Thesis submitted in partial fulfilment of the degree of Doctor of Clinical Psychology (DClinPsych)

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The thesis I am submitting is entirely my own work except where otherwise indicated.

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ABSTRACTS

Systematic Review of the Literature

Differential Reinforcement to treat self-injurious behaviour in autistic children: A systematic review of methodology and outcomes

Purpose. Self-injurious behaviour (SIB) in autistic children and young people (CYP) is common and associated with harmful consequences. Differential reinforcement (DR) is a behavioural intervention which has been used to treat SIB. The aim of the current review was to report on (1) participant and intervention characteristics, (2) methodological issues, and (3) outcomes of DR interventions for SIB in autistic CYPs.

Methods. A systematic literature review was conducted to identify studies reporting DR interventions for SIB in autistic CYPs. Seven databases were searched (PsychINFO, Embase, Medline, Web of Science, Scopus, CINAHL, Biosis Citation Index). Data regarding the participant, study design, intervention characteristics, and study outcomes was extracted. Study methodology was assessed using the SCED-scale and the presence of a functional assessment, stimulus preference assessment and follow-up data. A narrative synthesis was used to integrate the results.

Results. Fourteen studies were identified, all of which used single-case experimental design methodology. There was variation with regards to study methodology with most studies rated as methodologically 'adequate'. The discussion of outcomes was structured by the DR schedule used and whether the DR intervention was presented in isolation or alongside other treatments. Outcome data was mixed, with the most robust evidence being for differential reinforcement of alternative behaviour (DRA).

Conclusions. There were some relative methodological strengths and limitations of studies investigating DR as an intervention for SIB. The variation in methodological quality makes it

difficult to make definitive conclusions regarding the efficacy of DR interventions as a whole, though DRA had the strongest evidence. Clinical implications of this are discussed.

Service Improvement Project

Outcomes and experiences of a parent-led CBT group for anxious autistic children in a Children and Young People's Mental Health Service in England

Objectives: Autistic children and young people (CYP) are at a higher risk of developing anxiety. Research suggests that the mechanisms underlying anxiety in autistic CYP may be different from the dominant cognitive behavioural theories of anxiety (South & Rodgers, 2017). Nonetheless, many services continue to use therapies developed for neurotypical CYP. The aim of this project was to examine the efficacy and service-user experience of a parent-led CBT group to treat anxiety in autistic CYP in a Child and Adolescent Mental Health Service (CAMHS) in England.

Methods: An audit of routine outcome measures (ROMs) and post-intervention status was completed for children whose parents had attended the parent-led group (n = 33). Semi-structured interviews were conducted with eight parents, and a thematic analysis was conducted to understand the service-user experience of the group. Stakeholders (experts-by-experience, clinicians, and researchers) were consulted at key stages throughout the project.

Results: The completion rate of ROMs data was poor, but descriptive analysis of this and the post-intervention status data suggests that the majority of CYP attending the group did not experience a clinically significant change in anxiety. Results of the semi-structured interviews were grouped according to four themes and five subthemes, suggesting overwhelmingly that parents felt that the group was not the right fit for their autistic CYP (theme 1) and that their motivation for doing the group was for something other than gaining

from the intervention (theme 2), but that there were some components of the group (theme 3) and therapy non-specific factors (theme 4) that were helpful.

Conclusions: The parent-led CBT group was found to not be effective and, in some cases, harmful to autistic CYP. This is consistent with the idea that mechanisms, and therefore treatment, for autistic anxiety may be different. Strengths and limitations are discussed, and service-specific recommendations are made.

Theoretically-Driven Research Project

Overprotective parenting and anxiety in children born with a cleft lip and/ or palate and their parents

Objective: There is some evidence that children born with a cleft lip and/ or palate (CL/P) and their parents are at a slightly increased risk of developing anxiety. The intergenerational model of anxiety (IMA) suggests that overprotective parenting, and parental appraisals may contribute to this risk. This study compared levels of overprotective parenting, child anxiety, and parent anxiety, between children born with a CL/P and a control group. For the CL/P group, exploratory analyses were conducted to assess parent and child factors related to anxiety in the CL/P group.

Methods: A cross-sectional design was used. Participants (n = 129) consisted of parents of 8 – 12-year-old children born with a CLP (n = 63) or without a CL/P (the 'control group'; n = 66) recruited through the community. Participants completed an online survey containing measures of overprotective parenting, child anxiety, parent anxiety, and appraisals of their child's CL/P (CL/P group only).

Results: Participants in the CL/P group reported comparable overprotective parenting and parental anxiety, but higher levels of child anxiety than in the control group. Child anxiety in the CL/P group was related to parental anxiety, overprotection and certain parental appraisals,

as well as gender. Parent anxiety in the CL/P group was related to parental overprotection and parental appraisals regarding self-blame.

Conclusions: These findings suggest that children with a CL/P are at an increased risk of anxiety, but that overprotective parenting is similar between children born with and without a CL/P. The clinical and research implications for the relationship between CL/P appraisals and child and parent anxiety are discussed.

PAPER ONE: SYSTEMATIC REVIEW OF THE LITERATURE

Differential Reinforcement to treat self-injurious behaviour in autistic children: A systematic review of methodology and outcomes

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Suggested Journal: This paper is currently being prepared for submission for publication in the Journal of Autism and Developmental Disorders (see appendix A for submission guidelines). This journal was chosen because it is a leading journal in the field of autism and developmental disabilities and encourages research investigating psychotherapeutic treatment efficacy for co-occurring difficulties in autism.

Abstract

Purpose. Self-injurious behaviour (SIB) in autistic children and young people (CYP) is relatively common and associated with significant and harmful consequences. Differential reinforcement (DR) is a behavioural intervention which has been used to treat SIB. The aim of the current review was to report on (1) participant and intervention characteristics, (2) methodological issues, and (3) outcomes of DR interventions for SIB in autistic CYPs.

Methods. A systematic literature review was conducted to identify studies reporting DR interventions for SIB in autistic CYPs. Seven databases were searched (PsychINFO, Embase, Medline, Web of Science, Scopus, CINAHL, Biosis Citation Index). Data regarding the participant, study design, intervention characteristics, and study outcomes was extracted. Study methodology was assessed using the SCED-scale and the presence of a functional assessment, stimulus preference assessment and follow-up data (Tate et al., 2008) A narrative synthesis was used to integrate the results.

Results. Fourteen studies involving fifteen participants were identified, all of which used single-case experimental design methodology. There was variation with regards to study methodology with most studies rated as methodologically 'adequate'. The discussion of outcomes was structured by the DR schedule used and whether the DR intervention was presented in isolation or alongside other treatments. Outcome data was mixed, with the most robust evidence being for differential reinforcement of alternative behaviour (DRA).

Conclusions. There were some key themes in the relative methodological strengths and limitations of studies investigating DR as an intervention for SIB. The variation in methodological quality makes it difficult to make definitive conclusions regarding the efficacy of DR interventions as a whole, though DRA had the strongest evidence. Clinical implications of this are discussed.

Introduction

Autism Spectrum Condition is a relatively common lifelong neurodevelopmental condition associated with strengths and difficulties. Strengths can include high attention to detail, a strong sense of justice, and a tendency to develop intense interests in certain areas (Chow & Cooper, 2024). Autistic[1] individuals can also experience difficulties in areas such as social communication and sensory processing. Social constructionist ideas highlight that these difficulties can arise or be made worse by inflexible social norms, structures and narratives which place the blame on the autistic individual when they do not fit with the neurotypical way of being (Milton, 2012; Oliver, 1983). Given these societal challenges, it is unsurprising that autistic individuals are likely to display behaviours that are considered challenging, conceptualised as culturally abnormal behaviours which harm the individual or others around them (NICE, 2014). Self-injury is a particularly concerning yet common behaviour observed in autistic individuals, with prevalence estimates suggesting that 42% of autistic individuals engage in self-injurious behaviour (SIB; Steenfeldt-Kristensen et al., 2020). SIB refers to any self-directed behaviour that has the potential to result in physical injury and includes head banging, self-biting, skin scratching, hair pulling, hitting oneself against hard objects and the ingestion of inedible items (Steenfeldt-Kristensen et al., 2020). SIB is associated with multiple medical complications, including soft tissue damage (e.g. wounds and infections from skin picking), musculoskeletal issues (e.g. broken bones, swelling joints), gastrointestinal problems, and brain injury (Cantin-Garside et al., 2021; Fields et al., 2021; Hof et al., 1991; Hyman et al., 1990). It is therefore unsurprising that, among autistic children and young people (CYP), SIB is associated with higher rates of

[1] Identity-first language (e.g. “autistic individual”) is used throughout this report as research suggests that it is preferred by autistic individuals as it reflects the nature of the condition as central to their personality (Kenny et al., 2016).

psychiatric hospitalisation and is the most common cause of emergency hospital admissions (Kalb et al., 2012; Mandell, 2008). Psychosocially, SIB is associated with poor educational engagement, loneliness and isolation for the individual (Cantin-Garside et al., 2021; Miller & Misher, 2016) and poorer quality of life and lower perceived general health amongst parents and siblings (Hanson, 2024).

Historically, there have been ethical issues surrounding the management and treatment of SIB in autistic individuals. The Serious Case Review of Winterbourne View Care home in 2012 brought to light the institutional abuse and overuse of psychotropic medication, often prescribed to manage SIB in the absence of any underlying mental health need (Department of Health, 2012). This review led to several guidelines and programmes, including the STOMP-STAMP initiative, Transforming Care agenda, and the NHS long-term plan, all of which aim to reduce institutionalisation and overmedication whilst improving psychosocial and community support services for autistic individuals (with or without a co-occurring learning disability; Branford et al., 2019; Department of Health, 2012; NHS England, 2019). The current best practice guidelines for supporting autistic individuals therefore recommend that SIB be managed and treated using psychosocial interventions informed by a functional assessment of behaviour (NICE, 2013).

Differential reinforcement (DR) is one such behavioural-based procedure which involves selectively reinforcing specific behaviours whilst withholding reinforcement for other behaviours (Cooper et al., 2007). There are several DR procedures which have been applied to reduce SIB. Differential reinforcement of other behaviour (DRO) involves providing reinforcement contingent on the absence of SIB for specific intervals of time. Differential reinforcement of alternative behaviour (DRA) involves reinforcing a behaviour that is functionally equivalent to SIB. An example of DRA for a child engaging in head banging for sensory stimulation would be to reinforce an alternative method of achieving the

stimulation (e.g. bouncing, swinging, or spinning). Differential reinforcement of incompatible behaviour (DRI) involves selectively reinforcing any behaviour that is incompatible with SIB. For a child engaging in self-biting, a DRI intervention might involve reinforcing the use of a toy that is safe to bite. Differential reinforcement of low rates of behaviour (DRL) aims to reduce the frequency SIB without eliminating it altogether by providing reinforcement contingent upon a pre-determined low rate of the behaviour (Cooper et al., 2007). For a child who engages in skin scratching (a behaviour which is considered normal in moderation), DRL would involve only reinforcing a pre-determined rate of skin scratching which is deemed typical.

Most of the research investigating DR procedures adopts a single-case experimental design (SCED) which involves the systematic manipulation of an independent variable (e.g. DR) and measuring the impact of this on the dependent variable (e.g. SIB). By the use of a baseline phase, whereby the dependent variable is measured in the absence of independent variable, the participant acts as their own comparison which allows for the control of confounding variables (Matson, 2023). SCED designs allow clinicians and therapists to share valuable knowledge and experience, and are considered an important springboard for developing hypotheses and research questions for future studies (Wallace et al., 2022). However, there are some methodological issues that need to be considered when conducting SCED research on behavioural interventions. Tate et al (2008) describe many of these methodological considerations which include using a precise, repeatable and operational definition of the target behaviour, using a design that allows for cause-and-effect relationships to be examined (i.e. introducing one treatment component at a time, and using reversal designs, described as ‘the cardinal rule’), providing measures of inter-rater reliability (IRR), and assessing the generalisation of results. Other potential methodological considerations include (1) the use of functional assessment (FA), (2) the use of a stimulus preference

assessment (SPA), and (3) inconsistent follow-up data (Chowdhury & Benson, 2011; Schrader et al., 1983; Tate et al., 2008). With regards to the first issue, functional assessments (FAs) aim to understand the relationship between SIB and the environment and are based on the assumption that SIB is maintained because it serves a function to the individual (Iwata et al., 1994). The main hypothesised functions for SIB are (a) escape from demands, (b) social attention, (c) access to tangible items, and (d) automatic, whereby SIB serves an internal or sensory function to the individual (Iwata et al., 1994; Matson & Minshawi, 2007). Understanding the function of SIB allows for the intervention to be functionally equivalent to the problem behaviour (Cooper et al., 2007). For example, an individual engaging in SIB for automatic reinforcement is unlikely to respond to a DRO intervention providing access to tangible items contingent upon a lack of SIB for a period of time, however, they might be likely to benefit from a DRA procedure whereby they learn alternative, safer, means of receiving the sensory stimulation (Cooper et al., 2007). FAs can take many forms including experimental functional analyses (EFA) whereby possible maintaining variables are systematically manipulated and the impact on SIB is measured. They can also be completed via questionnaire, checklist or descriptive methods (e.g. ABC charts; Matson & Minshawi, 2007). FA's are considered a vitally important component of behavioural interventions, with a meta-analysis finding that the use of an FA is the only variable that predicts success of behavioural interventions (Didden et al., 1997). Despite this, in a review investigating DR procedures for 'problem behaviour' in intellectually disabled adults, only 32% of included studies used an FA (Chowdhury & Benson, 2011).

The second methodological issue relevant to behavioural interventions is the use of stimulus preference assessments (SPA). The success of any DR protocol is dependent upon the efficacy of the reinforcer. For example, providing praise and hugs as a reinforcer to a child who finds social attention and physical affection aversive would result in even the most

well designed and implemented DR procedure to be ineffective and would lead to the assumption that the intervention itself was ineffective when, in fact, it may have been effective if an appropriate reinforce had been used. Therefore, predicting the most valuable reinforcer is critically important to the success of any behavioural intervention (Matson, 2023). However, the lack of an SPA is commonly reported, with a systematic review finding that only 10% of studies investigating DR for problem behaviour in intellectual disabilities included an SPA (Chowdhury & Benson, 2011). SPA methods can be categorised as either indirect (via survey or interviews with parents or care staff) or direct (whereby an individual is observed engaging with stimuli that is presented in a systematic manner; Matson, 2023).

The third methodological issue pertains to the issue of follow-up data which is reportedly missing from half of studies investigating DR as an intervention for 'problem behaviour' in intellectual disabilities (Chowdhury & Benson, 2011). Whilst a DR intervention may be deemed efficacious during the treatment period itself, a lack of follow-up data prevents any conclusions regarding the social and clinical validity of findings.

The present review comes after other related reviews on the topic of DR which have found some evidence for the use of DR procedures in autistic and/or intellectually disabled individuals in clinical and applied settings (Chowdhury & Benson, 2011; MacNaul & Neely, 2018; Muharib & Walker, 2024; Petscher et al., 2009; Weston et al., 2018). In a review of DR interventions targeting 'problem behaviour' in adults with intellectual disabilities, Chowdhury & Benson (2011) concluded that there is reasonable evidence to support the use of DR procedures in these circumstances, however that there exist significant methodological flaws which limit the generalisability of findings.

The aim of the current review, therefore, is to examine the state of DR research specifically targeting SIB in autistic CYP, using the following questions:

1. What are the participant characteristics, target behaviours and intervention characteristics for SCED studies investigating DR interventions for SIB in autistic CYP?
2. What is the quality of the research and what are the common methodological issues? This question will be assessed via (a) the use of a validated quality appraisal tool and (b) a descriptive synthesis of the methodological issues pertinent to behavioural SCED research (i.e. use of functional analysis, use of SPA, and follow-up data).
3. What are the outcomes of studies involving DR interventions targeting SIB in autistic CYPs and can meaningful conclusions be drawn about the efficacy of DR interventions with SIB?

Methods

The review was registered on PROSPERO (ID: CRD42022315616) and the Preferred Reporting Items for Systematic Reviews and Meta Analysis guidelines (PRISMA; Moher et al., 2009) were used to inform the methods and reporting of the review (see appendix B).

Eligibility

Inclusion criteria were studies (a) published in English; (b) including participants who were autistic CYP aged 17 or under (c) using a differential reinforcement procedure which was explicitly named as DRA, DRO, DRI or DRL, either independently or as part of a behavioural intervention package (BIP), (d) treating SIB and, (e) published in peer-reviewed journals or as a doctoral thesis.

Exclusion criteria were (a) book chapters, existing reviews, conference abstracts or opinion pieces, (b) studies exclusively using an intervention other than DRA, DRO, DRI or

DRL (this includes functional communication training; FCT[2]), (c) studies targeting a behaviour other than SIB, (d) where the participant was 18 or over (e) where the participant did not have a formal diagnosis of autism and, (f) studies published in a language other than English.

Search strategy

A comprehensive search was conducted in May 2025 using PsychInfo, Embase Medline, Web of Science, Scopus, CINAHL and Biosis Citation Index. The search terms used to search abstracts, titles and key concepts were as follows:

((“autis*” OR “Asperg*” OR “ASD” OR “ASC”) AND (“Child*” OR “Adolescen*” OR “Teen*” OR “Young person” OR “Young people” OR “Youth”) AND (“Differential reinforcement” OR “DRO” OR DRI” OR“DRL” OR “DRA”)).

Selection Process

A title and abstract screen was conducted by the first author and was duplicated by a second rater for 20% of studies, with excellent agreement between the two raters ($\kappa = 0.88$). The first author completed a full-text screen which was carried out in duplicate by the second rater for 25% of the papers. This resulted in excellent agreement ($\kappa = 0.94$). The PRISMA diagram for the selection process is displayed in figure 1.

Data extraction

The following data was extracted[3]: (1) location, (2) participant characteristics (number of eligible participants, age, sex, ethnicity, comorbid diagnoses) (3) target behaviour, (4) study design, (5) study setting, (6) the interventionist, (7) the specific DR approach used and (8) any other interventions it was used alongside, (9) the use of an FA, (10) the use of an SPA

[2] A decision was made not to include FCT as part of this review for the following reasons; (1) FCT is considered a variation rather than an essential form of DRA (Matson, 2023), (2) a clinician consultation highlighted that FCT is considered distinct to DRA in clinical practice and, (3) the presence of an existing review investigating FCT to treat SIB in autistic CYP (Tejada-Flores & Paredes-Gonzales, 2024).

[3] This data can be provided upon request to the first author.

and, where no SPA was conducted, information on the reinforcer, (11) intervention procedures, (12) interobserver agreement methods and result and, (13) results and, (14) whether DR data was presented in isolation (Y/N). Data for 50% of the studies was extracted in duplicate, with excellent inter-rater reliability ($\kappa = 0.94$).

Quality Assessment

The SCED-Scale was used on the basis that it was developed specifically for SCED research and it has shown excellent IRR for both individual and consensus ratings (see appendix C; Tate et al., 2008). The SCED-scale is an 11-item scale where each item is rated as present or absent, and a total score of 0-10 is derived (item 1 is not included in the total score). Each study was assigned a quality description based on the SCED-scale score: studies scoring between 0-3 were classified as 'weak', studies scoring between 4-6 were classified as 'adequate', and scores between 7-10 were classified as 'strong' (Tate et al., 2008). Study quality was assessed by two independent raters and there was excellent agreement ($\kappa = 0.95$).

Data synthesis

Data for all research questions was presented as a descriptive synthesis. This was deemed the most appropriate method for research questions one and two, and the lack of raw data or effect size calculations within the studies meant that a meta-analysis or reporting of effect size calculations would not be possible for research question three.

For research question two, data was synthesised according to the SCED-scale and other important methodological variables. For research question three, studies were grouped according to the DR procedure used and discussions were focused around (a) studies that present DR data in isolation (allowing for stronger causal attributions to be made about the efficacy of DR interventions) and, (b) studies that combine DR data with that of other behavioural interventions.

Results

Identification of studies

As shown in figure 1, the initial search identified 1320 studies which, after the removal of 670 duplicates, led to the initial title and abstract screening of 650 studies. After the exclusion of 416 studies at this stage, a total of 234 papers were sought for retrieval and 232 papers were reviewed in full. A total of 14 papers were identified for inclusion in the final review.

Research question 1. Characteristics of participants, target SIB and the intervention

The review included 14 studies published between 1985 and 2024, including fifteen participants. Table 1 presents the characteristics of participants, target SIB and the intervention. Participants ranged in age from four to seventeen, and 66% (n = 10) of participants were male. All participants were autistic, and the majority had at least one other neurodevelopmental, physical or mental health diagnosis (n = 9) with the most common diagnosis being intellectual disability (n = 5). Ethnicity data was not consistently reported. Target behaviours were pica (n = 4), self-biting (n = 3), self-hitting/ punching (n = 3), skin picking (n = 2), head banging (n = 1), joint dislocation (n = 1), and operant vomiting (n = 1). Most interventions took place in the participant's home (n = 6), followed by school/ day treatment centres (n = 3), residential settings (n = 2), behavioural clinics (n = 2) or was not reported (n = 2). Data on the interventionists was limited, with most papers referring to 'the therapist/ experimenter' without describing qualifications or professions. DRA was the most used intervention (n = 7), followed by DRO (n = 5), and DRL (n = 1) and DRI (n = 1).

