmission]	Version1	09 May 2015
IRAS Checklist XML [Checklist_20052015]		20 May 2015
Non-validated questionnaire [Frequency of Anger Episodes]	Version1	20 March 2015
Non-validated questionnaire [Therapist Log]	Version1	20 March 2015
Non-validated questionnaire [Post Intervention Interview Questions]	Version1	20 March 2015
Other [Response Email for Validation]		02 April 2015
Participant consent form	Version2	29 April 2015
Participant consent form [Consent Form]	Version2	20 May 2015
Participant Information Sheet (PIS) [Child PIS]	Version2	01 March 2015
Participant Information Sheet (PIS) [Child PIS retrospective]	Version2	01 March 2015
Participant information sheet (PIS) [PIS]	PIS_revised	09 May 2015
Participant information sheet (PIS) [PIS retrospective version]	Version2	20 May 2015
REC Application Form [REC_Form_01042015]		01 April 2015
Referee's report or other scientific critique report [d]	Version2	20 March 2015
Research protocol or project proposal [Protocol_Version1_27_02_2015]	Version2	09 May 2015
Response to Request for Further Information		20 May 2015
Summary CV for Chief Investigator (CI) [CI Summary CV]	Version1	
Summary CV for student [Student Summary CV]	Version1	20 March 2015
Validated questionnaire [SDQ]	Version1	20 March 2015
Validated questionnaire [Goal Progress Chart]		

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

After ethical review

Reporting requirements

The attached document *"After ethical review – guidance for researchers"* gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports

Re-issue 17.06.15 – reinstate child PIS documents into checklist

- Notifying the end of the study

The HRA website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

User Feedback

The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the application procedure. If you wish to make your views known please use the feedback form available on the HRA website:
http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance/

HRA Training

We are pleased to welcome researchers and R&D staff at our training days – see details at
http://www.hra.nhs.uk/hra-training/

15/WM/0140	Please quote this number on all correspondence

With the Committee's best wishes for the success of this project.

Yours sincerely

██████████

pp
Mr Paul Hamilton
Chair

████████████████████████

Enclosures: "After ethical review – guidance for researchers" SL-AR2

Copy to: Sponsor - Dave Wilson
             R&D Contact - Karen Ignatian

Appendix 7: Participant Information Sheet

Project Title: The feasibility, acceptability and effectiveness of a family Cognitive Behaviour Therapy intervention aimed at families of children with a diagnosis of Autism Spectrum Disorder and presenting with significant anger management difficulties

Investigator: Miss Sohini Shah, Trainee Clinical Psychologist

Principal Investigator: Professor David Skuse

We would like to invite you and your family to take part in our research study. Before you decide we would like you to understand why the research is being done and what it would involve for you. With your consent, one of our team will contact you in the next few weeks by telephone to answer any questions you may have. Please take time to read the following information.

Ask us if there is anything that is not clear. Take your time to decide whether or not you want to take part.

What is the purpose of the study?
We want to find out more about what treatment programmes are helpful for children with an Autism Spectrum Disorder (ASD) and anger management difficulties. We offer a treatment programme at the Social and Communication Disorders Clinic aimed specifically to help families of children with an ASD and showing difficulties managing anger. The purpose of the study is to conduct some research on this treatment programme. We want to find out the following things:

➢ whether this treatment programme is *effective* in reducing the number of anger episodes displayed by children and also to see whether it is linked to a reduction in parental stress levels
➢ whether the treatment programme is *acceptable* to the families involved. It also aims to find out their view on how we can improve it
➢ whether it is *feasible* to conduct this individualised treatment programme in eight to ten sessions guided by a manual.

Why have we been chosen?
You have also been chosen for the study because you are going to take part in the treatment programme that we offer at the SCDC and because we think that you meet the requirements to participate in the study. We are aiming to get about ten families to take part in this study.

Do we have to take part?
It is up to you. If you do decide to take part we will give you this information sheet to keep and ask you to sign a consent form for yourself and on behalf of your child. You are free to withdraw your consent at any time, without giving reason. This will not affect the standard of care you or your child receives or whether you can receive the treatment programme.

