Family Functioning and Mental Health in Families with an Autistic Child or a Child with Intellectual Disability

by

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Thesis Overview

This thesis volume is submitted by Elizabeth Hawkins for the research component of the Clinical Psychology Doctorate at the University of Birmingham.

The first chapter is a meta-analytic review of family functioning in families with an autistic child and/or a child with intellectual disability, compared to families with a typically developing child. The second chapter is an empirical study of family functioning and mental health in autistic young people and their parents. The third chapter contains two press releases that summarise the review and empirical study for a lay audience.

A second volume of the thesis consists of five clinical practice reports (CPRs) based on clinical work completed while on placements throughout the doctorate. This volume has not been submitted to maintain the confidentiality of clients and services. CPR1 consisted of a formulation from cognitive-behavioural and psychodynamic perspectives of a client experiencing dissociation. CPR2 presented a service evaluation of an adult mental health service's waiting times. CPR3 described a case study of work with an older adult and staff within a care home. Due to the pandemic, CPR4 consisted of an academic exercise based on single case experimental designs. CPR5 consisted of an abstract detailing leadership skills developed during facilitation of group supervision for assistant psychologists.

Dedication

This thesis is dedicated to the Ant(h)onys who have so significantly influenced my own experience of family.

Acknowledgements

I am hugely grateful for the expert guidance offered by my supervisor, Dr Caroline Richards, research tutor, Dr Chris Jones, and appraisal tutor, Dr Michelle Fisher, particularly in providing a compassionate and flexible safety net during disruptions to training.

The thesis was conducted alongside fellow doctoral candidate, Lucy Licence, whose significant contribution to the planning and recruitment of the empirical study was invaluable. I would also like to acknowledge the support Lucy and master's placement student Emily Warren in the extensive meta-analysis screening.

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1.1 Abstract

Rationale

Previous meta-analyses have identified poorer outcomes in individuals in families with an autistic child (FwASC) and/or a child with intellectual disability (FwID) but have failed to consider collective family functioning. This quantitative meta-analytic review explored how the functioning of FwASC and/or FwID compared to the functioning of families with a typically developing child (FwTD).

Methods

A systematic search of the literature was conducted to identify studies reporting family functioning differences between FwTD and FwASC and/or FwID. The effect sizes were meta-analysed using a random effects model. The impact of influential primary studies and risk of biases were controlled for. Sub-group analyses and meta-regressions were conducted to explore moderating variables.

Results

There were 24 studies reporting 28 samples, with a total of 14169 participants. A significant difference between the FwASC, FwID and FwID+ASC compared to the FwTD groups was found (X^2=20.83, p<.01), so the neurodevelopmental groups were considered separately for the analysis. After correction for biases, the FwASC/FwTD analysis returned a synthesised standardised mean difference (SMD) of 0.35 and the FwID/FwTD analysis returned a synthesised SMD of 0.22, interpreted as small but significant effect sizes. For FwASC

compared to FwTD, mothers reported poorer family functioning than fathers; no other methodological or sample characteristics moderated the effects.

Discussion

The findings demonstrate that FwASC and FwID report poorer family functioning compared to FwTD, and that this effect was largest for FwASC. This suggests syndrome-specific effects on family functioning need to be further explored, with future research including children alongside their parents as informants.

1.2 Introduction

1.2.1 Autism and Intellectual Disability

Neurodevelopmental conditions, including autism spectrum conditions (ASC)[1] and intellectual disability (ID), are a group of conditions with a childhood onset (American Psychiatric Association, 2013). ASC had an estimated prevalence of 2.25% in England in 2018/19, rising from 0.97% in 2010/11 (McConkey, 2020). ASC are characterised by differences in social communication and restricted, repetitive behaviours, and the current diagnosis is labelled autistic spectrum disorder in the DSM-V (American Psychiatric Association, 2013). ID is characterised by reduced intellectual functioning, typically with an IQ score below 70, and associated difficulties in adaptive functioning, with an estimated childhood prevalence in the UK of 2.5% (Office for National Statistics, 2019; Public Health England, 2016). ASC and ID co-occur at high rates, with 20-70% of autistic individuals estimated to have a co-occurring ID (Thurm et al., 2019) and research suggests ASC and ID may both form part of a complex genetic neurodevelopmental continuum (Morris-Rosendahl

[1] The term autism spectrum conditions (ASC) has been chosen over the diagnostic term autism spectrum disorder (ASD) to reflect the view that autism is a difference rather than a dysfunction (Baron-Cohen, 2015). This is consistent with the neurodiversity perspective (Singer, 1998, as cited in McGee, 2012) and the deficit-as-difference conception of autism (Kapp et al., 2013). The identity-first phrasing ‘autistic children’ is also used, as it is the preferred term by the UK autism community (Kenny et al., 2016).

& Crocq, 2020). While ID and ASC are separate diagnoses in the DSM-V and ICD-11, there can be significant phenotypic overlap. Both diagnoses share a defining feature of delayed early development and reduced adaptive functioning, and both are associated with higher rates of comorbidities, such as behaviours that challenge and poor physical and mental health compared to the general population (Matson & Shoemaker, 2009). The high co-occurrence and aspects of shared aetiology and phenotype provide a rationale for considering ASC and ID *both* as separate diagnostic groups and when co-occurring as part of a neurodevelopmental spectrum of neurodiversity. Doing so is particularly important as individuals with co-occurring diagnoses are currently underrepresented in research (Thurm et al., 2019).

Children with neurodevelopmental conditions should be considered within their familial contexts, as chronic conditions affect the individual themselves, other family members and their interpersonal relationships. As one mother described "the whole family had autism, not just the child" (Lutz et al., 2012, p.210). These families can therefore be considered more broadly as neurodiverse or neurodivergent families[2]. In the UK, the National Institute for Health and Care Excellence (2013, 2018) recommends that services for individuals with ASC or ID include support for families and carers. It is therefore essential for research to support service design with family-level understanding; several researchers, however, have identified a lack of research and policy in this area (Cridland et al., 2014; Tint & Weiss, 2016).

1.2.2 Family Functioning

A family unit can be defined by legal, biological, economic, residential, caring, or emotional relationships. While families increasingly self-define, the household definition is common within research largely for practical reasons (Wollny et al., 2010). Family

[2]An autistic individual can be considered as neurodivergent, and a neurodivergent family contains at least one family member who can be considered neurodivergent. A neurodiverse family would be considered to contain both neurodivergent and neurotypical members.

functioning encompasses the social and structural characteristics of the global family environment, including interactions, relationships, cohesion, conflict, adaptability, organisation and communication (Lewandowski et al., 2010). The degree of family functioning can be measured as a range from positive to negative (Cridland et al., 2014). Good functioning is conceptualised as having clear roles, open and straight-forward communication, and well-regulated affect. Families with poorer functioning may be more chaotic, unfocused and dysregulated, or over-controlled, rigid and restricted (Alderfer et al., 2008).

The importance of family functioning for a child can be understood using Bronfenbrenner's (1977) ecological systems theory model (Figure 1), which views a child's biopsychological development as influenced by a complex system of dynamic relationships at multiple environmental levels. Microsystems, such as the family or school systems with direct contact with the child, are the most influential. These microsystems interact to form interconnected mesosystems, nested within wider society (exosystem) and culture (macrosystem). Family functioning is situated within the microsystem with the family as the unit of measurement and analysis (Greenlee et al., 2018), rather than focusing on individuals (e.g. child, sibling, parent), family sub-systems (e.g. parent-child, marital), or mesosystems (e.g. school-family, neighbourhood-family). Ecological systems theory, alongside family systems-based theories widely used in psychology (Wollny et al., 2010), acknowledge bi-directional and interdependent relationships within the family, distinguishing it from measures of a unidirectional influence, such as caregiver burden. These theories understand the family as interrelated, transactional, and as greater than the sum of its individuals or subgroups.

Several models of family functioning have been developed, influenced by a variety of social and systemic theories. In a systematic review of family measures relevant to paediatric

populations, Alderfer et al. (2008) identified thirteen measures of general family functioning clustered in seven theoretical groups. Measures fall into observational or clinician-rated interviews and self-report questionnaires, with questionnaires being the more commonly used

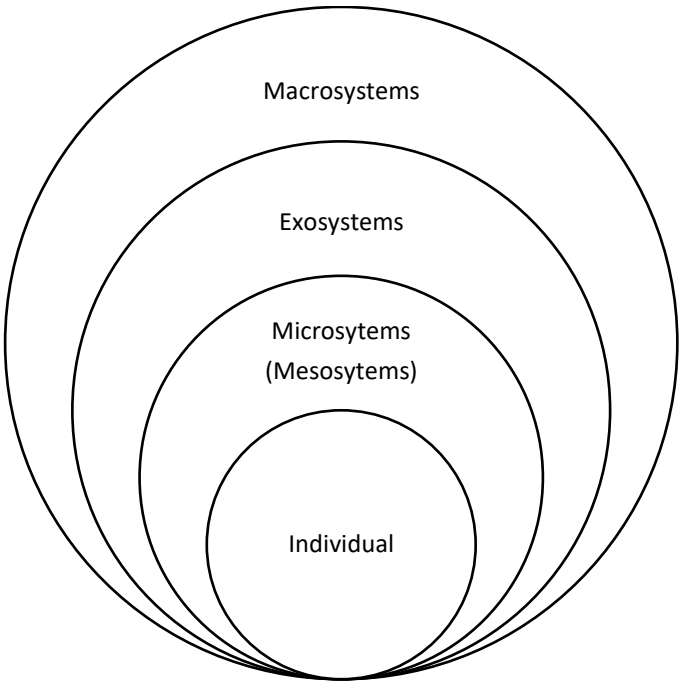

Figure 1: The ecological systems theory model (Bronfenbrenner, 1977)

format as they are short, unintrusive and quick. Most measures aim to assess family organisation, cohesion, communication, sometimes as separate scales or within a single general functioning index reflecting overall adaptation (Alderfer et al., 2008). Three self-report questionnaires (The Family Assessment Device, Epstein et al., 1983; the Family Assessment Measure-III, Skinner et al., 2000; and the Family Relationship Index of the Family Environment Scale, Moos & Moos, 2009) were assessed as well-established, demonstrating use in at least two peer-reviewed articles by different teams with manuals and evidence of good statistical reliability and validity. Another four self-report measures were judged as 'approaching well-established', including the Family Adaptability and Cohesion Evaluation Scale (Olson, 1991). No gold-standard measure has been identified, however, and there are therefore a range of measurement tools covering the concept of family functioning.

Connections between family and child functioning are complex, dynamic and bi-directional (Wollny et al., 2010). Healthy family functioning has been linked to better individual outcomes for autistic children, including improved behaviour, mental health, and quality of life (Greenlee et al., 2018), as well as for parents in neurodevelopmental families, including improved wellbeing, marital happiness, parenting competence and acceptance, and reduced stress (Bonis & Sawin, 2016; Bristol, 1984; Corcoran et al., 2015; Glidden, 2012). Longitudinally, family cohesion, rather than the child's cognitive ability, predicts changes in adaptive functioning in children with Down's syndrome (Hauser-Cram et al., 1999), and family adaptability predicts maternal depression and behaviours that challenge in autistic children (Baker et al., 2011). These studies demonstrate the importance of understanding the family as a collective even when individual-level outcomes are of clinical interest. Turnbull et al. (2007) further highlight the importance of measuring positive dimensions and constructs in family research to move the field away from a focus on individual-level deficits and risk factors, consistent with a neurodiversity perspective. As such, family functioning research offers an alternative approach, which may help elucidate further understanding, protective factors, and potential systemic intervention targets to support both improved family and individual-level outcomes.

1.2.3 Prior Reviews of the Literature

Previous meta-analyses in the field have focused on parent subsystem factors, reflecting a tendency in the literature to focus on the effect of the child on the outcomes of others (Greenlee et al., 2018). These meta-analyses show that parents of autistic children report increased stress, depression and anxiety compared to parents of both typically developing (TD) children and children with ID with small to large effect sizes (Dunn et al., 2019; Hayes & Watson, 2013; Schnabel et al., 2019; Singer, 2006; Yirmiya & Shaked, 2005). A further meta-analysis found poorer mental health in mothers of children with ID compared

to fathers, although paternal mental health worsened with increased disability severity (Dunn et al., 2019). The functioning of siblings has also been meta-analysed (Shivers et al., 2019), with siblings of autistic children reporting more negative outcomes in a wide range of areas compared to siblings of TD children and siblings of children with other neurodevelopmental conditions, including ID. While Shivers et al. (2019) found that family functioning with respect to siblings was similar across neurodevelopmental groups, there were few studies available to include in this meta-analysis. Additionally, the construct was defined at the level of the parent-child sub-system (e.g., attachment) rather than the functioning of the collective family system as defined within ecological and family systems theories. There is therefore a need to synthesise data on family functioning in ASC and ID, as previous meta-analyses have focused on individual or dyad-level outcomes. The common findings in these meta-analyses of poorer individual outcomes for family members of autistic children compared to those families with a child with ID demonstrates the need to compare neurodevelopmental group data by diagnostic subgroups. These prior meta-analyses also share a common theme of focusing on negative outcomes, highlighting a need to synthesise the literature of positive outcomes to prevent bias in research.

The collective functioning of the family system in neurodevelopmental conditions has been examined in narrative, systematic and scoping reviews, but not synthesised via meta-analysis. One systematic review of families of children with ID found impacts across the family sub-systems, highlighting that functioning was negatively affected by increased demands, reduced resources and changed family roles, and positively affected by strong family ties and emotional support (Lima-Rodríguez et al., 2018). A scoping review of wellbeing in families with an autistic child identified poorer outcomes in these families compared to both TD and ID control groups, and suggested this may be due to confounding variables, such as behaviours that challenge and socioeconomic factors (Tint & Weiss, 2016).

A further scoping review found the current literature provides mixed support for family level processes correlating with behavioural, mental health, quality of life and autism severity outcomes (Greenlee et al., 2018).

Turnbull et al. (2007) highlight an overall lack of diverse and representative samples within the field of family functioning, with the majority of studies focused on mothers (Hayes & Watson, 2013). Other methodological issues identified include a focus on cross-sectional research using small convenience samples (Glidden, 2012), inconsistent findings, unclear risk and protective factors, lack of exploration of gender and age variables, and a lack of conceptual models (McStay et al., 2015). There is therefore a need to move beyond the existing narrative reviews and synthesise statistically the data on family functioning in families with children with neurodevelopmental conditions. This meta-analytic approach will confer opportunities to further explore how participant variables and methodological choices moderate family functioning outcomes, which may help elucidate risk and protective factors, screening and intervention targets, and best methodological practice for future research.

In summary, ASC and ID are neurodevelopmental conditions with significant overlap and commonalties, particularly in their impact on the whole family. Previous meta-analyses in this area have identified poorer outcomes for individuals within these families but have failed to consider the collective family system. In the present meta-analysis, ASC and ID will be considered jointly *and* separately where possible, to account for their co-occurrence but differential outcomes identified in prior narrative reviews. Family-level and methodological variables will be explored as potential moderators of effect sizes and to evaluate the overall quality of the literature.

This review aims to answer the following research question: How does the functioning of families with an autistic child (FwASC) and/or a child with intellectual disability (FwID) compare to the functioning of families with a typically developing child

(FwTD)? In keeping with prior research, it is hypothesised that FwASC and/or FwID will report poorer family functioning than FwTD. The review also aims to explore whether methodological or participant characteristics moderate any differences in family functioning between FwASC, FwID and FwTD.

1.3 Methods

1.3.1 Identifying Primary Studies

1.3.1.1 Search of Electronic Databases

The review was pre-registered on Prospero (CRD42020170820; Hawkins et al., 2020). The search and review were conducted according the 2009 PRISMA guidelines (Moher et al., 2009; Appendix 1)[3]. A systematic search of the literature was carried out on 14th May 2020 using the databases Ovid PsycInfo, EBSCO ERIC, Web of Science, and Cochrane Central[4]. The search terms (Table 1) were used to identify relevant constructs in titles and abstracts, with reference to the PICO (population, intervention/exposure, comparator, outcome) search strategy (Richardson et al., 1995). Specific search terms were identified through consideration of previous reviews in these populations, including Richards et al. (2015), Shivers et al. (2019) and Surtees et al. (2018). Search terms were combined with *OR* and constructs were combined with *AND*. A scoping search was conducted prior to this and included individual-level free text search terms (e.g., parent, mother, father), which were subsequently removed as they did not yield additional relevant papers.

[3] The 2020 guidelines were not published when the review was started, but updates to the guidelines have been considered where possible.

[4] Research in autism and intellectual disabilities crosses disciplines and research databases. Databases were selected to focus on psychosocial outcomes, and those with a more medical focus (e.g., Ovid EMBASE, PubMed) were excluded as they were likely to result in a higher number of irrelevant articles.

Table 1: The strategy and terms used to search the databases Ovid PsycInfo, EBSCO ERIC, Web of Science and Cochrane Central for papers on family functioning in autism and intellectual disability populations

Construct	Free Text Search Terms[5]
Autism / intellectual disability	Autis* OR Asperger* OR ASD OR "Pervasive developmental disorder" OR PDD* OR "Learning disab*" OR "Intellectual disab*" OR "Intellectual disturbance*" OR "Developmental disab*" OR "Mental retar*" OR "Mental* handicap*" OR "Mental* deficien*" OR "Mental incapac*" OR Idiocy OR oligophrenia
	AND
Child	Child* OR Adolescen* OR Youth OR Young OR Teen* OR P?ediatric*
	AND
Family	Famil* OR Household*
	AND
Functioning	Function* OR Relation* OR System* OR Resilienc* OR Satisfaction OR "Quality of Life" OR Adapt* OR Well?being OR Adjustment OR Environment OR Cohesi* OR Conflict* OR Climate OR Coping OR Communication OR Flexibil*

1.3.1.2 Inclusion Criteria

Full inclusion/exclusion criteria are detailed in Table 2. At the title/abstract screening stage, empirical quantitative English-language papers that involved cohort research with neurodevelopmental families and family functioning were included. At full-text screening, this was further refined to include peer-reviewed papers that included a measure of family functioning, and that reported group level comparison data for ASC and/or ID groups compared to a TD group.