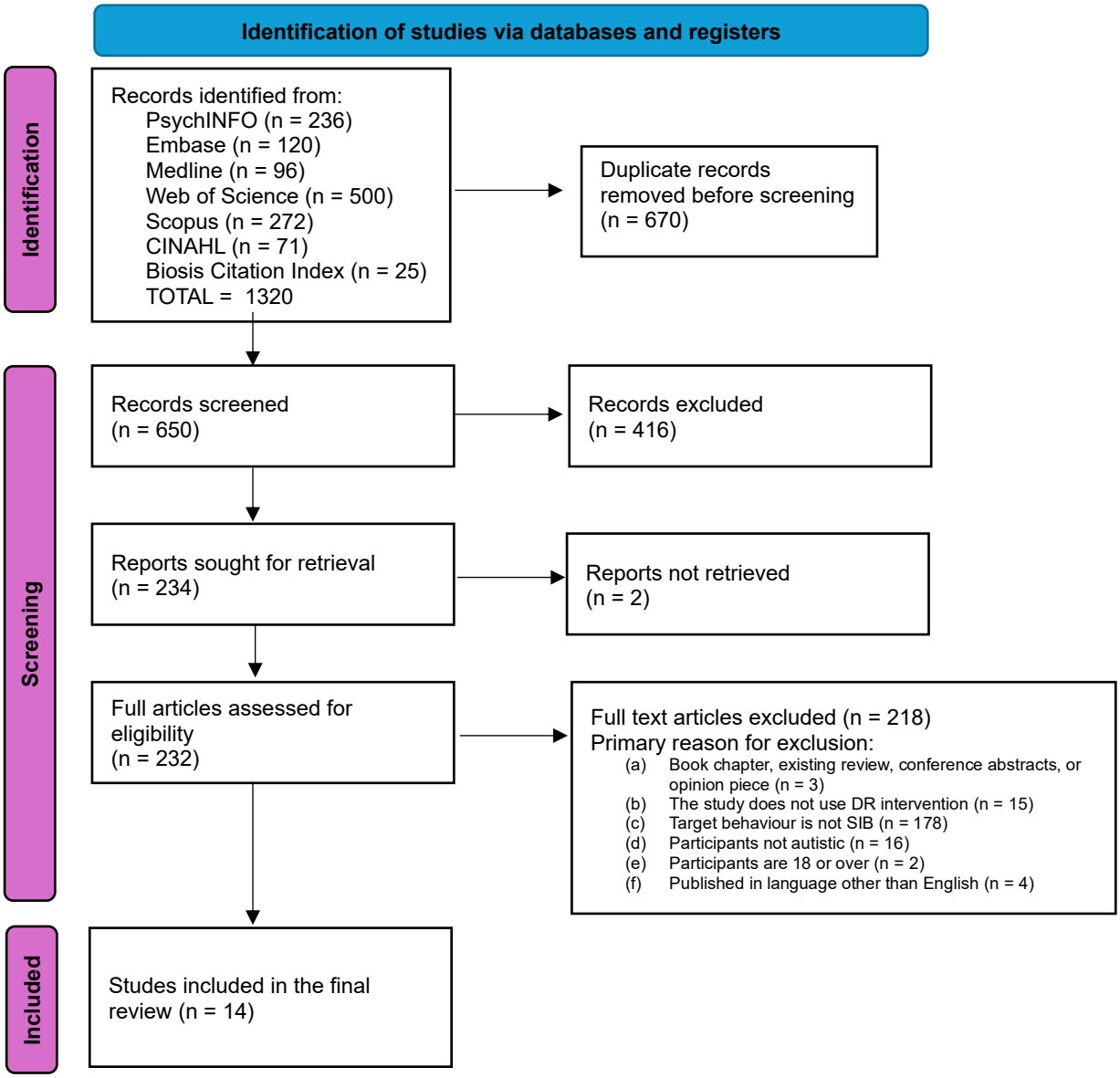

Figure 1. PRISMA diagram detailing search strategy.

Research question 2. Quality appraisal and methodological critique

The quality appraisal and methodological critique will be guided by (1) the SCED scale (Tate et al., 2008) and (2) the other important methodological variables for behavioural SCED research, namely the use of FA, SPA, and follow-up reporting.

The SCED Scale

As shown in table 2, the total SCED scores of included studies ranged from 2-7. Most studies (n = 9; 64%) were classified as having adequate methodologies, a further 21% (n = 3) were classified as having strong methodologies and the remaining 14% (n = 2) were classified as methodologically weak.

Clinical History. Just under one third of studies (2, 4, 12, 13) reported sufficient information regarding the clinical history of the participant (29%; n = 4). The lack of reporting on ethnicity by the other ten studies was the primary contributing factor to them not meeting this criteria, and is therefore a significant limitation of the generalisability of findings in these studies.

Table 1. Study Characteristics

Abbreviations: DRO – Differential reinforcement of other behaviour. DRA – Differential reinforcement of alternative behaviour. DRL – Differential reinforcement of low rates of behaviour. DRI – Differential reinforcement of incompatible behaviour. USA – United States of America. CRT – Competing Response Training. FAIR-T - Functional Analysis Informant Record for Teachers. FAOF - Functional Assessment Observation Form. BIRS – Behaviour Interview and Reinforcement Survey. QABF - Questions about Behavioural Function. MSTO – Movement suppression time out. FM-DRO – fixed-momentary DRO. VM – variable-moment DRO. FAST – Functional Analysis Screening Tool. PSPA – Paired stimulus preference assessment. NCR – Non-contingent reinforcement. MAS – Motivation assessment scale. MSWO – Multiple-stimulus without replacement. RAISD – Reinforcer Assessment for Individuals with Severe Disabilities. VSR – Variable schedule of reinforcement. OCD – Obsessive compulsive disorder. RIRD – Response interruption and redirection. PSPA – Paired stimulus preference assessment. CSA – Competing stimulus assessment.

Study ID (Authors, year)	Location	Participant characteristics	Target behaviour(s)	Study setting	Intervention ist	Intervention Components	Study design	DR treatment data presented in isolation? (Y/N)	Functional Assessment (Y/N, method and results)	Stimulus Preference Assessment (Y/N, method)	Inter-rater Reliability	Results & Conclusions
DRO												
1. Cavalari et al., 2014	USA	• n = 1 • Sex: F • Age: 17 • Diagnoses: Autism • Ethnicity: not reported	Skin picking	School	Classroom staff (inferred: profession/ training not reported)	DRO CRT	Reversal design with fading component. A-B-A-C (inferred) A= baseline B = Treatment (DRO + CRT) C = Fading Follow-up: "many weeks later"	No (DR data for fading is presented in isolation but not for treatment)	Yes Questionnaires: FAIR-T (teacher), FAOF (classroom staff), BIRS (self-report). Informal parent interview Hypothesised function: Escape and automatic	Yes Indirect: Questionnaire - BIRS (self-report). Informal parent and classroom staff interview.	Yes IRR calculated for 8% of sessions. Results: Agreement on SIB = 75%. Agreement on non-occurrence of SIB: 100%.	DRO + CRT (B): ⇓ SIB. Reversal to baseline (A): ⇑ SIB. Treatment fading and follow-up (C): ⇓ SIB. Conclusion: The treatment package was effective and durable over time. It is unclear as to the unique contributions of DRO in the treatment phase. When DRO was implemented in isolation for the fading component, treatment gains were maintained.
2. Lang et al., 2009	USA	• n = 1 • Sex: F • Age: 17 • Diagnoses: Aspergers, Borderline Intellectual Disability • Ethnicity: White	Skin picking	Home (data collected at monthly clinic visits).	Mother and self-delivered	DRO Antecedent control (bandages)	Reversal design with fading component. A-B-A-C A= Baseline B = Intervention (DRO + Antecedent control) C = Fading Follow-up: 4 months	No	Yes Informal interview with participant and her mother. Questionnaire: QABF (mother). Hypothesised function: Automatic	No (reinforcer was arbitrary)	Yes IRR calculated for 100% of data points. Results: Agreement on SIB = 100%	DRO + Antecedent control (B): ⇓ SIB to 0. Reversal to baseline (A): ⇑ SIB. Treatment fading: Initial ⇑ SIB followed by ⇓ SIB to 0. Follow-up: 0 SIB. Conclusion: The treatment package was effective and durable over time. Unclear about the unique contribution of DRO.
3. Rolider & Van Houten., 1985	Canada	• n = 1 • Sex: M • Age: 9 • Diagnoses: Autism • Ethnicity: Not reported	Biting self and mouthing objects (often swallowing)	Home	Parents	DRO Punishment (MSTO)	A multiple baseline across behaviours design and follow-up. A-B-BC (inferred) A = Baseline B = DRO C = MSTO Follow-up: 57 days.	Yes	No	No (reinforcer was arbitrary)	Yes IRR calculated twice during each condition. Results: Agreement on SIB = 95%	DRO alone (B): No change in SIB DRO + punishment (BC): ⇓ SIB to near zero levels. Follow-up: 0 SIB Conclusion: DRO is not effective on its own. The treatment package (DRO + punishment) was effective. Unclear whether DRO provides any additional benefit over and above punishment.

Study ID (Authors, year)	Location	Participant characteristics	Target behaviour(s)	Study setting	Intervention ist	Intervention Components	Study design	DR treatment data presented in isolation? (Y/N)	Functional Assessment (Y/N, method and results)	Stimulus Preference Assessment (Y/N, method)	Inter-rater Reliability	Results & Conclusions
4. Wilder et al., 2023	USA	• n = 1 • Sex: F • Age: 11 • Diagnoses: Autism & Turner Syndrome • Ethnicity: White	Banging chin against objects	Home	Experimenter (profession not/ training not reported)	DRO (FM-DRO was compared against VM-DRO)	Multielement reversal design Initial baseline (A) was followed by alternating FM-DRO (B), VM-DRO (C) and baseline (A) conditions.	Yes	Yes Questionnaire: FAST (caregiver). EFA (screening for automatic reinforcement only, no other conditions) Hypothesised function: Automatic	Yes Direct and indirect (PSPA guided by caregiver interview)	Yes IRR calculated for 30% of sessions across each condition, Results: Agreement on SIB = 95.6%	FM-DRO and VM-DRO: ⇓ SIB compared to baseline Conclusions: DRO schedules (both FM-DRO and VM-DRO) are effective in reducing SIB.
5. Baker et al., 2010	USA	• n = 1 • Sex: M • Age: 8 • Diagnoses: Autism, mild esophagitis, stomach ulcers, extensive tooth decay. • Ethnicity: not reported	Operant Vomiting	Not reported	Parent/ caregivers (inferred)	DRO Punishment (time-out, contingent mouthwash) Extinction (visual screen)	Design not named. Procedure as follows: • DRO + time-out • Punishment • Extinction • Follow-up (4 months).	No	Yes EFA (inferred) Hypothesised function: Automatic	Not completed (reinforcer not described)	Not completed	DRO+ time-out: Not change in SIB Contingent mouthwash + extinction: ⇓ SIB Conclusion: DRO and time-out were not effective in reducing SIB.
DRA												
6. Cummings & Carr, 2005	USA/ Canada	• n = 1 • Sex: M • Age: 8 • Diagnoses: Benign Joint Hypermobility Syndrome, 'autistic disorder'. • Ethnicity: Not reported	Joint dislocation	Home	A trained adult therapist (profession not reported)	DRA NCR FCT Escape Extinction Curricular Revision	Multiple treatment reversal design with generalisation. A-B-C-A-D-BC-A-E-F-A-BF-BCDEF A = Baseline B = NCR C = FCT D = Extinction E = Curricular Revision F = DRA Follow-up: 8 months. Generalisation: New setting and therapist	Yes	Yes EFA Questionnaire: MAS (parents and nanny). Hypothesised function: Escape (and possibly automatic).	Yes Direct and indirect: Direct observation, MSWO preference assessment, RAISD questionnaire (father).	Yes IRR calculated for 30% of sessions Results: Agreement on SIB = 95%	DRA only (F): ⇓ SIB comparable to other treatment components. NCR+DRA (BF): Even more effective in ⇓ SIB. Combination of all components (BCDEF): ⇓ SIB to near zero levels. Generalisation: SIB remained low and participant was re-admitted to school. Follow-up: Successful maintenance. Conclusions: DRA is effective but the combined treatment package was required to eliminate SIB altogether.
7. Devlin et al., 2011	Ireland	• n = 1 • Sex: M • Age: 9 • Diagnoses: Autism, Moderate-severe learning disability • Ethnicity: Not reported	Biting fingers	School	Experimenter (student completing MA in Applied Behavioural Analysis under supervision)	DRA VSR Extinction	Alternating treatments design (BIP compared to SIT), with initial baseline and final best treatment phase.	No	Yes Questionnaire: FAST-revised (not reported who completed it) Hypothesised function: Escape and access to tangible items.	No (reinforcer matched the function)	Yes IRR calculated for 20% of sessions. Results: Agreement on SIB was 98%	BIP: ⇓ SIB Conclusions: BIP including DRA is effective. Unclear about unique contributions of DRA.

Study ID (Authors, year)	Location	Participant characteristics	Target behaviour(s)	Study setting	Intervention ist	Intervention Components	Study design	DR treatment data presented in isolation? (Y/N)	Functional Assessment (Y/N, method and results)	Stimulus Preference Assessment (Y/N, method)	Inter-rater Reliability	Results & Conclusions
8. Fritz et al., 2017	USA	• n = 1 • Sex: M • Age: 7 • Diagnoses: Autism, OCD. • Ethnicity: Not reported	Hitting self	Day Treatment Centre	Therapist (Profession/ training not reported)	DRA NCR	Nonconcurrent multiple baselines across participants design A-B-C-BD (inferred) A = Baseline B = NCR C= NCR thinning D =DRA	No	Yes EFA <u>Hypothesised function</u>: Access to tangible items	No (reinforcer was function matched)	Yes IRR calculated for 18% to 100% of sessions during each condition. Results: Agreement for SIB in baseline = 92% Agreement for SIB in NCR = 99% Agreement for SIB in NCR + DRA = 98%.	NCR (B): ⇓ SIB to 0 NCR thinning (C): ⇑ SIB. NCR+DRA: ⇓ SIB to 0 NCR thinning+ DRA: 0 SIB <u>Conclusion</u>: BIP including DRA + NCR is effective in reducing SIB. DRA was an essential component to ensure that NCR thinning was effective.
9. Mitteer et al., 2015	USA	• n = 1 • Sex: F • Age: 6 • Diagnoses: Autism, Pica • Ethnicity: Not reported	Pica	Not reported	Therapist (profession/ training not reported)	DRA Punishment ('facial screen').	Intervention reversal design A-B-C-CD-C-CD A & B = Baselines C = DRA D = Punishment	Yes	Yes EFA (screening for automatic reinforcement only) <u>Hypothesised function</u>: Automatic	No (reinforcer was function matched)	Yes IRR calculated for 33% of sessions. Results: Agreement for SIB = 98%.	DRA alone (C): No change in SIB DRA+ Punishment (CD): ⇓ SIB Reversal to DRA alone (C): ⇑ SIB. Reversal to DRA + Punishment: (CD): ⇓ SIB <u>Conclusion</u>: DRA alone was not effective. DRA + punishment was effective. Not clear whether DRA added any unique benefits to punishment alone.
10. Slocum et al., 2017	USA	• n = 1 • Sex: F • Age: 13 • Diagnoses: Autism, Pica • Ethnicity: Not reported	Pica	Behaviour Analysis Research Clinic	Therapist (profession/ training not reported)	DRA	Reversal design A-B-A-B A: Baseline B: DRA	Yes	Yes EFA screening (for automatic reinforcement only) <u>Hypothesised function:</u> Automatic	No (reinforcer was function matched)	Yes IRR calculated for 20.6% of sessions. Results: Agreement for SIB = 97.5%	DRA(B): ⇓ SIB Reversal to baseline (A): ⇑ SIB. Reversal to DRA(B): ⇓ SIB to low levels. <u>Conclusion</u>: DRA was effective in reducing SIB to low levels. Reversal design allows for inferences about causality to be made.
11. Taylor, 2020	Australia	• n = 1 • Sex: M • Age: 4 • Diagnoses: Autism, Pica, food selectivity • Ethnicity: Not reported	Pica	Home	A trained, doctoral-level behaviour analyst	DRA RIRD	Multielement design with reversal to baseline. Summary of procedures: • Baselines (x 2) • RIRD+DRA • Reversal to baselines • RIRD+DRA • Generalisation • Follow-up (3 months)	No	Yes EFA <u>Hypothesised function</u>: Automatic	Yes Direct (PSPA and CSA).	Yes IRR calculated for 38% of sessions across phases and conditions. Results: Agreement for SIB = 95% to 100%	RIRD+DRA: ⇓ SIB Reversal to baseline: ⇑ SIB. Reversal to RIRD+DRA: ⇓ SIB Generalisation & follow-up: SIB remained low <u>Conclusion</u>: RIRD+DRA was effective in reducing SIB. Unclear as to whether DRA provided any unique effects.

Study ID (Authors, year)	Location	Participant characteristics	Target behaviour(s)	Study setting	Intervention ist	Intervention Components	Study design	DR treatment data presented in isolation? (Y/N)	Functional Assessment (Y/N, method and results)	Stimulus Preference Assessment (Y/N, method)	Inter-rater Reliability	Results & Conclusions
12. Thomas et al., 2024	USA	• n = 1 • Sex: M • Age: 7 • Diagnoses: Autism, Pica, Sickle Cell Disease • Ethnicity: Black	Pica	Outpatient behavioural psychology clinic	Parents (supervised by clinical psychologist)	DRA RIRD Punishment (Response cost) FCT	Reversal design with follow-up. A-B-A-B-C (inferred) A= Baseline B = DRA + RIRD+ Punishment C = DRA + RIRD + Punishment + FCT D = Generalisation Follow-up: 1-7 months.	No	Yes EFA Hypothesised function: Automatic	Yes Direct (MSWO preference assessment)	Yes IRR calculated for 40.4% of sessions. Results: Agreements for SIB = 96.5%.	DRA + RIRD + Punishment (B): ⇓ SIB Reversal to baseline (A): ⇑ SIB. Reversal to DRA + RIRD + Punishment (B): ⇓ SIB to near zero levels DRA + RIRD + Punishment + FCT: SIB remained at near zero Generalisation & follow-up: SIB remained at near zero Conclusions: Treatment package containing DRA is effective in reducing SIB. Reversal design allows for inferences around causality. Not clear about the unique role of DRA.
DRL												
13. McClean & Grey, 2012	Ireland	• n = 2 • Sex: M (P1 & P2) • Age: P1 - 15, P2 - 17 • Diagnoses: Severe intellectual disability (P1 & P2), ADHD (P1), Bipolar depression (P2) Ethnicity: Not reported	Hand biting (P1) Punching head, hand biting (P2)	Residential placement	Not reported	DRL Antecedent control ('low arousal environment', 'predictability') Rapport building (e.g. follow, name, wait praise protocol). FCT	Multiple element baseline design A-B-BC-BCD-BCDE-BCDEF A: Baseline B: Antecedent control (low arousal environment) C: Rapport building D: Antecedent control (predictability) E: FCT F: DRL Follow-up: 3 years.	No	Yes Descriptive functional assessments. Questionnaire: QABF (completed by frontline keyworkers) Hypothesised function (P1 & P2): Escape	No (reinforcer not reported)	Yes P1: IRR checks completed for a minimum of 30 minutes each week during baseline (A) and low arousal (B), at 9 months and follow-up. Results: Agreement on SIB was 94% to 100% P2: IRR checks completed for 30 minutes every day at baseline and 9 months. Results: Agreement on SIB was 88% to 100%.	P1: BIP was effective in ⇓ SIB. Addition of DRL to BIP (comparison of BCDE and BCDEF): ⇓ SIB P2: BIP was effective in ⇓ SIB. Addition of DRL to BIP (comparison of BCDE and BCDEF): No difference in SIB Follow-up (P1 & P2): Low rates of SIB remained. Conclusions: The addition of DRL to BIP resulted in lower SIB for one participant, but had no effect for the other.
DRI												
14. Underwood et al., 1989	USA	• n = 1 • Sex: M • Age: 12 • Diagnoses: 'Profound mental retardation' Ethnicity: Black	Hitting head	Residential placement	Not reported	DRI Mild interruption	Design not reported. Procedures as follows: • Mild interruption • Mild interruption + DRI • Mild interruption • Mild interruption +DRI	No	No	No (reinforcer was arbitrary)	Yes IRR calculated for 29% of sessions Results: Agreement on SIB was 94%	Mild interruption + DRI: ⇓ SIB compared to mild interruption alone Conclusions: DRI in addition to mild interruption was an effective intervention for SIB. It is assumed that DRI provided unique benefits when compared to mild interruption alone.

Target Behaviour. All but two studies defined the target behaviour with operational precision. Of the two studies that did not, one study (2) measured the target behaviour (skin picking) by proxy (number of skin sores) which is not considered a sensitive nor reliable measure. The other study (13) did not provide the operational definition of the SIB.

Design. Around one fifth of papers (21%) (n = 3) met this criterion by using a reversal design (4, 6, 10). The remaining 11 papers combined intervention components as part of a BIP which prevents conclusions regarding individual components. It is worth noting that two studies (3, 9) partly adhered to this criterion by introducing DR in isolation but then failed to introduce other behavioural interventions in isolation. These studies therefore allow for the unique contributions of DR interventions to be assessed.

Baseline. Most studies (n = 11) established an adequate baseline measure of the target behaviour. Of the studies that did not meet this criteria, one study (2) only established two baseline measures which is not considered to be enough of a reliable baseline. The other two studies did not report baseline data (5, 14).

Sampling behaviour during treatment. Thirteen of the studies met this criterion by employing a continuous measure of behaviour during treatment. One study (2), however, provided monthly data sampling which is not considered robust to normal variation.

Raw data *record.* All studies provided a representation of the variability of data by displaying data in graphical form.

IRR. All but one study, (5), reported some attempt at establishing IRR, with the number of sessions for which IRR was calculated ranging from 8 – 100%. Two studies (3, 13) did not provide information about proportion of sessions for which IRR was calculated. IRR was above 80% (considered strong), with the exception of one study (1) where it was 75% (considered moderate; Fink, 2010).

Independence of assessors. Two studies reported utilising independent assessors (11, 14) either by having a trained observer record behaviour live and via videos (11), or by having an observer seated behind a one-way mirror to record target behaviour. For the remaining studies, data was collected by the interventionist which may result in biases or errors in data collection (Tate et al., 2008).

Statistical analysis. None of the studies included in this review reported any statistical analysis of effect size calculations, thus preventing any conclusions about the significance or size of effects.

Replication. Over one third of the included studies (36%, n = 5; 3, 4, 7, 8, 13) demonstrated replication of treatment effects to other participants in the study (even if these participants did not meet inclusion criteria of this review).

Generalisation. Four studies made attempts to establish or measure generalisation. Two studies (6, 12) established generalisation to new settings and therapists, one study established that treatment effects generalised to actual pica items (as opposed to 'baited' pica items used during treatment; 11), and one study demonstrated that the participant's quality of life and mental health status had improved (13).