What will happen to me and my child if I take part?
If you make the decision for you and your child to take part in the study, then we will use the information from some of the questionnaires that our clinicians ask you to complete as part of your routine clinical care. Also we will ask you to fill in some questionnaires for the research. They will include the following:

- two tick box questionnaires about your child's behaviour before and after the programme (each takes less than 5 minutes)
- two additional questionnaires, one about your child's behaviour and one about your stress levels. We'll ask you to complete these in the middle of treatment and 6 weeks after the treatment programme (each takes 10 minutes)
- At the very end of the intervention it will be helpful to hear about your experiences of the intervention and so the researcher will ask you some questions about your experience of the treatment programme over the telephone. We anticipate that this should take about 20-30 minutes.

We anticipate that it will take you less than an hour and a half to complete these questionnaires over the course of the study. You are welcome to complete these questionnaires during the middle part of the treatment sessions whilst the therapist will be seeing your child on a 1:1 basis.

What are the potential benefits?
Although we anticipate benefits from the intervention, we cannot guarantee that participating in the study will help families. However the information and feedback provided by you for the study could help us improve the intervention and help similar families with children with an ASD and anger management difficulties in the future.

What are the possible disadvantages and risks of taking part?
We do not anticipate any risk will come to families from taking part in this study. A potential burden could be the time and energy taken to fill out outcome questionnaires. To minimise this burden, we have purposely designed the study so that it will not take longer than an hour and a half in total over the course of the study to complete the research questionnaires.

What if there is a problem?
If you have a concern about any aspect of this study, you should ask to speak to the researcher (Sohini Shah) who will do their best to answer your questions ███████████████████. If you wish to complain, or have any concerns about any aspect of the way you have been approached or treated by members of staff you may have experienced due to your participation in the research, National Health Service or UCL complaints mechanisms are available to you. You can contact the Great Ormond Street Hospital Patient Liaison Service by phone ███████████ or e-mail ███████████████.

In the unlikely event that you are harmed by taking part in this study, compensation may be available. After discussing with your researcher, please make the claim in writing to the [Dr William Mandy] who is the Chief Investigator for the research and is based at [University College London].

Will my taking part in this study be confidential?
All data from this study will be kept in accordance with the Data Protection Act 1998. The paper results of the study will be kept in a locked filing cabinet where only the researcher and clinicians in your child's direct healthcare team will be able to access them. An electronic copy of data will also be stored. This electronic data will not be linked to you or your child's name, address or date of birth and instead it will be linked to a unique participant code. Authorised persons from regulatory authorities, the sponsor (UCL Joint Research Office) or the NHS Trust may request access to study and source data for study governance purposes.

What will happen to the results of the research study?

We plan to publish the results of this study in a peer reviewed journal and we intend to present it at relevant conferences. We hope that the results of this study will be used for an application for an even larger controlled study of the treatment programme. If you are interested in the results of the study, we are happy to send you a short report of the results in lay language and / or a copy of the paper (whatever you prefer).

Who has reviewed the study?

All research in the NHS is looked at by independent group of people, called a Research Ethics Committee, to protect your interests. This study has been reviewed and given favourable opinion by West Midlands - Egbaston Research Ethics Committee.

Further Information and Contact Details

If you would like to contact me, the researcher and find out more about the study and/or ask any questions about it then please feel free to e-mail me (⬛⬛⬛⬛⬛⬛⬛⬛⬛⬛) and I will get back to you as soon as possible.

Appendix 8: Participant Information Sheet & Assent form- Child version

Project Title: The feasibility, acceptability and effectiveness of a family CBT intervention aimed at families of children with a diagnosis of Autism Spectrum Disorder and presenting with significant anger management difficulties

Investigator: Miss Sohini Shah, Trainee Clinical Psychologist

Why are we doing this research study?

A research study is a way to learn more about people. We would like to find out more about a treatment programme that we offer for children with an Autism Spectrum Disorder who find it difficult to manage angry feelings.

Why am I being asked to be in the study?

We are inviting you to be in this study because you and your family are going to take part in a treatment programme that we offer at Great Ormond Street Hospital. We want to study whether this treatment programme is helpful to you and your family.