The results of the systematic search are presented in a PRISMA flow chart (Figure 2; Moher et al., 2009). The search yielded 16375 articles; 2 additional articles were later identified from reference lists of included articles. Duplicates were removed using the duplicate finder within EndNote X9, leaving 12850 articles. Article titles and abstracts were screened against the exclusion criteria. The most common reasons for exclusion were not including family-level data (N=4116), not empirical research (N=4096), qualitative or case studies (N=1983), and not ASC or ID (N=1574). A second researcher independently

[5] Ovid PsycInfo: truncation $, wildcard ?, no exact phrase syntax used; EBSCO ERIC: truncation *, wildcard ?, " " exact phrase; Web of Science: truncation *, wildcard $, no exact phrase syntax used; Cochrane Central: truncation *, wildcard ?, " " exact phrase

Table 2: The inclusion and exclusion criteria for each stage of the meta-analysis screening

Inclusion Criteria	Exclusion Criteria	Justification
	Title and Abstract Screening	
Study written in English language	Study not written in English language	Researchers are English-speaking and unable to translate papers
Human studies	Animal studies	
Empirical research	Non-empirical research (e.g., books, review papers, reports)	
Quantitative research	Qualitative research	A gap in the literature for a quantitative meta-analysis has been identified
Study includes a neurodevelopmental condition group (i.e., autism and/or intellectual disability, or associated conditions, or a non-specified neurodevelopmental condition group)	The study does not include a neurodevelopmental group, or specifies a separate neurodevelopmental diagnosis (e.g., ADHD, tic disorder)	The scoping search identified that autism and/or intellectual disability are sometimes subgroups within broader neurodevelopmental condition groups
Cohort research, including a comparison group	Non-group research (e.g., case studies)	To allow comparison between groups
Includes family functioning or other family-level data	Does not include family functioning or family-level data	The scoping search identified that family functioning measures are not always named explicitly in the abstract
Full-text available	Full-text not available	
	Full-text screening	
Published in a peer review journal	Dissertations	It was not possible to include grey literature within the scope of this review. To mitigate this statistical analysis of publication bias was included in the analysis
Includes a measure of family functioning	Does not include a measure of family functioning	
Reports group level data for autism, intellectual disability, and/or typically developing children	Does not report group level data, uses normative data or data from another study as comparison group, or authors not able to provide additional data	
Study reports group number, mean and standard deviation, or these are obtainable through backward calculation or contact with authors; or study reports data to calculate effect size	Study does not report group number, mean and standard deviation, and these are not obtainable through backward calculation or contact with authors	To enable the calculation of standardised mean difference or Cohen's d

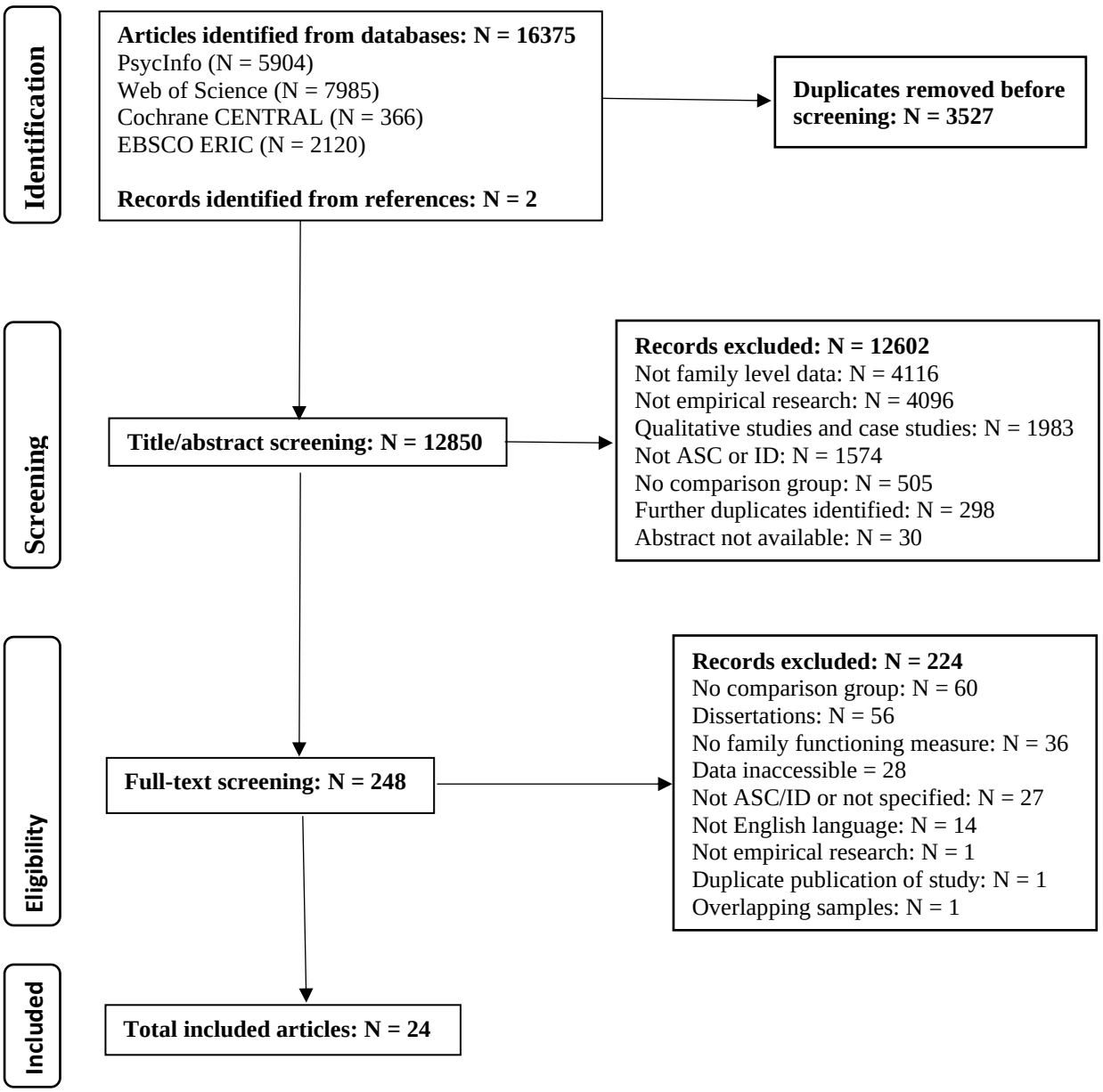

Figure 2: PRISMA flow diagram of the literature search, adapted from Moher et al. (2009)

completed title/abstract screening for a random 10% sample of the 12850 articles. Inter-rater reliability assessed using Cohen's kappa was 0.72, which indicated a substantial agreement (Landis & Koch, 1977; McHugh, 2012). Disputes were resolved by full text screening and all twelve disputed papers were excluded.

The remaining 248 articles were reviewed against the full-text exclusion criteria. A 20% sample was also independently reviewed by a second researcher. Inter-rater reliability

assessed using Cohen's kappa was 0.85, which indicates an almost perfect agreement (Landis & Koch, 1977; McHugh, 2012). Disputes were resolved by a change to the exclusion criteria, clarifying that the comparison group could not be normative data. A further dispute about the Family Orientated Personal Evaluation Scale (McCubbin & McCubbin, 1996) was resolved by reviewing the questionnaire and agreeing to include it.

The main reasons for exclusion at full-text screening were no comparison group (N=60), dissertations (N=56), no family functioning measure (N=36), not ASC/ID or unspecified (N=27), and not English language (N=14). A further 29 papers did not report accessible data; attempts were made to contact authors resulting in one additional paper being included (Cahill et al., 2010), and the remaining 28 papers were excluded. One article (Cuzzocrea et al., 2013) was excluded, as the sample appeared to overlap with another included paper by the same authors based on reported sample demographic data, and the authors could not be contacted for clarification. The final number of articles that satisfied the inclusion criteria for the meta-analysis was 24. Further details about inclusion/exclusion criteria, including excluded measures, are documented in Appendix 2.

1.3.2 Quality Rating and Data Extraction

1.3.2.1 Data Extraction

Data relating to methods, participants and outcomes were extracted by the lead author. A second researcher independently replicated the data extraction on a random 20% sample, and there were no disputes. Where possible, the group N, family functioning score mean and standard deviation (group or pooled) were extracted, and the standardised mean difference (SMD) and standard error were calculated (Borenstein, 2009) using an effect size calculator (Jones, 2021; R Core Team, 2021). Where these data were not reported, results of statistical tests (e.g., t or F statistics) were used to calculate Cohen's d as a measure of mean difference

effect size. A positive effect size was arbitrarily selected to mean greater family functioning in TD controls compared to the ASC or ID groups.

The following study level data were extracted: groups, outcome measure, who completed outcome measure and country. The following information was extracted about the children: type of neurodevelopmental condition, group N, recruitment and screening strategy, gender, age, ethnicity, and level of functioning. The following information was extracted about the parents: marital status, gender, age, ethnicity, and level of education.

1.3.2.2 Risk of Bias Assessment

All papers included cross-sectional data and there is a lack of consensus about how to assess for risk of bias in cross-sectional studies. No tool has been used consistently within similar reviews in the field, with many meta-analyses discussed in Section 1.2.3 neglecting to state if or how bias was assessed (e.g. (Hayes & Watson, 2013; Schnabel et al., 2019; Shivers et al., 2019; Singer, 2006; Yirmiya & Shaked, 2005). An in-press systematic review by Wang et al. (2019) due to be published in the Australian National Health and Medical Research Council's Guidelines for Guidelines highlights the need for the creation of a tool for observational studies that contains the following domains: selection, exposure, outcome assessment, confounding, analysis, selective reporting, conflicts of interest, and other. As such, a set of quality criteria (Table 3) based on these domains was adapted from the JBI Checklist for Analytical Cross Sectional Studies (Moola et al., 2020), a tool recommended by Ma et al. (2020). Low risk of bias categories were scored 2 points and unclear risk categories scored 1 point, giving a maximum total of 14 points; the overall risk of bias is reported as a percentage quality index score. The risk of bias assessment for individual papers is shown in Table 4 and a summary across the studies is shown in Table 5.

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Table 3: The risk of bias assessment criteria.

Bias Type	Risk of Bias Assessment
Selection	Low risk: • The characteristics of the source population, setting and sample are clearly described and without evidence of bias. • The recruitment method is from a general population (e.g., schools, local/national database). Unclear risk: • The characteristics of the source population, setting and sample are not clearly described. • The recruitment process is not clearly reported. • The recruitment process relied on convenience sampling (e.g., flyers) or is through general support services. High risk: • The characteristics of the source population, setting and sample show evidence of bias. • The recruitment process is inappropriate. • The recruitment process relied on target sampling (e.g., recruitment from support services for behaviour/mental health etc).
Exposure	Low risk: • Autism/ID has been verified by trained researchers or qualified professionals. Unclear risk: • Participants self-report receiving a diagnosis, or a screening tool has been used. High risk: • Autism/ID has not been verified by trained researchers or qualified professionals, or no information is given.
Outcome assessment	Low risk: • The outcome measures are clearly defined and used as intended. • The outcome measures are valid and reliable detailed in the paper itself or the Alderfer (2008) review. • The outcome measures are implemented consistently across all participants. • There is no evidence of different motivational biases between groups (e.g., anonymity, confidentiality, rewards, blinding). Unclear risk: • The outcome measures are not clearly defined. • The outcome measures' validity and reliability are not reported in this or other review papers. • It is unclear whether the measure was implemented consistently across all participants. • It is unclear whether groups might have different motivational biases. High risk: • The outcome measure is not defined or not used as intended (e.g., one subscale, translated) • The outcome measures are not valid and reliable. • The outcome measures are not implemented consistently across all participants. • There is evidence of different motivational biases between groups.

Bias Type	Risk of Bias Assessment
Confounding	Low risk: • The characteristics of each group and any significant differences are clearly reported. • Confounding factors have been identified and strategies to deal with these have been clearly stated and are appropriate (e.g., matching/ stratifying sampling). Unclear risk: • The characteristics of each group and any significant differences are not clearly reported. • It is unclear whether confounding factors have been identified, clearly stated or are appropriate. High risk: • The characteristics of each group or any significant differences have not been measured. • Confounding factors have not been identified, or no/ inappropriate strategies to deal with these have been stated.
Statistical / Analysis	Low risk: • Appropriate statistical testing is used and clearly reported. Unclear risk: • It is unclear what statistical testing is used. High risk: • Inappropriate statistical testing is used.
Reporting	Low risk: • The results of all measures reported in the methods are clearly reported for all participants. • The results of the family functioning measure are reported as per the manual. Unclear risk: • The results of all measures reported in the methods are not clearly reported (e.g., narrative report rather than statistical report). High risk: • The results of all measures reported in the methods are not reported for all participants. • The results of the family functioning measure are not reported as per the manual.
Conflicts of Interest	Low risk: • The authors clearly report no conflict of interest (e.g., through declaring funding source), or report how conflicts of interest were appropriately managed. Unclear risk: • It is unclear whether there are any conflicts of interest. High risk: • There is evidence of conflicts of interest that have not been appropriately managed.

Table 4: The risk of bias assessment for included papers

Paper	Selection Bias	Exposure Bias	Outcome Bias	Confounding Bias	Statistical Bias	Reporting Bias	Conflict of Interest Bias	Quality Index Score (%)
Al-Krenawi et al., 2011								86
Becker et al., 1997								64
Blacher et al., 1997								71
Cahill et al., 2010								50
Cuzzocrea et al., 2011								57
Duvekot et al., 2017								79
Emerson, 2003								57
Gau et al., 2012								79
Heiman & Berger, 2008								79
Kissel & Nelson, 2016								64
Kostiukow et al., 2019								71
Lynch et al., 1993								86
Manor-Binyamini, 2016								93
Nuske et al., 2018								57
Pisula & Porebowicz-Dorsmann, 2017								86
Rao & Beidel, 2009								93
Rodrigue et al., 1992								93
Schneider et al., 2019								79
Sivberg, 2002								57
Sullivan et al., 2012								79
Tsibidaki & Tsamparli, 2009								93
van Riper et al., 1992								71
von Gontard et al., 2002								86
Walton, 2019								86

Red = high risk of bias; amber = unclear risk of bias; green = low risk of bias

Table 5: The risk of bias summary for included papers showing N and percentage of papers in each risk category for each domain

Bias	Low risk N (%)	Unclear risk N (%)	High risk N (%)
Selection	7 (29)	14 (58)	3 (13)
Exposure	10 (42)	11 (46)	3 (13)
Outcome	15 (63)	3 (13)	6 (25)
Confounding	19 (79)	3 (13)	2 (8)
Statistical	23 (96)	0 (0)	1 (4)
Reporting	22 (92)	0 (0)	2 (8)
Conflict of Interest	9 (38)	14 (58)	1 (4)

Overall, the selection bias was unclear within the studies, with 14 of the 24 papers rated as unclear, commonly because of reliance on convenience sampling. Exposure bias was generally low or unclear, with 10 papers verifying ASC/ID through professional or researcher diagnosis (low risk) and 11 relying on self-report or screening tools (unclear risk). Outcome bias was mixed, with 15 papers assessed as low risk for using a clearly defined, valid, reliable measure consistently as intended across all participants. A range of family functioning measures were included in the meta-analysis, shown in Appendix 2

Confounding bias was generally low, with 19 papers assessed as low risk, and the characteristics of samples generally clearly reported. Statistical and reporting biases were both overwhelmingly judged to be low, as most commonly raw data in the form of group means, standard deviations and sample sizes were available to meta-analyse. More than half of the papers were assessed as unclear risk with respect to conflicts of interest; however, it seems likely that this is due to a lack of reporting, particularly in older studies due to change in publishing standards, rather than a high level of conflict within this field.

Overall, there was a mixed level of bias across the studies included in the meta-analysis. No studies were assessed as low risk across all domains; 13 of the 24 papers were assessed as high risk in at least one domain. There was a notable risk of outcome bias, and unclear risk due to selection, exposure, and conflict of interest biases. Due to the low number of studies in this field, studies with medium to high risk of bias were included and statistical

analyses were undertaken to incorporate the risk of bias analysis within the pooled effect sizes.

1.3.3 Data Analysis

Where family functioning data were recorded at multiple time points for the same participants, the baseline data were selected to better represent the general population and maximise sample N. Studies that included both maternal and paternal reports of family functioning were included, with maternal and paternal data as independent comparisons in the main analysis, and separately in the sub-group analysis on this variable. While this may have caused subtle complications regarding random-effects analyses, the effect is likely to be trivial (Higgins et al., 2021). For studies that included autism, ID and TD groups, the autism and ID groups were combined to create a single pair-wise comparison for the main analysis of neurodevelopmental families compared to FwTD, and included as separate groups where possible for the sub-group analysis of this variable (Higgins et al., 2021). In papers where multiple family functioning outcome measure domains were reported, the average effect was calculated for equivalent scales (Borenstein et al., 2009; McKenzie et al., 2021). For domains that were not equivalent, either clinically or numerically, cohesion was selected as the most clinically relevant outcome across a variety of measures, as judged by the researchers based on previous reviews (McKenzie et al., 2021; McStay et al., 2015). All measures were interpreted as linear dimensions[6].

To test the hypothesis that FwASC and FwID report poorer family functioning than FwTD, the effect sizes (SMD or Cohen's d) measuring the difference in family functioning between the groups were meta-analysed. The extracted data were analysed using a bespoke meta-analysis programme (Jones, 2020; R Core Team, 2021). A random effects model was

[6] One measure, the FACES-III, was originally designed to measure extremes of family functioning, but its interpretation has since been revised and the authors recommend re-interpretation of prior studies based on a linear dimension (Olson, 1991).

selected to account for variation attributable to the neurodevelopmental groups as well as for variation attributable to other factors, such as methodological variation, measurement error and uncontrolled confounding variables. The model was calculated using the generic inverse variance method, which allows for a range of effect size formats and weights studies based on sample size and variance.