Other Methodological Variables

Functional assessment. Twelve of the fourteen included studies (86%) used some form of FA. Of these twelve, five studies (42%; 5, 6, 8, 11, 12) utilised a full EFA and one of these also utilised a parent questionnaire (6). A further 3 studies (4, 9, 10) reported apriori hypotheses that the target behaviour was automatically reinforced and therefore completed an 'EFA screening for automatic reinforcement'. Four studies utilised questionnaires, either in isolation (7, 13) or accompanied by interviews with parent (1) or participants (2).

Stimulus preference assessment. Five studies reported completing a SPA (1, 4, 6, 11, 12). Of these, two used a direct SPA (11, 12), one used an indirect SPA (1) and two used a

combination of direct and indirect (4, 6). Where no SPA was completed, information about the reinforcer chosen suggests that reinforcers were function-matched for four of the studies (7, 8, 9, 10), and arbitrary in three of the studies (2, 3, 14). Three of the studies did not report any information about the reinforcer (5, 13).

Follow-up. Over half (n = 8) of the studies reported follow-up data. The length of follow-up varied from 3 months (2, 11), 4 months (5), 7 months (12), 8 months (6), and 3 years (13). Two studies did not report on the specific amount of time for follow-up (1, 3).

Table 2. SCED-Scale scores and qualitative descriptions.

Note: Item 1 is not included in the total score. Brackets in item three indicate that DR interventions were implemented in isolation and so causal inferences about DR can be made, but that other behavioural interventions were added to the DR intervention without having been presented in isolation first and, thus, these studies do not adhere to the 'cardinal rule'.

Study ID (Authors, year)	1. Clinical history	2. Target behaviours	3. Design	4. Baseline	5. Sampling behaviour during treatment	6. Raw data record	7. Inter-rater reliability	8. Independence of assessors	9. Statistical Analysis	10. Replication	11. Generalisability	TOTAL SCORE	Qualitative description of quality
1. Cavalari et al., 2014		✓		✓	✓	✓	✓					5	Adequate
2. Lang et al., 2009	✓					✓	✓					2	Weak
3. Rolider & Van Houten., 1985		✓	(✓)	✓	✓	✓	✓			✓		6	Adequate
4. Wilder et al., 2023	✓	✓	✓	✓	✓	✓	✓			✓		7	Strong
5. Baker et al., 2010		✓			✓	✓						3	Weak
6. Cummings & Carr, 2005		✓	✓	✓	✓	✓	✓				✓	7	Strong
7. Devlin et al., 2011		✓		✓	✓	✓	✓			✓		6	Adequate
8. Fritz et al., 2017		✓		✓	✓	✓	✓			✓		6	Adequate
9. Mitteer et al., 2015		✓	(✓)	✓	✓	✓	✓					5	Adequate
10. Slocum et al., 2017		✓	✓	✓	✓	✓	✓					6	Adequate
11. Taylor, 2020		✓		✓	✓	✓	✓	✓			✓	7	Strong
12. Thomas et al., 2024	✓	✓		✓	✓	✓	✓				✓	6	Adequate
13. McClean & Grey, 2012				✓	✓	✓	✓			✓	✓	6	Adequate
14. Underwood et al., 1989	✓	✓			✓	✓	✓	✓				5	Adequate

Research question 3. What are the outcomes of studies involving DR interventions targeting SIB in autistic CYPs?

Differential Reinforcement of Other Behaviour (DRO)

DRO data presented in isolation. Two studies (3, 4) found conflicting results for the use of DRO in isolation. One study (3) found that DRO in isolation was ineffective for the treatment of self-biting in an autistic nine-year old male but, when combined with a punishment procedure (movement suppression time out[4]) led to a reduction in self-biting which was maintained at follow-up. Ethical issues arise from the fact that the rationale for using punishment is not described in the study, and it is not clear whether guidelines regarding the use of punishment were followed (Pokorski & Barton, 2021). This study was classified as methodologically adequate, but there was no report of an FA or SPA, and the reinforcer chosen was arbitrary. It is therefore possible that the inefficacy of the DRO intervention was due to inappropriate reinforcers and/ or the treatment not targeting the function of the behaviour. These methodological limitations pose a significant threat to the validity of the conclusions that DRO alone was not effective.

The other study (4) implementing DRO in isolation provides more methodologically sound evidence, as it is classified as 'strong' and includes the use of an FA and SPA. This study found that two different DRO schedules were equally as effective in reducing the chin-banging of an eleven-year-old girl. There was no follow-up data, so it is not clear how effective this intervention was over time.

DRO data combined with other behavioural interventions. A total of three studies (1, 2, 5) used DRO alongside other interventions, and found mixed results. One study (1) found that DRO in combination with competing response training (CRT) was effective in the treatment of skin-picking in a 17-year-old autistic female. Results were maintained during a fading

[4] Movement suppression time out involved the participant being guided to stand facing the corner for two minutes contingent upon self-biting

component where DRO was presented in isolation and the reinforcement schedule was thinned, and at follow-up ‘many weeks later’. The reversal design strengthens the conclusion that the BIP was effective in reducing SIB. The study was classified as methodologically adequate, and the inclusion of a FA and SPA also strengthen the validity of the findings. Another study (2) concluded that DRO in combination with an antecedent control was effective in reducing the number of skin-picking wounds in a seventeen-year-old autistic female. This study was classified as methodologically weak and, whilst an FA was completed, the reinforcer was arbitrary and did not reflect the proposed function of the behaviour. Caution is therefore recommended when interpreting the results of this study. Finally, another study also classified as methodologically weak (5), reported that DRO in combination with time-out (punishment) was ineffective in the treatment of operant vomiting of an eight-year-old autistic male. Similarly to study 3, this study raises ethical questions regarding the use of punishment procedures (Pokorski & Barton, 2021). The use of an EFA is not clear, and no SPA is reported. It is therefore possible that the inefficacy of the DRO intervention is, in part, due to inappropriate reinforcers. These methodological flaws significantly limit the validity of any conclusions from this study.

Differential Reinforcement of Alternative Behaviour (DRA)

DRA data in isolation. The three studies using DRA in isolation (6, 9, 10) report mixed findings. In a multiple treatment reversal design, one study (6) used a BIP and adhered to the ‘cardinal rule’ by only introducing one component at a time. Results found that DRA was the most effective individual treatment at reducing levels of joint dislocation in an eight-year-old autistic male, but that a combination of all individual interventions was the most effective, leading to near zero levels of SIB. This study was methodologically strong, and included an EFA, SPA, and an 8-month follow-up where treatment gains were maintained.

Two studies reported the use of DRA in isolation for the treatment of pica. One study (9) used a reversal design which demonstrated that DRA in isolation was not effective but, when combined with a punishment procedure ('visual screen'[5]), it reduced pica. Similarly to the other studies describing punishment procedures, there was no evidence that guidelines had been followed with regards to the use of punishment (Pokorski & Barton, 2021). On the SCED scale, this study was classed as adequate. It included a screening EFA (for automatic reinforcement only) but did not report an SPA, though the reinforcer was classified as function-matched which is considered superior to an arbitrary reinforcer. The lack of follow-up prevents conclusions about the efficacy of the intervention over time.

The other study (10) used a reversal design to show that DRA was an effective stand-alone intervention for the treatment of pica in a 13-year-old autistic female. Methodologically, this study was classified as adequate and included an EFA screen for automatic reinforcement, but did not report an SPA, though the reinforcer was function matched. Follow-up data was not reported so it is not clear how robust the results were over time.

DRA data combined with other behavioural interventions. Four studies combined DRA data with that of other behavioural interventions (7, 8, 11, 12). All these studies found that DRA in combination with other behavioural interventions was effective in reducing SIB.

One study (7) found that a BIP (which included DRA, variable schedules of reinforcement, and extinction) was more effective than a sensory integration training package in reducing finger biting in a nine-year old autistic male. It is unclear about the unique contribution of DRA within the intervention. The study was classed as methodologically adequate. It did report that an FA was completed in the form of a questionnaire. No SPA was reported but the

[5] Visual screen involved hands being placed over the participant's hands and face, covering her eyes for 30 seconds.

reinforcer was classified as function matched. There was no follow-up data reported so it is unclear as to the long-term efficacy of the BIP.

One study (8) investigating the use of NCR and DRA in the treatment of self-hitting in a 7-year-old autistic male found that NCR alone was effective in eliminating SIB, but that DRA was required to be able to thin the NCR schedule of reinforcement (when NCR thinning occurred without DRA, rates of SIB increased). Whilst this study did not adhere to 'the cardinal rule' (i.e. DRA was not presented independently), it does provide strong evidence that DRA provided an essential component allowing the NCR schedule to be thinned, though it is not clear how robust these findings were over time. The study was classified as methodologically adequate and included a full EFA. No SPA was conducted but the reinforcer was function matched.

Another study (11) reports the use of DRA and response interruption and redirection (RIRD) in the treatment of pica in a four-year-old autistic male. A reversal design found that DRA and RIRD in combination reduced pica. These results generalised to actual pica items (as opposed to the baited items used in previous conditions) and pica remained low at three-month follow-up. This study was methodologically strong and included a full EFA and SPA. Finally, another study (12) investigated the use of a BIP in the treatment of pica of a seven-year-old autistic male. This study used a reversal design and found that a BIP including DRA, RIRD, and punishment (response cost) was effective in reducing pica to near zero levels. This study generalised results to a new setting and therapist, and results were maintained at a seven-month follow-up. However, of note, the use of punishment was not justified and there was no evidence that guidelines were followed for this (Pokorski & Barton, 2021). Methodologically, this study was classified as adequate and he inclusion of an EFA and direct SPA provide further confidence in the methodology.

Differential Reinforcement of Low Rates (DRL)

Only one study (13) used a BIP where treatment components including antecedent control, FCT, and DRL, were sequentially added to treat hand-biting and/ or head-banging in two autistic males. DRL was the final component to be added. For one participant (P1) DRL reduced levels of SIB, whereas for the other participant (P2) it had no effect. Treatment effects of the BIP were generalised to measures of quality of life and social inclusion, and follow-up data suggests that BIP provided long-term effects. Methodologically, this study was classified as adequate. It reported the use of questionnaires as an FA but did not use an SPA or report on the reinforcer used.

Differential Reinforcement of Incompatible Behaviour (DRI):

Only one study (14) investigated DRI for the reduction of SIB in autistic CYP. This study combined DRI with mild interruption in the treatment of head hitting in a 12-year-old autistic male. There was no baseline condition, but the authors concluded that mild interruption alone was not effective. When DRI was combined with mild interruption, a reduction in SIB was observed. It is therefore assumed that DRI provides some added benefit when compared to mild interruption alone. This study was classed as methodologically adequate but is limited by the fact that it did not include a functional assessment, SPA or follow-up data.

Discussion

This systematic review aimed to investigate the methodology and outcomes of DR interventions in the treatment of SIB for autistic CYP. All fourteen included studies used a SCED but there was substantial variation in terms of methodological rigour and outcomes. However, there were some key themes with regards to methodological strenghts and limitations which could inform clinical practice and research.

Relative methodological strengths included the fact that most studies reported sufficient information on the operational definition of the target behaviour, baseline data, sampling of behaviour, presentation of data, and IRR measures. The most prevalent methodological limitation, present in all studies, was the lack of statistical analyses or effect size calculations, making it difficult to quantify the size of the treatment effect, and to compare this between studies. It is strongly recommended that future SCED research investigating DR interventions utilise one of the recommended effect size statistic, such as PND (percentage of non-overlapping data; Mastropieri & Scruggs, 1985) or, for interventions targeting SIB, PZD (percentage of zero data; Scotti et al., 1991).

The use of the interventionist as the data collector for the majority of studies risks biased or incorrect data reporting which represents another significant methodological limitation (Tate et al., 2008). It could be assumed that independent data collection was often not possible due to most studies being carried out in real-life settings often by parents or care staff and where the resource and equipment for secondary data collection are less available. Nonetheless, it is recommended that future SCED research attends to the risk of data collection bias by having an independent data collector be present in sessions or watching via video recording (Taylor, 2020; Underwood et al., 1989).

A further methodological limitation of most studies was the use of designs which do not allow for causal inferences to be made. Namely, most studies using a BIP did not adhere to 'the cardinal rule' and, thus, did not allow for the efficacy of individual treatment components to be established. Whilst this is reflective of clinical practice, whereby the individual need may warrant the use of a BIP as opposed to a singular intervention, it remains one of the largest limitations of behavioural SCED research (Matson, 2023). To obtain the most definitive evidence regarding the efficacy of DR interventions in this area, it is recommended that DR interventions are implemented in isolation and, where a BIP is

indicated, the ‘cardinal rule’ be adhered to. Where BIPs are used, it is recommended that an alternating treatment design (ATD) is used as this not only answers the question of whether an intervention is effective, but also which intervention is most effective (Follette, 2001).

The issue of generalisation and follow-up was relatively neglected by many of the included studies which calls into question the robustness and social validity of interventions that were regarded as successful. One of the hallmarks of a successful behavioural intervention is that it the outcome has been generalised to new contexts, behaviours and other aspects of the individuals life with a view to improving long-term maintenance and social validity (Arnold-Saritepe et al., 2023). This issue is of particular importance given the difficulties that some autistic individuals have generalising knowledge and skills (Arnold-Saritepe et al., 2023). It is therefore recommended that future research investigating DR interventions for SIB in autistic CYP build in a generalisation and follow-up procedure into the protocol.

It was promising to find that 86% of the included studies completed an FA, and that close to half of those completed an EFA which is considered to be the gold standard of FA (Matson, 2023). There was an emerging trend where, in three of the more recently published studies, an ‘EFA screening for automatic reinforcement’ was used, which involved applying the ‘alone’ condition of an EFA, in the absence of other conditions. This EFA procedure is recommended as an efficient alternative to a full EFA where there is an existing suspicion that SIB is automatically reinforced, and has been found to have good predictive validity (Querim et al., 2013). Given the significant role that FA’s play in predicting the success of behavioural interventions, the finding here that the majority of studies used some form of FA provides confidence in the results, and caution is recommended when interpreting the results of studies that did not use an FA (Didden et al., 1997).

A final methodological limitation was the lack of SPAs, with just over one third of studies reporting an SPA. This figure is lower than a similar systematic review, finding that only 10% of studies included an SPA, but it remains concerning that so few studies pay attention to the value of the reinforcer (Chowdhury & Benson, 2011). It is argued that an intervention is only as effective as the reinforcer chosen, and failure to identify a highly valued reinforcer for a DR intervention threatens the validity of results, particularly where no effect is found, because it is not clear if the intervention was not effective or just that the reinforcer selected was not valued by the individual (Karsten A.M. & Carr J.E., 2009; Matson, 2023).

There was variability amongst studies with regards to study outcomes. Outcomes for DRO were mixed, but when taking into account study quality, the two studies that found no effect for DRO did not complete an EFA or an SPA and it is therefore possible that the inefficacy of DRO was due to an inappropriate reinforcer and/ or the treatment not targeting the function of the behaviour. The DR procedure with the strongest evidence was DRA, with six of the seven studies finding that DRA interventions either in isolation or in combination with a BIP were effective in reducing SIB. DRA has been described as 'the ideal intervention' because it aims to provide the individual with an alternative, safer means of accessing function-matched reinforcement and therefore inherently considers long-term maintenance (Petscher et al., 2009). This is evidenced in this review by the fact that all the included study's using DRA report the use of an EFA, and reinforcers were either identified via an SPA or were classified as function matched. Translating to clinical practice, these findings suggest that DRA may be a promising intervention for SIB in autistic CYPs.

Both DRL and DRI procedures were only used by one study each, both of which found some support for the use of DR procedures.

Strengths and limitations

This systematic review is the first to examine the methodological quality and outcomes of DR procedures treating SIB in autistic CYPs. The variation in study quality, alongside the limited availability of data investigating the specific and unique contributions of DR to BIPs limits our ability to draw firm conclusions, but it is hoped that the findings and recommendations provide a foundation for future research and clinical practice.

The inclusion and exclusion, particularly the requirement that studies explicitly name which differential reinforcement procedure they are using (i.e. DRO, DRA, DRL, DRI) may have limited the findings. This is because DR is considered a relatively common foundational behavioural intervention procedure and, as such, some studies were noted to make reference to having used DR but did not present enough information for the specific procedures to be inferred, and were therefore excluded (Matson, 2023).

Another significant limitation of this review includes the fact that it includes only SCED research, which not only raises methodological issues as previously discussed, but also raises concerns regarding publication bias. Research has found that SCED studies with small effect sizes are less likely to be submitted and accepted for publication (Shadish et al., 2016), and that behavioural intervention SCED's in particular are subject to issues with publication bias (Sham & Smith, 2014)

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PAPER TWO: SERVICE IMPROVEMENT PROJECT

Outcomes and experiences of a parent-led CBT group for anxious autistic children in a Children and Young People's Mental Health Service in England

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Suggested Journal: This paper is currently being prepared for submission for publication in Psychology and Psychotherapy: Theory, Research and Practice (see appendix D for submission guidelines). This Journal was chosen because it focuses on work which has direct implications for the practice of psychological therapy, and the target audience is practicing therapists as well as researchers.

[6] At the time of conducting this project, Dr Alice Farrington was the Principal Clinical Psychologist in the CAMHS Anxiety and Depression Team, Children Young People and Families, Berkshire Healthcare NHS Foundation Trust.

Abstract

Objectives: Autistic children and young people (CYP) are at a higher risk of developing anxiety. Research suggests that the mechanisms underlying anxiety in autistic CYP may be different from the dominant cognitive behavioural theories of anxiety (South & Rodgers, 2017). Nonetheless, many services continue to use therapies developed for neurotypical CYP. The aim of this project was to examine the efficacy and service-user experience of a parent-led CBT group to treat anxiety in autistic CYP in a Child and Adolescent Mental Health Service (CAMHS) in England.

Methods: An audit of routine outcome measures (ROMs) and post-intervention status was completed for children whose parents had attended the parent-led group (n = 33). Semi-structured interviews were conducted with eight parents, and a thematic analysis was conducted to understand the service-user experience of the group. Stakeholders (experts-by-experience, clinicians, and researchers) were consulted at key stages throughout the project.

Results: The completion rate of ROMs data was poor, but descriptive analysis of this and the post-intervention status data suggests that the majority of CYP attending the group did not experience a clinically significant change in anxiety. Results of the semi-structured interviews were grouped according to four themes and five subthemes, suggesting overwhelmingly that parents felt that the group was not the right fit for their autistic CYP (theme 1) and that their motivation for doing the group was for something other than gaining from the intervention (theme 2), but that there were some components of the group (theme 3) and therapy non-specific factors (theme 4) that were helpful.

Conclusions: The parent-led CBT group was found to not be effective and, in some cases, harmful to autistic CYP. This is consistent with the idea that mechanisms, and therefore treatment, for autistic anxiety may be different. Strengths and limitations are discussed, and service-specific recommendations are made.

Introduction

Autism is a lifelong neurodevelopmental difference manifesting in strengths and some difficulties. Strengths often observed in autistic[7] individuals include detailed-oriented patterns of thinking, a strong sense of justice, and the ability to develop highly focused interests and skills (Chow & Cooper, 2024). Autistic individuals are also likely to experience difficulties in areas of social interaction, communication and sensory processing (American Psychiatric Association, 2013). Autism is associated with various co-occurring internalising and externalising difficulties and it is commonly these difficulties that lead many families to seek help from NHS services, rather than the condition itself (Rodgers et al., 2017). Anxiety is the most commonly co-occurring mental health difficulty experienced by autistic individuals, with prevalence research suggesting that 40% of autistic CYP meet criteria for an anxiety disorder (Simonoff et al., 2008; Vasa et al., 2020). This co-occurring anxiety has been found to be a stronger predictor of quality of life in autistic individuals than the core characteristics of autism (Adams et al., 2020; Adams & Emerson, 2020; Ozsivadjian et al., 2012).

Research investigating the mechanisms underlying this increased risk suggests that autistic CYP are more likely to experience co-occurring anxiety disorders, such as those outlined by the DSM ("traditional anxiety"), *and* are at an increased risk of so-called "atypical anxiety" manifestations which are thought to be associated with features of autism itself (Kerns et al., 2014). Mechanisms underlying traditional anxiety presentations have been well researched with the general consensus suggesting that they are caused and maintained by a combination of cognitive factors (e.g. a tendency to overestimate threat and underestimate

[7] Research has found that autistic people prefer to use identity-first language because they view autism as a defining feature of their personality and feel that using person-first language (e.g. 'person with autism') diminishes the positive aspects of autism, and perpetuates the idea that autism is somehow 'wrong' (Kenny et al., 2016). We have therefore chosen to use identity-first language throughout this report.

one's ability to cope), physiological experiences of anxiety via activation of the autonomic nervous system, and behavioural responses (e.g. responding to these symptoms by avoidance and employing safety behaviours; (Creswell, Parkinson, et al., 2017; López et al., 1999). The experience of atypical anxiety has been less comprehensively researched. One model suggests that intolerance to uncertainty (IU) is a key factor moderating the relationship between autism-related factors (sensory sensitivity, alexithymia, rigidity of thought) and anxiety. This atypical anxiety model suggests that autism is marked by sensory over- and under-responsivity which increases uncertainty for external and internal stimuli, thus contributing to IU which, itself, leads to increased anxiety. These sensory processing differences are thought, in part, to contribute to the experience of alexithymia (difficulties understanding and labelling emotions) and this leads to confusion and uncertainty about emotional experiences, consequently increasing anxiety (Griffin et al., 2016; South & Rodgers, 2017). This model suggests that rigidity of thought (cognitive inflexibility) also contributes to IU by creating uncertainty about whether rules will be followed, leading to further anxiety (South & Rodgers, 2017).

In a community sample of autistic CYP, it is estimated that 15% present with atypical anxieties, 17% present with traditional anxieties and a further 31% experience a combination of traditional and atypical anxiety (Kerns et al., 2014). The distinction between these types of anxiety is important because it dictates the treatment approach that should follow. For traditional anxiety disorders, there is a wealth of evidence suggesting that adapted CBT approaches are effective in reducing anxiety in autistic CYPs (Perihan et al., 2020). For atypical anxieties, however, there is evidence to suggest that where IU is high, disorder-specific CBT interventions are less efficacious and, instead, interventions directly addressing IU may be more effective (Keefer et al., 2017; Rodgers et al., 2017). There is, at present, a

lack of research or guidance on how to intervene when an individual experiences a combination of traditional and atypical anxieties.