If I am in the study, what will happen to me?

If you decide that you want to be part of this study, you will be asked if a researcher can use some information collected from your parents about how you are getting on. Your parents will give us this information by completing some forms.

What are the risks of taking part in the study?

We do not think that any harm can come to you or your family as part of taking part in this study.

Will the study help me?

Although we think the treatment programme might help you, we cannot promise that the study will help. But the study can help us to understand whether the treatment programme is helpful to you and your family. It can help us get some ideas of how we can change it in the future to make it more helpful.

Do I have to be in this study?

You do not have to be in this study, if you do not want to be. If you do not want to be in this study, you can still take part in the treatment programme offered at our clinic. If you decide that you don't want to be in the study after we begin, that's OK too. We are discussing the study with your parents and you should talk to them about it too.

What happens after the study?

When we have finished this study we will write a report about what was learned. This report will not include your name or that you were in the study.

If you decide you want to be in this study, please sign your name below:

Name: Date:

_____   _____

Name of person taking assent: Date:

_____   _____

Centre Number:

Study Number:

Participant Identification Number for this trial:

Appendix 9: Parent Consent Form

UCL

Great Ormond Street
Hospital for Children
NHS Foundation Trust

Centre Number:

Study Number:

Participant Identification Number for this trial:

CONSENT FORM

Title of Project: The feasibility, acceptability and effectiveness of a family Cognitive Behaviour Therapy intervention aimed at families of children with a diagnosis of Autism Spectrum Disorder and presenting with significant anger management difficulties

Name of Researcher: Sohini Shah

Principal Investigator: Professor David Skuse

Please initial box

1. I confirm that I have read the information sheet dated 29.04.2015 (Revised Version 2) 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 and my child's participation is voluntary and that we are free to withdraw at any time without giving any reason, without my medical care or legal rights being affected.

3. I understand that relevant sections of my child's medical notes and data collected during the study will be looked at by the researcher and may be looked at by individuals from regulatory authorities, the Sponsor (UCL Joint Research Office), or from the NHS Trust, where it is relevant to my taking part in this research. I give permission for these individuals to have access to these records.

4. I agree to take part, with my child, in the above study.

_____   _____   _____
Name of Participant   Date   Signature

_____   _____   _____
Name of Person   Date   Signature
taking consent

When completed: 1 for participant; 1 for researcher site file; 1 (original) to be kept in medical notes.

Appendix 10: Intervention manual

Session	Aim	Activities in Session	Between Session Project
1	Assessment and Motivation to change	• Introductions • 'All about me' worksheet 1. *(Hobbies/favourites/strengths)* • 'All about me' worksheet 2. *(what makes me feel happy, angry, afraid and relaxed).* Involve family members as well. • Normalizing anger. Anger is adaptive. • What makes family members angry and how do they deal with it? • Consider ways that child responds to anger. (Explore incentives to change by considering pro/cons of current way of responding. • Hopes for therapy (Goals based Outcomes- outcome measure worksheet)	Family Storyboard (simple) - Trigger What made us family feel angry? -Response/behaviour How did we deal with it?
2	Affective Education & Socialisation to CBT model	• Review of homework (Involving whole family. Framed as a 'quiz'. What responses worked well and which did not?) • Drawing on the homework task to explore anger: - Effects on body, behaviour and thinking (using CAT-kit) - Explore different intensities of emotion (using thermometer in CAT-kit) - Explain CBT model (in a very concrete way) Recognising the warning signs of anger	Family Storyboard (enhanced) (triggers, thoughts, feelings, behaviours) Worksheet: 'The early warning signs'
3	Applying CBT model to personal examples & starting to explore alternative ways of responding	• Review of homework Recent examples • Exploring the connections between thoughts, feelings, behaviours and body sensations for feeling angry and for relaxed • Using comic strip conversations to explore alternative ways of responding and 'what would make me feel better'	Family Storyboard (enhanced +) (triggers, thoughts, feelings, behaviours that make them feel better)
4	Behavioural Techniques	• Review homework • Behavioural techniques to manage anger (generate from child and family members) • Introduce a 'toolbox' (empty shoebox) to fill with tools to be used in conjunction with behavioural techniques.	Decorate toolbox Practice using a few techniques from the toolbox with encouragement from parents/teachers. Experiment to establish most effective and favourite