Heterogeneity in an effect size can result from methodological variation, measurement errors, or confounding participant characteristics. A high level of heterogeneity reflects variation in the effect that cannot be attributed to true variation in a population. Higgins I^2 is a commonly used measure of heterogeneity (Borenstein et al., 2009; Higgins et al., 2003), expressed as a percentage ranging from 0-100 with higher values indicating higher levels of heterogeneity. Problematic heterogeneity, where studies cannot be considered to be of the same population, has been defined as a Higgins I^2 value above 75% to reflect a literature with methodological variation that would be expected to contribute to variation.

The impact of disproportionately influential studies was explored. The random effects model was calculated with each of the primary studies removed in turn and change in weighted average effect size (i.e., influence) and the change in heterogeneity (i.e., discrepancy) was examined. Highly influential and discrepant studies were reviewed and excluded where risk of bias was identified, and a corrected meta-analytic effect size calculated. Risk of bias was accounted for using a quality effects model (Tritchler, 1999) which extends the random effects model with the addition of the risk of bias ratings to the weightings (quality index score in Table 4). This model can be interpreted as the synthesis that could be obtained if all the studies were of the same methodological quality as the best study in the review.

Publication bias results from the tendency for statistically significant results to be published over and above those with non-significant results. Small study bias is the tendency

for studies with smaller sample sizes to show greater variability in their measurement of family functioning due to power and precision effects. These biases are inter-related, as small studies are more likely to report non-significant results and therefore less likely to be published. These biases can be identified using a funnel plot, which plots the magnitude of the study's effect size estimate against a measure of the study size or precision (the square root of the study's sampling variances). The 95% confidence interval of the expected distribution of effect sizes is shown as an inverted funnel, and the overall meta-analytic effect is shown as a vertical line. In the absence of publication bias, small samples with greater variability will scatter more widely at the bottom of the plot, whereas larger samples will lie closer to the overall meta-analytic effect line. In the absence of bias, it would be expected that the plots resemble the symmetrical inverted funnel. The funnel plots were visually inspected; however this method can be susceptible to inaccuracies and biases (Terrin et al., 2005) and Egger et al.'s (1997) statistic was therefore also calculated to assess for publication and small study biases. Rosenthal's (1979) failsafe number was also calculated to represent the number of studies averaging null results that would be needed to reduce the meta-analytic effect size to non-significance.

1.4 Results

In total, 24 papers were included in the meta-analysis. The study-level and sample-level participant variables of included studies are summarised in Table 6.

1.4.1 Families with Autism and/or Intellectual Disability

The distribution of primary study effect sizes relative to those expected in a normal distribution is shown in Figure 3. There was no evidence of substantial non-normality in the distribution within the primary studies, so the variance of the true effect (tau^2) was calculated using the DerSimonian & Laird (1986) estimator method.

Table 6: A summary of the study-level and participant sample-level variables of included studies

Paper	Outcome Measure (reporter)	Recruitment *Screening*	ID/ASC (NDD N / TD N)	Child Characteristics NDD Group / TD Group			Parent Characteristics NDD Group / TD Group			
				Age Mean *Range*	% Male	Diagnoses (%) *Functioning*	% Race / Ethnicity	% Two-parent household	Age Mean *Range*	% Post-School Qualifications
Al-Krenawi et al., 2011	FAD (parents)	Schools & community in Israel *School professional*	ID (300/100)	- *<5-22 / -*	68 / -	*Receiving support 93%*	Bedoiun-Arab 100 / 100	76 / 81	- *30-60+ / 30-60+*	3 / -
Becker et al., 1997	F-COPES (mothers)	Agencies, clinics & community in US *BSID*	ID (30/30)	1 / 1 -	67 / 57	Mental delay 63 Known syndrome 37	Caucasian 100 / 97 Native American 0 / 3	100 / 100	30 / 30 -	
Blacher et al., 1997	FES (mothers or grandmothers)	Regional centres, flyers, peer nomination in US *Professionals, self-report, or service access*	ID (148/101)	11 / 8 -		*VABS mean 31.9*	Latina 100 / 100	62 / 79	41 / 37 -	4 / -
Cahill et al., 2010	SCORE-28 (ID parents; TD children)	Disability services & schools in Ireland	ID (146/132)	- *- / 15-20*	- / 79	Physical and mental disability 100			- / 38	- / 54

Paper	Outcome Measure (reporter)	Recruitment *Screening*	ID/ASC (NDD N / TD N)	Child Characteristics NDD Group / TD Group			Parent Characteristics NDD Group / TD Group			
				Age Mean *Range*	% Male	Diagnoses (%) *Functioning*	% Race / Ethnicity	% Two-parent household	Age Mean *Range*	% Post-School Qualifications
Cuzzocrea et al., 2011*	FACES-III (mothers and fathers)	Rehab centres in Italy	ID+ASC (16/16)	7 / 7 -		Down's syndrome 44 West syndrome 6 Angelman syndrome 6 Autism 44			45 / - -	50 / -
Duvekot et al., 2017	FAD (primary caregiver)	Child services in Netherlands *ADOS-2, SRS-2, 3Di-sv*	ASC (92/74)	- *1.5-18 / 1.5-18*						
Emerson, 2003	FAD-GF (mothers)	Stratified sample from a survey in UK *Parent/teacher report, school attendance*	ID (245/9481)	- *5-15 / 5-15*	72 / 49	*Mean SDQ* *Parent 16.6 / 8.1* *Child 14.1 / 10.1* *Teacher 15.3 / 6.1*	White 94 / 93 Black 3 / 2 South Asian 2 / 3 Other 2 / 2	71 / 29		10 / -
Gau et al., 2012*	FACES-III Chinese version (mothers & fathers)	Services & schools in Taiwan *Professional diagnosis, ADI-R, teacher-report*	ASC (151/113)	8 / 8 -	87 / 61			100 / 97	Fathers 40 / 41 - Mothers 37 / 38 -	Fathers 63.4 / - Mothers 45 / -

Paper	Outcome Measure (reporter)	Recruitment *Screening*	ID/ASC (NDD N / TD N)	Child Characteristics NDD Group / TD Group			Parent Characteristics NDD Group / TD Group			
				Age Mean ***Range***	**% Male**	**Diagnoses (%)** ***Functioning***	**% Race / Ethnicity**	**% Two-parent household**	**Age Mean** ***Range***	**% Post-School Qualifications**
Heiman & Berger, 2008	FES (parents)	Asperger's organisation & schools in Israel *Professional diagnosis and IQ assessment*	ASC (33/45)	13 /11 -		Asperger's 100 *Mainstream education with support 100%*			49 / 40 -	
Kissel & Nelson, 2016	FAM-III (parents)	Services and schools in US *Parent-report*	ASC (48/16)	9 / 9 -	96 / 63	*GARS-2 Severe behaviours 69%* *Less severe behaviours 31%*	Caucasian 83 / 88 Latino 6 / 0 Asian Pacific Islander 0 / 6 Other 10 / 6	88 / 100	41 / 40 -	76 / -
Kostiukow et al., 2019	FACES-IV (parents)	Poland	ASC (70/70)						- *25-45*	- *25-45*
Lynch et al., 1993	FES (parents)	School programs & paediatric clinics in US	ID (9/10)	13 / 14 -		*Moderate to profound 100*	Caucasian 75 / 92			
Manor-Binyamini, 2016	FACES-III (mothers)	Schools in Israel *Professional diagnosis*	ID+ASC (160/80)	- *12-21 / -*	64 / -	Autism 23 ID 66 Genetic & chromosomal syndrome 9 Down's syndrome 2	Druze 100 / 100	97 / 98	- *30-50+ / 30-50+*	3 / -

Paper	Outcome Measure (reporter)	Recruitment *Screening*	ID/ASC (NDD N / TD N)	Child Characteristics NDD Group / TD Group			Parent Characteristics NDD Group / TD Group			
				Age Mean *Range*	% Male	Diagnoses (%) *Functioning*	% Race / Ethnicity	% Two-parent household	Age Mean *Range*	% Post-School Qualifications
Nuske et al., 2018	PedsQL™ (parents)	Services & community In Australia *ADOS-2, SCQ*	ASC (43/28)	3 / 4 -	76 / 73	*Low 52%* *Moderate 14%* *High 34%*				
Pisula & Porebowicz-Dorsmann, 2017*	FAM-III (mothers and fathers)	Services & schools in Poland *Parent-report, professional diagnosis*	ASC (49/52)	10 / - *- / 5-17*	88 / 89	Asperger's 80 Autism with no ID 20 *Attending mainstream school 100%*		100 / 100	42 / - -	
Rao & Beidel, 2009	FES (parents)	Treatment clinic, medical centre & community in US *ADI-R, KBIT-2*	ASC (15/14)	- *8-14 / 8-14*	100/ / 100	*High functioning 100%*	Caucasian 93 / 93 Hispanic 7 / 7		43 / 42 -	
Rodrigue et al., 1992	FACES-III (fathers)	Universities, parent groups, schools in US *Professional diagnosis, VABS*	ASC & ID (20/20/20)	11 / 12 / 4^	60 / 55	*VABS mean 28 / 3.9 / -* ^	Caucasian 90 / 90	100 / 100	40 / 41 / 37 -	

Paper	Outcome Measure (reporter)	Recruitment *Screening*	ID/ASC (NDD N / TD N)	Child Characteristics NDD Group / TD Group			Parent Characteristics NDD Group / TD Group			
				Age Mean *Range*	% Male	Diagnoses (%) *Functioning*	% Race / Ethnicity	% Two-parent household	Age Mean *Range*	% Post-School Qualifications
Schneider et al., 2019	Bespoke score (parents)	National survey dataset in US *Parent-report*	ASC (468/600)	12 / 12 -	69 / 69		Combined group White 79 Biracial 7 African American 7 Asian 4 American Indian 1 Pacific Islander 0.4 Other 2			
Sivberg, 2002[+]	FARS (parents)	Sweden	ASC (37/37)	- *1-26 / 1-26*	53 / 61	Asperger's 24 High-level autism 32 Intermediate level autism 19 Low-level autism 24		86 / 86	43 / 39 -	
Sullivan et al., 2012	FACES-IV (mothers)	Support services & schools in US *Parent-report*	ASC (50/34)	10 / 10 -	83 / 39	Asperger's 62 Autism 38	Predominantly Caucasian			76 / -
Tsibidaki & Tsamparli, 2009*	FACES-III (mothers & fathers)	Disability association archives & municipality directory in Greece *Professional diagnosis*	ID (30/30)	8 / - -	50 / -			100 / 100	39 / 39 -	Fathers 17 / - Mothers 30 / -

Paper	Outcome Measure (reporter)	Recruitment *Screening*	ID/ASC (NDD N / TD N)	Child Characteristics NDD Group / TD Group Age Mean *Range*	% Male	Diagnoses (%) *Functioning*	% Race / Ethnicity	Parent Characteristics NDD Group / TD Group % Two-parent household	Age Mean *Range*	% Post-School Qualifications
van Riper et al., 1992	FAM-III (parents)	Disability & community organisations in US	ID (34/41)	- *3-10 / 3-10*		Down's syndrome 100 *Mild to moderate 100%*		100 / 100	Mothers 36 / - - Fathers 38 / -	
von Gontard et al., 2002	F-COPES (parents)	Support groups, services & schools in Germany *K-ABC, Raven test*	ID (49/32)	9 / 11 -	100 / 100	Fragile X syndrome 100 *Special education 94%* *Mean IQ 47* *CBCL borderline to clinical range 90%*		90 / 75		
Walton, 2019	FACES-IV (parents)	Academic registry & online in US *SCQ*	ASC (112/123)	11 / 9	79 / 49	*Not in full-time mainstream classes 72%*	American Indian 4 / 0 Asian 3 / 3 Black African American 6 / 5 Pacific Island 1 White 85 / 89 Other 2 / 3		40 / 39 -	47 / -

* Both mothers and fathers completed questionnaire, so parent N is double child N; + Parent N = 66; ^ ASC / ID / TD

ADI-R = Autism Diagnostic Interview-Revised; ADOS-2 = Autism Diagnostic Observation Schedule-2; ASC = autism spectrum conditions; BSID = Bayley Scales of Infant Development; CBCL = Child Behaviour Checklist; 3Di-sv = Developmental, Dimensional and Diagnostic Interview; F-COPES = Family Orientated Personal Evaluation Scale; FACES = Family Adaptability and Cohesion Evaluation Scale; FAD = Family Assessment Device; FAM = Family Assessment Measure; FARS= Family Relations Scale; FES = Family Environment Scale; GARS-2 = Gilliam Autism Rating Scale – Second Edition; ID = intellectual disability; K-ABC = Kaufman Assessment Battery for Children; KBIT-2 = Kaufman Brief Intelligence Test; ND = neurodevelopmental; Peds-QL = Pediatric Quality of Life Family Impact Module: The Family Functioning Scale; SCORE-28 = Systemic Clinical Outcome and Routine Evaluation; SCQ = Social Communication Questionnaire; SDQ = Strengths and Difficulties Questionnaire; SRS-2 = Social Responsiveness Scale; TD = typically developing; VABS = Vineland Adaptive Behaviour Scales

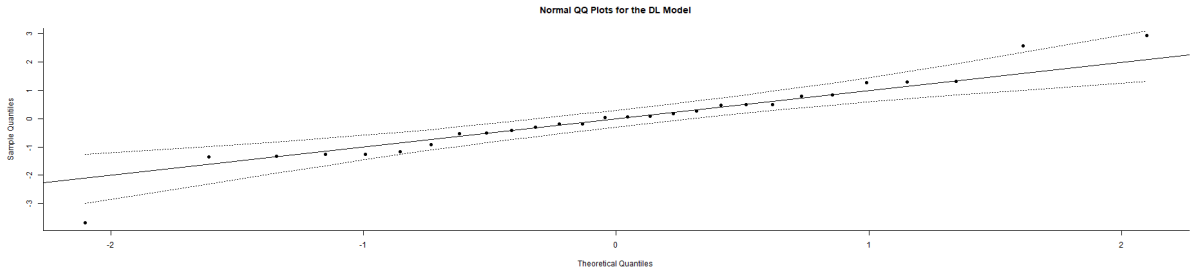

Figure 3: The Quantile-Quantile plot of the distribution of standardised mean differences within the primary studies

1.4.1.1 The Omnibus Test

There were 24 studies reporting 28 samples and a total of 14169 participants. A random effects model was calculated using the generic inverse variance method. The forest plot is shown in Figure 4. The random effects model suggested a weighted average SMD=0.23 (z=3.57, p<.01) and a 95% confidence interval of between 0.11 to 0.36. However, there was a significant difference between three groups (FwASC, FwID, FwASC+ID) compared to the FwTD groups (X^2=20.83, p<.01). Therefore, the neurodevelopmental condition groups were considered separately for further analysis.

Three papers reporting on four samples and 364 participants were analysed for differences in family functioning between combined FwASC and FwID groups (FwASC+ID) compared to FwTD (third section of Figure 4). Both the fixed and random effects model suggested a weighted average SMD=-0.17 (z=-1.66, p=.0966) and a 95% confidence interval of between -0.37 and 0.03. An effect of this size would be considered trivial and non-significant. Due to the small number of studies, no further analyses were conducted on the data for FwASC+ID. Data from two papers (Cuzzocrea et al., 2011; Manor-Binyamini, 2016) could not be disaggregated into ASC or ID subgroups and so were excluded from the further sub-group analyses; a third (Rodrigue et al., 1992) provided subgroup data and so was included as two separate samples in each subgroup analysis.

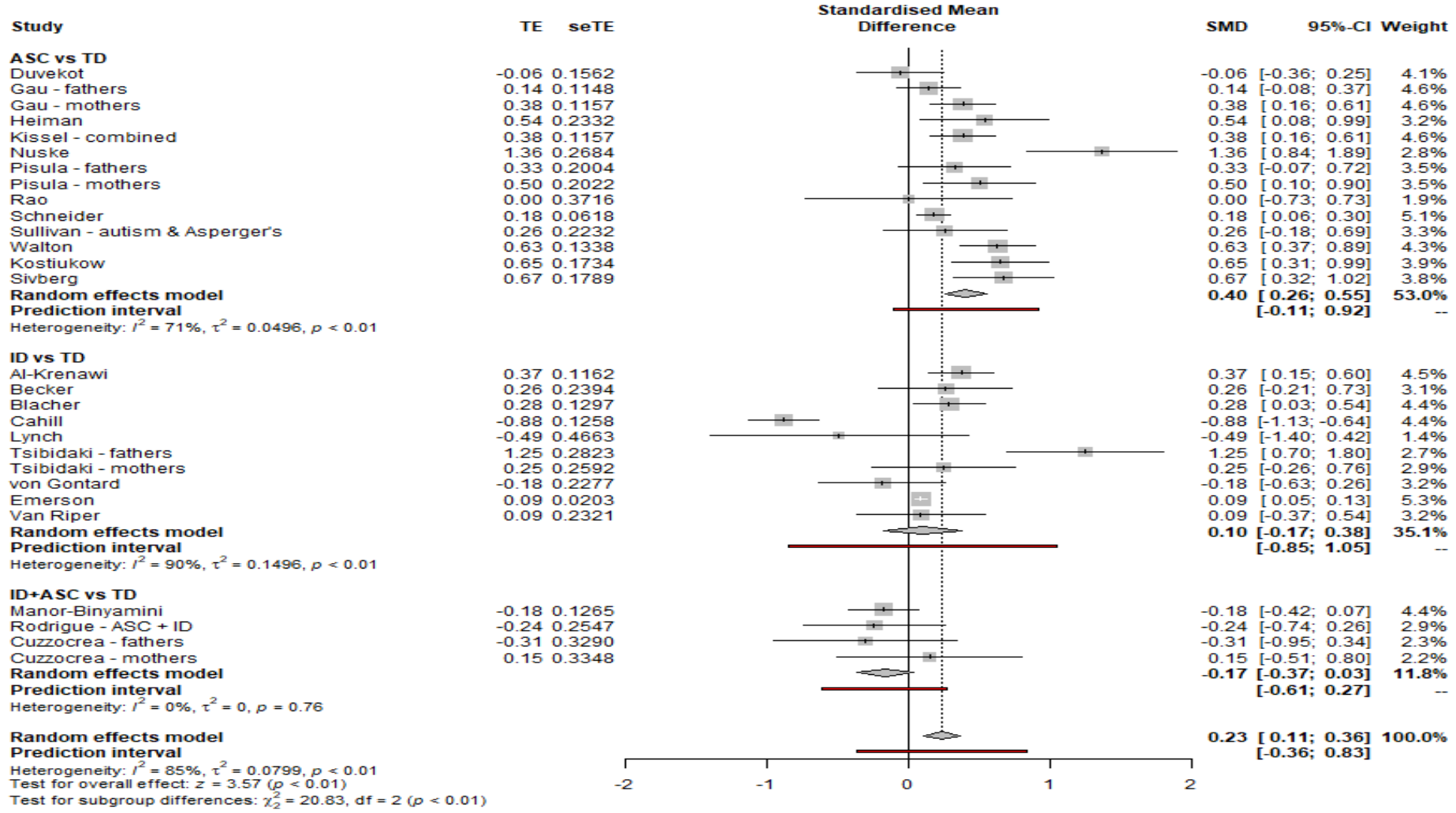

Figure 4: Forest plot of the standardised mean difference between family functioning of typically developing children and children with autism and/or intellectual disabilities.