Relevant Guidelines and Legislation

The Autism Act was brought into legislation in 2009 and was followed in 2021 by a strategy for children and young people, focusing on increasing awareness, reducing health and care inequalities and increasing access to community services, amongst other things (*The Autism Act*, 2009). Ten years after the Autism Act was put into legislation it was reviewed by the All Party Parliamentary Group on Autism (APPGA) who recommended that community-based mental health services include autism-adapted, accessible counselling and low-level psychological therapy, with a view to intervening early in the development of mental health problems (All Party Parliamentary Group on Autism, 2019). This report also recommended not excluding individuals from services on the basis of an autism diagnosis alone.

Service Context

This project was commissioned by the Anxiety and Depression (A&D) pathway of a regional CAMHS service. The A&D pathway is a Getting More Help level service providing CBT interventions for children and young people with diagnoses of anxiety disorders, depression and/ or PTSD (Wolpert et al., 2014). For children under the age of 12 with an anxiety presentation (or older if it is felt that appropriate to their developmental level), the A&D pathway usually offer a low-intensity parent-led CBT-based group intervention, 'Helping Your Child with Fears and Worries' (HYC; see appendix E). When delivered for neurotypical populations, HYC has been found to be a clinically- and cost-effective treatment for anxiety disorders in children aged between five and twelve years old (Creswell, Violato, et al., 2017).

The A&D pathway receives a high number of referrals for autistic children and a significant majority (86%) of those attending the HYC group are parents of autistic children (diagnosed or suspected). As the service is commissioned to treat children with a traditional anxiety disorder (i.e. as diagnosed by the DSM-5), it is assumed that those autistic children whose parents attend the HYC group are likely to be experiencing a combination of traditional and atypical anxieties. The service is under significant resource pressure, with average waiting times for individual treatment being over two years whilst the waiting time for the HYC group is three to four months. The service therefore offers the HYC group as a means of early intervention but have recognised that it may not be meeting the needs of autistic children.

Research questions

The aim of the project was to understand the extent to which the group was meeting the needs of anxious autistic children and to make recommendations to the service about what changes could be made to improve the service provision for this group. The project addressed the following two research questions:

Research question 1: How effective is the HYC group is in treating anxiety in children with diagnosed or suspected autism?

Research question 2: What are the experiences of parents of autistic children of the HYC group?

Methods

Design

The project employed a mixed methods design, with research question one using quantitative methods and research question two using qualitative methods.

Participants

To address the first question, routinely collected data was gathered from the electronic health records of children whose parents had (1) attended one of the previous four HYC groups and, (2) who had diagnosed or suspected autism. This resulted in a total participant pool of 33 children with pre-intervention ages ranging from 9-15 years. Table 1 displays the participant characteristics.

To address the second research question, the thirty-three participants were contacted by clinicians in the A&D team to discuss the project. Fifteen participants gave consent for their contact details being shared with the trainee clinical psychologist. Of these, three parents agreed but then did not attend the interview, three parents declined to take part and one parent could not be contacted. This resulted in a total of eight parents completing the semi-structured interviews. Characteristics of these participants are displayed in table 2.

Table 3. Participant characteristics for research question 1 (n = 33)

		Percentage of participants (number of participants)
Gender	Male	61% (n = 20)
	Female	39% (n = 13)
Diagnostic status	Diagnosed	73% (n = 24)
	Suspected (on waiting list for autism assessment)	27% (n = 9)
Ethnicity	White British	76% (n = 25)
	White – Any other white background	6% (n = 2)
	Mixed – White Asian	3% (n = 1)
	Mixed – any other mixed background	3% (n = 1)
	Asian or Asian British - Indian	3% (n = 1)
	Not recorded	9% (n = 3)
Group attended	Group 1: May 2021	24% (n = 8)
	Group 2: November 2021	18% (n = 6)
	Group 3: January 2022	15% (n = 5)
	Group 4: September 2022	42% (n = 14)

Table 4. Participant characteristics for research question 2 (n = 8)

		Percentage of participants (no. of participants)
Gender	Male Female	50% (n = 4) 50% (n = 4)
Diagnostic status	Diagnosed Suspected (on waiting list for autism assessment)	63% (n = 5) 38% (n = 3)
Ethnicity	White British Mixed – White Asian Not recorded	63% (n = 5) 25% (n = 2) 12.5% (n = 1)
Group attended	Group 1: May 2021 Group 2: November 2021 Group 3: January 2022 Group 4: September 2022	38% (n = 3) 13% (n = 1) 25% (n = 2) 25% (n = 2)

Ethics and Governance

The project was considered audit and, as such, NHS approval was not required. The researcher had an honorary contract with Berkshire NHS Foundation Trust (BHFT) where the project was taking place and, as such, had legitimate access to the data. A data protection impact assessment (DPIA) was completed (see appendix F). Informed written consent was gathered for all participants.

Measures

Research question 1

The *Revised Children's Anxiety and Depression Scale-Parent Version (RCADS-P)* is a 47-item questionnaire measuring anxiety and depression in CYP and is routinely used by the service to assess outcomes of the HYC group. The RCADS-P shows strong internal consistency and convergent validity. It has shown good discriminant validity and is able to discriminate well between different traditional anxiety disorders (Ebesutani et al., 2010). Pre- and post-intervention scores of the 'total anxiety' subscale of the RCADS-P were available on electronic health records for nineteen of the thirty-three participants.

Post-intervention status was obtained by reviewing electronic health records and coding the status of the child after the group as either (1) discharged from CAMHS ('Discharged'), (2) waiting for individual treatment in the A&D team ('Treatment Waiting'), (3) 'Referred to Another CAMHS Service' (4) 'Moved Out of Area' or (5) receiving concurrent 1:1 CBT which continued post-intervention ('Concurrent Individual Treatment').

Research question 2

A semi-structured interview schedule (see appendix G) was co-produced with clinicians in the A&D team and Professor Cathy Creswell, whose team developed the HYC intervention.

Procedure

For research question one, assistant psychologists in the A&D team collated the pre- and post-intervention RCADS-P scores and the post-intervention status from the health records of the thirty-three participants. For research question two, eight semi-structured interviews were conducted over Microsoft Teams. The interviews varied in length from 30 to 78 minutes. The researcher listened back, transcribed and then pseudonymised the interview data.

Data analysis

Owing to the small participant numbers, and the issue of missing raw data, it was not possible to conduct formal statistical analyses. Quantitative data for research question 1 is therefore analysed descriptively.

For research question 2, qualitative data was analysed using reflexive thematic analysis methodology, using the six stages outlined by Braun et al (2019). Data analysis was approached from a deductive perspective where the researcher's existing knowledge and experience guided the development of codes and themes (Braun & Clarke, 2006). The

researcher initially coded the data and identified preliminary themes using NVivo 12. Three experts-by-experience (parents of autistic children with anxiety) recruited through the service's participation panel were consulted to review these preliminary codes and themes. Following this, all three researchers met to review and refine final themes.

Development of recommendations

The three experts-by-experience provided consultation on the translation of themes into recommendations. The primary author met with clinicians in the service to present the findings and co-produce the recommendations.

Researcher perspective

The first author (RB), who conducted and analysed the qualitative data, is a 30-year-old white British female who has a professional and personal interest in the mental health of autistic CYP. RB had previous experience delivering the HYC intervention on a 1:1 basis with parents of autistic children, and had some pre-determined ideas based on both experience and theory about the experience of parents of autistic children of the HYC group. RB kept a reflective log throughout the data collection and analysis process and also conducted bracketing interviews (see appendix H).

Results

Research question 1

RCADS-P

Pre- and post-intervention RCADS-P total anxiety scores were available for nineteen of the thirty-three participants, characteristics of whom are displayed in table 3. The scoring programme used by the service does not specify t-scores above 80. As the majority of t-scores were >80, it is not possible to make any meaningful comparisons between individual t-scores and we therefore report on clinical severity as opposed to individual scores. As shown

in figure 1, pre-intervention RCADS-P scores suggest that seventeen of the total nineteen children were experiencing severe anxiety, meeting the clinical cut-off for an anxiety disorder before completing the intervention. Post-intervention data, displayed in figure 2, suggests that this did not change, with seventeen of the total nineteen participants' RCADS-P scores being classified as severe at post-intervention. Seventeen of the nineteen participants had the same pre- and post-intervention RCADS-P clinical severity classification. One participant's classification went from severe (pre-intervention) to normal (post-intervention), and another participant's classification went from borderline (pre-intervention) to severe (post-intervention).

Table 5. Characteristics of participants for whom RCADS-P total anxiety data is reported (n = 19)

		Percentage of participants (number of participants)
Gender	Male	63% (n = 12)
	Female	37% (n = 7)
Diagnostic status	Diagnosed	74% (n = 14)
	Suspected (on waiting list for autism assessment)	26% (n = 5)
Ethnicity	White British	79% (n = 15)
	Mixed – White Asian	5% (n = 1)
	Asian or Asian British - Indian	5% (n = 1)
	Not recorded	11% (n = 2)
Group attended	Group 1: May 2021	32% (n = 6)
	Group 2: November 2021	21% (n = 4)
	Group 3: January 2022	11% (n = 2)
	Group 4: September 2022	37% (n = 7)

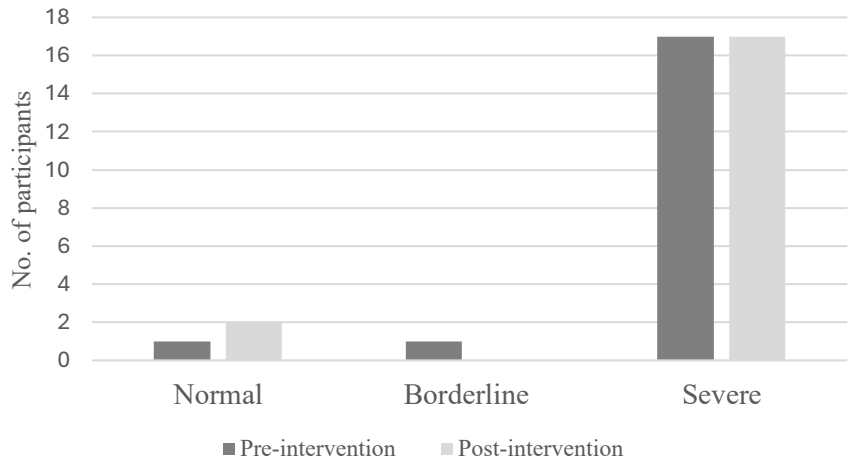

Figure 2. Pre- and post-intervention RCADS-P classifications (n = 19)

Post-intervention status.

Post-intervention status was available for all thirty-three participants. As shown in figure 2, the majority of participants continued to require intervention, either being placed on the waiting list for individual therapy within the team (n = 21; 64%), being referred to another CAMHS service (n = 4; 12%), or receiving concurrent individual 1:1 CBT which continued post-intervention (n = 1; 3%), resulting in a total of 79% (n = 26) of participants requiring further intervention. Five participants were discharged (15%), with notes suggesting that at least one of these participants went on to receive private therapy. Post-intervention status was not available for the 6% of participants who moved out of area (n = 2).

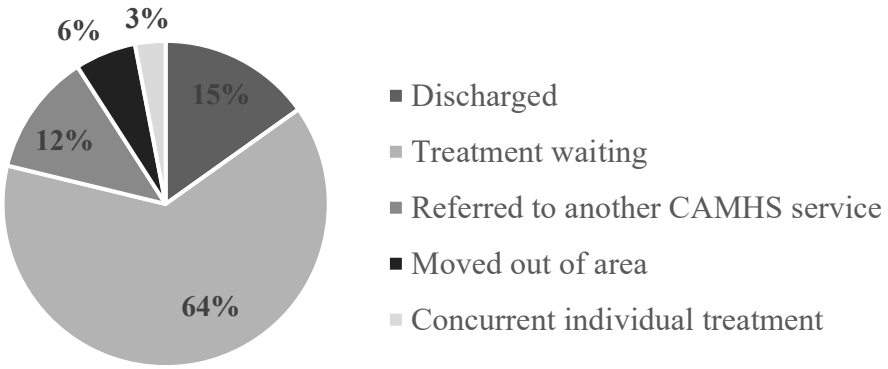

Figure 3. Post-intervention status for participants in research question 1 (n = 33).

Research question 2

Four themes and five sub-themes were developed from the eight parent interviews. A summary of these is displayed in table 4.

Table 6. Key themes and subthemes derived from the participant interviews

THEME	SUBTHEMES
1. *"They're not even talking our language":* The group was not the right fit	1a. *"Trying to get her to engage in these things can be really triggering":* The child found it difficult to engage in the process 1b. *"It's not a one size fits all":* The group was not tailored for our situation 1c. *"There's not the right brain fit":* Autistic brains work differently
2. *"You're going through the motions":* Motivations for doing the group	N/A
3. *"We were encouraged not to avoid":* Helpful changes to parents' understanding and responding	N/A
4. *"The atmosphere was quite calm and relaxing":* There were therapy non-specific factors that were helpful	4a. *"We're not alone":* Connecting with other parents 4b. "*The course reassured us":* Feeling contained and validated

1. **"They're not even talking our language": The group was not the right fit**

1a. "Trying to get her to engage in these things can be really triggering": The child found it difficult to engage in the process

Four participants identified that talking with their child about their fears and worries caused distress; "*There's a lack of being able to reason with them and they go into complete flight mode*" (P3). Participants reported finding it difficult to be able to work with their child to understand their anxiety (e.g. identifying triggers, cognitive, and physiological aspects of the problem) "*often the cause of anxiety is not necessarily… a tangible thing for autistics so*

for him it can just be… an everyday thing that he's used to doing but because he's got dry skin and it's itchy it's taking up the sensory part of his brain" (P4). Four participants reported that their child was resistant to the goal-setting and rewards part of the group "*Having a set goal gives him such a huge level of demand... it's kind of counterproductive*" (P4).

1b. "It's not a one size fits all": The group was not tailored for our situation

All participants endorsed the theme that the approach was not tailored for them or their child: "*it did feel a little bit like.. I got 10% out of it and I had to sit through 80% of stuff that was actually not going to be helpful*" (P1). Participants reported that the approach was too lightweight; "*It's like a tiny little sticking plaster for a gaping open wound and it's.. not enough and it's actually bordering on inappropriate*" (P6). As a result, some participants reported that they would pick and choose what they would take from the group; "*so we were trying to just pick out, pick and choose, what was going to be best for us*" (P7).

1c. "There's not the right brain fit": Autistic brain works differently

Six participants endorsed the idea that the approach did not align with the "*autistic brain*". Some participants reported that the focus on exposure was not helpful, and could actually increase distress "*the CBT model makes you feel like… if I expose enough.. it will improve and sometimes, actually, it can make it worse*" (P7); "*you cannot push CBT as hard.. especially when there's such a distressing response to it*" (P1). Participants identified "*sensory issues*" (P5), "*fixed thinking*" (P6), "*resistance to change of routine*" (P1) and "*social cues [being] misunderstood*" (P6) as issues that are central to their child's distress that weren't addressed.

2. "You're going through the motions": Motivations for doing the group

Four participants endorsed the idea that they were motivated to do the group for reasons other than benefiting from the intervention itself. Some parents did the group

knowing it wasn't the right fit because it was all they were offered; "*I'm thinking.. this doesn't seem right but unfortunately it was the only thing that was available*" (P4). Some participants reported that they felt they had to do the group in order to receive further support from CAMHS "*you're going through the motions because you don't wanna be taken off some CAMHS list because you hope that at some point there might be something, right?.. We have to jump through these hoops because we don't wanna be, like d-listed, or overlooked*" (P6). One parent reported that their main motivation was to be seen by a psychiatrist; "*we'll take what we can from it, but the end goal for us is for her to be seen by the psychiatrist afterwards and to stay in the system because she obviously needs that level of intervention*" (P7).

3. "We were encouraged not to avoid": Helpful changes to parents' understanding and responding

Some participants reported finding it helpful to learn about the cycle of anxiety and how their behaviour may unintentionally reinforce aspects of anxiety; "*I think prior to the sessions, you would always be not necessarily endorsing but going along with it, just to almost try and make your life a little bit easier.. and I think the lightbulb in the session was that.. that's not the best approach.. so we're now trying to challenge more of that in a constructive way*" (P8). Some parents found the importance placed on validating feelings helpful; "*in fact [my child] and I were talking about validations of feelings just a couple of days ago because she was saying that she can remember when she was younger and she'd say something and I'd… completely brush it under the carpet thinking that's what the best thing to do was and actually, through this course, it was kind of.. you've got to listen and you've got to empathise*" (P2). Some participants reported that learning to gently question their child's anxious predictions was helpful; "*just talking to him about.. what's the worst thing that could happen… that really diffuses things for him*" (P5). Some participants noticed

that they have learnt to be more proactive, rather than reactive, in relation to their child's anxiety; "*just maybe thinking it through a bit and kind of being a bit more.. planned and rationale rather than reactive to the situation*" (P5).

4. **"The atmosphere was quite calm and relaxing": There were therapy non-specific factors that were helpful**

4a. "We're not alone": Connecting with other parents

All participants reported some positive benefit from connecting with other parents in the group. Some participants reported finding the experience normalising and reassuring; "*it's quite nice to hear other people going through the same struggles.. you don't feel like such a failure*" (P3). Participants found it helpful to learn from other parents; "*just hearing those examples of what somebody's tried and it's worked can mean more as a parent than the expert who, you know, you respect, but they don't necessarily have a child at home that does this and they don't know what it's actually like to deal with every day*" (P7).

4b. "The course reassured us": **Feeling contained and validated**

Four participants reported experiencing benefit from some of the process factors. Some participants reported feeling reassured by the group process; *"that's where the link between techniques and the reassurance from the course was really important"* (P8). A proportion of participants identified that, whilst the group was not the right fit for them, the facilitators were validating which made it a positive experience; "*there was a good… sensitivity to the fact that a lot of us have been waiting for a really long time and that these sessions aren't necessarily what we want*" (P7). Many participants reported positive feelings regarding the atmosphere of the group, describing the experience as "*welcoming… gentle*" (P7); "*calm*" (P8); and identified that the facilitator was "*relaxed… laid back*" (P10), "*nice*" (P6) and "*made sure everyone was engaging*" (P1).

Discussion

The project aimed to assess (1) the efficacy of the HYC group for anxious autistic children and (2) the experiences of parents of autistic children who have attended the group. Results suggest that the HYC group did not result in any change to the clinical severity of anxiety, as measured by the RCADS-P total anxiety, from pre- to post-intervention. For most participants, their pre-intervention classification of 'severe' anxiety remained at post-intervention. The high proportion of 'severe' classifications amongst participants raises the question about the appropriateness of an intervention that was developed for mild-moderate anxiety (Creswell, Violato, et al., 2017; Halldorsson et al., 2019) and suggests that a more individualised and formulation-led approach may be preferential. For example, in the treatment of anxiety in adults, the National Institute for Health and Care Excellence (NICE) guidelines suggest that for clinically severe anxiety in the context of other comorbidities, more specialised CBT interventions should be offered (as opposed to group interventions that may be offered for mild-to-moderate difficulties; NICE, 2011). Post-intervention status data supports this notion in finding that the vast majority of participants continued to require some form of further intervention after attending the group. Taken together, this data suggests that for autistic children with severe anxiety, the HYC group is not a clinically- or resource-effective intervention.

Analysis of the qualitative data identified four themes and five subthemes. All participants endorsed the idea that the group did not feel like the right fit for their child, either because their child couldn't engage in the process, it was too lightweight, or it was not the right fit for the autistic brain. The reasons given by participants for why the group was not the right fit were remarkably consistent with the model of autistic anxiety, and included issues related to sensory processing, fixed/ rigid thinking, and difficulties identifying triggers and internal cues of anxiety (alexithymia; South & Rodgers, 2017), all of which are not addressed

in the HYC group. Consistent with this, parent identified that there were aspects of the CBT approach that they knew would not be helpful for their child, particularly exposure and goal setting, and thus felt like they were "going through the motions" in order to complete the group and get more personalised support. Whilst some participants did identify some key ingredients of the group that were helpful, centred around the theme of changing their understanding and patterns of responding to their child, there was no specific ingredient that was endorsed as particularly helpful for this group. Importantly, all participants reported benefiting from the therapy non-specific processes within the group, particularly connecting with other parents within a supportive and warm atmosphere. The finding that the parents identified some benefit to the group process, despite the overwhelming consensus that it was not the right fit, highlights the importance of therapy non-specific factors in psychological therapy and supports the notion that these factors are the most powerful predictor of treatment outcomes, over and above that of the treatment content itself (Chatoor & Kurpnick, 2001).

Strengths and limitations

Key stakeholders, including experts by experience, clinicians and researchers, were consulted throughout the project and were included in the co-development of the interview schedule, review of preliminary themes and the co-production of recommendations. This involvement improves our confidence that the outcomes and recommendations will meet the needs of service-users and are realistic for the service.

Limitations of the project include the fact that clinical classifications, as opposed to t-scores, were reported for the RCADS-P. The ranges for these classifications are large which limited our ability to make nuanced interpretations of pre- to post-intervention change. The high rate of missing matched pre- and post-intervention RCADS-P scores (fourteen of the thirty-three participants) means that the outcomes should be interpreted with caution.

Conducting formal statistical analyses on this data was not possible due to the small sample size and lack of specificity of t-scores. Future research could focus on improving the quality of the quantitative data to include specific t-scores, and ensuring a full set of data is available for formal statistical analysis.

Participants who took part in the interviews were self-selected and were not offered any reimbursement for taking part. It is therefore possible that the data is biased, reporting only on the experience of a group of participants who have particular types of experience with the HYC group or service (e.g. those parents who have had difficulty accessing support from CAMHS).

Recommendations

Recommendations were co-produced with the researchers, three experts-by-experience, and clinicians in the A&D team and are displayed in table 5.