		Examples of tools: - Physical techniques (timeout, exercise, walking, other activities to release emotional energy) - Relaxation techniques (deep breathing, music, solitude, stress ball, rubrics cube, structured relaxing task/chore) - Family/teachers/others (reassurance, compliments, space)	techniques Note down difficulties to discuss next session.
5	Cognitive Techniques	• Review homework • Cognitive Restructuring Introduce cognitive techniques and alternative ways of thinking Practice the techniques, in combination with anger thermometer. Link alternative ways of thinking and responding with concrete examples of consequences (e.g., getting on well with parents, being able to join in favourite activity etc.) and also link alternative thoughts with alternative forms of responses	Practice Thought Record with 'alternative thought' column and 'alternative way of responding' column
6	Cognitive Techniques	Cognitive Restructuring	
7	Generalising the learning and relapse prevention		Family and teachers continue to encourage use of effective techniques Make techniques accessible (e.g., wallet size cards, sticky notes on fridge)
8	Blueprint & relapse prevention	Bringing it all together What makes me angry? What are the early warning signs? What makes me feel better? (behavioural) What makes me feel better? (Cognitive)	
9	Spill-over session		
10	Spill-over session		

Appendix 11: Reliable change criteria for the Strength and Difficulties Questionnaire (SDQ) and Parenting Stress Index-Short Form (PSI-SF)

Outcome Measure	Subscale (range of scores)	Mean	Standard Deviation (SD)	Standard Error	Reliability Coefficients (Cronbach's alpha)	Reliable Change Criterion	Clinically Significant Cut-off
SDQ	Total difficulties (0-40)	8.2	5.8	3.48	0.82	+/- 6.82	17
	Behaviour Difficulties (0-10)	1.5	1.7	1.46	0.63	+/-2.87	4
	Emotional Symptoms (0-10)	1.9	2.0	1.62	0.67	+/-3.18	5
	Hyperactivity (0-10)	3.2	2.6	1.76	0.77	+/-3.46	7
	Peer Problems (0-10)	1.4	1.7	1.58	0.57	+/-3.09	4
	Prosocial Behaviour 0-10)	8.6	1.7	1.42	0.65	+/-2.79	4
	Impact (0-10)	0.10	1.20	0.66	0.85	+/-1.29	2
PSI-SF	Total Impact (0-120)	114.17	13.93	3.11	0.95	+/- 8.63	
	Parent Distress (0-60)	32.17	6.11	1.93	0.90	+/- 5.36	
	Parent-Child Dysfunctional Interaction (0-60)	35.17	7.00	2.32	0.89	+/- 6.44	
	Difficult Child (0-60)	46.83	5.12	1.77	0.88	+/- 4.92	

Appendix 12: Annotated example of coding

1. What changes, if any, have you noticed in your child since the programme started?*

P: Um…yeah I have noticed, he is, he is calmer, all round I'd say he is a bit calmer, however, there is still issues to do with um, his anger. It's not completely resolved, but um…I think it sort of helped in terms of teaching me maybe more of a way of what- how to sort of go about things with him and discuss things and situations if they arise. Um…so it kind of in a way has given me a bit of confidence. It was… [I: Okay, that's really…] And what I was doing before, it wasn't so much as like really really changing what I was doing but sort of understanding that- how much it's helping him you know along the right lines of discussing things and going through [inaudible] needs to through a lot of um the injustices of what's happened and he really needs you to sort of understand how he's feeling rather than trying to tell him how things should be. It's helped in terms of that, definitely.

I: That's really good to know, that will be really helpful for our researchers. So I just wondered if you could explain to me what you mean by um how he's calmer, if you could explain a little bit more?

P: How he's calmer? He just seems um… to not get- he does get worked up about things, he still- he still does get angry but he's calmer in terms of, he hasn't been hitting himself and he hasn't been getting so so distressed like he was before. He gets distressed but it's easier to calm him down and it's easier to talk through rather than getting so angry.