Positive standardised mean difference represents better family functioning for the typically developing families

1.4.2 Families with Autism

1.4.2.1 The Omnibus Test

The differences in family functioning between FwASC and FwTD were calculated using a random effects model and the generic inverse variance method. There were 13 studies reporting 15 samples and with a total of 2837 participants. The random effects model suggested a weighted average standardized mean difference of SMD=0.38 (z=4.93, p<.0001) and a 95% confidence interval of between 0.23 to 0.53 (see Figure 5). An acceptable level of heterogeneity in the primary studies was observed (tau^2=0.0542, Higgins I^2=71%, Q=49.00, df=14, p<.0001), suggesting that the estimates of difference between FwASC and FwTD in the primary studies are not markedly biased by the presence of uncontrolled or confounding factors.

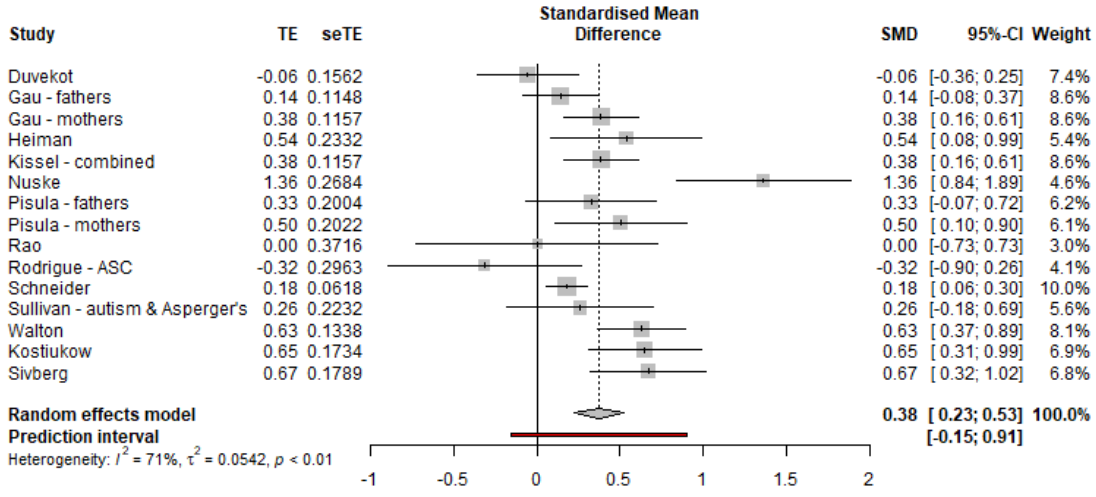

Figure 5: Forest plot of the standardised mean difference between functioning of families with an autistic child compared to families with a typically developing child

1.4.2.2 The Impact of Influential Primary Studies

A Baujat et al. (2002) plot (Figure 6) shows the influence of each study on the overall effect (vertical axis) and the discrepancy of the study with the rest of the literature (horizontal

axis). The study by Nuske et al. (2018) was identified to be both influential and discrepant from the other primary studies. This study was therefore reviewed for risk of bias. Nuske et al. (2018) used the Pediatric Quality of Life Family Impact Module: The Family Functioning Scale to measure family functioning, which was modified to suit the needs of the study. No other studies used this measure, and the study was judged to have a high risk of outcome bias due to modifications. Therefore, given the discrepancy and influence of this study and given the concerns regarding the measurement of family functioning, Nuske et al. (2018) was omitted from subsequent analysis.

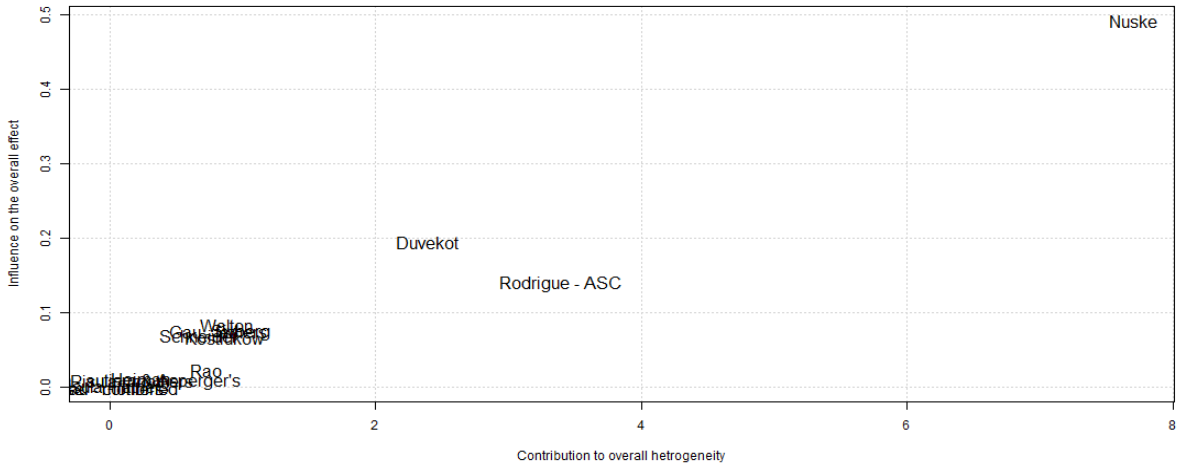

Figure 6: Baujat diagnostic plot of sources of heterogeneity in the autistic sub-sample

The random effects model was recalculated without Nuske et al. (2018). The corrected random effects model reported a synthesis of SMD=0.33 (95% CI 0.20 to 0.46) and I^2 reduced to 61%. The corrected random effects model evidences an approximately 13% decrease in SMD relative to the uncorrected estimate. This corrected estimate is shown in a forest plot in figure 7.

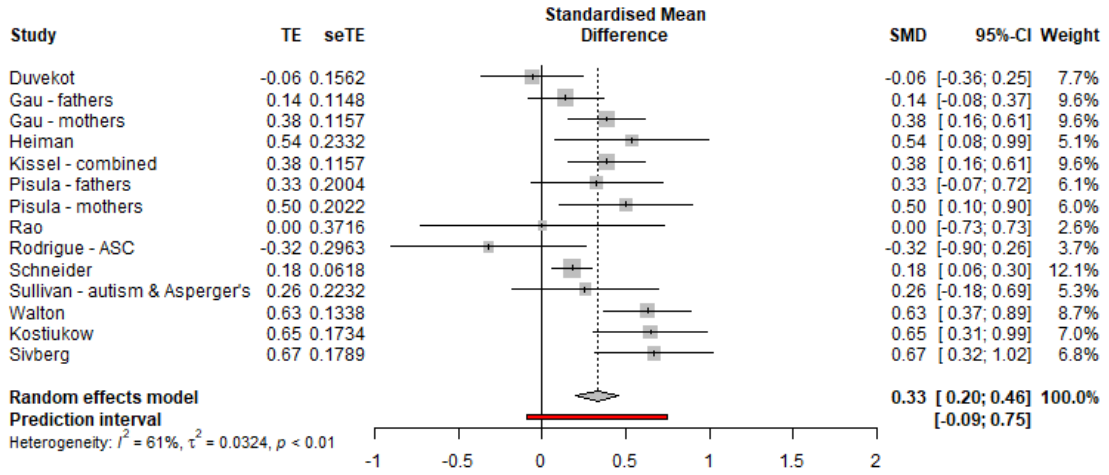

Figure 7: Forest plot of the standardised mean difference between functioning of families with an autistic child compared to families with a typically developing child, after removal of overly influential or discrepant studies

1.4.2.3 The effect of risk of bias in the primary studies

The quality effects model reported a synthesis of SMD=0.35 (95% CI 0.20 to 0.50), a 6% increase relative to the previous random effects estimate. The inclusion of the risk of bias assessment therefore results in relatively little change to the overall effect size estimate.

1.4.2.4 The impact of publication and small study biases

The shaded area of the funnel plot (Figure 8) is the area associated with small sample sizes and non-significant results; if there is an absence of studies in the area then it is likely there is some publication bias, and the effect size has been over-estimated.

The presence of a result within the shaded circle alongside the symmetrical inverted funnel suggests there is no visual evidence of publication bias in the distribution of SMDs. Egger et al.'s (1997) test of funnel plot asymmetry was also calculated and did not indicate asymmetry (t=1.28, df=13, p=.2214). Rosenthal's (1979) failsafe N calculated that 415 studies would be required to reduce the observed SMD=0.35 to non-significance, suggesting that the observed effect is robust to studies missing due to publication bias. Therefore, no adjustment for publication bias and small study effects was undertaken.

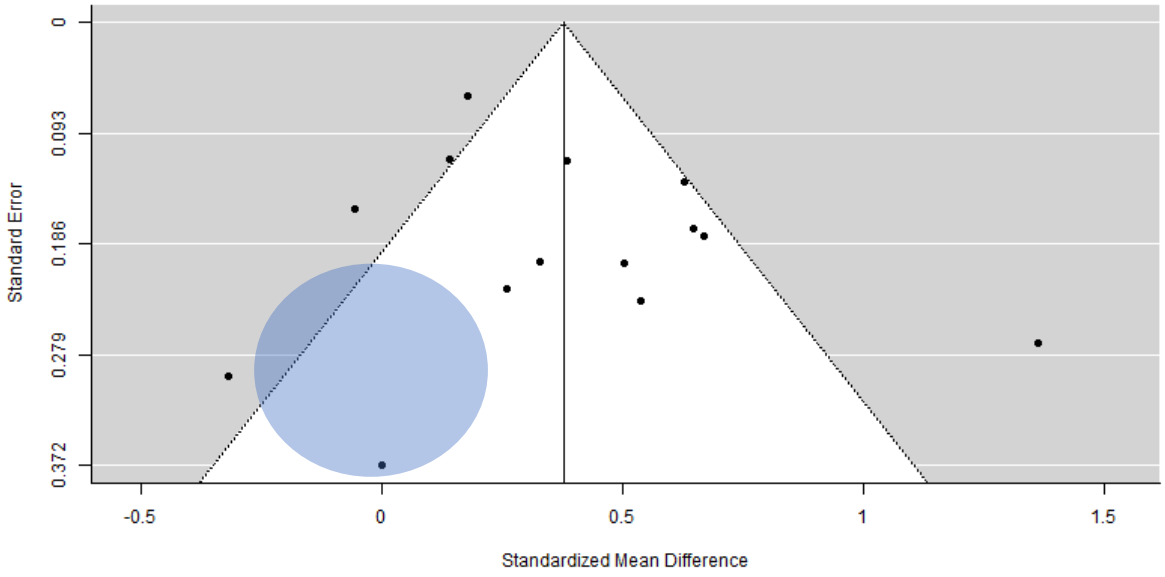

Figure 8: A funnel plot of the standardised mean differences for the autistic sub-sample

1.4.2.5 In Summary: Families with an Autistic Child

The results support the hypothesis that FwASC report poorer family functioning than FwTD. Twelve studies were included in the model, after the removal of one overly discrepant and influential study with a high risk of outcome bias. After correction for risk of bias, the model reported a synthesis of SMD=0.35, which is a small but significant effect size (Sawilowsky, 2009). An acceptable level of heterogeneity was found, and no adjustments for publication or small study biases were required.

1.4.3 Families with a Child with Intellectual Disability

1.4.3.1 The Omnibus Test

The difference in family functioning between FwID and FwTD was calculated using a random effects model and the generic inverse variance method. There were 10 studies reporting data from 11 samples, with a total of 11048 participants. The random effects model suggested a weighted average SMD=0.08 (z=0.63, p=.5316) and a 95% confidence interval

of between -0.18 and 0.34. An effect size of this magnitude would be considered trivial and non-significant. An unacceptable level of heterogeneity in the primary studies was observed (Higgin's I^2=89%, tau^2=-.1433, Q=89.38, df=10, p<.0001).

1.4.3.2 The Impact of Influential Primary Studies

The Baujat et al. (2002) plot (Figure 9) demonstrates each study's influence over the result and discrepancy from the literature. The study by Cahill et al. (2010) is shown to be both influential and discrepant from the rest of the literature, and was therefore reviewed for risk of bias. The groups in Cahill et al. (2010) had different members of the family report on family functioning (parents for ID, adolescents for TD) and some of the data were obtained from a separate review paper, suggesting there is a high risk of outcome and reporting bias for this particular research question. Given these concerns, Cahill et al. (2010) was omitted from subsequent analysis. While the father sub-sample from Tsibidaki & Tsamparli (2009) was also discrepant, the influence on the overall effect is smaller, so this sample was retained.

The random effects model was recalculated, and the results are presented in a forest plot in Figure 10. The corrected random effects model reported a synthesis of SMD=0.21 (p<.01, 95% CI 0.02 to 0.39) and I^2 reduced to 69%, a 163% increase in SMD relative to the uncorrected estimate. This corrected result can now be considered a small and significant effect size with an acceptable level of heterogeneity.

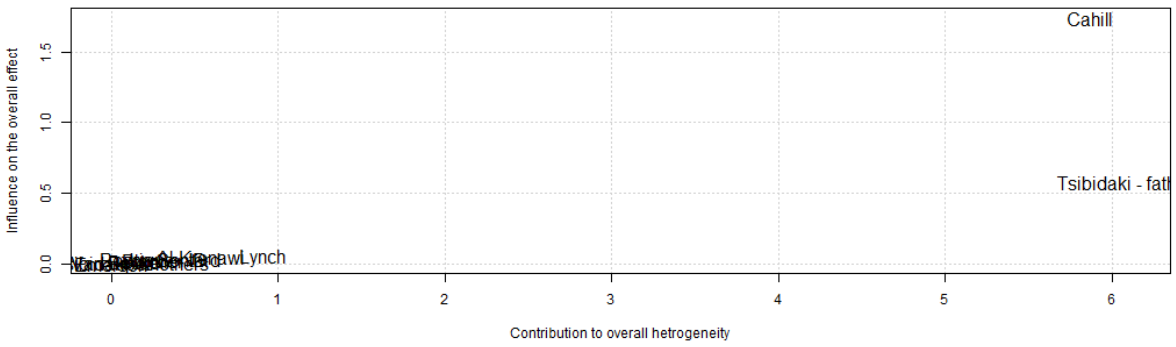

Figure 9: Baujat diagnostic plot of sources of heterogeneity for the intellectual disability sub-sample

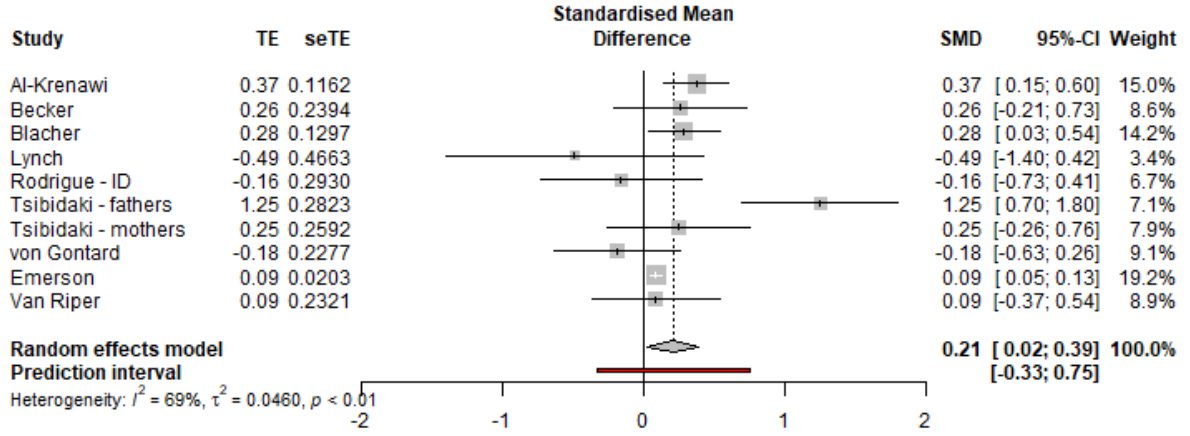

Figure 10: Forest plot of the standardised mean difference between functioning of families with a child with an intellectual disability compared to families with a typically developing child, with influential and discrepant studies omitted

1.4.3.3 The effect of risk of bias in the primary studies

The quality effects model was calculated using the total score from the risk of bias ratings reported in Table 4. The quality effect model reported a synthesis of SMD=0.22 (95% CI 0.03 to 0.41), a 5% increase relative to the uncorrected random effects estimate. The addition of the risk of bias assessment therefore results in relatively little change in the weighted average of these studies.