Table 7. Service recommendations

KEY FINDING	RECOMMENDATIONS
Overwhelmingly, parents felt that the group wasn't the right fit for their child. Some parents reported harmful effects of exposure when it was not personalized to their child's needs. Parents reported doing the group for reasons other than wanting to benefit from the intervention itself ('going through the motions'). Routine outcome monitoring suggests that the group was not a clinically- or resource-effective intervention for anxiety.	It is recommended that the HYC no longer be offered in a group format for autistic children with anxiety. Instead, it is recommended that the intervention offered be based on a thorough assessment and formulation of their difficulties. This would allow for a more tailored experience, taking into account the autistic experience. Based on the model of autistic anxiety, it is recommended that the assessment and formulation process consider the nature and impact of sensory processing, alexithymia, rigidity of thought, intolerance to uncertainty. It is recommended that the following standardised measures could be used to guide this formulation: **Anxiety Scale for Children- Autism Spectrum Disorder (ASC-ASD;** - For 8 – 16 year olds - Parent- and child-report version available - Provides total anxiety score - Four subscales: Performance anxiety, anxious arousal, separation anxiety, uncertainty **Intolerance of Uncertainty Scale for Children** (Comer et al., 2009) - For 7 – 17-year-olds - Parent- and child-report versions available - Provides a total IU score
Participants reported that the group didn't address issues related to the 'autistic brain' anxiety. There was confusion as to how the CBT approach mapped on to the autistic experience. Participants did report benefiting from some aspects of the intervention, namely connecting with other parents, the atmosphere of the groups, and learning about their child's anxiety and how to change their responses.	It is recommended that the service provide workshops for parents on issues that were brought up by parents as having been missed from the group, as follows: - Sensory processing - Difficulties with interoception and alexithymia - Difficulties with demands - Intolerance to uncertainty - Masking

	It is recommended that these workshops be co-produced and co-facilitated by experts-by-experience. It is also suggested that parents are offered the opportunity to attend drop-ins to review their learning and strategies from these workshops. These workshops could be offered either alongside, instead of, or before 1:1 intervention. It is recommended that these workshops be closely audited, using a service-specific feedback form and a standardised measure, such as the parent self-efficacy scale which is recommended by the Child Outcomes Research Consortium (www.corc.uk; (Woolgar et al., Unpublished data). Given the resource pressures that the service is under, it is recommended that the service make a case for additional funding to set up and audit these groups as recommended above.
Routine outcome measures suggest that most children were experiencing clinically significant anxiety at pre-treatment.	It is recommended that the service offer intervention matched to the level of severity of anxiety. Low-intensity interventions, such as HYC even when delivered as a 1:1 intervention have not been developed to treat severe anxiety. As outlined by NICE, more intensive, personalized 1:1 interventions are recommended for the treatment of severe anxiety. It is therefore recommended that the service make a case for the provision of additional resources to be able to meet this recommendation and adhere to NICE guidelines (NICE, 2011).
Matched pre- and post-intervention outcome measures were not available for many participants. T-scores were above 80 for the majority of participants which limits the services ability to make any meaningful conclusions about the efficacy of interventions.	It is recommended that the service improve their use of routine outcome measures and add more ecologically valid and sensitive outcome measures to their battery, such as: - Goal based outcomes - Child Anxiety Impact Scale (CAIS; Langley et al., 2004)

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PAPER THREE: THEORETICALLY-DRIVEN RESEARCH PROJECT

Overprotective parenting and anxiety in children born with a cleft lip and/ or palate and their parents

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Suggested Journal: This paper is currently being prepared for submission for publication in the Journal of Pediatric Psychology (see appendix J). This Journal was chosen as it publishes papers addressing the interaction between psychological and physical well-being of children, adolescents and their family.

[8] At the time of conducting this project, Dr Alice Farrington was the Principal Clinical Psychologist in the CAMHS Anxiety and Depression Team, Children Young People and Families, Berkshire Healthcare NHS Foundation Trust.

Abstract

Objective: There is some evidence that children born with a cleft lip and/ or palate (CL/P) and their parents are at a slightly increased risk of developing anxiety. The intergenerational model of anxiety (IMA) suggests that overprotective parenting, and parental appraisals may contribute to this risk. This study compared levels of overprotective parenting, child anxiety, and parent anxiety, between children born with a CL/P and a control group. For the CL/P group, exploratory analyses were conducted to assess parent and child factors related to anxiety.

Methods: A cross-sectional design was used. Participants (n = 129) consisted of parents of 8 – 12-year-old children born with a CLP (n = 63) or without a CL/P (the 'control group'; n = 66) recruited through the community. Participants completed an online survey containing measures of overprotective parenting, child anxiety, parent anxiety, and appraisals of their child's CL/P (CL/P group only).

Results: Participants in the CL/P group reported comparable overprotective parenting and parental anxiety, but higher levels of child anxiety than in the control group. Child anxiety in the CL/P group was related to parental anxiety, overprotection and certain parental appraisals, as well as gender. Parent anxiety in the CL/P group was related to parental overprotection and parental appraisals regarding self-blame.

Conclusions: These findings suggest that children with a CL/P are at an increased risk of anxiety, but that overprotective parenting is similar between children born with and without a CL/P. The clinical and research implications for the relationship between CL/P appraisals and child and parent anxiety are discussed.

Introduction

Cleft lip and/ or palate (CL/P) is the most common congenital anomaly of craniofacial structure, affecting 1 in 1,000 – 1,500 live births globally, and is more common in males than females (CRANE Report, 2023; World Health Organization, 2006). There are different types of CL/P with different prevalence rates. An isolated cleft palate (CP) is a gap in the soft and/ or hard palate in the roof of the mouth (42% of cases). An isolated cleft lip (CL) is a separation of the top lip which can extend up to the nostril (25% of cases). A cleft lip *and* palate (CLP) occurs in 30% of cases and is classified as either a unilateral CLP (affecting just one side of the mouth; 21% of cases) or bilateral (affecting both sides of the mouth; 9% of cases). A submucous CP is where there is a cleft in the muscles which is covered by the lining of the mouth (3% of cases; CRANE Report, 2023).

CL/P is usually treated with surgery within the first year of life, with other surgeries sometimes required for speech, aesthetic and functional purposes (Chadha & Beale, 2023). Various complications are associated with different cleft types. A CP is associated with feeding, speech, ear and hearing difficulties (Smallridge et al., 2015), whereas a CL can lead to visible difference and is therefore associated with poorer appearance satisfaction. A CLP is associated with a combination of the above as well as an increased risk of psychosocial distress (Branson et al., 2024; Kelly & Shearer, 2020; Mossey et al., 2009; Namdar et al., 2022). For these reasons, National Health Service (NHS) guidelines state that CL/P be treated by a multi-disciplinary team (MDT) made up of specialist nurses, speech and language therapists, dentists, audiologists, surgeons, and clinical psychologists (NHS England, 2013).

There is some indication in the research that children born with a CL/P are at an increased risk of psychological difficulties, including low self-esteem, anxiety, and depression (Branson et al., 2024; Cheung et al., 2007; Demir et al., 2011; Millar et al., 2013). Specifically, those born with a CL/P are more likely to experience separation anxiety disorder as children, and social anxiety disorder as adolescence (Demir et al., 2011; Tyler et al., 2013).

However, there is also evidence to the contrary, that the impact of being born with a CL/P on psychological wellbeing is generally low (Branson et al., 2024; Stock & Feragen, 2016). The discrepency in the literature may be because adjustment to a CL/P is impacted by a complex interaction between CLP-related factors (e.g. speech dificulties, poor appearance satisfaction; Demir et al., 2011; Tyler et al., 2013), and non-CL/P related factors, such as lower socioeconomic status, maternal stress, and a comorbid condition (Berman et al., 2000; Feragen & Stock, 2014; Lund et al., 2018; Tyler et al., 2013). The role of gender has also been investigated, with mixed findings depending on the outcome being measured. Boys with a CL/P are at increased risk of behavioural difficulties compared to girls, and girls report higher levels of emotional difficulties compared to boys, a finding that is similar to the general population (Feragen et al., 2015; Feragen & Stock, 2016).

The outcomes with regards to parental mental health are also highly variable, with some research finding that parents of children born with a CL/P are at an increased risk of depression, anxiety, post-traumatic stress and reduced quality of life compared to parents of children without a CL/P, whereas other research has found no difference in psychological outcomes between these groups (Stock et al., 2024, 2025; Vasile et al, In preparation).

There is evidence to suggest that parental anxiety in this group is driven by CL/P-related concerns, with feeding and speech concerns being highest amongst parents of children born with a CP and appearance concerns being highest amongst parents of children born with a CL (Niinomi et al., 2021; Srivastav et al., 2021). Qualitative research highlights that parents feel anxious, guilty and a strong emotional urge to protect their child from invasive treatments associated with a CL/P (Nelson et al., 2012). Parents anticipate that their child will be stigmatised throughout life for their perceived difference and, as a result, find it hard to balance the desire to protect their child from stigma with the need to encourage independence (Nelson et al., 2012).

The stress/ coping model can help explain why having a child born with a CL/P is associated with anxiety in some, but not all, parents (Lazarus & Folkman, 1984). This model suggests that parental anxiety would be strongest when threat appraisals outweigh coping appraisals, and is supported by research suggesting that parental appraisals of their child's CL/P predicts variance in parental wellbeing, independently of demographic and social support variables (Shuttlewood et al., 2014).

The Intergenerational Model of Anxiety

The presence of heightened levels of anxiety in some children born with a CL/P and their parents is an important area of study as research and theory suggests a link between parent and child anxiety (Aktar, 2022; Creswell et al., 2010). The intergenerational model of anxiety (IMA), displayed in figure 1, suggests that parent anxiety is related to appraisals that overestimate threat and underestimate coping and that these, in turn, lead to fear-enhancing and lack of autonomy-granting behaviours which includes overprotective parenting practices. These parenting practices inadvertently promote anxious cognitive styles in children which lead to feelings of anxiety (Aktar, 2022).

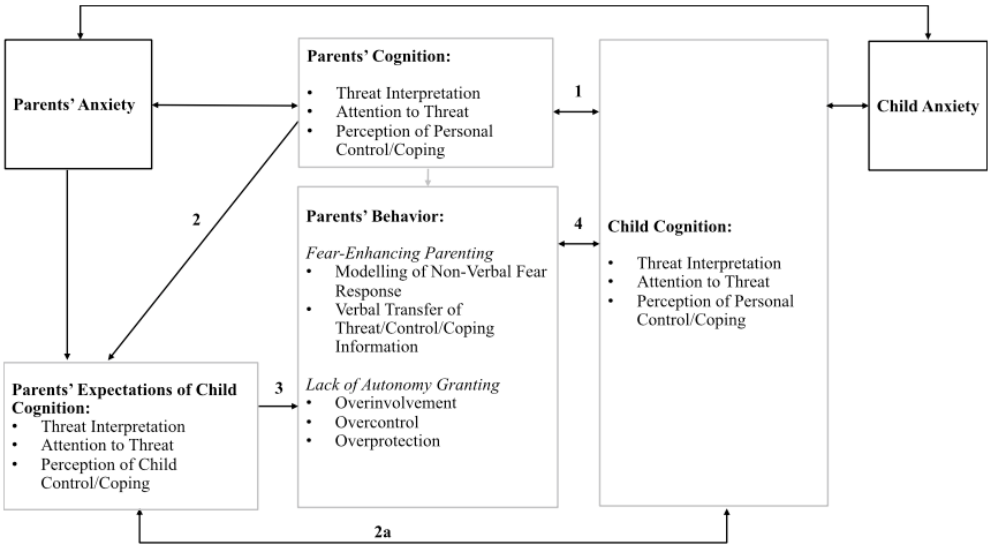

Figure 1. An updated conceptual model of the intergenerational model of anxiety (Aktar, 2022)

The IMA and CL/P

The IMA has not been evaluated in children born with a CL/P specifically, however, longitudinal research has implicated negative parental appraisals as a causal contributing factor to poorer parental quality of life (Stock et al., 2025). Research also suggests that parent-child interactions in the context of child chronic illness are characterised by higher levels of overprotection (Khargekar et al., 2016; Pinquart, 2013). Qualitative data highlights that parents struggle to promote independence due to concerns that their child will be stigmatised and are less able to cope with life transitions (Nelson et al., 2012).

Research aims and questions

The current study aims to examine the role of overprotective parenting and parental appraisals and the relationship between parent and child anxiety in children born with a CL/P. The aims of this project are in line with recently published research priorities; to understand the impact of a CL/P on individual and family wellbeing, and to understand factors that predict psychosocial adjustment in this group (Hotton et al., 2025).

Research question 1

The primary research question asks whether parents of children born with a CL/P ('the CL/P group') report more overprotective parenting compared to parents of children without a CL/P ('the control group'). It is hypothesised that parents in the CL/P group will report higher levels of overprotective parenting compared to parents in the control group. Further correlational analyses will be conducted to explore what factors are associated with overprotective parenting.

Research question 2

The second research question asks whether rates of anxiety are higher amongst children in the CL/P group (question 2a) and their parents (question 2b) compared to the

control group. It is hypothesised that parent and child anxiety in the CLP group will be higher than the control group.

Research question 3

Research question 3 asks what factors are associated with parent and child anxiety in the CL/P group. As this question is exploratory in nature, no specific hypotheses are made.

Methods

Design

This study used a cross-sectional design, following the STROBE (Strengthening the Reporting of Observational Studies in Epidemiology) guidelines (see appendix K).

Ethics

Ethical approval was obtained from the University of Oxford's Central University Research Ethics Committee (reference: R83406/RE001; see appendix L).

Experts-by-experience

Three parents of children with a CL/P were recruited through the Cleft Lip and Palate Association (CLAPA) and acted as consultants by providing insights and feedback regarding the study design and materials.

Participants

Participant recruitment was carried out in the United Kingdom between December 2022 and August 2024. Participants were recruited via social media and word of mouth using the inclusion and exclusion criteria displayed in table 1. A power calculation using G-power indicated that a total sample size between 102 (for power of 0.8) and 176 (for power of 0.95) is required to detect a medium effect size of $d = 0.5$ for research question one. A final sample size of 129 was achieved, with 63 participants in the CL/P group and 66 in the control group.

Table 8. Participant inclusion and exclusion criteria

	Inclusion criteria	Exclusion criteria
CL/P group	Parents of children who are aged between 8 and 12 years of age and were born with a CL/P Parents able to speak English and consent to the study	Parents of children who have: - A CL/P associated with a syndrome - A submucous CL/P - Autism spectrum disorder - Attention deficit hyperactivity disorder - A learning disability - A genetic syndrome - A chronic physical health condition requiring specialist review and/ or treatment
Control group	Parents of children who are aged between 8 and 12 years Parents able to speak English and consent to the study	Parents of children who have: - Autism spectrum disorder - Attention deficit hyperactivity disorder - A learning disability - A genetic syndrome - A chronic physical health condition requiring specialist review and/ or treatment

Measures

Child anxiety: *The Revised Children's Anxiety and Depression Scale-Parent Version (RCADS-P)* is a 47-item parental-report questionnaire measuring anxiety and depression in children and young people between the ages of 8 – 18 years old (Chorpita et al., 2000). Parents rate each item on a 4-point Likert scale (0 = never; 1 = sometimes; 2 = often; 3 = always) according to how true each statement is for their child. The scale provides age-corrected t-scores for common childhood anxiety disorders (separation anxiety, generalised anxiety, panic disorder, social phobia, obsessive compulsive disorder) and a total anxiety score. In previous research, the RCADS-P has displayed good convergent validity in non-mental health settings (Donnelly et al., 2019). For the current sample, the RCADS-P total anxiety showed excellent internal consistency for both the control (α = .95) and CL/P groups (α = .96)

Parent anxiety: The Generalized Anxiety Disorder (GAD-7) questionnaire consists of seven self-report items asking about anxiety symptoms over the past two weeks. Respondents rate the frequency of various symptoms from 'not at all' to 'nearly every day' (Spitzer et al., 2006). The GAD-7 has shown good reliability and good construct, criterion and procedural validity (Spitzer et al., 2006). For the current sample, the GAD-7 showed excellent internal consistency for both the control (α = .92) and CL/P groups (α = .90).

Parent overprotection: *The Parental Overprotection Measure* (OPM; Clarke et al., 2013) is a 19-item self-report questionnaire containing situation-specific questions, designed to assess the extent to which parents engage in behaviours that restrict a child's independence. Parents rate the extent to which the statement represents their typical behaviour on a 5-point Likert scale (0 = not at all; 4 = very much; see appendix L). The OPM has shown very good internal consistency and predictive validity in a sample of 7 – 12 year olds (Clarke et al., 2013). In the current research, the OPM showed very good internal consistency (α = 0.89 for both groups)

Parent appraisals of CL/P: The *Parental Appraisals of Cleft-Questionnaire (PAC-Q)* is a 30-item parent-report of appraisals of their child's CL/P (Shuttlewood et al., 2014). Parents rate the extent to which they agree with statements on a scale of 0 – 100 (0 = "I do not believe this statement at all"; see appendix M). The items map on to five subscales, listed here with Cronbach's alpha for the current sample; (1) Child's Self-Confidence (α = .77), (2) Self-blame (α = .87), (3) Growth through Challenge (α = .89), (4) Perceived Manageability (α = .45), (5) Impact of Treatment (α = .71). In other research, the PAC-Q has shown good test-retest reliability and good convergent and divergent validity (Shuttlewood et al., 2014).

In addition to the abovementioned measures, table 2 displays the list of background and demographic data that was collected.

Table 9. Demographic and background data collected

All participants (CL/P and control group)	CL/P group only
Child's date of birth Child's gender Child's ethnic group Whether the child had transitioned to secondary school in the past 12 months (Y/N) Presence of siblings Parent gender Parent relationship to child Family set-up	Type of cleft How supported the parent felt by the cleft team (0 – 100; 100 = very supported)

Procedures

An online study format was used, with Qualtrics hosting the entire study. The study advert was posted alongside the link to the study on CLAPA's website and social media accounts, as well as the professional social media accounts of the study team.

Data Analysis

Data was analysed using Statistical Package for the Social Sciences (SPSS v29). Group comparisons of demographic data were conducted using Chi-squared for categorical data and the Mann-Whitney U test for child age, as this was non-normally distributed. So as not to violate the chi-squared assumption regarding minimum expected observations, ethnicity was coded as 'white' vs 'other', and family set-up was coded as 'two parent family' vs 'other'. It was not possible to conduct comparisons for 'relationship to child' and 'parent gender' variables as these would have violated the minimum expected observation assumption. Tests for underlying assumptions were carried out and there was found to be some degree of non-normality in some of the continuous measures (see appendix O) but it was deemed that parametric tests for group comparisons to be sufficiently robust, due to the Central Limit Theorem (Field, 2009).

Analysis of covariance (ANCOVA) tests were used to assess the between group differences hypothesised in research questions 1, 2a and 2b. For all analyses, group (CLP vs

control) was entered as a fixed factor and gender as a covariate to account for group differences in gender. Total OP scores (for research question 1), RCADS-P scores (for research question 2a) and GAD-7 scores (for research question 2b) were added as dependent variables. Additionally, to test for group differences between the proportion of participants with clinically significant anxiety symptoms, scores on the RCADS-P and GAD-7 were coded as either above or below their recommended clinical cut-off of 70 and 10 respectively. A chi-squared analysis was conducted to compare the proportion of participants who scored above the clinical cut-off for these measures between groups.

The validity of the IMA was assessed in research question 1, whereby Spearman's rho correlations were conducted across both groups to assess the relationship between OP and RCADS-P scores, and OP total scores and GAD-7 scores. For the CL/P group only, a Spearman's rho correlation was conducted between OP total scores and each of the PAC-Q subscales.

For research question 3, data from the CL/P group only was included in Spearman's rho correlations between RCADS-P scores, GAD-7 scores and other measures (parental OP scores, PAC-Q scores). Finally, a t-test was used to analyse gender differences on anxiety within the CL/P group, where gender was entered as the independent variable and RCADS-P t-scores were entered as the dependent variable.

Effect sizes are reported as Cohen's d (*d*) for t-tests, partial Eta-squared for ANCOVAs (η_p^2), Phi (φ) for chi-squared tests and Spearman's rho (r_s) for correlations. Cohen's guidelines are used to interpret the effect sizes as small, medium or large (Cohen, 1988).

Results

Participant characteristics

Participant characteristics are summarised in table 3. There were no significant differences between the CL/P and control groups in terms of child's age, child ethnicity, presence of siblings, family set-up or whether they had transitioned to secondary school in the last 12 months. Child gender differed significantly between groups, $\chi^2(1) = 5.97$, p = 0.02, with the CL/P group having a higher proportion of boys to girls than the control group. Within the CL/P group, 10% (n = 6) had a CP, 21% (n = 13) had a CL, 41% (n = 26) had a unilateral CLP and 29% (n = 18) had a bilateral CLP.

Research question 1

There was no significant difference between the CL/P (M = 28.68, SD = 12.54) and control groups (M = 28.35, SD = 11.67) on parental overprotection, $F(1,126) = .436$, p = .65. However, for the CL/P group, parental overprotection did significantly positively correlate with parental appraisals regarding *self-blame* (PAC-Q subscale 2; $r_s = .30$, p = <.018), with a small to medium effect size. For the sample as a whole, parental overprotection was significantly related to child anxiety $r_s = .31$, p = <.001, and parental anxiety, $r_s = .37$, p = <.001, both with a small-to-medium effect size.