I: I'm really pleased to hear that it's been so helpful for you.

P: Yeah, and it's a credit really a lot to (participant) as well, um… how he's able to sort of communicate um well with himself. So…

P1 YP is calmer

P1 Easier to talk through rather than getting angry

P1 still issues, not comletely resolved

P1 Teach [parent] new ways to deal with the problem

P1 Given [parent] confidence about ability to manage YP's difficulties

Category: CHANGE\Process of change
Code: P1 Given [parent] confidence about ability to manage YP's difficulties
Date: 21/03/2016
Coder: Admin

P1 needs you to know how he's feeling

P1 hasn't been hitting himself

P1 calmer, not hitting, talks things through easier

P1 hasn't been getting as distressed

P1 Easier to talk through rather than getting angry

Category: CHANGE\Positive changes\Systemic change/Communication
Code: P1 Easier to talk through rather than getting angry
Date: 21/03/2016
Coder: Admin

P1 child able to communicate with himself

P: Yes, there is um, change with me because um, I used to get frustrated as well, with dealing with all these issues. But I realised that I?m a bit calmer and I get to understand how to deal with this problem and the fact that I understand his problem make me a bit very calm you know? I?m okay with myself now, yeah.

P5 Parent gets less frustrated when dealing with [YP's] issues

P5 Parent feels calmer and gets less frustrated in dealing with YP's anger

P5 Understanding YP's problem helped parent feel calmer (less frustrate

And what <u>specific ideas</u>, if any, have you gotten from therapy so far?)

3. Thinking about your child, has anything changed for the <u>worse</u> for you since the programme started?*

P: For the worse? No, it has got better, from I start this programme, my entire life is a bit better compared to what it was, yeah.

P5 parent linking positive changes to starting the programme

Category: Therapy-change link
Code: P5 parent linking positive changes to starting the programme
Date: 21/03/2016
Coder: Admin

4. Thinking about your child, is there anything that you <u>wanted</u> to change that hasn?t since the programme started?*

P: To tell you the truth, the programme is- the programme is great. There?s nothing that I could think about that um, should change. Everything about the programme is fantastic. It?s the best opportunity I get for me and my son and I would recommend it to anyone, very very good.

P5 Wouldn't change anything about it. Would recommend it to anyone

Appendix 13: Initial stage of sorting codes into potential themes and subthemes

CHANGE — *Potential Theme*

Positive changes — *Potential subtheme*

- P1 easier to calm [child] down
- P1 child able to communicate with himself
- P1 not as angry when talking about incidents
- P8 Young person has learnt vocabulary to talk about emotions
- P8 YP has more vocabulary to talk about it
- P5 Increased awareness. when he get annoyed, he realises he is getting annoyed
- P2 difficult to conclusively describe the changes
- P2 YP seemed to take it more seriously, that she needed to not lose her temper
- P4 More aware of techniques she could use to help frustration
- P4 no longer worry about YP getting frustrated on trains

Improvements in behaviour/anger

- P1 calmer, not hitting, talks things through easier
- P1 hasn't been hitting himself
- P4 no frustration in class (short lived)
- P4 no more physical aggression in school
- P4 Behaviour at school improved, definitely
- P1 YP is calmer
- P5 Less angry and frustrated since the programme
- P8 Tantrums don't go on as long
- P8 Tantrums are less frequent
- P1 hasn't been getting as distressed
- P2 he is aware of his anger
- P3 more aware of his anger
- P4 Able to tolerate travelling on tubes/trains. Changed totally

Parent changes

- P4 Felt better able to cope, because we were doing better
- P5 Parent feels calmer and gets less frustrated in dealing with YP's anger problems

Systemic change/Communication

- P1 child more readily talking to parent after school about incidents
- P1 Easier to talk through rather than getting angry
- P8 As a family, talk about emotions rather than lose our tempers
- P3 Communication can tell parents when he is getting angry
- P8 As a family, we talk through it more and describe whats going on, rather than lose out tempers
- P1 not so angry and talking more to parent about it