1.4.3.4 The impact of publication and small study biases

The funnel plot of the study SMDs is presented in Figure 11, showing a symmetrical inverted funnel effect suggesting minimal evidence of publication bias, although there is a notable absence of studies in the shaded circle which can indicate potential vulnerability to the ‘file drawer effect’ (Choi & Lam, 2016). Egger et al.'s (1997) test of funnel plot asymmetry was calculated and did not indicate asymmetry (t=1.00, df=8, p=.3461). Rosenthal's (1979) failsafe N was calculated as 65, suggesting that the observed effect is robust to studies missing due to publication bias. Therefore, no simulation of and adjustment for publication bias and small study effects was undertaken.

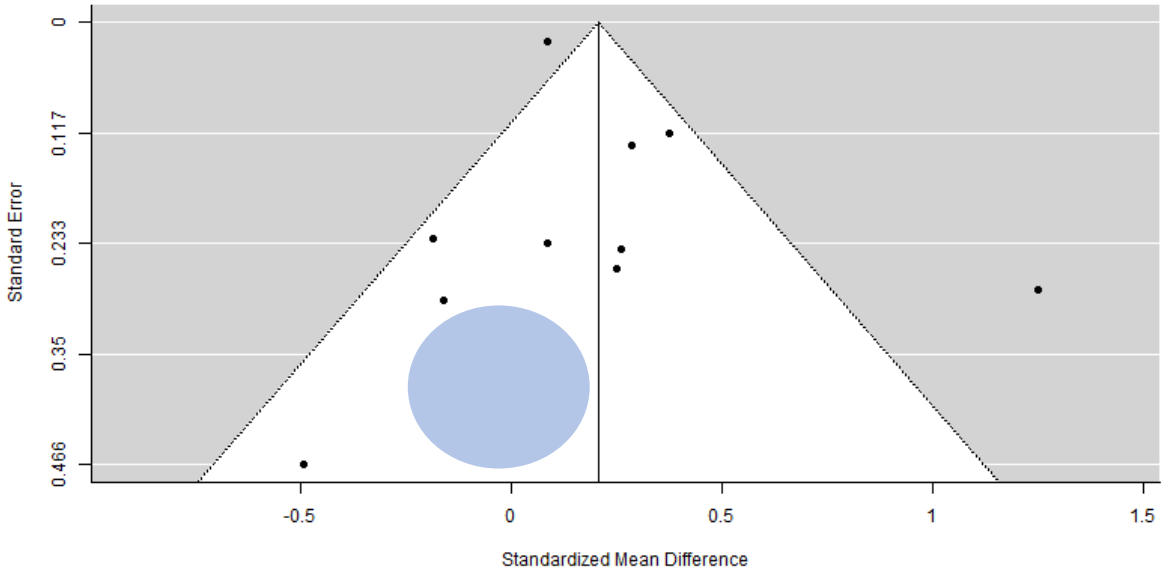

Figure 11: A funnel plot of the standardised mean differences for the intellectual disability sub-sample

1.4.3.5 In Summary: Families with Children with Intellectual Disability

The results support the hypothesis that FwID report poorer family functioning than FwTD. Nine studies were included in the model, after the removal of one overly discrepant and influential study with a high risk of outcome bias. After correction for risk of bias, the model reported a synthesis of SMD=0.22, which is a small but significant effect size (Sawilowsky, 2009). An acceptable level of heterogeneity was found, and no adjustments for publication or small study biases were required. The difference between FwASC and FwTD was greater than the difference between FwID and FwTD, although both effect sizes were of the same small magnitude and the confidence intervals overlap.

1.4.4 Subgroup analyses and Meta-regressions

To further explore the impact of study level covariates upon family functioning in the overall body of literature for ASC and ID, a series of subgroup analysis based on parent gender, neurodevelopmental child gender (samples above or below the median proportion of boys in the overall samples), outcome measure and administration, and study location were

conducted, and the results are shown in Table 7. The subgroup analyses were conducted on all studies with available data, except for Cahill et al. (2010) and Nuske et al. (2018) given their exclusion for being overly influential and discrepant in the overall analyses. The FwASC and FwID were considered jointly to maximise sample power, as well as separately given the significant difference between the two groups found in the prior results. A significant difference was found between fathers and mothers for FwASC compared to FwTD, with mothers reporting a greater difference in family functioning than fathers (X^2=4.56, p=.03). No other significant differences were found.

A series of meta-regressions were also run to explore putative differences in the continuous variables of neurodevelopmental child age, parent age, parent education, family marital status, and study publication year. The results are shown in Table 8. No significant effects were found.

In summary, when comparing FwASC to FwTD, mothers report poorer family functioning compared to fathers. No other methodological or sample characteristics moderated the effect.

Table 7: The subgroup analyses for parent gender, neurodevelopmental child gender, outcome measure and administration, and study location

Moderator	Neuro-developmental group	Level	SMD	95% CI	k	X^2	p
Parent gender	ASC+ID	Fathers	0.23	-0.19 to 0.66	5	0.03	.86
		Mothers*	0.28	0.13 to 0.42	13		
	ASC	Fathers	0.19	-0.01 to 0.38	2	4.56	**.03**
		Mothers*	0.44	0.32 to 0.55	7		
	ID	Fathers	Insufficient number of samples				
		Mothers					
Neurodevelopmental child gender+	ASC+ID	Above median	0.32	0.17 to 0.46	9	0.49	.48
		Below median	0.24	0.07 to 0.40	9		
	ASC	Above median	0.31	0.20 to 0.43	6	0.47	.17
		Below median	0.42	0.13 to 0.71	4		
	ID	Above median	0.14	-0.10 to 0.37	3	1.67	.49
		Below median	0.57	-0.05 to 1.20	3		
Outcome measure	ASC+ID	FACES	0.28	-0.05 to 0.51	11	2.73	.60
		FAD	0.14	-0.06 to 0.34	3		
		FAM-III	0.36	0.19 to 0.52	4		
		FES	0.24	-0.07 to 0.54	4		
		Other	0.24	-0.05 to 0.54	4		
	ASC	FACES	0.41	0.20 to 0.61	5	7.33	.12
		FAD	-0.06	-0.36 to 0.25	1		
		FAM-III	0.40	0.22 to 0.57	3		
		FES	0.35	-0.16 to 0.85	2		
		Other	0.35	-0.08 to 0.87	2		
	ID	FACES	0.74	-0.24 to 1.72	2	2.03	.73
		FAD	0.21	-0.07 to 0.48	2		
		FAM-III	0.09	-0.37 to 0.54	1		
		FES	0.03	-0.69 to 0.74	2		
		Other	0.03	-0.40 to 0.47	2		
Location	ASC+ID	Europe	0.30	0.09 to 0.52	11	0.25	.88
		US	0.24	0.09 to 0.39	10		
		Asia, Middle East, Australasia[7]	0.24	0.00 to 0.46	5		
	ASC	Europe	0.41	0.12 to 0.70	5	0.32	.85
		US	0.33	0.13 to 0.53	5		
		Asia, Middle East, Australasia	0.31	0.10 to 0.52	3		
	ID	Europe	0.31	-0.15 to 0.76	4	0.16	.69
		US	0.21	0.01 to 0.40	4		

*Including mixed samples with over 80% mothers

+The median percentage of boys in the samples was calculated and studies classified as having a male gender percentage above or below this median

[7] The number of papers was not sufficient to disaggregate 'Asia, Middle East & Australasia' into subgroups, but it is acknowledged that it is problematic to include this large and diverse geographical area as one subgroup in the analysis.

Table 8: The meta-regressions of continuous variables

Moderator	Neuro-developmental group	Coefficient	SE	Z	p
Mean age of child with ASC and/or ID	ASC+ID	-0.00	0.03	-0.01	.99
	ASC	0.01	0.04	0.28	.78
	ID	-0.03	0.06	-0.54	.59
Mean age of the parent of the child with ASC and/or ID	ASC+ID	0.00	0.02	-0.08	.93
	ASC	0.01	0.03	0.52	.60
	ID	0.03	0.16	0.49	.62
Proportion of parent sample with post-school qualifications	ASC+ID	-0.00	0.00	-0.49	.62
	ASC	-0.01	0.01	-0.93	.35
	ID	0.01	0.02	0.39	.69
Proportion of two-parent households	ASC+ID	0.00	0.00	0.41	.68
	ASC	-0.02	0.01	-1.49	.13
	ID	0.01	0.01	0.92	.36
Publication year	ASC+ID	0.01	0.01	1.44	.15
	ASC	-0.01	0.01	-0.57	.57
	ID	0.03	0.02	1.70	.09

1.5 Discussion

This review was the first meta-analysis to statistically synthesise collective family functioning of families with an autistic child and/or a child with ID compared to families with a TD child. The review also evaluated whether methodological or participant characteristics moderated observed differences in family functioning. The findings demonstrate that families with an autistic child and/or a child with ID report poorer family functioning than families with a TD child. This difference was larger for families with an autistic child than for families with a child with ID, although the magnitude of the effect size was small for both groups and the overlapping confidence intervals do not provide statistical evidence of different effect sizes. These results are consistent with previous meta-analyses finding poorer outcomes for parents and siblings in neurodevelopmental families (Dunn et al., 2019; Hayes & Watson, 2013; Schnabel et al., 2019; Shivers et al., 2019; Singer, 2006; Yirmiya & Shaked, 2005). The present meta-analysis extends these findings from individual to collective family-level outcomes.

For families with an autistic child, mothers also reported a greater difference in family functioning compared to typically developing families than fathers. A previous review found

that mothers of children with ID report poorer mental health and well-being compared to fathers (Dunn et al., 2019). While the results were not replicated in the ID group, this may be due to the small number of studies available. No other significant moderators were identified, which again may be a function of the small number of studies available with subcategories with numerous levels, raising the risk of type II errors.

1.5.1 Implications for Theory, Research and Clinical Applications

There is ongoing debate about the extent to which ASC and ID form part of a complex genetic neurodevelopmental continuum (Morris-Rosendahl & Crocq, 2020) or exist as discrete diagnoses, with some phenotypical overlap and co-occurrence (American Psychiatric Association, 2013). Both families with an autistic child or a child with an ID reported poorer family functioning than families with TD children. Therefore, regardless of aetiology, the presence of either of these neurodevelopmental conditions confers risk for reporting poorer family functioning outcomes. There was a significant difference in the chi-square analysis reported on the core effect for all groups, which suggests the level of risk may be mediated by the specific condition the child is diagnosed with and that there may be condition-specific risk factors. Even within ID, syndrome-specific outcomes have been identified; for example, Down syndrome is associated with improved family outcomes whereas the severity of autism symptomology predicts poorer family outcomes, likely mediated by the extent of behaviours that challenge (Blacher et al., 2010; Ilias et al., 2018). Further research is therefore needed to explore potential syndrome-specific explanatory factors in the identified family functioning outcomes, and a meta-analysis directly comparing families with an autistic child to families with a child with ID would be a useful extension of this current review. The group differences identified in this meta-analysis suggest that separate diagnoses may be a more useful theoretical construct and research grouping within the field of family functioning. This highlights the importance of better characterisation of

combined neurodevelopmental groups and better reporting of outcomes by diagnostic sub-group. Researchers should also consider including a measure of behaviours that challenge given its possible role in mediating the link between autism characteristics and family outcomes.

These results provide further support for the existing guidance that services for individuals with ASC or ID include support for families and carers (NICE, 2013, 2018). As all informants in the present meta-analysis were parents, it is also unclear whether these ratings of poorer family functioning replicate when children themselves report on their collective family functioning. Importantly, child and parent ratings in the typically developing population correlate poorly (Olson, 2011). As such, future research should include the perspective of the child alongside their parents and services should also be reminded of the need to assess families based on information provided by different family members, with a particular focus on facilitating the perspective of the child.

Family functioning instruments have largely been developed with neurotypical populations and are often decades old (Turnbull et al., 2007). They also offer implied criteria for family units being described as well or unwell, without consideration of subjectivity, culture (Wollny et al., 2010) or neurodiversity. For example, families with an autistic child may communicate and display cohesion in a different way to families with a typically developing child (Greenlee et al., 2018), be happier with greater levels of enmeshment or disengagement (Walton & Tiede, 2020), and neurodevelopmental family units may be more likely to include adult-aged children. It can therefore be questioned whether these measures are appropriate for modern families or for neurodevelopmental families, both in terms of the construct itself and its measurement. This could suggest that neurodiverse/neurodivergent families may have a *different* rather than *worse* experience of family functioning. These results could also be a function of the measurement properties of the measures used. This

needs to be explored in future research, both to inform the development and validation of family functioning measures for families with an autistic child and potentially to adapt theories and models to better describe healthy family functioning for these families. It is essential that the voices of neurodivergent children and families are involved in co-production at all stages of future research in this field to avoid further potential stigmatisation through conducting neurotypical research with neurodiverse groups.

Further threats to validity exist for family functioning measurement. Most measures do not offer clinical cut offs and family dysfunction is difficult to identify except in its extreme form (Glidden, 2012). Therefore, whilst this review identified statistical differences in family functioning between families with an autistic child and/or a child with ID compared to families with a TD child, these data should not be interpreted as evidence of family dysfunction in the neurodevelopmental condition families. As such, it is difficult to translate the observed effect sizes from a wide range of outcome measures into clinically meaningful interpretations. This highlights a need for measures that can be used to meaningfully assess families within services and for research purposes. The most recent identified review of family measures (Alderfer et al., 2008) is now 14 years out of date, and the field would benefit from an update to identify a potential gold-standard measure of the collective concept of family functioning. The FACES-IV questionnaire (Olson, 2011) is one such measure updated since the review that now includes clinical interpretations as well as an updated range of scales specifically targeted at identifying extremes of family dysfunction. Future research in neurodevelopmental families should consider using measures such as the FACES-IV with particular attention to the clinical categorisations.

One key limitation of the literature available for meta-analysis was that it largely relied on small convenience samples, raising concerns about the representativeness of the samples to the general population. It could conceivably be both that families with poorer

functioning are known to support services and more likely to be invited to participate in research, but also that families with poorer functioning have greater barriers to research participation. The impact of this common recruitment strategy on the overall effect size is therefore difficult to judge. Given the overall mixed level of bias in the primary studies, the results of this meta-analysis should therefore be interpreted cautiously. However, the studies included are representative of the current literature, and highlight a need for future research to ensure higher standards, particularly around participant selection (i.e., moving beyond convenience-based samples) and choice of outcome measures.

Turnbull et al. (2007) criticised the field for lacking diversity and representative samples, and this limits the generalisation of results. This review, for example, highlights a focus in the literature on school-age boys from white/Caucasian two-parent families. The field would therefore benefit from greater inclusion of more diverse families. There is an over-representation of mothers as reporters, consistent with the findings of previous reviews (e.g. Hayes & Watson, 2013). Given that mothers of children with neurodevelopmental disorders also experience poorer mental health (Dunn et al., 2019), this raises a risk of inflation of the results found here as the mother is the shared rater of her own mental health and the collective family functioning (Yorke et al., 2018). The sub-group differences between mothers and fathers combined with the discrepancy of Cahill et al. (2010) based on reporter differences demonstrates the potential for differences in individual perspectives. This review cannot account for intra-familial differences in reporting on the basis of the existing literature (Wollny et al., 2010). There is therefore a need for research to move beyond single-report family functioning questionnaires towards synthesising multi-perspective measures (Greenlee et al., 2018). Observational and clinician-rated interviews are already available to provide an outsider perspective (Alderfer et al., 2008), but there is a notable gap in understanding of family functioning from the perspective of the child. Child-appropriate and neurodiverse-

appropriate family functioning measures should be adapted or developed and then validated to progress research in this field. This recommendation is consistent with recent calls to engage in an ontological shift towards understanding the subjective experience of autistic people themselves within their relational social reality (Milton, 2012). The Kvebaek Family Sculpture Technique (Cromwell et al., 1980), while excluded from this review as it primarily measures sub-system relationships within the family (e.g. parent-child) rather than the functioning of the family as a whole system, offers a potentially more accessible route to assessing a neurodivergent child's perspective through symbolic objects.

1.5.2 Threats to the Meta-analysis Validity

Tint & Weiss (2016) concluded that syntheses in the field of family wellbeing are hampered by varied measurement techniques and a lack of common theoretical direction. Family functioning often forms an implicit part of many measures and overlaps with other constructs (Wollny et al., 2010), and the literature search offered a wide range of possible measures. In response to these criticisms, the inclusion criteria were theoretically grounded in ecological systems theory (Bronfenbrenner, 1977) and limited to measures exclusively and explicitly measuring the psychological construct of family functioning. The wide range of family functioning measures included, with some studies using multiple measures or sub-scales, required decisions to be made about equivalency based on clinical and numerical rationales that have been documented throughout. High inter-rater reliability statistics were found on 20% screening and data extraction procedures, which supports the reliability of these criteria in practice.

Neurodevelopmental populations were also poorly defined and described in the literature, with difficulty comparing across studies due to changing conceptualisations of ASC and ID as well as variability in presentations (Karst & Van Hecke, 2012). Despite an extensive and systematic literature search, it is feasible that some papers were omitted,

although the absence of publication bias in the results offers some reassurance. While studies were separated into ASC and ID subgroups based on the sample descriptions, less than half of the studies screened participants based on professional or researcher diagnosis. This raises concerns about the validity of analysing them as separate groups, although the differences found are consistent with prior research findings in both magnitude and direction. The quality effects model was also similar in magnitude to the random effects model, suggesting risk of bias does not have a large impact on the overall meta-analytic effect size.

The sub-group analyses were limited by a small number of studies, raising the likelihood of type II errors. There was an under-representation of fathers, girls, and countries outside of Europe and the US, and important moderating variables such as socioeconomic status or behaviour that challenges (Tint & Weiss, 2016) were inconsistently reported. A small number of studies using a wide range of outcome measures also limits comment on differences in measurement between these questionnaires. Although the absence of publication bias means the inclusion of grey literature would be unlikely to change the overall findings, its inclusion may have increased the power of the subgroup analyses.

1.5.3 Conclusions

In conclusion, the results of this meta-analysis show that families with an autistic child and/or a child with ID report poorer family functioning than families with a TD child. For families with an autistic child, mothers also reported a greater difference in family functioning compared to typically developing families than fathers, but no other significant moderators were identified. These results provide guidance for researchers regarding recruitment and reporting and identify a future direction for the field focusing on outcome measures and neurodevelopmental condition-specific factors. The child's perspective is notably lacking in these results, and this should therefore be a key focus for researchers and services alike.