Table 10. Participant characteristics

		Control	CL/P	Total
Child age (years)		M = 9.9 SD = 1.23	M = 10.05 SD = 1.52	M = 9.9 SD = 1.38
Child gender	Girl	53% (n = 35)	31.7% (n = 20)	42.6% (n = 55)
	Boy	47% (n = 31)	68.3% (n = 43)	57.4% (n = 74)
Child Ethnicity	White	81.8% (n = 54)	88.9% (n = 56)	85.3% (n = 110)
	Mixed/ multiple	7.6% (n = 5)	3.2% (n = 2)	5.4% (n = 7)
	Asian/ Asian British	6.1% (n = 4)	4.8% (n = 3)	5.4% (n = 7)
	Black/ African/ Caribbean/ Black British	3% (n = 2)	3.2% (n = 2)	3.1% (n = 4)
	Other	1.5% (n = 1)	0% (n= 0)	0.8% (n = 1)
Secondary school transition in last 12 months	Yes	9.2% (n = 6)	12.7% (n = 8)	10.9% (n=14)
	No	90.8% (n = 59)	87.3% (n = 55)	89.1% (n = 114)
Presence of siblings	Yes	86.4% (n = 57)	84.1% (n = 53)	85.3% (n = 110)
	No	13.6% (n = 9)	15.9% (n = 10	14.7% (n = 19)
Relationship to child	Mother	90.9% (n = 60)	93.7% (n = 59)	92.2% (n = 119)
	Father	7.6% (n = 5)	6.3% (n = 4)	7% (n = 9)
	Stepmother	1.5% (n = 1)	0% (n = 0)	0.8% (n = 1)
Parent gender	Female	92.4% (n = 61)	93.7% (n = 59)	93% (n = 120)
	Male	7.6% (n = 5)	6.3% (n = 4)	7% (n = 9)
Family set-up	Two-parent family	80.3% (n = 53)	85.7% (n = 54)	82.9% (n = 107)
	Single parent	13.6% (n = 9)	9.5% (n = 6)	11.6% (n = 15)
	Other	6.1% (n = 4)	4.8% (n = 3)	5.4% (n = 7)

Research question 2

For research question 2a, there was a significant difference between the CL/P group (M = 57.38, SD = 16.88) and control group (M = 52.52, SD = 12.10) on child anxiety, F (1,126) = 5.11, p = .026, with a small effect size (η_p^2 = .039). Clinical levels of anxiety also differed significantly between the groups, χ^2 (1) = 6.48, p = 0.01, whereby 24% of the CL/P group scored above the clinical cut-off for anxiety, compared to 8% of the control group with a small effect size (φ = .24).

With regards to specific areas of anxiety, there were significant group differences on RCADS-P separation anxiety subscale scores, F(1, 126) = 4.78, p = .007, with the CL/P group scoring significantly higher (M = 58.06, SD = 17.24) than the control group (M = 51.94, SD = 11.84) with a small-to-medium effect size (η_p^2 = .057). There were also significant differences between the CL/P (M = 54.52, SD = 12.68) and control (M = 50.85, SD = 9.67) groups on generalised anxiety scores, F(1, 126) = 4.024, p = 0.5, with a small effect size (η_p^2 = .031). All other group comparisons between RCADS-P subscale scores were non-significant.

For research question 2b, there was no significant difference in parental anxiety between the CL/P (M = 5.95, SD = 4.76) and control groups (M = 7.09, SD = 5.52), F(1,126) = 1.26, p = .264. Consistent with this, there was also no significant difference between groups on clinically significant symptoms of anxiety, χ^2 (1) = 2.63, p = .11. These results demonstrated that 21% of parents in the CLP group reported clinically significant levels of anxiety, compared to 33% of parents in the control group.

Across the two groups, parental and child anxiety were found to be significantly positively correlated, r_s = .38, p = <.001, with a medium effect size.

Research question 3

With regards to understanding what is related to child and parent anxiety in the CL/P group, correlation analyses found that child anxiety was significantly correlated with parental overprotection (r_s = .376, p = .002), with a medium effect size. Child anxiety was also significantly correlated with parental anxiety (r_s = .286, p = .023), with a small to medium effect size. Child anxiety significantly correlated with appraisals regarding *child's self-confidence* (PAC-Q subscale 1; r_s = .445, p = <.001; medium effect size) and *perceived manageability* (PAC-Q subscale 4; r_s = -.245, p = .05; small effect size). The direction of the

PAC-Q correlations confirms a relationship between more negative parental appraisals and higher child anxiety. Child anxiety did not correlate with any other subscales of the PAC-Q.

Child anxiety was also found to differ significantly between genders, $t(61) = 1.73$, p = 0.04, with a small-to-medium effect ($d = .469$) of girls (M = 62.7, SD = 13.8) scoring significantly higher for anxiety than boys (M = 54.9, SD = 17.7).

For the CL/P group, parental anxiety was significantly positively correlated with parental overprotection ($r_s = .404$, p = .001) with a medium effect size. Parental anxiety was also significantly positively correlated with appraisals regarding *self-blame* (PAC-Q subscale 2; $r_s = .37$, p = .003) with a medium effect size. No other subscales of the PAC-Q were significantly correlated with parental anxiety.

Finally, a significant positive correlation between parental feelings of support from the CL/P team and the PAC-Q subscale *perceived manageability* ($r_s = .51$, p = <.001; large effect size) suggests that parents' stronger feelings of support from the CL/P team are related to positive appraisals about CL/P manageability.

Discussion

This research aimed to understand parental factors in relation to child anxiety in those born with a CL/P. Results found that parents of children with a CL/P reported similar levels of overprotection and anxiety as those in the control group, and that appraisals regarding self-blame were significantly correlated to both overprotection and parental anxiety in parents of children born with CL/P. Children born with a CL/P had higher levels of anxiety than children without a CL/P, and this was driven by significantly higher levels of separation anxiety and generalised anxiety in this group. Child anxiety was associated with parental overprotection, parental anxiety, negative parental appraisals regarding child's self-confidence and the perceived manageability of the cleft, and female gender.

The current project is the first to quantify overprotective parenting in parents of children born with a CL/P and compare this to a control group and, whilst the similarity of self-reported overprotection between the groups was contrary to our hypothesis, it leads to the conclusion that parents of children born with a CL/P are able to promote developmentally appropriate independence just as much as parents of children without a CL/P.

The finding that parental anxiety was similar between the CL/P and control group was contrary to our hypothesis and some of the literature but, given the similarity between the two groups in terms of overprotection, is in-line with what would be predicted by the IMA (Aktar, 2022). It is possible that the discrepancy between our findings and some of the literature is due to the complexity of adjustment to a CL/P, which is complicated by various dynamic and static risk and protective factors including social, cultural, physical and psychological factors (Stock et al., 2020). One possible factor when considering parental outcomes is the age of the child. Qualitative data suggests that the first year of life is experienced as the most distressing for parents (Sischo et al., 2016; Stock & Rumsey, 2015), whilst longitudinal research has found that parental quality of life significantly improves over the course of time (Stock et al., 2025). It is therefore possible that the risk of anxiety fluctuates over the course of parenting a child with a CL/P, and that the 8-12 year old age bracket represents a period of lower risk for parental anxiety, however longitudinal research would be required to test this hypothesis.

The context where this research was conducted is also worth considering. In the UK, children and families are supported by a specialist CL/P team which includes clinical psychology (Hotton et al., 2023). The significant relationship between how supported parents felt by their child's CL/P team and positive appraisals about treatment manageability suggest that CL/P teams provide a sense of manageability and containment for parents, and it is therefore possible that this support protects parents from anxiety (Lazarus & Folkman, 1984;

Stock et al., 2024). We would therefore recommend caution in generalising these findings to other contexts or countries whereby cleft care may be different to the UK.

The finding that parental appraisals regarding self-blame were significantly correlated to parental anxiety is consistent with previous research (Stock et al., 2024, 2025). The additional finding that self-blame appraisals were correlated with parental overprotection provides novel information on the mechanisms by which parental anxiety, appraisals and behaviours may be linked. The IMA would lead to the interpretation that parental anxiety and self-blame appraisals are bidirectionally linked, and that self-blame appraisals are one of the factors that cause overprotective parenting (Aktar, 2022), however, more research would be required to determine causality.

The finding that children born with a CL/P are at a small but significantly increased risk of anxiety, and that nearly a quarter of these children experience clinically significant levels of anxiety is an important addition to the mixed literature on this topic. This finding is consistent with other literature suggesting that most children born with a CL/P have similar psychological outcomes to their peers, but that a minority of children will experience emotional difficulties (Pinckston et al., 2020). The finding that child anxiety significantly correlated with parental anxiety, parental overprotection and certain negative parental appraisals provides some indication of possible pathways to anxiety in children born with a CL/P.

Clinical Implications

Clinical implications will be discussed according to the Paediatric Psychosocial Preventative Health Model (PPPHM) which proposes a hierarchy of needs and associated interventions (universal, targeted, and specialist) within paediatric settings (Kazak et al., 2024). Based on the findings of this study, a Universal intervention in the form of clinical and psychoeducation to assess and challenge unhelpful appraisals regarding self-blame, child's

self-confidence, and the perceived manageability of CL/P is recommended. In the UK, it is possible that this intervention be provided by routine contact with CL/P clinical nurse specialists. Where distress is elevated, as was the case for 24% of the children with a CL/P in this study, clinical psychology interventions drawing on techniques from cognitive behaviour therapy (CBT) and compassion focused therapy (CFT) are recommended. CFT has been found to be effective in in reducing self-blame, self-criticism and shame in parents (Gilbert, 2014; Sirois et al., 2019) and parent-led CBT whereby negative appraisals and overprotective parenting practices have been targeted has been found to be clinically- and cost-effective in reducing child anxiety (Creswell, Violato, et al., 2017). However, the evidence for the efficacy of psychological interventions in the field of CL/P is mixed and further research is therefore required (Norman et al., 2015).

Limitations

The fact that some comorbid conditions were excluded from this study means that these results can't be generalised to the general population of children born with a CL/P. This is important because it is reported that 40% of children born with a CL/P have at least one other condition, and that comorbidity represents a significant risk factor for psychological difficulties in this group (Feragen & Stock, 2014).

Furthermore, there is a possible issue of bias by using parent-report of child anxiety which may be contaminated by parental anxiety and appraisals (Manley & Francis, 2022). However, the RCADS-P has demonstrated similar sensitivity and specificity compared to gold-standard diagnostic interview, and has shown good agreement with the RCADS child version (Serafimova et al., 2021).

The sample had an unrepresentative distribution of cleft types, namely an increase in CLP and decrease in CP compared to the wider cleft population (CRANE Report, 2023) which may threaten the validity of the findings given that there is some evidence to suggest

that parental concerns differ significantly depending on the type of cleft (Niinomi et al., 2021), and that children born with a CP have fewer adjustment problems than those born with a CLP (Feragen & Stock, 2016).

The cross-sectional design used here meant that inferences about causal relationships were limited, thus, the direction of the relationships between parental factors and child anxiety are unclear. The fact that the project was powered only for the primary research question meant that it lacked sufficient power to conduct mediational analyses to understand the direct and indirect pathways to anxiety in children with a CL/P.

Future research

It is recommended that future research use longitudinal designs and mediational analysis to investigate the interaction of parent and child factors over time. Furthermore, it is recommended that research focuses on the efficacy of intervention studies targeting the appraisals and parenting behaviours identified as possible contributors to parent and child anxiety. Such studies could look at intervention at various stages of the PPPHM, assessing the impact of Universal psychoeducational approaches on parental appraisals and behaviours, as well as more specialist CFT and CBT approaches targeting parental appraisals and behaviours.

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EXECUTIVE SUMMARY OF THE TDRP

Why this research was conducted

Cleft lip and/ or palate (CL/P) is a birth defect where the lip and/ or the roof of the mouth do not form properly during foetal development. Previous studies have found that children born with a CL/P and their parents may be more likely to experience anxiety. The specific reasons for this increased risk are not yet clear. Research suggests there might be some CL/P related factors (e.g. difficulties with speech and feeding, appearance concerns, parental guilt) and some non-CL/P related factors (e.g. lower socioeconomic status and comorbid conditions) that increase the risk of anxiety for some, but not all, children born with a CL/P.

The intergenerational model of anxiety (IMA) can help understand the processes underlying anxiety in children born with a CL/P and their parents. This model suggests that anxiety in parents is related to certain thinking styles (appraisals) such as a tendency to overestimate danger or threat and underestimate their child's ability to cope. These appraisals lead parents to engage in overprotective parenting practices which can cause anxious thinking styles in children and lead to subsequent anxiety. For parents of children with a CL/P, there is some evidence to suggest that negative appraisals lead to poorer quality of life, and that they struggle to promote independence in their children. This study was therefore conducted to examine some of the processes in the IMA in children born with a CL/P and their parents.

Our research questions were:

Research question 1: Are parents of children born with a CL/P more overprotective compared to parents of children born without a CL/P?

Research question 2: Are rates of anxiety higher amongst children born with a CL/P and their parents compared to children born without a CL/P?

Research question 3: What factors are associated with parent and child anxiety in the CL/P group?

How the study was carried out

Parents of children aged between 8 and 12 years old born with (n = 63) and without (n = 66; 'the control group') a CL/P were recruited through word of mouth and social media to complete an online questionnaire. This questionnaire contained questions about their background and information about their child, alongside measures of child anxiety, parent anxiety, parent overprotection, and parent appraisals of CL/P.

What we found:

Research question 1: Parents of children born with a CL/P reported comparable levels of overprotective parenting to the control group. Parental appraisals of 'self-blame' regarding the CL/P were related to parental overprotection in the CL/P group.

Research question 2: Children born with a CL/P group were reported to have higher levels of anxiety compared to the control group; 24% of children born with a CL/P scored above the clinical cut-off for anxiety compared with 8% in the control group. There was no difference in parental anxiety between parents of children born with a CL/P and the control group.

Research question 3: Anxiety in children born with a CL/P was related to gender (girls were more anxious than boys), and to negative parental appraisals regarding the child's self-confidence and their ability to manage the child's CL/P. Anxiety in parents of children born with a CL/P was related to appraisals regarding self-blame.

Conclusions

The findings here suggest that parents of children born with a CL/P can promote developmentally appropriate autonomy just as much as parents of children born without a CL/P. In line with this, parents of children with a CL/P reported comparable levels of anxiety to those parents of children born without a CL/P. Children born with a CL/P, however, were

found to be at an increased risk of anxiety. Negative parental appraisals regarding the child's CL/P were related to both child and parent anxiety and may therefore represent a specific mechanism contributing to individual differences in anxiety for this group.

Clinical implications

These findings suggest that an early intervention for all parents of children with a CL/P (a 'Universal intervention') targeting the negative appraisals linked to child and parent anxiety might be an effective use of resource, as this could prevent these appraisals becoming entrenched and contributing to anxiety in the future. In the United Kingdom, where families are supported by specialist CL/P teams, this intervention could be implemented as part of routine contact with specialist CL/P nurses. For those families showing elevated levels of anxiety, targeted interventions based on models of cognitive behavioural therapy or compassion focused therapy are recommended.

CONNECTING REFLECTIVE NARRATIVE

This connecting narrative provides a reflection on the three projects presented in this thesis. Whilst different in scope, there were some key themes in my motivations and reflections which will be discussed before reflecting on each of the projects individually.

One theme that has tied together my previous experience working in CAMHS is the role of parents or carers (hereafter referred to as parents) and the opportunity to intervene early by supporting and empowering parents from an early stage. This led to my Theoretically-Driven Research Project (TDRP) and Service Improvement Project (SIP) ideas, which were both on the theme of understanding and targeting parent-child interactions in the context of child anxiety. The findings from my TDRP have encouraged me to include parents in my clinical work more frequently, paying particular attention to the role of parental appraisals in child distress. The finding from my SIP that parents continued to attend a group due to therapy non-specific factors, despite feeling that it wasn't helpful, has encouraged me to actively consider the importance of the therapeutic relationship with parents as well as children, even in the context of individual child work. My Systematic Review of the Literature (SRL) was born out of my motivation to examine whether individual behavioural interventions (as opposed to higher intensity positive behaviour support) could be implemented by parents as a way of intervening early, as well as skilling up and empowering parents. The mixed results and methodological issues meant that firm conclusions regarding the efficacy of differential reinforcement as a stand-alone intervention could not be made. This finding has led me to reflect on feelings of frustration, anxiety, and a lack of containment when, as a clinician, I am working with issues such as SIB where there is no clear evidence-based intervention. I have wondered about the parallel process that happens for families who look to their clinical team to contain them and provide evidence-based

support and how it might lead to understandable feelings of frustration and anxiety when the team cannot provide this containment and support.

Another connecting theme for all my projects is the social model of disability which asserts that distress is often caused or exacerbated by inflexible social norms, ideologies and structures. From speaking to parents of children with a cleft lip and/ or palate (CL/P) for the TDRP patient and public involvement (PPI), it was clear that distress in this group is often driven by concerns of visible difference and social stigma, irrespective of the medical concerns. The finding that self-blame appraisals were linked to parent anxiety makes me wonder about the impact of societal narratives around the choices of mothers during pregnancy which could lead to, or entrench, the feelings of blame and related distress experienced by parents. Understanding and supporting autistic children, a topic that connects my SIP and SRL, has received much attention in the literature on the social model of disability. The findings in my SIP that the manualised CBT intervention was inappropriate for autistic children but that parents felt unable to access anything more appropriate highlights the social and structural factors that contribute to distress in this population. In completing the literature search for this project, I was struck by the lack of research into this topic despite anxiety and autism co-occurring so frequently. My SRL also highlighted some issues related to the social model of disability, including the fact that so many of the papers that I came across during screening targeted stereotyped behaviours (e.g. hand flapping) under the rationale that these behaviours were 'socially unacceptable'. This clearly highlights how inflexible and inaccurate social norms can lead to harmful clinical practice when left unchecked.

Theoretically-Driven Research Project

Prior to completing this project, I had not had any experience working with CL/P, and the PPI input was therefore particularly insightful not only in helping me co-develop the

project but also having the issues I had been reading about academically brought to life by the real-life stories and journeys of parents of children with a CL/P.

I was fortunate enough to be linked with a leading CL/P charity who helped with recruitment by advertising on their social media platform. Here, I learnt the importance of multi-agency working in research and how the skills I often use in clinical practice (rapport building, professionalism, organisation, and communication) are transferrable to the field of research. This experience has left me excited to contribute to research in my career going forwards.

Service Improvement Project

My prior experience working with autistic children with anxiety, and my knowledge of the literature in this area, contributed to some strongly held beliefs on this topic which led to feelings of tension as I was trying to be objective during data collection and analysis. My experience completing this project helped me recognise that it is not possible to be truly objective and that it is therefore important in qualitative research to notice and reflect on our biases and observations. I therefore used research supervision, bracketing interviews and PPI involvement in the development of themes to be collaborative and transparent about the analytic process.

Systematic Review of the Literature

Prior to training, I had used the results of many an SRL to inform my clinical practice, but I had not actually completed one myself. This meant that there were some aspects of the process that presented some challenges. Namely, I had assumed that the decisions regarding screening papers would come naturally from the inclusion and exclusion criteria, and I had not anticipated the volume of papers which were ambiguous and therefore required joint decision making with supervisors, second raters, and transparency with regards to reporting this. This has been a helpful learning experience not only for any future SRLs that I complete,

but also it has encouraged me to be more critical when reading published SRLs by paying close attention to the decisions regarding inclusion and exclusion criteria and how decisions were made with regards to ambiguous papers.

ACKNOWLEDGEMENTS

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To all my wonderful friends and colleagues on the course, in particular Jerica, Becca, Ruby, and Jade. One of the benefits of having integrated into so many cohorts has been the opportunity to grow and develop with you, riding all the ups and downs of this course together. I'm so grateful to be finishing training with such an inspirational group of colleagues and friends.

Finally, to all those who have supported me outside work. Thank you to my lovely group of friends for bringing laughter and joy to my life. Stuart, thank you for filling my life with happiness and love, and for keeping the house running without hesitation or complaint

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APPENDICES

Appendix A. SRL: Author Guidelines for the Journal of Autism and Developmental Disorders[9]

Instructions for Authors

Editorial procedure
Double-Anonymous Peer Review

MANUSCRIPT FORMAT

All JADD manuscripts should be submitted to Editorial Manager in 12-point Times New Roman with standard 1-inch borders around the margins. Please disregard the suggestion of 10-point font in the Text section below.

APA Style

APA Publication Manual standards must be followed.

As of January 20, 2011, the Journal has moved to a double-anonymous review process. Therefore, when submitting a new manuscript, DO NOT include any of your personal information (e.g., name, affiliation) anywhere within the manuscript. When you are ready to submit a manuscript to JADD, please be sure to upload these 3 separate files to the Editorial Manager site to ensure timely processing and review of your paper:

A title page with the running head, manuscript title, and complete author information.
Followed by (page break) the Abstract page with keywords and the corresponding author e-mail information.
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Types of papers

Articles, Commentaries Brief Reports, Letters to the Editor

[9] Only guidelines that are relevant to this submission are included here. For the full guidelines, see: https://link.springer.com/journal/10803/submission-guidelines#Instructions%20for%20Authors_Types%20of%20papers

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- A Letter to the Editor/Commentary is 6 or less double spaced pages with shorter references, tables and figures.
    - Style sheet for Letter to the Editor:
    - A title page with the running head, manuscript title, and complete author information including corresponding author e-mail information
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        - - Line 1: "Letter to the Editor"
        - - Line 6: Text begins; references and tables, figure caption sheet, and figures may follow (page break between each and see format rules)

Review your manuscript for these elements

1. Order of manuscript pages:
- Title Page with all Author Contact Information & Abstract with keywords and the corresponding author e-mail information.
- Anonymized Abstract, manuscripts and References without contact information
- Appendix
- Figure Caption Sheet
- Figures
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2. JADD submissions should include:
- A structured abstract with the Purpose, Methods, Results, and Conclusion.
- COI and other author statements placed on the title page.
- No more than 40 double-spaced pages, including double-spaced references (with hanging indents), tables, and figures.
- Tables and Figures placed at the end of the manuscript with callouts in the text.

Manuscript Submission

Manuscript Submission

Submission of a manuscript implies: that the work described has not been published before; that it is not under consideration for publication anywhere else; that its publication has been approved by all co-authors, if any, as well as by the responsible authorities – tacitly or explicitly – at the institute where the work has been carried out. The publisher will not be held legally responsible should there be any claims for compensation.

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JADD Inclusive Language Guide (Download pdf, 134 kB)

Title Page

Please make sure your title page contains the following information.

Title

The title should be concise and informative.

Author information

- The name(s) of the author(s)
- The affiliation(s) of the author(s), i.e. institution, (department), city, (state), country
- A clear indication and an active e-mail address of the corresponding author
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For authors that are (temporarily) unaffiliated we will only capture their city and country of residence, not their e-mail address unless specifically requested.

Large Language Models (LLMs), such as ChatGPT, do not currently satisfy our authorship criteria. Notably an attribution of authorship carries with it accountability for the work, which cannot be effectively applied to LLMs. Use of an LLM should be properly documented in the Methods section (and if a Methods section is not available, in a suitable alternative part) of the manuscript. The use of an LLM (or other AI-tool) for "AI assisted copy editing" purposes does not need to be declared. In this context, we define the term "AI assisted copy editing" as AI-assisted improvements to human-generated texts for readability and style, and to ensure that the texts are free of errors in grammar, spelling, punctuation and tone. These AI-assisted improvements may include wording and formatting changes to the texts, but do not include generative editorial work and autonomous content creation. In all cases, there must be human accountability for the final version of the text and agreement from the authors that the edits reflect their original work.