Process of change

Ideas gained from the programme

- P8 Realised he didn't have much of a vocabulary to discuss [losing temper]
- P5 Never watched for signs or known why he upset. Now I learn about watchung for signs before he gets upset
- P1 Anger scale was helpful. Vocabulary for different levels of anger
- P2 Parent pointing out to YP early signs of anger
- P1 Teach [parent] new ways to deal with the problem
- P1 Given [parent] confidence about ability to manage YP's difficulties
- P8 Talking about/ awareness of the process and spiral of anger
- P8 being more conscious of the process [of losing our temper]
- P8 (as a family?) Learning a vocabulary to talk about anger
- P5 Understanding the need to watch the signs [of anger] before he gets angry
- P5 Parent learnt to stop talking to him if I see signs annd realise he's going to get angry
- P5 Parent gets less frustrated when dealing with [YP's] issues
- P5 Understanding YP's problem helped parent feel calmer (less frustrated)
- P5 Parent learnt to see early signs of anger and rather than carrying on talking, she now stops talking
- P5 Yp increased insight and awareness- when he get annoyed, he realises he is getting annoyed

Helpful processes

- P1 cognitive strategy Use phrase with child- life is full of better things to remember overall picture
- P8 talking about it, escalating nature of anger, what can do differently
- P1 feeling as if he is being understood and heard
- P1 acknowledging his feelings helps calm him down
- P5 Meeting someone that understands your child and talking about his problem, it makes it good
- P5 Someone to listen to him (his needs and what he is facing)
- P1 For parent to know they were doing the right thing already (open communication with YP)
- P1 Helped parent gain perspective
- P1 Reinforcing importance of hearing young person and understanding
- P1 Helpful for parent to know a few steps to use
- P1 Taking a wider perspective, a step back helped parent to be able to help young person
- P2 Giving importane to the problem, evem though didn't produce magical results
- P4 worked because new person to please
- P4 Using reporting improvements to therapist as motivator to find ways to calm down
- P4 Therapist as 3rd person for child to speak about dealing with incidents
- P1 Using the anger scale. Jointly learning language for different levels of anger and what to do at each level
- P2 The corse has played a part in giving her more awareness
- P4 more aware of techniques she could use to help frustration.
- P4 YP aware of behavioural strategy- stress ball
- P4 Awareness of cognitive strategy (think of happy place)
- P4 YP retained an awareness of behavioural (stress ball) and cognitive strategies (think of a happy place) she could use
- P4 using behavioural strategies to manage frustrations on public transport

Not completely resolved
- P1 still issues, not comletely resolved
- P1 Skills are still developing
- P2 not a major transformation of eveything suddenly being easier
- P2 Occasionally able to use techniques and strategies
- P2 Hasnt translated into permenant change
- P2 hasn't resulted in permenant change
- P4 still gets frustrated (shouting and screaming) when she thinks she can't do somthing
- P4 after programme finished, behaviours have come back
- P4 after programme, we've regressed
- P4 Improved communication stopped ater programme stopped
- P4 frustrations have come abck, less time in classroom

Novelty of content. Prior knowlege of content/strategies
- P1 what I was doing before, reassuring I was going along the right lines
- P1 feel better knowing I am doing the right thing. we are doing a good job
- P2 already knew contents of programme, nothing new
- P4 Learning new information/strateges
- P4 Parents learning something new that never tried before all of Sons life

Child's angry feelings need validating
- P1 needs you to know how he's feeling

Barriers
- P1 not learnt straight away, need for repetition
- P1 Parent isnt always there to help (e.g with peers)
- P1 fixed thinking, hard for child to take on board what parent is saying
- P1 takes time to establish habits, programme is short
- P3 Doesn't want to use toolbox. feels different from other kids
- P3 Didn't achive much in sessions. B followed his own agenda

Hopes
- P8 I'd like him to be more thoughtful about the way he comes across
- P2 Hope the programme has given her vocabulary and a way of approaching problem solving emotions

Related to ASD deficits
- P8 Hard for him to be more thoughtful about how he comes accross- theory of mind deficits
- P2 ASD means she doesnt have much control over how she reacts

- P2 difficult to direct from the outside, attributed to her ASD

Feasibility of measure
- P8 Unsure about accuracy of them
- P8 We travel alot and have lots on so hard to complete a measure for the whole week
- P5 Easier to understand tally chart. Couldn't understand PSI and SDQ
- P1 Unrealiable
- P1 Hard to complete tally when dealing with instances
- P1 would recap at the end of the week but nto 100! accurate
- P2 difficult to capture outbursts at school
- P3 difficult to trace numerous incidents.
- P3 too much going on
- P3 get frustrated by forms and get a skewed opinion of the day. Prefer a diary form to tick box forms.