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CHAPTER 2 - FAMILY FUNCTIONING AND MENTAL HEALTH IN AUTISTIC YOUNG PEOPLE WITHOUT INTELLECTUAL DISABILITY AND THEIR FAMILIES

2.1 Abstract

Rationale

Families with autistic children report poorer family functioning, but it is unknown to what extent autism itself or the identity or mental health of the informant may explain this finding. There is a lack of research from the perspective of autistic children. This study is therefore the first to explore collective family functioning from the perspective of both autistic young people and their parents, and to explore the associations between autism characteristics, mental health, and family functioning.

Method

Participants were N=115 autistic young people (mean age 19.81, SD 2.85) and N=50 parents (mean age 47.52, SD 5.35). Within these two samples, 25 parent-child dyads were identified. Participants in this cross-sectional online survey were recruited through convenience sampling. The study was a cross-sectional online survey design. The primary outcome measure was a self-report measure of collective family functioning (the Family Adaptability and Cohesion Evaluation Scale) completed by all participants. Young people reported on their own autism characteristics, depression, anxiety, and self-harm. Parents reported on their own depression, anxiety, and stress.

Results

Both parents and young people reported overall balanced levels of family functioning but low levels of family satisfaction. In the total samples, young people tended to report poorer family functioning than parents, but no significant correlations in family functioning scores were found between the parent and young person dyads in the subsample. Parent-reported family

functioning was predicted by parent anxiety and stress. Young person-reported family functioning was predicted by their depression scores. Self-harm and autism characteristics were not associated with family functioning. No significant associations were found across dyads (i.e., child mental health with parent-reported family functioning, and vice versa). Poorer collective family functioning was better predicted by the informant's poorer mental health rather than autism characteristics, highlighting a need for mental health support for families. The discrepancy between parent and child reported collective family functioning highlights the need for multiple perspectives on family experiences.

2.2 Introduction

The current diagnosis of autistic spectrum disorder in the DSM-V (American Psychiatric Association, 2013) encompasses previously separate sub-diagnoses, including Asperger's syndrome and Pervasive Developmental Disorder. Autism spectrum conditions (ASC, see Section 1.2 for further detail) often co-occur with intellectual disability (ID; reduced intellectual functioning and difficulties in adaptive functioning), but nevertheless it is estimated that up to 80% of autistic people do not have a co-occurring ID (Fombonne, 2003; Ghirardi et al., 2018; Rydzewska et al., 2019). There is therefore a need for research focusing specifically on the needs and experiences of autistic people without co-occurring ID.

Autism affects both the individual themselves and their interpersonal and familial relationships. The importance of family functioning is discussed in Section 1.2.2, highlighting the importance of adopting a whole family perspective for families with autistic children to optimise outcomes for both families and individuals. The results of the previous meta-analysis (Section 1.4) revealed poorer family functioning in families with an autistic child compared to typically developing children, but current research and policy with these families is sparse (Cridland et al., 2014; Tint & Weiss, 2016). Therefore, there is a need for

further family-level research in families with an autistic child to increase understanding and improve support.

While no gold-standard family functioning model or measure has been identified (Alderfer et al., 2008), the Circumplex Model of Marital and Family Systems (Olson et al., 1979) is one of the most widely used, with cohesion and flexibility as its two central dimensions. The model defines cohesion as "the emotional bonding that family members have towards each other" (Olson, 2011, p.65), and ranges from disengaged to enmeshed. Flexibility is defined as "the quality and expression of leadership and organisation, role relationship and relationship rules and negotiations" (Olson, 2011, p.65), and ranges from chaotic to rigid. Communication within the family system is viewed as a facilitating dimension allowing families to alter their levels of cohesion and flexibility as required. Overall, the model hypothesises that balanced levels of cohesion and flexibility represent healthy family functioning. These constructs are often targeted in systemic family therapies, although no systematic research on these therapies for families of autistic children has been conducted (Walton & Tiede, 2020).

The previous meta-analysis found that families with an autistic child and/or a child with ID both report poorer family functioning than families with a typically developing child (Section 1.4). Previous research suggests autism-specific factors are linked to family functioning above and beyond other neurodevelopmental conditions. In children with ID and attention-deficit/hyperactivity disorder, higher levels of co-occurring autism characteristics independently predict poorer family outcomes (Green et al., 2016). This association may be explained in part by the co-occurrence of behaviours that challenge, emotional outbursts, mental health difficulties, and the lower socioeconomic status of families with children with multiple neurodevelopmental conditions (Blacher et al., 2010; Green et al., 2016; Ilias et al., 2018; Nuske et al., 2018). Research with families of autistic children without co-occurring ID

is therefore one strategy for understanding the role that autism-specific mechanisms may have in poorer family outcomes, as well as to identify the specific clinical needs of these families.

There is an overwhelming tendency for family functioning to be reported by mothers (Greenlee et al., 2018; Hayes & Watson, 2013). Mothers of autistic children report poorer family functioning than fathers when compared to typically developing families (Section 1.4.4), and mothers of children with ID report poorer mental health than fathers (Dunn et al., 2019). These findings raise the risk of inflation of mental health and poor family functioning outcomes through shared rater effects (Yorke et al., 2018), where poor maternal mental health may be an explanatory factor in mothers reporting poorer collective family functioning. There is therefore a need for research on family functioning to include multiple informants to ensure a whole-family perspective. In typically developing families, parent and child reports of family functioning can have a low level of agreement (Palmer et al., 2016). The perception of family functioning from the perspective of autistic children and young people themselves is lacking and Walton & Tiede (2020) urged future research to include this perspective to further understand the relationship between child and parent reported family functioning in families with autistic children.

The possibility that mental health may be an explanatory factor in family functioning reports also highlights a need for combining these constructs within research. Rates of depression, suicide and self-harm are greater in autistic adults and those with high autistic traits than in the general population; this risk may be particularly heightened in adolescents and in autistic people without co-occurring ID (Baghdadli et al., 2003; Bennett, 2016; Cassidy & Rodgers, 2017; Dell'Osso et al., 2016, 2019; Gulsrud et al., 2018; Maddox et al., 2017; Richards et al., 2012; Shattuck et al., 2007; Veenstra-VanderWeele, 2018). It is estimated that 70-80% of autistic young people experience poor mental health, particularly

depression and anxiety (Lever & Geurts, 2016; Simonoff et al., 2008; Strang et al., 2012); this compares to an estimated one in seven (approximately 14%) of adolescents globally (Unicef, 2021). Autism-specific risk factors are present in mental health outcomes (Cassidy & Rodgers, 2017), and autism itself may affect the way that mental health is experienced (Crane et al., 2019). Given the social communication differences in autism and the resultant double empathy problem that affects mutual understanding and communication between neurotypical and neurodivergent people (Milton, 2012), autistic people may be particularly vulnerable to social risk factors for poor mental health, such as isolation, exclusion, loneliness and a lack of social support (Cassidy et al., 2014; Hedley et al., 2017; Hedley, Uljarević, Foley, et al., 2018; Hedley, Uljarević, Wilmot, et al., 2018). Given these autism-specific differences in mental health and high rates of mental health difficulties in autistic young people, further research in this area is a research priority to facilitate better understanding of the aetiology, risk and protective factors, and potential preventative, screening, and intervention targets. It is, however, unknown to what extent poor mental health may have a causal role in family functioning in this population.

In the general population, research across many cultures has implicated poor family functioning in the onset, maintenance and persistence of poor mental health outcomes for young people (Arbuthnott & Lewis, 2015; Berryhill & Smith, 2020; Fortune et al., 2016; Guberman & Manassis, 2011; Hughes et al., 2008; Hurd et al., 1999; Law & Shek, 2013; Lewis et al., 2015; McGinn et al., 2010; Tatnell et al., 2014; Wei, 2016; Zhao & Yiyue, 2018). Family dysfunction may be one of the environmental mechanisms for inter-generational transmission of poor mental health (Daches et al., 2018). Healthy family functioning acts as a protective factor that may offset the effects of other stressors, leading Webb (2002) to suggest that the absence of familial protective factors may be most pertinent for childhood outcomes. The success of treatment for depression and anxiety in adolescents is

contingent upon healthy family functioning, while treatment itself may improve family functioning (Dardas, 2019; Feeny et al., 2009; Jongerden & Bogels, 2015; Keeton et al., 2013). It is likely that the relationship between mental health and family functioning is bidirectional, complex, and inter-related with individual factors (Baetens et al., 2014; Claes et al., 2015; Crowell et al., 2008; Jutengren et al., 2011; Lo et al., 2016; Palmer et al., 2016; Sim et al., 2009). However, these purported bidirectional associations are yet to explored in autistic young people and their families.

The mental health of parents is also highly relevant for family functioning in families with an autistic child. Several meta-analyses have found that parents of autistic children experience high levels of psychological distress, depression and anxiety compared to parents of typically developing children and children with ID (Dunn et al., 2019; Hayes & Watson, 2013; Schnabel et al., 2019; Singer, 2006; Yirmiya & Shaked, 2005; Section 1.2.3). In families of autistic pre-schoolers, parental depression mediates the relationship between behaviours that challenge and family functioning (Jellett et al., 2015), and extreme patterns of family functioning are related to poorer outcomes in depression, happiness and satisfaction with family life in parents of autistic children (Walton & Tiede, 2020). However, despite these associations and data from the previous meta-analysis showing that parents of autistic children report poorer family functioning (Section 1.4.2), there is a lack of research exploring the putative link between parent mental health variables and parental reports of family functioning in families with an autistic child.

In summary, current research on family outcomes in families of autistic children is sparse. It is known that poorer family functioning is associated with poorer individual outcomes, and that family functioning is poorer for families with an autistic child when compared to typically developing families or other neurodiverse/neurodivergent families. This suggests there are autism-specific risk factors and highlights a need to explore family

functioning in the families of autistic children without co-occurring neurodevelopmental diagnoses such as ID, who form the majority of the autistic population. There is also a lack of research including the autistic child's perspective on their collective family functioning. It has been suggested that mental health affects how someone reports their family functioning, and mental health is poorer for both autistic young people and their parents. It is therefore crucial to explore the extent to which autism characteristics and mental health can explain family functioning reports from multiple family members.

As such, this study will be the first to consider family functioning from the perspective of both the autistic young person (YP) and their parents. It will also explore the associations between family functioning, YP autism characteristics and YP and parent mental health, with dyads of parents and YP used to explore associations within families. The study aimed to answer the following research questions:

1. How do autistic YP and their parents describe their family functioning? It is hypothesised that there will be a significant difference and a positive association between parent-reported and YP-reported family functioning.
2. Is there an association between YP autism characteristics or mental health (depression, anxiety, self-harm) and YP or parent-reported family functioning? It is hypothesised that there will be an association between YP autism characteristics and YP and parent-reported family functioning. It is also hypothesised that there will an association between YP mental health and YP and parent-reported family functioning.
3. Is there an association between parental mental health (depression, anxiety, and parenting stress) and YP or parent-reported family functioning? It is hypothesised that there will be a negative association between parent mental health and parent-reported family functioning. It is also hypothesised that there will be an association between parent mental health and YP-reported family functioning in parent/YP dyads.
4. Do YP autism characteristics, YP mental health (depression, anxiety, self-harm) or parent mental health (depression, anxiety, stress) influence the associations between parent-reported and YP-reported family functioning? It is hypothesised that there will be significant positive associations between parent-reported and YP-reported family functioning when these variables are controlled for.

5. Do YP autism characteristics, YP mental health and parent mental health predict a model of YP- or parent-reported family functioning? It is hypothesised that YP autism characteristics and mental health will predict YP-reported family functioning. It is also hypothesised that parent mental health will predict parent-reported family functioning.

2.3 Method

2.3.1 Participants

Parents and YP were recruited via university research databases, schools, university disability services, autism charities, and social media. Participants could take part on their own or alongside their dyad YP/parent. All participants were offered a £5 voucher or payment as reimbursement for their time. It was estimated using a G*Power statistical power analysis (Faul et al., 2009) that 138 participants would be required to detect a medium effect size for a multiple linear regression[8].

YP participants self-reported[9] a diagnosis of autism, and parents provided an informant reported diagnosis for their child. Participants were required to be aged 14-25 (YP) or have an autistic child in this age range (parents), reside in the UK, be fluent in English, and able to complete an online survey. A co-occurring diagnosis of ID was an exclusion criterion cited in the promotional materials and information sheets (Appendix 4). To validate this criterion, YP participants were asked about mainstream school attendance and education level, and the ability to use a computer and understand the online questionnaire was also used as a proxy, consistent with other research in this population (e.g. Moseley et al., 2019).

[8] The regression was selected for the power analysis as this would involve the greatest number of potential independent variables, and therefore greater participant numbers to be sufficiently powered to detect a significant effect size.

[9] It was beyond the scope of the project to confirm diagnoses through direct assessment or access to medical histories, as this added burden for participants and researchers may have reduced overall participant numbers. This methodological choice is also consistent with other survey-based studies in this area (e.g. Maddox et al., 2017).

Participants awaiting an autism diagnosis (N=2) were excluded, alongside any participants who did not complete the family functioning questionnaires.

The YP sample included 115 autistic young adults, of whom 25 participated as a dyad with an identified parent/caregiver. Demographic data is reported in Table 9. The YP sample was predominantly White/Caucasian, single, living with parents, attended mainstream school, educated to A-Level, currently a student, and living with a health condition; 51% of the sample reported a mental health condition. The dyad subsample of YP were significantly younger than the rest of the sample, less likely to have reached higher levels of education, and more likely to have been diagnosed at a younger age.

The parent sample included 50 parents/caregivers[10], of whom 25 participated as a dyad with an identified autistic YP. Demographic data is reported in Table 10. The parent sample was predominantly married parents living with the young person at least some of the time, female, White/Caucasian, educated to college level or above, employed with a household income above the UK median, and with no physical health, mental health, or developmental conditions; 34% of the sample reported a mental health condition. No significant differences were found between the dyad parent subsample and the full parent sample.

The scores for the dyad sample were compared to the rest of the sample using t-tests or Mann-Whitney tests to explore whether there were differences between these self-selecting groups (Table 12, Section 2.4.1). The dyad samples reported consistently better family functioning and lower mental health scores than the rest of the sample. Statistically significant differences were found between YP and parent-reported cohesion ratios, as well as YP-reported flexibility ratio, FCS, FSS and BDI scores.

[10] The parent/caregiver sample will hereafter be called the parent sample, but it should be noted that any legal caregiver was invited to participate, and one participant described themselves as a sibling guardian.

Table 9: The young person demographic data for the whole group and the dyad subsample, including statistical differences between the whole group and dyad subsample

	Whole sample (N=115)		**Dyad sample (N=25)**		**Dyad / Non-Dyad Group differences**
	N (%)	**Mean (SD)**	**N (%)**	**Mean (SD)**	**χ^2 (p-value)**
Age (years)	-	19.81 (2.85)	-	17.26 (2.56)	U=1577.5 (<.001)
Gender					
Female	52 (45)	-	8 (32)	-	
Male	51 (44)	-	14 (56)	-	2.29 (.32)
Prefer not to say/prefer to self-describe	12 (10)	-	3 (12)	-	
Transgender					
No	97 (84)	-	22 (88)	-	
Yes	16 (14)	-	3 (12)	-	0.69 (.71)
Prefer not to say	2 (2)	-	0 (0)	-	
Autism diagnosis age (years)	-	13.71 (5.67)	-	11.78 (4.81)	U=1208 (.03)
Autism diagnosis professional					
Clinical psychologist	20 (17)	-	2 (8)	-	
Paediatrician	20 (17)	-	5 (20)	-	
Psychiatrist	18 (16)	-	4 (16)	-	6.07 (.42)
Specialist autism team	30 (26)	-	9 (36)	-	
Speech and language therapist	2 (2)	-	0 (0)	-	
Other/Unknown	25 (22)	-	5 (20)	-	
Ethnicity					
Asian/Asian British	2 (2)	-	0 (0)	-	
Jewish	1 (1)	-	0 (0)	-	2.72 (.44)
Mixed/multiple ethnic groups	4 (3)	-	2 (8)	-	
White/Caucasian	108 (94)	-	23 (92)	-	
Living with caregiver					
Yes	83 (70)	-	21 (84)	-	2.23 (.14)
No	32 (30)	-	4 (16)	-	
Relationship status					
Cohabiting	7 (6)	-	1 (4)	-	
Single	101 (88)	-	23 (92)	-	0.52 (.77)
Other/Prefer not to say	7 (6)	-	1 (4)	-	

	Whole sample (N=115)		Dyad sample (N=25)		Group differences
	N (%)	Mean (SD)	N (%)	Mean (SD)	χ^2 (p-value)
Education (%)					
No qualifications	13 (11)	-	8 (32)	-	
GCSEs or equivalent	20 (17)	-	9 (36)	-	
A-Levels or equivalent	53 (46)	-	7 (28)	-	26.77 (<.001)
Undergraduate	17 (15)	-	0 (0)	-	
Postgraduate	11 (10)	-	1 (4)	-	
Unsure/prefer not to say	1 (1)	-	0 (0)	-	
School attendance					
Home schooled	2 (2)	-	0 (0)	-	
Mainstream	102 (89)	-	23 (92)	-	
SEN unit within a mainstream school	5 (4)	-	0 (0)	-	3.24 (.52)
SEN school	5 (4)	-	2 (8)	-	
Other	1 (1)	-	0 (0)	-	
Employment status					
Employed full-time	5 (4)	-	1(4)	-	
Employed part-time	7 (6)	-	0 (0)	-	
Self-employed	3 (3)	-	0 (0)	-	5.62 (.47)
Student	87 (76)	-	23 (92)	-	
Not employed	9 (8)	-	1 (4)	-	
Other/prefer not to say	4 (3)	-	0 (0)	-	
Household income (%)					
Above the UK median	46 (40)	-	10 (40)	-	
Below the UK median	43 (37)	-	7 (28)	-	2.00 (.37)
Unsure/Prefer not to say	26 (23)	-	8 (32)	-	
Health condition					
Yes	69 (60)	-	15 (60)	-	
No	39 (34)	-	10 (40)	-	2.29 (.32)
Prefer not to say	7 (6)	-	0 (0)	-	
Familial autism					
Yes	30 (26)	-	6 (24)	-	
No	82 (71)	-	18 (72)	-	0.29 (.86)
Prefer not to say	3 (3)	-	1 (4)	-	
Covid shielding					
Yes	30 (28)	-	6 (24)	-	
No	80 (68)	-	18 (72)	-	0.09 (.96)
Unknown	5 (4)	-	1 (4)	-	