Abstract

Please provide a structured abstract of 150 to 250 words which should be divided into the following sections:

- Purpose (stating the main purposes and research question)
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- Results
- Conclusion

For life science journals only (when applicable)
Trial registration number and date of registration for prospectively registered trials
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Keywords
Please provide 4 to 6 keywords which can be used for indexing purposes.

Statements and Declarations

The following statements should be included under the heading "Statements and Declarations" for inclusion in the published paper. Please note that submissions that do not include relevant declarations will be returned as incomplete.

- **Competing Interests**: Authors are required to disclose financial or non-financial interests that are directly or indirectly related to the work submitted for publication. Please refer to “Competing Interests and Funding” below for more information on how to complete this section.

Please see the relevant sections in the submission guidelines for further information as well as various examples of wording. Please revise/customize the sample statements according to your own needs.

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Text Formatting

Manuscripts should be submitted in Word.

- Use a normal, plain font (e.g., 10-point Times Roman) for text.
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- Use the equation editor or MathType for equations.
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Please use no more than three levels of displayed headings.

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Footnotes to the text are numbered consecutively; those to tables should be indicated by superscript lower-case letters (or asterisks for significance values and other statistical data). Footnotes to the title or the authors of the article are not given reference symbols.

Always use footnotes instead of endnotes.

Acknowledgments

Acknowledgments of people, grants, funds, etc. should be placed in a separate section on the title page. The names of funding organizations should be written in full.

Body

- The body of the manuscript should begin on a separate page. The manuscript page header (if used) and page number should appear in the upper right corner. Type the title of the paper centered at the top of the page, add a hard return, and then begin the text using the format noted above. The body should contain:
- Introduction (The introduction has no label.)
- Methods (Center the heading. Use un-centered subheadings such as: Participants, Materials, Procedure.)
- Results (Center the heading.)
- Discussion (Center the heading.)

Headings

Please use no more than three levels of displayed headings.

Level 1: Centered

Level 2: Centered Italicized

Level 3: Flush left, Italicized

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Center the label “Footnotes” at the top of a separate page. Footnotes can be used to give additional information, which may include the citation of a reference included in the reference list. They should not consist solely of a reference citation, and they should never include the bibliographic details of a reference. They should also not contain any figures or tables.

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Please always use internationally accepted signs and symbols for units (SI units).

Scientific style

- Generic names of drugs and pesticides are preferred; if trade names are used, the generic name should be given at first mention.
- Please use the standard mathematical notation for formulae, symbols etc.:Italic for single letters that denote mathematical constants, variables, and unknown quantities Roman/upright for numerals, operators, and punctuation, and commonly defined functions or abbreviations, e.g., cos, det, e or exp, lim, log, max, min, sin, tan, d (for derivative) Bold for vectors, tensors, and matrices.

References

Citation

Cite references in the text by name and year in parentheses. Some examples:

- Negotiation research spans many disciplines (Thompson, 1990).
- This result was later contradicted by Becker and Seligman (1996).
- This effect has been widely studied (Abbott, 1991; Barakat et al., 1995; Kelso & Smith, 1998; Medvec et al., 1999).

Authors are encouraged to follow official APA version 7 guidelines on the number of authors included in reference list entries (i.e., include all authors up to 20; for larger groups, give the first 19 names followed by an ellipsis and the final author's name). However, if authors shorten the author group by using et al., this will be retained.

Reference list

The list of references should only include works that are cited in the text and that have been published or accepted for publication. Personal communications and unpublished works should only be mentioned in the text.

Reference list entries should be alphabetized by the last names of the first author of each work.

Journal names and book titles should be italicized.

If available, please always include DOIs as full DOI links in your reference list (e.g. “https://doi.org/abc”).

- Journal article Grady, J. S., Her, M., Moreno, G., Perez, C., & Yelinek, J. (2019). Emotions in storybooks: A comparison of storybooks that represent ethnic and racial groups in the United States. Psychology of Popular Media Culture, 8(3), 207–217. https://doi.org/10.1037/ppm0000185
- Article by DOI Hong, I., Knox, S., Pryor, L., Mroz, T. M., Graham, J., Shields, M. F., & Reistetter, T. A. (2020). Is referral to home health rehabilitation following inpatient rehabilitation facility associated with 90-day hospital readmission for adult patients with stroke? American Journal of Physical Medicine & Rehabilitation. Advance online publication. https://doi.org/10.1097/PHM.0000000000001435
- Book Sapolsky, R. M. (2017). Behave: The biology of humans at our best and worst. Penguin Books.
- Book chapter Dillard, J. P. (2020). Currents in the study of persuasion. In M. B. Oliver, A. A. Raney, & J. Bryant (Eds.), Media effects: Advances in theory and research (4th ed., pp. 115–129). Routledge.
- Online document Fagan, J. (2019, March 25). Nursing clinical brain. OER Commons. Retrieved January 7, 2020, from https://www.oercommons.org/authoring/53029-nursing-clinical-brain/view

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A Graduate Student's Guide to Determining Authorship Credit and Authorship Order, APA Science Student Council 2006

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Appendix B. SRL: PRISMA Checklist

Section and Topic	Item #	Checklist item	Location & Comments
TITLE			
Title	1	Identify the report as a systematic review.	Title and Abstract (P9)
ABSTRACT			
Abstract	2	See the PRISMA 2020 for Abstracts checklist.	Abstract (P10)
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of existing knowledge.	Introduction (P11-P15)
Objectives	4	Provide an explicit statement of the objective(s) or question(s) the review addresses.	Research questions (P16)
METHODS			
Eligibility criteria	5	Specify the inclusion and exclusion criteria for the review and how studies were grouped for the syntheses.	Methods (P16 – P17)
Information sources	6	Specify all databases, registers, websites, organisations, reference lists and other sources searched or consulted to identify studies. Specify the date when each source was last searched or consulted.	Methods (P17)
Search strategy	7	Present the full search strategies for all databases, registers and websites, including any filters and limits used.	Methods (P17)
Selection process	8	Specify the methods used to decide whether a study met the inclusion criteria of the review, including how many reviewers screened each record and each report retrieved, whether they worked independently, and if applicable, details of automation tools used in the process.	Methods (P17)
Data collection process	9	Specify the methods used to collect data from reports, including how many reviewers collected data from each report, whether they worked independently, any processes for obtaining or confirming data from study investigators, and if applicable, details of automation tools used in the process.	Methods (P17-P18)
Data items	10a	List and define all outcomes for which data were sought. Specify whether all results that were compatible with each outcome domain in each study were sought (e.g. for all measures, time points, analyses), and if not, the methods used to decide which results to collect.	Methods (P17 – P18)
	10b	List and define all other variables for which data were sought (e.g. participant and intervention characteristics, funding sources). Describe any assumptions made about any missing or unclear information.	Methods (P17 – P18) and Table 1
Study risk of bias assessment	11	Specify the methods used to assess risk of bias in the included studies, including details of the tool(s) used, how many reviewers assessed each study and whether they worked independently, and if applicable, details of automation tools used in the process.	Methods (P18)
Effect measures	12	Specify for each outcome the effect measure(s) (e.g. risk ratio, mean difference) used in the synthesis or presentation of results.	N/A
Synthesis methods	13a	Describe the processes used to decide which studies were eligible for each synthesis (e.g. tabulating the study intervention characteristics and comparing against the planned groups for each synthesis (item #5)).	N/A – all studies were eligible for each synthesis.
	13b	Describe any methods required to prepare the data for presentation or synthesis, such as handling of missing summary statistics, or data conversions.	N/A
	13c	Describe any methods used to tabulate or visually display results of individual studies and syntheses.	N/A
	13d	Describe any methods used to synthesize results and provide a	Methods (P18)

Section and Topic	Item #	Checklist item	Location & Comments
		rationale for the choice(s). If meta-analysis was performed, describe the model(s), method(s) to identify the presence and extent of statistical heterogeneity, and software package(s) used.	
	13e	Describe any methods used to explore possible causes of heterogeneity among study results (e.g. subgroup analysis, meta-regression).	N/A
	13f	Describe any sensitivity analyses conducted to assess robustness of the synthesized results.	N/A
Reporting bias assessment	14	Describe any methods used to assess risk of bias due to missing results in a synthesis (arising from reporting biases).	N/A
Certainty assessment	15	Describe any methods used to assess certainty (or confidence) in the body of evidence for an outcome.	Results (P20 – P29)
RESULTS			
Study selection	16a	Describe the results of the search and selection process, from the number of records identified in the search to the number of studies included in the review, ideally using a flow diagram.	PRISMA diagram (P20)
	16b	Cite studies that might appear to meet the inclusion criteria, but which were excluded, and explain why they were excluded.	N/A
Study characteristics	17	Cite each included study and present its characteristics.	Table 1 (P22 – P26) Results (RQ1: P19)
Risk of bias in studies	18	Present assessments of risk of bias for each included study.	Table 2 (P30) Results (RQ2 and RQ3; P20 – P35)
Results of individual studies	19	For all outcomes, present, for each study: (a) summary statistics for each group (where appropriate) and (b) an effect estimate and its precision (e.g. confidence/credible interval), ideally using structured tables or plots.	N/A
Results of syntheses	20a	For each synthesis, briefly summarise the characteristics and risk of bias among contributing studies.	Results (RQ2 and RQ3: P20- P35)
	20b	Present results of all statistical syntheses conducted. If meta-analysis was done, present for each the summary estimate and its precision (e.g. confidence/credible interval) and measures of statistical heterogeneity. If comparing groups, describe the direction of the effect.	N/A
	20c	Present results of all investigations of possible causes of heterogeneity among study results.	N/A
	20d	Present results of all sensitivity analyses conducted to assess the robustness of the synthesized results.	N/A
Reporting biases	21	Present assessments of risk of bias due to missing results (arising from reporting biases) for each synthesis assessed.	N/A
Certainty of evidence	22	Present assessments of certainty (or confidence) in the body of evidence for each outcome assessed.	N/A
DISCUSSION			
Discussion	23a	Provide a general interpretation of the results in the context of other evidence.	Discussion (P35 – P39)
	23b	Discuss any limitations of the evidence included in the review.	Discussion (P35 – P39)
	23c	Discuss any limitations of the review processes used.	Discussion (P35 – P39)
	23d	Discuss implications of the results for practice, policy, and future research.	Discussion (P35 – P39)
OTHER INFORMATION			
Registration and protocol	24a	Provide registration information for the review, including register name and registration number, or state that the review was not registered.	Methods (P16)

Section and Topic	Item #	Checklist item	Location & Comments
	24b	Indicate where the review protocol can be accessed, or state that a protocol was not prepared.	Methods (P16)
	24c	Describe and explain any amendments to information provided at registration or in the protocol.	N/A
Support	25	Describe sources of financial or non-financial support for the review, and the role of the funders or sponsors in the review.	N/A
Competing interests	26	Declare any competing interests of review authors.	N/A
Availability of data, code and other materials	27	Report which of the following are publicly available and where they can be found: template data collection forms; data extracted from included studies; data used for all analyses; analytic code; any other materials used in the review.	Methods (P17)

Appendix C. SRL: Quality Appraisal Tool

The SCED Scale (Tate et al., 2008)

Item	Definition
1. Clinical History	The study provides critical information regarding demographic and injury characteristics of the research subject that allows the reader to determine the applicability of the treatment to another individual.
2. Target behaviours	The paper identifies a precise, repeatable and operationally defined target behaviour that can be used to measure treatment success.
3. Design	The study design allows the for the examination of cause-and-effect relationships to demonstrate treatment efficacy.
4. Baseline	To establish that sufficient sampling of behaviour had occurred during the pre-treatment period to provide an adequate baseline measure.
5. Sampling behaviour during treatment	To establish that sufficient sampling of behaviour during the treatment phase has occurred to differentiate a treatment response from fluctuations in behaviour that may have occurred at baseline.
6. Raw data record	To provide an accurate representation of the variability of the target behaviour.
7. Inter-rater reliability	To determine if the target behaviour measure is reliable and collected in a consistent manner.
8. Independence of assessors	To reduce assessment bias by employing a person who is otherwise uninvolved in the study, to provide an evaluation of the patients.
9. Statistical Analysis	To demonstrate the effectiveness of the treatment of interest by statistically comparing the results over the study phases.
10. Replication	To demonstrate that the application and results of the therapy are not limited to a specific individual or situation (i.e., that the results are reproduced in other circumstances – replicated across subjects, therapists or settings).
11. Generalisation	To demonstrate the functional utility of the treatment in extending beyond the target behaviours or therapy environment into other areas of the individual's life.

Appendix D. SIP: Author Guidelines for Psychology and Psychotherapy: Theory, Research, and Practice[10]

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Appendix E. SIP: Overview of HYC group

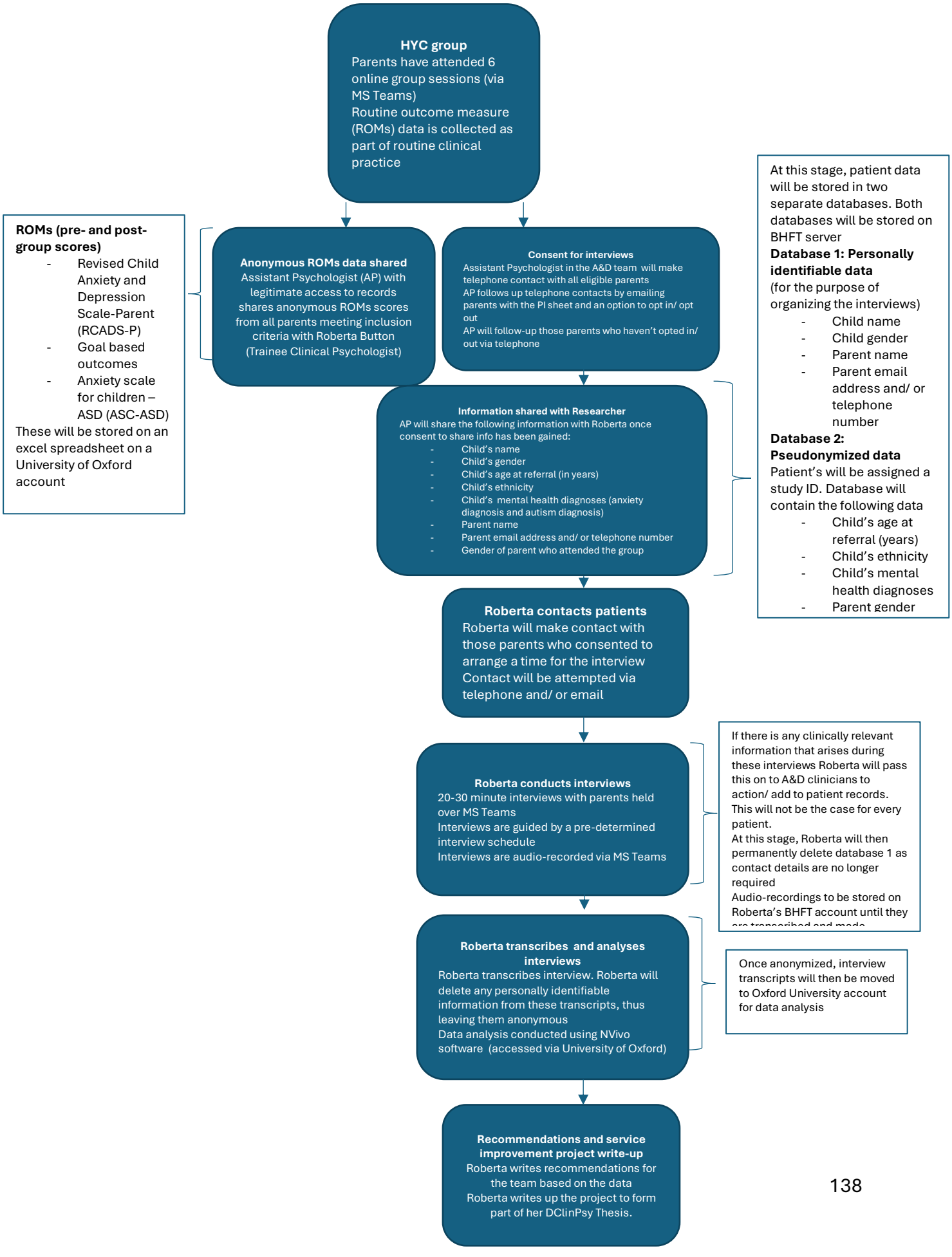
Prior to start of treatment

- Parent reads: Part 1, Chapter 6 (how to use this book) and Chapter 7 (setting goals) of Helping Your Child with Fears and Worries (Creswell & Willetts, 2019).

Session number	Format	What is covered in session?	Homework
1	Face-to-face	Philosophy of programme (why CBT and why parental approach) Psychoeducation How anxiety develops and is maintained Treatment goals	Reflect on treatment goals Be on the lookout for maintenance factors Complete handout (maintenance of anxiety problems – part 2) Read chapters 8 and 9 of the book
2	Face-to-face	What is my child thinking? What does my child need to learn? Promoting independence and 'having a go' Identifying rewards	Read chapter 10 of the book Encourage independence and 'having a go' behaviour (optional handout) Reflect on rewards (complete handout)
3	Face-to-face	Step-by-step plan	Finalise step-by-step plan with child (handout) Start implementing step-by-step plan Monitor progress of step-by-step plan (using handout)
4	Telephone (15-20 minutes)	Checking in and reviewing homework Make changes to step-by-step plan as necessary	Continue implementing step-by-step plan Read chapter 11 of book
5	Face-to-face	Checking in and reviewing homework Problem solving approach	Continue using step-by-step plan Continue recording progress

Session number	Format	What is covered in session?	Homework
			Use problem solving approach (handout)
6	Telephone (15-20 minutes)	Reviewing progress	Continue using step-by-step plan Continue using problem solving approach Work towards long-term goals (handouts)
		4-week break	
7	Face-to-face	Follow-up appointment Review progress: discharge, monitor or refer elsewhere	N/A

Appendix F. SIP: Data Protection Impact Assessment (DPIA)



Appendix G. SIP: Interview schedule

***Warming question*:** Tell me about your child and how you came to attend the group?

Overall perceptions and experiences of the HYC group

1. What was the group like?

2. As a parent of an autistic child (or a child awaiting an assessment for autism), do you think the group was helpful for you and your child?
- Probe: Do you feel you and your child are better off as a result of the group?
- Probe: If you could go back in time, knowing what you know now, would you have done the group? Why?
- Probe: Did you feel that the group was the right fit for your child and their specific needs related to autism/ potential autism?
- Probe: What did you think of the parts of the group that focused on autism?

3. What was the impact of the group?
- Probe: Has the group changed your perception of your child's anxiety? What have you learnt about your child's anxiety?
- Probe: Was there any change in your understanding of autism and anxiety?
- Probe: Have you changed how you react when your child is anxious?/ How you manage your child's anxiety
- Probe: Have you noticed any change in your child's anxiety as a result of the group?
- Probe: Would you recommend the group to other people?

4. What did you like about the group?

5. What did you not like so much about the group?

Specific components of the HYC group that were more/ less helpful

6. What do you remember was helpful about the group?
- Probe: Did you get a chance to try out any of the strategies? How did that go?
- Probe: Were there any strategies that were particularly helpful for your child?
- Probe: What, if anything, do you continue to use from the group?
- Probe: Do you think that applies more or less because of your child's specific needs related to autism/ potential autism?
- Probe: If you had to recommend one of the components/ strategies to a friend/ family member in a similar situation to you, what would it be?
- Probe: Remind parent of modules by screensharing summary of the HYC group

7. Can you tell me if there was anything in the group that wasn't helpful?
- Probe: From the modules I just mentioned, did you get a chance to try out any of the strategies? How did that go? Were there any strategies that didn't work so well for your child?
- Probe: Do you think that applies more or less because of your child's specific needs related to autism/ potential autism?

- Probe: Is there anything in the group that you think should be dropped? Why is that?

Barriers and facilitators to being able to engage in the HYC group

8. Can you think of anything that made it easier for you to do the group?
- Probe: What, if anything, made it easier for you to *attend and take part in the groups*? What, if anything, made it easier for you to take in all the *suggestions/ strategies*? What, if anything, made it easier for you to do the *in-between session tasks*?
- Probe: Do you think that applies more or less because of your child's specific needs related to autism/ potential autism?

9. Can you think of anything that got in the way of you being able to do the group?
- Probe: What, if anything, got in the way of you being able to *attend and take part in the groups*? What, if anything, got in the way of you being able to take in all the *suggestions/ strategies*? What, if anything, got in the way of you being able to do the *in-between session tasks*?
- Probe: Do you think that applies more or less because of your child's specific needs related to autism/ potential autism?

Perceptions on how the HYC group could be improved (for those with autism?)

10. What could we do to improve the HYC group for parents of an autistic child (OR a child waiting for an autism assessment) like you?
- Probe: If you were in charge of the group, would you make any changes for autistic children (or children with potential autism)? If so, what would they be?
- Probe: Is there anything you wish you'd known before the group?
- Probe: Was there anything you wish the group spent more time on?
- Probe: Was there anything you wish the group spent less time on?
- Probe: What would have been helpful to know from the beginning?

11. Do you think your child has any specific difficulties that contribute to their anxiety that weren't covered in the group?

Appendix H. SIP: Notes from bracketing interview

Bracketing interview conducted on 20th February 2023 with a Trainee Clinical Psychologist colleague who provided the following summary

We discussed that your motivation to do this project came after have previously delivered the project in an autism team. You noticed a lot of the group doesn't fit for autistic children, and the definition of anxiety in autism isn't the same as for neurotypical children, eg. Sensory sensitivities. However, there can still be some benefits to this group for autistic children. You want to see experiences from parents around what works and what was unhelpful. You shared that you hold some beliefs about the CBT model of anxiety not fitting for autistic anxiety.