Negative changes
- P8 Nothing changed for the worse
- P5 Everything has been positive
- P2 More outbursts but this attributed to onset of puberty
- P4 Nothings changed for the worse

Acceptability of programme

Expectations (not met)
- P2 Didn't work in the way I had hoped
- P2 Parent had not solution as to getting YP to engage in techniques at home
- P4 would have liked to have seen changes continue
- P1 Child got frustrated by being asked to write thoughts on paper, ripped up homework exercises
- P1 Sometimes felt rushed because so much to go through
- P1 Overloaded with too much (content of the programme)
- P2 Didn't work in the way I had hoped.
- P3 Wouldn't recommend if ADHD

Positive aspects
- P5 Wouldn't change anything about it. Would recommend it to anyone
- P4 YP Enjoyed creative aspect of programme. Craft activities and coloured pens
- P4 Agenda for each session
- P8 Good that it was directed at my Son, he hasn't had that yet
- P8 I think he benefited from talking about his emotions without worrying
- P8 Practical, focused on solutions not endlessly talking

Therapy-change link
- P5 Parent acknowledging reductions in problem behaviour since the programme
- P5 parent linking positive changes to starting the programme
- P5 From the programme, theres alot of changes with YP
- P5 YP started to open up more, to talk to us
- P5 THe change is within the therapy because...
- P5 its the signs I've learn within your thapy that make a change to my Son because rather than carrying on talking whilst he gets upset,
- P5 I (parent) learnt more by coming to the programme
- P1 Acknowledgement of his feelings that was made in teh therapy really makes a difference to him
- P3 Changes (awareness of anger) not caused by therapy

Feasibility of programme
- P1 Hard for parent to implement structured nature of technique in everyday life, so used in more scattered way
- P1 Alot to cover in short number of sessions. felt rushed, overwhelmed
- P1 Hard to put new ideas into practice when dealing with incidents in the moment
- P1 Overlaoded. realisitically can only implement so many strategies otherwise overloaded with too much
- P1 Sometimes felt rushed

Improvements to programme
- P1 A small booklet/printout to recap what was covered in sessions (tailor made)
- P1 Focus on 1-2 strategies and master one first

- P1 Therapist to pick out and recommend one strategy that is suited to you
- P2 Printed booklet
- P2 A diary, no guarantees she would fill it in though
- T2 address inattention would have been more helpful
- P2 Something to make it more visual for YP

Structurig of sessions
- P4 Intensive at start then longer gaps between sessions to review
- P4 sessions during term time
- P4 Initially weekly but then gradually less frequent lessons spread over longer period of time
- P1 wish for ongoing sessions?

Generalizing
- P4 Can explain the strategies she could use but won't use them
- P4 Imprvements with outbursts on tube, in classroom but not when thinks she can't do something

Motivation/ability to use techniques
- P2 had difficulty engaging with the techniques
- P2 Parent had no solution to getting her to engage in the techniques at home
- P3 Difficult to stay focused on topics. Inattention
- P3 inattention

Appendix 14: Note regarding author's background and theoretical orientation

I am a clinical psychology trainee in my mid-twenties. I have previously worked in an ASD assessment clinic which adopted a holistic and multi-systemic approach to interventions with this population. However, I had not implemented a behavioural or CBT-based intervention with this population prior to commencing this research project. I was familiar with CBT techniques and came to this project open-minded without any pre-conceived assumptions as to how CBT would work for this population. Given that I implemented the clinical work with the majority of families, I was to an extent invested in the idea of the intervention being successful. Thus, I was mindful of this when interpreting the qualitative results.