Table 10: The parent demographic data for the whole group and the dyad subsample, including statistical differences between the whole group and dyad subsample

	Whole sample (N=50)		**Dyad sample (N=25)**		**Group differences**
	N (%)	**Mean (SD)**	**N (%)**	**Mean (SD)**	**χ^2 (p-value)**
Age (years)					
Parent	-	47.52 (5.35)	-	47.00 (5.83)	t=-0.68 (.50)
YP	-	17.33 (2.68)	-	17.26 (2.56)	U=267.5 (.51)
Gender					
Female	44 (83)	-	22 (88)	-	
Male	3 (6)	-	2 (8)	-	1.33 (.72)
Prefer not to say/prefer to self-describe	3 (4)	-	1 (4)	-	
Transgender					
No	47 (94)	-	23 (92)	-	
Yes	1 (2)	-	1 (4)	-	1.02 (.60)
Prefer not to say	2 (4)	-	1 (4)	-	
Relationship to young person					
Parent	49 (98)	-	25 (100)	-	1.02 (.31)
Sibling guardian	1 (2)	-	0 (0)	-	
Living with caregiver/YP					
Yes	48 (96)	-	24 (96)	-	2.01 (.37)
No	2 (4)	-	1 (4)	-	
YP autism diagnosis age (years)	-	11.44 (4.77)	-	11.78 (4.81)	t=0.36 (.72)
Ethnicity					
Asian/Asian British	1 (2)	-	0	-	
White/Caucasian	45 (90)	-	25 (100)	-	
White/Hispanic	1 (2)	-	0	-	4.35 (.23)
Prefer not to say	2 (4)	-	0	-	
Education (%)					
GCSEs or equivalent	4 (8)	-	2 (8)	-	
A-Levels or equivalent	17 (34)	-	10 (40)	-	1.06 (.79)
Undergraduate	17 (24)	-	7 (28)	-	
Postgraduate	12 (24)	-	6 (24)	-	
Relationship status					
Cohabiting	4 (8)	-	1 (4)	-	
Divorced/separated	3 (6)	-	0 (0)	-	
Married	37 (74)	-	20 (80)	-	3.31 (.19)
Single	5 (10)	-	2 (8)	-	
Other/Prefer not to say	1 (2)	-	2 (8)	-	

	Whole sample (N=50)		Dyad sample (N=25)		Group differences
	N (%)	Mean (SD)	N (%)	Mean (SD)	χ^2 (p-value)
Employment status					
Employed full-time	8 (16)	-	5 (20)	-	
Employed part-time	17 (34)	-	9 (36)	-	
Self-employed	6 (12)	-	1 (4)	-	
Student	2 (4)	-	1 (4)	-	5.37 (.50)
Not employed	7 (14)	-	3 (12)	-	
Homemaker/carer	8 (16)	-	4 (16)	-	
Other/prefer not to say	2 (4)	-	2 (8)	-	
Household income (%)					
Above the UK median	27 (54)	-	15 (60)	-	
Below the UK median	19 (38)	-	10 (40)	-	4.39 (.11)
Unsure/Prefer not to say	4 (8)	-	0 (0)	-	
Health condition					
Yes	23 (46)	-	12 (48)	-	
No	21 (42)	-	10 (40)	-	0.09 (.96)
Prefer not to say	6 (12)	-	3 (12)	-	
Parent autism diagnosis					
Yes	2 (4)	-	1 (4)	-	
No	47 (94)	-	24 (96)	-	1.02 (.80)
Prefer not to say	1 (2)	-	0 (0)	-	
Covid shielding					
Yes	12 (24)	-	4 (16)	-	1.75 (.19)
No	38 (76)	-	21 (84)	-	

2.3.2 Design

The study was a cross-sectional survey design. The primary outcome was a self-report measure of collective family functioning completed by both YP and parent participants. Predictor variables included YP autism characteristics, YP mental health (anxiety, depression, self-harm), and parent mental health (depression, anxiety, and parental stress).

2.3.3 Materials

The measures completed by all participants are summarised in Table 11. The YP survey included a demographic questionnaire, alongside measures of self-harm, autism characteristics, depression, anxiety, and family functioning. The parent survey included a demographic questionnaire, alongside measures of depression, anxiety, parenting stress and family functioning. The research took place during the Covid-19 pandemic, where many families may have had to shield[11] for an extended period, during which time school and work closures and absences were common. Therefore, the survey also asked participants to report on their shielding status to explore this in the analysis.

Table 11: The measures completed by participants

Construct	Measure	
Demographics	Bespoke (Appendix 5)	
Family functioning	The Family Adaptability and Cohesion Evaluation Scale (FACES-IV) package – FACES-IV (cohesion and flexibility subscales), Family Communication Scale (FCS), Family Satisfaction Scale (FSS)	
	Parent Measures	**Young Person Measures**
Autism	-	Autism-Spectrum Quotient (AQ) – Adult version
Depression	Patient Health Questionnaire (PHQ-9)	Beck Depression Inventory (BDI)
Anxiety	Generalised Anxiety Disorder Assessment (GAD-7)	Anxiety Scale for Autism – Adults (ASA-A; aged 16-25) Anxiety Scale for Children – Autism Spectrum Disorder (ASC-ASD; aged 14-15)
Self-harm	-	The Non-Suicidal Self-Injury Assessment Tool (NSSI-AT)
Parenting stress	The Parenting Stress Scale (PSS)	-

[11] The UK shielding programme meant that people who were considered clinically extremely vulnerable were advised to stay at home and minimise any contact with others outside their household and was in place for much of the time the survey was open to participants.

2.3.3.1 Autism

Autism characteristics were measured using The Autism-Spectrum Quotient questionnaire (AQ; Baron-Cohen et al., 2001) which is well-established for autism screening[12] and has been used in clinical and non-clinical populations with adequate or good reliability and validity (Auyeung et al., 2008; Baron-Cohen et al., 2001; Woodbury-Smith et al., 2005). The 50-item questionnaire asks participants to rate their agreement on a 4-point Likert scale, giving a score of 0-50, with higher scores indicating greater autism characteristics. The adult version, which has a suggested cut-off of 32 (borderline 26+), was completed by all YP participants.

2.3.3.2 Family functioning

Family functioning was assessed using The Family Adaptability and Cohesion Evaluation Scale (FACES-IV) package (Olson, 2011), based on the Circumplex Model of Marital and Family Systems (Section 2.2). It was completed by both YP and parents. The package of questionnaires measures family cohesion (balanced cohesion, disengaged, and enmeshed subscales), flexibility (balanced flexibility, rigid and chaotic subscales), communication and satisfaction (Figure 12), with a total of 62 5-choice items rating agreement or satisfaction. Higher scores indicate greater levels of the construct; higher scores on balanced cohesion, balanced flexibility, communication, and satisfaction are therefore interpreted as healthier family functioning, whilst higher scores on the disengaged, enmeshed, rigid and chaotic subscales indicate poorer family functioning. Given these differences in directionality, the manual recommends combining scores for research purposes into balanced/unbalanced cohesion, flexibility, and total ratios to indicate the level of functional versus dysfunctional behaviour perceived in the family system; ratios below 1 are considered

[12] Participants were not excluded based on the AQ results, as they had confirmed an ASC diagnosis in the demographic questionnaire and the AQ is a screening tool rather than a diagnostic tool.

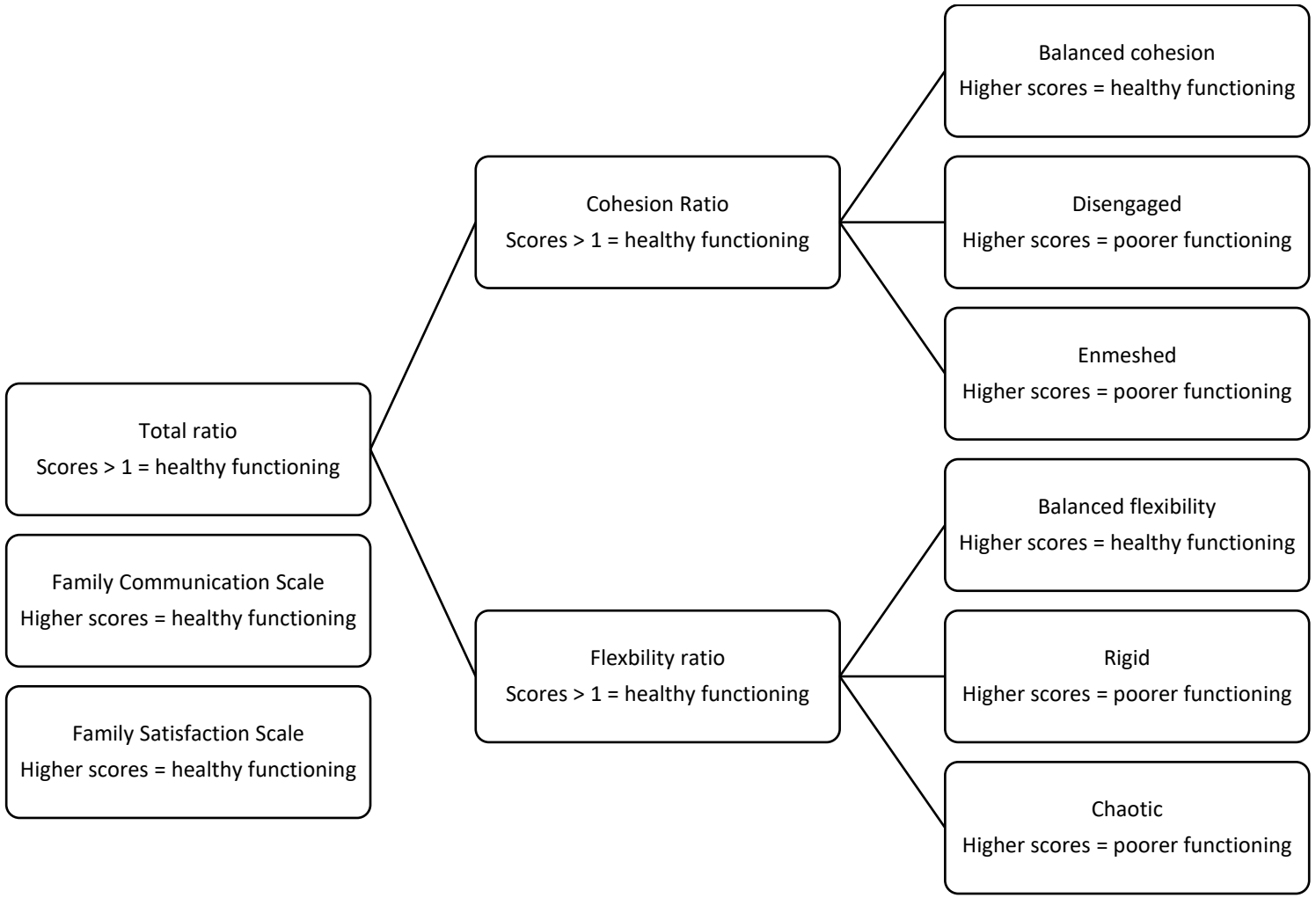

Figure 12: The dimensions and ratios measured by the Family Adaptability and Cohesion Evaluation Scale (FACES-IV)

unbalanced, and above 1 are considered balanced. The Family Communication Scale (FCS; Olson & Barnes, 2010) and the Family Satisfaction Scale (FSS; Olson, 2010) scores range from 10-50. The FACES-IV has been found to be reliable and has high levels of concurrent, construct and discriminant validity (Olson, 2011), and a previous version of the questionnaire was labelled as 'approaching well-established' in a systematic review of family functioning measures (Alderfer et al., 2008).

The reliability of the subscales in the current study was assessed using Cronbach's alpha (Appendix 7). Internal consistency assessed by Cronbach's (1951) alpha was within acceptable range (0.71-0.90; Tavakol & Dennick, 2011) for most subscales. The parent-reported enmeshed subscale was below the commonly used threshold of 0.7, suggesting some items may be poorly inter-related or the subscale may be heterogenous in its dimensions.

2.3.3.3 Young Person Measures

Depression was measured using the Beck Depression Inventory (BDI-II; Beck et al., 1961). Participants were asked to rate 21 items on a Likert scale of 0-3 for severity, giving a score range of 0-63. Higher scores indicate greater depressive symptom severity, with scores over 20 indicating depression in non-clinical populations. The measure demonstrates high internal consistency, content validity, construct validity, concurrent validity, and criterion validity (Beck et al., 1988; Jackson-Koku, 2016), and has been found to be a valid measure of depressive symptoms in autistic adults (Williams et al., 2021).

Anxiety was measured using the Anxiety Scale for Autism - Adults (ASA-A, aged 16+; Rodgers et al., 2020) or Anxiety Scale for Children – Autism Spectrum Disorder – Child version (ASC-ASD, aged 14-16; Rodgers et al., 2016). The 24-item 4-choice child scale rates anxiety frequency and has good internal consistency, validity, and test-retest reliability. The 20-item 4-choice adult version also measures anxiety frequency, has an indicative cut-off of

28, and promising psychometric properties. The raw scores were standardised into Z scores for combination in the analysis.

The Non-Suicidal Self-Injury Assessment Tool (NSSI-AT) assesses multiple features of self-harm (Whitlock et al., 2014). The first question, a behaviour-based yes/no screening question, was used in this study. Test-retest reliability and concurrent, convergent and discriminant validity were found to be adequate in a young adult sample (Whitlock et al., 2014).

2.3.3.4 Parent Measures

Parent depression was measured using the Patient Health Questionnaire (PHQ-9; (Kroenke et al., 2001). Participants rated the frequency of their symptoms using a four-choice Likert scale for nine items. Scores range from 0-27, with higher scores indicating higher levels of depression symptomology. Internal and test-retest reliability are rated as excellent, and good criterion, construct and external validity (Spitzer et al., 2014).

Parent anxiety was measured using the General Anxiety Disorder Assessment (GAD-7; Spitzer et al., 2006). Participants rated the frequency of their symptoms using a four-choice Likert scale for seven items. Scores range from 0-21, with higher scores indicating higher anxiety symptoms. The questionnaire demonstrates good reliability and good criterion, construct, factorial, and procedural validity.

Parenting stress was measured using The Parenting Stress Scale (PSS; Berry & Jones, 1995). The five-choice Likert scale asks participants to rate their agreement on 18 items exploring the positives and negatives of parenting. Scores range from 18 (low stress) to 90 (high stress). It demonstrates good internal consistency and test-retest reliability, and adequate construct, convergent, concurrent and divergent validity (Algarvio et al., 2018; Berry & Jones, 1995; Harding et al., 2020; Leung & Tsang, 2010; Pontoppidan et al., 2018; Zelman & Ferro, 2018).

2.3.4 Procedure

Promotional materials, information sheets, and consent forms can be found in Appendix 4. Flyers were shared on online social media platforms. Participants of prior university studies who had given permission to be contacted about further research were contacted directly by email after being screened as eligible by the researchers. Schools, university disability services, and autism charities were contacted by email and asked to share flyers and survey links with families using their services. The promotional materials described the survey as exploring wellbeing to minimise bias in recruitment. Both surveys were administered online through Qualtrics. The order of the questionnaires was randomised for participants to overcome order effects.

2.3.5 Data Analysis

All data were assessed for normality using the Shapiro & Wilk (1965) Test of Normality due to its greater power with small sample sizes (Appendix 7; Ghasemi & Zahediasl, 2012). These results informed the choice of parametric or non-parametric statistical tests (i.e., t-test vs Mann Whitney U tests for between group differences, and Pearson's vs Spearman rho's correlations) throughout. Valid mean substitution (Dodeen, 2003) was used to replace missing data (three data points in the parent survey, twelve in the YP survey) to maintain the sample size and power. As imputation can skew the data and reduce its variance, a threshold was set at 75% of complete data. Research question 1 was analysed using the FACES-IV package spreadsheet provided in the manual, including comparison against normative data[13]. Research questions 1-4 were explored using bivariate and partial correlations using SPSS Statistics v27. Research question 5 was explored using

[13] The manual provided a mean, N (FACES-IV N=465, FCS/FSS N=2465), and standard deviation for the normative samples. T-tests were selected to compare between the current samples and the normative data even where data did not meet all assumptions for parametric tests, as the descriptive data required for non-parametric testing was not available. No demographic information was available for the normative sample. Results should therefore be interpreted with caution.

hierarchical multiple linear regression; these analyses were completed for the whole samples as the dyad sample was small, so parent and YP variables were explored separately. Demographic variables (age, gender, ethnicity, living with dyad, relationship status, education level, employment status, household income, autism diagnosis age, health conditions, covid shielding) were condensed into dummy variables[14]. The enter method was used, with demographic variables added as controls in the first step, and other variables added in stages based on the strength of correlations found in the previous results. Variables were included or excluded based on significant coefficient values, significant change statistics, and an increase in the adjusted R^2. The Holm-Bonferroni method (Holm, 1979)[15] was used to adjust p-values based on the number of tests completed to minimise type I errors caused by multiple testing, and these were calculated using an Excel programme (Boustani, 2020). In this method, the p-values are ranked numerically from smallest to largest, and adjusted alpha thresholds of significance (dependent on the number of tests within that family) are assigned to each ranked p-value. For example, for a family of four tests, the adjusted alpha thresholds are 0.013, 0.017, 0.025 and 0.05, with the smallest p-value compared to 0.013 and the largest p-value compared to 0.05.