You shared that you really like the program and you feel a strong sense of loyalty to it. Your instinctual hope is to still say the group is good for autistic children but with some changes (eg. Making modules about sensory/intolerance of uncertainty/blend with a different ASD anxiety group.). You don't want to get rid of the CBT approach, but also acknowledge that it can't be used without any adaptions. You hope the project can help to fit for young people showing more autistic-type anxiety and anxiety that fits a CBT-type model (In the middle of a venn diagram)

Based on interviews you have already conducted you were surprised that parents thought one particular module of the group was helpful, as this was different to your experience of the group and you weren't expecting parents to find this module helpful. Lots of the information has been confirming your previously held beliefs, eg:

- CBT approach doesn't fit for all types of anxiety in autism (e.g. sensory needs)
- Feels very rushed (eg. Goals you progress thorough)
- Most parents named sensory as something that is not considered at all.
- It specifically mentions at the start of the book for the module that this approach its not appropriate for autistic children.
- Some parents mentioned they felt they had to do the group as a first line intervention and if they didn't engage they wouldn't be offered anything else.

Things that haven't been said yet that you were expecting to be said includes parents haven't spoken much about the graded exposure element of the group, as you expected this to be the main mechanism of change. Even parents who benefitted from the group haven't spoken about the graded exposure. What parents took away from the group was different to what you anticipated.

You shared that the impact if the outcome was that the intervention doesn't work is that it would make more work for you as a trainee to think about recommendations to the service. You would also feel frustrated about having done a lot of work on this program which would feel like it was for nothing, and you would feel less in control and unsure about what the outcome will be.

We discussed the difficulty in managing the role shifting between being a "therapist" and being an "interviewer." We discussed that this is not a bad thing as it is important to develop a rapport with interviewees. At times you will share own position to validate parents and normalize their experiences.

We discussed your relationship with the researcher who developed the group, and you considered whether this may impact on how you feel about the results. You shared that you have a sense that this individual is very open to the project and keen to hear the results as they are aware it's not quite right for autistic young people. This is potentially why you feel you are holding on to the intervention, and you plan on sharing results with their team. Ideally you would like to be able to create an adapted group to be able to disseminate to the service and the lab. Feeding back could be difficult if the outcome is the group is not appropriate.

For you, a good outcome would be for parents to still hold onto it being helpful, but to be able to make some reasonable adjustments (Eg. Adding in a module about autism-specific elements). You would like to identify some concrete changes (eg. changing the tone of the sessions from a facilitator point of view, Taking longer, sharing that if this doesn't work you're not a failure.)

A bad outcome for you would be having to scrap the group all together. You shared some concerns that every parent says something different so you are worried there will be no clear themes. You shared that you worry sometimes you have been influenced by what previous participants have said and you may ask later participants more about this. You shared that you have been reflecting on this as you have been going along and trying to be more neutral. Generally speaking you feel you have a lack of experience with qual analysis and every interview you are thinking about how to come up with themes and hold worries about how and when to identify themes.

Appendix I. SIP: Lay Summary

Background: Anxiety commonly co-occurs with autism and there is research to suggest that anxiety is caused, maintained and presents differently in autistic individuals. However, to treat anxiety in autistic individuals, many services use manualised cognitive behavioural therapy (CBT) approaches.

Aims. This service improvement project was carried out in a Child and Adolescent Mental Health Service (CAMHS) in the United Kingdom. The service offered a manualised CBT-based parent-led therapy group for anxious children under 12, but noticed that a large proportion of parents who attend the group had autistic children. The service were keen to understand the outcomes for autistic children and how parents of autistic children experienced the group.

Methods. We looked at the pre- and post-treatment scores on anxiety questionnaires, and at the post-treatment status (i.e. what happened to them after the group) of parents who attended one of the therapy groups. We also conducted interviews with eight parents of autistic children who had attended a recent group.

Results. The questionnaire scores and post-intervention status suggest that the group was not effective in reducing anxiety in autistic children, with many children requiring further therapy. The interviews were analysed using reflective thematic analysis and four key themes, and five subthemes were generated. Theme 1 highlighted that the group was not the right fit for autistic children, and this was split into three subthemes, suggesting that the child found it hard to engage in the intervention (1a), that the group was not tailored to the individual circumstances of autistic children (1b), and that it was not the right fit for the "autistic brain" (1c). Theme 2 suggested that many parents attended the group as a means to an end, to be offered a different intervention for example. Theme 3 highlights that, despite these

challenges, parents identified some helpful learning points from the group. Theme 4 also highlights that parents benefited from therapy non-specific factors, such as connecting with other parents (subtheme 4a) and feeling reassured and validated by the group facilitators (subtheme 4b).

Recommendations. Based on these results, a number of recommendations were co-produced with service-users and clinicians. Most notably, it was recommended that the group no longer be offered as standard, and instead the service focus their resource on providing specific workshops on issues that are highlighted in the literature, and by parents, as contributing to their child's anxiety (e.g. sensory differences). Recommendations were also made to use autism-specific outcome monitoring, and to offer 1:1 tailored interventions to autistic children experiencing anxiety.

Appendix J. TDRP: Author guidelines for the Journal of Pediatric Psychology[11]

Instructions to Authors

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References:

- Bernard, R. S., Cohen, L. L., & Moffett, K. (2009). A token economy for exercise adherence in pediatric cystic fibrosis: A single-subject analysis. Journal of Pediatric Psychology, 34, 354-365.
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- Rapoff, M., & Stark, L. (2008). Editorial: Journal of Pediatric Psychology statement of purpose: Section on single-subject studies. Journal of Pediatric Psychology, 33, 16-21.

Review articles

1. Topical Reviews: Topical reviews summarize contemporary findings, suggest new conceptual models, or highlight noteworthy or controversial issues in pediatric psychology. Topical reviews are not intended to provide short data summaries or syntheses. Rather they are intended to foster new ways of thinking about a topic area and provide a direction for future research or practice.
2. Systematic reviews and Meta-Analyses: Systematic reviews provide a research synthesis of a body of literature using an explicit methodology to minimize bias and ensure conclusions are made from reliable findings. Authors of systematic reviews that do not include a meta-analysis must provide a clear justification in the manuscript explaining why such an analysis is not included for all or relevant portions of the report. Please note the PRISMA should be submitted with your manuscript.
3. Scoping Reviews: Scoping reviews determine the scope of a body of literature on a particular topic and identify the volume of the literature (i.e., available studies), and provide an overview of its focus. These are particularly helpful for emerging evidence. Please note the PRISMA-ScR should be submitted with your manuscript.

Please consult this editorial (New Guidelines for Publishing Review Articles in JPP) which further describes guidelines for review articles.

Invited commentaries

Commentaries are invited on all topics of interest in pediatric psychology, and the page length and scope should be discussed with the Editor. Un-invited commentaries will not be considered.

Reporting Guidelines

JPP requires that the relevant reporting guidelines be used for the following studies:

- Randomized trials: CONSORT
- Pilot and feasibility trials: CONSORT extension
- Non-randomized trials: TREND
- Scoping reviews: PRISMA-ScR
- Systematic reviews: PRISMA

Editable checklists for reporting guidelines may be found on the EQUATOR network site. All intervention studies (RCTs and non-randomized trials) will undergo an additional review for transparent reporting conducted by the JPP Assistant Editor for Transparent Reporting. Review comments will be provided on the corresponding checklist. Authors will be required to address any identified reporting issues prior to publication.

Please clearly indicate the page numbers where each checklist item is reported in the manuscript. Please upload this checklist as supplementary material when you submit your manuscript for consideration. If a component of a checklist was not included in the manuscript, an explanation of the rationale for exclusion should be provided. Adherence to these reporting requirements provides standardization, ensures that important information has been included, and facilitates the peer review process.

We publish the final version of all required checklists as supplementary material. Thus, a final version of your CONSORT/TREND/PRISMA checklist will be requested as supplementary material prior to final acceptance of your manuscript. Please note the checklist should be reference in the methods section of your manuscript.

Organizing and Preparing Manuscripts

The Journal of Pediatric Psychology offers authors high-quality online publication. To ensure rapid and efficient publication, please follow the instructions below.

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Type of Manuscript	Length Limit (Text, exclusive of title page, abstract, figures/tables, and references)	Total Number Figures/Tables	Maximum Number of references
Original	5,000 words (20 pages)	5	50
Reviews:			
• Topical	2,000 words	2	30
• Systematic/Scoping	6,250 words (25 pages)	8	Unlimited
Commentaries			
• Student	1,000 words	0	12
• General (Invited)	1,500 words	0	12

General Formatting

1. File format. Please save the main manuscript file as a .doc format.
2. Font size and type. Please use a standard font that is compatible with Windows, such as Times New Roman or Arial. Font size should be 12 pt.
3. Double-spacing. Submissions should be double-spaced throughout, with margins of at least 1 inch and font size of 12 points (or 26 lines per page, 12-15 characters per inch).
4. Naming Files. When naming your files, please use simple file names and avoid special characters and spaces. If you are a Macintosh user you must type the three-letter extension at the end of the file name you choose (e.g. .doc, .rtf, .tif).

Manuscript Formatting

The American Psychological Association Publication Manual (7th edition) should be used to guide manuscript formatting, with the exception of the title page and abstract as noted below. Authors MUST format their references per the American Psychological Association Publication Manual (7th edition) . Failure to do so will cause their manuscript to be unsubmitted until this is corrected.

Title Page

In addition to the APA Manual, the academic degrees of authors should be placed on the title page following their names.

Abstract

A structured double-spaced abstract of not more than 250 words should be included. The abstract should include the following parts:

- a. Objective (brief statement of the purpose of the study)
- b. Methods (summary of the participants, design, measures, procedure)
- c. Results (the primary findings of this work)
- d. Conclusions (statement of implications of these data)

Body of the Manuscript

- a. Introduction
- b. Methods - Informed consent and ethical treatment of study participants: Authors should indicate in the Method section of relevant manuscripts how informed consent was obtained and report the approval of the study by the appropriate Institutional Review Board(s).
- c. Results
- d. Discussion - Clinical relevance of the research should be incorporated into the manuscripts. There is no special section on clinical implications, but authors should integrate implications for practice, as appropriate, into papers.

Acknowledgements

Add appropriate acknowledgements, including information on the funding sources as noted below:

Funding

Details of all funding sources for the work in question should be given in a separate section entitled "Funding." The following rules should be followed:

- The sentence should begin: "This work was supported by . . ."
- The full official funding agency name should be given, i.e. "the National Cancer Institute at the National Institutes of Health" or simply "National Institutes of Health," not "NCI" or "NCI at NIH" (full RIN-approved list of UK funding agencies)
- Grant numbers should be complete and accurate and provided in parentheses as follows: "(grant number xxxx)."Multiple grant numbers should be separated by a comma as follows: "(grant numbers xxxx, yyyy)"
- Agencies should be separated by a semi-colon (plus ‘and’ before the last funding agency)
- Where individuals need to be specified for certain sources of funding the following text should be added after the relevant agency or grant number "to [author initials]."

Oxford Journals will deposit all NIH-funded articles in PubMed Central. See the Complying with funder policies page for details. Authors must ensure that manuscripts are clearly indicated as NIH-funded using the guidelines above.

Tables and Figures

Tables

Tables should be included as separate pages using acceptable formats (e.g., .doc files).

Figures

Figure resolution should be no less than 300 dpi for halftone color (photo) images, 600 dpi for combination halftones, and 1200 dpi for line art. Most standard figure formats are acceptable, such as .jpg, .gif, .tif, or .eps format. Please follow this link for useful information on preparing your figures for publication.

Figure accessibility and alt text

Incorporating alt text (alternative text) when submitting your paper helps to foster inclusivity and accessibility. Good alt text ensures that individuals with visual impairments or those using screen readers can comprehend the content and context of your figures. The aim of alt

text is to provide concise and informative descriptions of your figure so that all readers have access to the same level of information and understanding, and that all can engage with and benefit from the visual elements integral to scholarly content. Including alt text demonstrates a commitment to accessibility and enhances the overall impact and reach of your work.
Alt text is applicable to all images, figures, illustrations, and photographs.
Alt text is only accessible via e-reader and so it won't appear as part of the typeset article.
Detailed guidance on how to draft and submit alt text.

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As an author, you must obtain permission for any material used within your manuscript for which you are not the rightsholder, including quotations, tables, figures, or images. In seeking permissions for published materials, first contact the publisher rather than the author. For unpublished materials, start by contacting the creator. Copies of each grant of permission should be provided to the editorial office. The permissions agreement must include the following:

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Diversity, Equity, and Inclusion Considerations

The Journal of Pediatric Psychology (JPP) and Clinical Practice in Pediatric Psychology (CPPP) both identified enhancing reporting practices for diversity, equity, and inclusion (DEI) in pediatric psychology as a top priority (Duncan, 2023; Modi, 2023). Building upon a guidance for reporting on race and ethnicity in JPP articles (Palermo et al., 2021), we recently published an editorial focused on recommendations on inclusive language and transparent reporting relating to diversity dimensions. Our current efforts strive to address a broader set of diversity dimensions, using the ADDRESSING Framework (Hays, 2016) as guidance, with the goal of enhancing the rigor and inclusiveness of pediatric psychology science. Several important existing checklists and guidelines on inclusive language and transparent reporting relating to DEI exist (American Psychological Association, 2023b; Buchanan et al., 2021; Letzen et al., 2022; Matsui et al., 2020; Miller et al., 2019; Williford, Sweenie, et al., 2023). However, no comprehensive guidelines were available that captured the specific context of pediatric psychology (e.g., developmental/family considerations), while also being broadly applicable across the field (e.g., not focused on a specific condition or symptom). Acknowledging that this is a rapidly evolving area of science, the updated guidelines represent a living document that will exist online within the Instructions for Authors of both journals, which will be updated yearly. Please find the first iteration as a guidance to

implement optimal DEI reporting practices (See Table 1), as well as examples of how these apply to the manuscript types published in our journals (see Table 2).
Table 1. Diversity Dimensions Checklist
Table 2. Application of Diversity Dimensions Checklist in Study Design and Manuscript Preparation
Please see full editorial.

Appendix K. TDRP: STROBE Checklist for cross-sectional studies

	Item No	Recommendation	Location
Title and abstract	1	(*a*) Indicate the study's design with a commonly used term in the title or the abstract	P74
		(*b*) Provide in the abstract an informative and balanced summary of what was done and what was found	P74
Introduction			
Background/rationale	2	Explain the scientific background and rationale for the investigation being reported	P73 – P78
Objectives	3	State specific objectives, including any prespecified hypotheses	P78 – P79
Methods			
Study design	4	Present key elements of study design early in the paper	P79
Setting	5	Describe the setting, locations, and relevant dates, including periods of recruitment, exposure, follow-up, and data collection	P80
Participants	6	(*a*) Give the eligibility criteria, and the sources and methods of selection of participants	P79 – P80 (including table 1)
Variables	7	Clearly define all outcomes, exposures, predictors, potential confounders, and effect modifiers. Give diagnostic criteria, if applicable	P80 – P82 (including table 2)
Data sources/ measurement	8	For each variable of interest, give sources of data and details of methods of assessment (measurement). Describe comparability of assessment methods if there is more than one group	P80 – P82 (including table 2)
Bias	9	Describe any efforts to address potential sources of bias	P80 – P82 (Standardised measures were employed. Efforts were taken to recruit participants from similar pools)
Study size	10	Explain how the study size was arrived at	P79
Quantitative variables	11	Explain how quantitative variables were handled in the analyses. If applicable, describe which groupings were chosen and why	P82 – P83
Statistical methods	12	(*a*) Describe all statistical methods, including those used to control for confounding	P82 – P83
		(*b*) Describe any methods used to examine subgroups and interactions	P82 – P83
		(*c*) Explain how missing data were addressed	N/A
		(*d*) If applicable, describe analytical methods taking account of sampling strategy	N/A
		(*e*) Describe any sensitivity analyses	N/A
Results			
Participants	13	(a) Report numbers of individuals at each stage of study—eg numbers potentially eligible, examined for eligibility, confirmed eligible, included in the study, completing follow-up, and analysed	P79
		(b) Give reasons for non-participation at each stage	N/A
		(c) Consider use of a flow diagram	N/A

Descriptive data	14	(a) Give characteristics of study participants (eg demographic, clinical, social) and information on exposures and potential confounders	P84 – P85
		(b) Indicate number of participants with missing data for each variable of interest	N/A
Outcome data	15	Report numbers of outcome events or summary measures	N/A
Main results	16	(*a*) Give unadjusted estimates and, if applicable, confounder-adjusted estimates and their precision (eg, 95% confidence interval). Make clear which confounders were adjusted for and why they were included	P84 – P87
		(*b*) Report category boundaries when continuous variables were categorized	P83
		(*c*) If relevant, consider translating estimates of relative risk into absolute risk for a meaningful time period	N/A
Other analyses	17	Report other analyses done—eg analyses of subgroups and interactions, and sensitivity analyses	P84 – P87
Discussion			
Key results	18	Summarise key results with reference to study objectives	P87 – P88
Limitations	19	Discuss limitations of the study, taking into account sources of potential bias or imprecision. Discuss both direction and magnitude of any potential bias	P90 – P91
Interpretation	20	Give a cautious overall interpretation of results considering objectives, limitations, multiplicity of analyses, results from similar studies, and other relevant evidence	P87 – P91
Generalisability	21	Discuss the generalisability (external validity) of the study results	P87 – P91
Other information			
Funding	22	Give the source of funding and the role of the funders for the present study and, if applicable, for the original study on which the present article is based	N/A (not reported for the doctoral thesis)

Appendix L. TDRP: Ethics approval correspondence

From: MSD Ethics <ethics@medsci.ox.ac.uk>
Date: Wednesday, 30 November 2022 at 14:09
To: Roberta Button <roberta.button@hmc.ox.ac.uk>
Cc: Matthew Hotton <matthew.hotton@hmc.ox.ac.uk>
Subject: R83405/RE001: Investigating Parenting and Its Relation to Child and Parental Anxiety in Children with a Cleft Lip.....-Approval & Letter

Dear Roberta,

Thank you for submitting the documentation for your proposed research study to MS IDREC for ethical review.

The study titled 'Investigating Parenting and Its Relation to Child and Parental Anxiety in Children with a Cleft Lip and/ or Palate' has been granted ethical approval, under reference R83405/RE001, for a period of 18 months commencing today (30th November 2022).

Please find a formal letter of approval attached.

For our records, please send clean versions of all documents (comments removed and tracked-changes accepted), with the version numbers re-set to 1.0 for each, and with the ethics approval reference inserted.

Best wishes
Leah

Appendix M. TDRP: Parental Overprotection Measure

POI: Parent Self-Report

Instructions: For each question put a mark in the circle that best applies to you ranging from **0-Not At All** to **4-Very Much**.

	Not At All				Very Much
	0	**1**	**2**	**3**	**4**
1. I comfort my child immediately when he/she cries	○	○	○	○	○
2. When playing in a park, I keep my child within a close distance of me (i.e., within about 30m)	○	○	○	○	○
3. I protect my child from criticism	○	○	○	○	○
4. I give my child extra attention when he/she clings to me	○	○	○	○	○
5. I would not allow my child to go out with family friends if I were not present	○	○	○	○	○
6. I almost always take my child to the doctor if he/she is unwell	○	○	○	○	○
7. I keep a close watch on my child at all times	○	○	○	○	○
8. I tend to be over-protective of my child	○	○	○	○	○
9. I try to anticipate and avoid situations where my child might do something risky	○	○	○	○	○
10. I try to protect my child from making mistakes	○	○	○	○	○
11. I do not allow my child to climb trees	○	○	○	○	○
12. I shelter my child from life's difficulties	○	○	○	○	○
13. When away from home I tend to panic if my child is out of my sight, even for a moment	○	○	○	○	○
14. I am reluctant for my child to play some sports for fear he/she might get hurt	○	○	○	○	○
15. I will only leave my child with close friends or relatives if I have to go out	○	○	○	○	○
16. I accompany my child on all outings	○	○	○	○	○
17. I shield my child from conflict	○	○	○	○	○
18. I do everything possible to protect my child from potential injury	○	○	○	○	○
19. I protect my child from his/her fears	○	○	○	○	○

Appendix N. TDRP: Parental Appraisals of Cleft Questionnaire (PAC-Q)

Parental Experience of Cleft

Listed below are different beliefs and attitudes which parents might sometimes have about their child's cleft. Please rate how much you agree or disagree with these statements. ***We would like to know what you think and feel, not what you may have thought in the past or what you think is the right answer***. Depending on the age of your child, some of these statements may be easier or harder to rate, but please rate all of them as best you can.

Please read each statement carefully and choose a rating out of 100 which best describes how much you usually believe the statement. Write the number in the box after the statement.

I do not believe this statement at all	**I am completely sure that this statement is true**
0	**100**

	Rate between 0-100
1. The cleft team will support me to care for my child.	
2. I will treat all my children the same.	
3. My child's cleft treatment will be distressing for him/her.	
4. The cleft is my fault.	
5. The cleft will make no difference to my child's self-esteem*	
6. The cleft will make my child a stronger person.	
7. My child will feel self conscious about the way they look.	
8. The cleft was caused by something I did.	
9. We will be able to access the medical care we need for our child.	
10. There is nothing I could have done to prevent my child's cleft*	
11. The cleft will make my child more caring and understanding of others.	
12. The cleft will not affect my child's quality of life*	
13. Other people in the family feel embarrassed or ashamed about my child's cleft.*	

14. My child will be just as confident as other children.*	
15. The cleft will bring us closer together as a family.	
16. My child is just the same as other children.	
17. I blame myself for my child's cleft.	
18. The cleft will make my child more determined in life.	
19. I should have done something to avoid this happening to my child.	
20. My child will never feel confident in a group.	
21. My child will be just as happy as other children*	
22. My child's cleft treatment will be distressing for me as a parent.	
23. My child will not feel different because of the cleft*	
24. The cleft has made me grow as a person.	
25. The cleft has had a positive impact on the way I see the world	
26. My child will have stronger friendships than other children.	
27. My child will find it difficult to cope with hospital visits for their cleft care.	
28. Having a cleft will help my child appreciate all that they and our family have.	
29. It was just chance that my child was born with a cleft*	
30. My child's cleft treatment will be difficult for our family.	

Appendix O. TDRP: Skewness and kurtosis of significantly non-normally distributed continuous data

	Skewness	Kurtosis
Child age	0.18	-1.136
RCADS-P	1	0.79
GAD-7	0.67	-.43
OP	0.53	-0.95
PACQ subscale 2	0.37	-0.39
PACQ subscale 4	-1.6	2.28
PACQ subscale 5	-0.5	-0.6