2.3.6 Ethics

The study proposal was discussed at a Patient & Public Involvement group (Birmingham Psychology Autism Research Team; B-PART) and feedback was incorporated into the procedure. The study was subsequently reviewed and approved by the University of Birmingham STEM ethics committee (Appendix 3). A best practice risk protocol and duty of care policy was in place (Appendix 6; Lloyd-Richardson et al., 2015; Singhal & Bhola, 2017). Participants were informed prior to the research study that questions about mental

[14] Dummy variables were explored as both dichotomous variables (e.g., female vs male and other) and wider categorical variables (e.g., female vs male vs prefer not to say/other).
[15] The Holm-Bonferroni correction was chosen due to its increased power over the Bonferroni correction, to minimise Type II errors.

health and self-harm would be included. Participants aged 14-15 required a password to access the survey, which was provided to parents after provision of parental consent. Items included in the BDI, NSSI-AT and PHQ-9 ask about self-harm and suicide. Research shows that asking about suicide results in small reductions in suicidal ideation and suicidal behaviour in typically developing populations, with adolescents reporting greater reductions than adults (Blades et al., 2018; Dazzi et al., 2014). Young adolescents and adults have also described it as cathartic, rewarding and therapeutic to take part in risk assessments or research on self-harm (Lockwood et al., 2018; Whitlock et al., 2013). For a small minority of participants, a temporary increase in stress and negative mood may occur; these participants report this distress is outweighed by the benefits of contributing to research (Biddle et al., 2013), and their mood returns to baseline within days with no long-term harm (Labott et al., 2013). Mood-elevation techniques are recommended (Lockwood et al., 2018), so the survey included suggestions and links to alternative websites as a distraction. The debrief included signposting to relevant resources and supporting organisations, including health and emergency services, and participants were offered contact with the research team. Parents were provided with resources on how to address the topic of mental health with their children (Oldershaw et al., 2008).

2.4 Results

2.4.1 Family Functioning

The FACES-IV data was used to assess the first research question (see Table 12 for descriptive data for all measures; see Table 13 for the FACES-IV subscale interpretations). The results can be interpreted as indicating overall balanced levels of cohesion, flexibility, and total family functioning, moderate to high levels of communication, but low levels of family satisfaction. Compared to the normative data (Table 14), parents reported greater

Table 12: The descriptive statistics for the measures completed by parents and young people

Measure	Parent Sample			Young Person Sample		
	Full Sample **Mean (SD) / Median*** **Range**	**Dyad Sample** **Mean (SD) / *Median*** **Range**	**Dyad / Non-Dyad Group Differences Test statistic (p)***	**Full Sample** **Mean (SD) / *Median**** **Range**	**Dyad Sample** **Mean (SD) / *Median**** **Range**	**Dyad / Non-Dyad Group Differences Test statistic (p)***
Cohesion Raw Score	*29* 18-34	-	-	*26* 8-35	-	-
Flexibility Raw Score	*25* 9-32	-	-	*22* 8-35	-	-
Disengaged Raw Score	*14* 9-27	-	-	*20* 9-31	-	-
Enmeshed Raw Score	15.44 (4.38) 8-25	-	-	*14* 7-26	-	-
Rigid Raw Score	20.46 (5.58) 10-35	-	-	*18* 10-32	-	-
Chaotic Raw Score	*15* 8-35	-	-	19.04 (5.58) 7-33	-	-
Cohesion Ratio	3.11 (1.46) 0.65-5.79	3.63 (1.38) 1.13-5.79	t=2.69 (.01)	*1.94* 0.37-6.80	2.84 (1.16) 0.94-5.33	U=624 (<.001)
Flexibility Ratio	1.70 (0.69) 0.33-3.52	1.88 (0.63) 0.97-3.52	t=1.90 (.06)	*1.20* 0.34-3.22	1.70 (0.66) 0.65-3.00	U=708.5 (<.01)
Total Ratio	2.41 (0.96) 0.49-4.22	2.76 (0.89) 1.15-4.22	t=2.75 (<.01)	*1.61* 0.36-4.49	2.26 (0.80) 0.79-4.17	U=653 (<.01)
FCS Total Score	*39* 18-49	39.52 (4.31) 33-49	t=1.70 (.10)	33.62 (8.37) 12-50	37.40 (6.03) 22-47	t=37.40 (<.01)
FSS Total Score	29.54 (6.94) 10-43	32* 10-37	U=256 (.27)	*32* 20-50	35.84 (5.94) 26-49	U=36 (<.01)
PHQ-9 Total Score	*6* 0-21	6* 0-18	t=-1.12 (.27)	-	-	-
GAD-7 Total Score	*6.5* 0-20	6* 0-19	U=321.5 (.86)	-	-	-
PSS Total Score	47.80 (11.34) 28-77	44.84 (9.18) 28-58	t=-1.91 (.06)	-	-	-
AQ Total Score	-	-	-	*33* 11-48	33.58 (8.60) 15-47	t=0.23 (.82)

Measure	Parent Sample			Young Person Sample		
	Full Sample Mean (SD) / Median* Range	**Dyad Sample Mean (SD) / Median* Range**	**Dyad / Non-Dyad Group Differences Test statistic (p)**	**Full Sample Mean (SD) / Median Range**	**Dyad Sample Mean (SD) / Median* Range**	**Dyad / Non-Dyad Group Differences Test statistic (p)**
BDI Total Score	-	-	-	*19* 1-48	9 1-48	U=1403 (.03)
ASA-A Total Score (age 16-25)	-	-	-	*33* 6-56	-	-
ASC-ASD Total Score (age 14-15)	-	-	-	*20* 17-53	-	-
Anxiety Standardised Scores	-	-	-	0.00 (1.00) -2-2	0.00 (0.98) -1.54-2.10	t=-1.33 (.19)

*T-tests and means are reported for parametric data, while Mann Whitney U tests and medians are reported for non-parametric data.

Table 13: The interpretation of the Family Adaptability and Cohesion Evaluation Scales for the whole samples

Subscale	Parent Interpretation	Young Person Interpretation
Balanced Cohesion	Very connected	Connected
Balanced Flexibility	Flexible	Flexible
Disengaged	Very low	Low
Enmeshed	Very low	Very low
Rigid	Low	Low
Chaotic	Very low	Low
FCS	High	Moderate
FSS	Low	Low

Table 14: The comparison of the Family Adaptability and Cohesion Evaluation Scale package scores with normative data

Scale	Parent Data					Young Person Data				
	Difference from normative mean	**t**	**df**	**p**	**d**	**Difference from normative mean**	**t**	**df**	**p**	**d**
Cohesion ratio	+0.7	4.85	513	<.001*[1]	0.58	-0.2	2.16	578	.03*[5]	0.20
Flexibility ratio	+0.1	0.99	513	.32[5]	0.15	-0.2	3.51	578	<.001*[1]	0.36
Total ratio	+0.4	3.78	513	<.001*[2]	0.49	-0.2	3.06	578	<.01*[3]	0.29
FCS	+1.9	1.47	2513	.14[4]	0.24	-2.6	3.01	2578	<.01*[4]	0.30
FSS	-8.0	6.58	2513	<.001*[3]	1.08	-4.1	5.05	2578	<.001*[2]	0.57

*Significant p-value after Holm-Bonferroni p-adjustment applied. Parent data and young person data were treated as two separate families of tests and the correction applied to each family separately. The p-values are annotated with their superscript rankings from 1-5, and the adjusted alpha thresholds were 0.01, 0.013, 0.017, 0.025, and 0.05.

cohesion and overall family functioning (medium effect sizes; Sawilowsky, 2009), but a significantly lower level of satisfaction (large effect size). These effects were statistically significant after correction for multiple tests. YP reported lower cohesion, flexibility, overall family functioning, communication, and satisfaction (small to medium effect sizes) compared to the normative sample, and these effects were statistically significant after correction for multiple comparisons.

To further explore the first research question, the YP and parent cohesion and flexibility ratios[16], FCS and FSS scores were compared to each other (Table 15). In both the full sample and the dyad subsamples, young people reported consistently lower scores on all family functioning scales. After correction for multiple comparisons, this was significant for all of the measures in the full sample, and for the FSS scores in the dyad subsample.

Table 15: The differences between the parent and young people reported family functioning scores in the full sample and the dyad sample

Family Functioning Measure	Full sample group differences between parents and young people		Dyad sample group differences between parents and young people	
	U	p	t	p
Cohesion ratio	1783.5	<0.001*[1]	2.18	0.03[2]
Flexibility ratio	1980	0.002*[3]	1.18	0.25[4]
FCS	1929.5	<0.001*[2]	1.43	0.16[3]
FSS	3639.5	0.007*[4]	U=456.5	0.005*[1]

*Indicates a significant p-value after correction for multiple comparisons, with the full sample and dyad sample treated as two separate families of tests for correction. The p-values are annotated with their superscript rankings from 1-4, and the adjusted alpha thresholds were 0.013, 0.017, 0.025, and 0.05.
Mann Whitney U tests used for non-parametric data, and t-tests used for parametric data

Finally, correlations were used to explore associations between the dyad parent and YP reported family functioning scores (Table 16). Positive correlations were found between all measures, with correlations that could be considered trivial/low (Hinkle et al., 1979) to moderate (Davis, 1971) in size. The associations explained low levels of the variation as

[16] The cohesion and flexibility ratios were used to explore correlations with other variables to allow for a more nuanced understanding of the results. The total ratio was not included in these analyses as doing so reduced statistical power, without furthering understanding.

measured by R^2 (range 0.64-16.00%). All correlations were statistically non-significant after correction for multiple tests.

Table 16: The correlations between the parent- and young person-reported family functioning scores

Measure	Correlation	R^2
Cohesion ratio	0.34*	11.56
Flexibility ratio	0.08	0.64
FCS	0.40*	16.00
FSS	0.27	7.29

*Significant at p<.05 level, but non-significant after correction for multiple comparisons. The adjusted alpha thresholds were 0.013, 0.017, 0.025, and 0.05.
The cohesion ratio, flexibility ratio and FCS report Pearson's correlation; the FSS reports Spearman's rho

In summary, the parent and YP participants reported balanced levels of family functioning. Family satisfaction was reported as low for both samples and was significantly lower than the normative FSS sample. Parents reported higher levels of cohesion and overall family functioning than the normative sample, while YP in this sample reported lower scores on all measures compared to the normative sample. When using the full sample data, YP reported lower scores on all family functioning measures than the parent sample. In the dyad subsample, YP reported lower family satisfaction. No significant associations were found between the YP and parent-reported family functioning data in the dyad subsample.

2.4.2 Young Person Autism Characteristics and Mental Health

To address the second research question, the descriptive statistics for YP AQ, BDI, Anxiety Z scores, and NSSI-AT were calculated (Table 12), and their associations with family functioning were explored using correlations (Table 17). The full sample was used for correlations between YP-only variables, and the dyad sample was used for comparisons with YP characteristics and parent-reported family functioning.

The median AQ score indicated that on average the sample scored over threshold for autism, and 82% of participants' individual scores were over the threshold. YP AQ scores were consistently negatively correlated with YP-reported family functioning, but results were

not statistically significant after correction for multiple comparisons. No significant correlations were found between YP AQ scores and parent-reported family functioning scores.

Based on the completed NSSI-AT data, 62% of the whole sample and 52% of the dyad sample had engaged in NSSI at some point. The difference in family functioning by presence or absence of NSSI was explored using independent samples t-tests[17] (Table 18). There were significant differences between YP-reported flexibility and family communication and YP NSSI, with all YP family functioning measures indicating healthier family functioning for the non-NSSI group with medium effect sizes. The difference in the YP-reported flexibility ratios between the NSSI and non-NSSI groups was significant after correction for multiple tests. The tests showed no significant differences between parent-reported family functioning and YP NSSI.

The median BDI score was below the cut-off of 20 used in non-clinical populations, and 49% of autistic YP scored above this threshold for depression. YP BDI scores were significantly negatively associated with all measures of YP-reported family functioning. No significant correlations were found between YP BDI scores and parent-reported family functioning scores. The median ASA-A completed by participants aged 16+ was above the clinical cut-off for impactful anxiety, and 64% of autistic YP scored above this threshold. The median ASC-ASD completed by participants aged 14-15 was at the clinical cut-off for impactful anxiety, and 71% of autistic YP scored above this threshold. YP anxiety scores were significantly negatively associated with YP-reported cohesion ratio, flexibility ratio, and FCS scores, but not FSS scores. No significant correlations were found between YP anxiety scores and parent-reported family functioning scores.

[17] Four outliers identified by SPSS were removed from the YP-reported family functioning dataset (two from each of the cohesion and flexibility ratios) for t-tests to meet the test assumptions.

Table 17: The correlations between young person autism characteristics and mental health with parent and young person reported family functioning

	Cohesion Ratio (Parent)	Flexibility Ratio (Parent)	FCS (Parent)	FSS (Parent)	Cohesion Ratio (YP)	Flexibility Ratio (YP)	FCS (YP)	FSS (YP)
AQ	-0.09	0.02	-0.41	-0.20^	-0.18^	-0.19^	-0.23^	-0.17^
BDI	0.13^	0.05^	-0.33^	-0.02^	-0.39*^	-0.35*^	-0.36*^	-0.31*^
Anxiety	0.24	0.41	0.02	0.03^	-0.29*^	-0.33*^	-0.25*	-0.13^

* Correlation is significant after correction for multiple tests, with the tests treated as two separate families (parent-reported family functioning, young person-reported family functioning). The adjusted alpha thresholds were: 0.004, 0.005, 0.005, 0.006, 0.006, 0.007, 0.008, 0.01, 0.013, 0.017, 0.025, and 0.05.
Tests reported are Pearson's correlations; tests reporting Spearman's rho are marked with ^

Table 18: The comparison between family functioning scores across the self-harming and non-self-harming young person sample

Family functioning measure	Parent-reported family functioning						YP-reported family functioning					
	Mean (SD)		t	df	p	d	Mean (SD)		t	df	p	d
	NSSI +	NSSI -					NSSI +	NSSI -				
Cohesion ratio	3.57 (1.42)	3.69 (1.40)	-2.14	23	.83[3]	-0.09	1.98 (1.38)	2.46 (1.30)	-1.83	112	.07[4]	-0.35
Flexibility ratio	1.89 (0.74)	1.88 (0.52)	0.029	23	.97[4]	0.01	1.21 (0.63)	1.56 (0.74)	-2.65	112	<.01*[1]	-0.51
FCS	37.92 (4.05)	41.25 (4.05)	-2.052	23	.05[1]	-0.82	32.28 (8.54)	35.70 (7.79)	-2.14	112	.04[2]	-0.41
FSS	29.77 (5.39)^	30.92 (7.59)^	96.50^	-	.32[2]	0.20	32.48 (7.51)	35.05 (6.29)	-1.88	112	.06[3]	-0.36

^ Data reported is median and Mann-Whitney test statistics, as data did not meet parametric assumptions of normality
*Test is significant after correction for multiple tests. Results were treated as two families of tests (parent-reported and young-person reported family functioning). The p-values are annotated with their superscript rankings from 1-4, and the adjusted alpha values were: 0.013, 0.017, 0.025 and 0.05.

In summary, the results demonstrated no associations between YP autism characteristics or mental health with parent-reported family functioning. No significant associations were found between YP autism characteristics YP-reported family functioning scores. Significant negative associations were found between YP BDI scores and all measures of YP-reported family functioning, and between YP anxiety scores and YP-reported cohesion, flexibility, and communication scores. The YP self-harming group reported a significantly lower flexibility ratio than the non-self-harming group.

2.4.3 Parent Mental Health

To address the third research question, the descriptive statistics for parent PHQ-9, GAD-7 and PSS scores were calculated (Table 12), and their associations with family functioning were explored using correlations (Table 19). The full sample was used for correlations between parent-only variables, and the dyad sample was used for comparisons with parent mental health and YP-reported family functioning.

The PHQ-9 and GAD-7 median values indicate mild levels of depression and anxiety in the parent sample; the range was from none to severe. Consistent negative correlations were found between all measures of parent mental health and all family functioning scores. PHQ-9 scores were significantly negatively correlated with parent-reported flexibility ratios. GAD-7 scores were significant correlated with parent-reported cohesion and flexibility. PSS scores were significantly correlated with parent-reported cohesion, flexibility, and communication. In summary, no significant correlations between parent mental health and YP-reported family functioning were found. Parent-reported flexibility ware significantly negatively associated with all measures of parent mental health, cohesion was associated with GAD-7 and PSS scores, and FCS scores were also significantly negatively associated with PSS scores.

Table 19: The correlations between parent mental health and parent- and young person-reported family functioning

	Cohesion Ratio (Parent)	Flexibility Ratio (Parent)	FCS (Parent)	FSS (Parent)	Cohesion Ratio (YP)	Flexibility Ratio (YP)	FCS (YP)	FSS (YP)
PHQ-9	-0.38^	-0.55*^	-0.02^	-0.22^	-0.18^	-0.10^	-0.21^	-0.31^
GAD-7	-0.46*^	-0.45*^	-0.21^	-0.37^	-0.20^	-0.11^	-0.08^	-0.09^
PSS	-0.56*	-0.44*	-0.58*^	-0.45	-0.17	-0.10	-0.04	-0.45

* Correlation is significant after p-value correction for multiple tests. Tests were considered as two separate families (parent-reported family functioning and young person-reported family functioning) for the purpose of correction. The adjusted alpha thresholds were: 0.004, 0.005, 0.005, 0.006, 0.006, 0.007, 0.008, 0.01, 0.013, 0.017, 0.025, and 0.05.

Parent-reported tests were 1-tailed; young-person reported tests were 2-tailed

Tests reported are Pearson's correlations; ^ indicates Spearman's rho was used as a non-parametric alternative

2.4.4 Exploring Family Functioning

To explore the fourth research question, the correlations between the YP- and parent-report family functioning measures were further explored using partial correlations[18], where YP and parent variables were controlled for (Table 20). The results between the bivariate and partial correlations were compared for changes in magnitude and significance. Controlling for parent mental health scores had minimal effect on the family functioning correlations, and no partial correlations were found to be statistically significant, and therefore no correction for multiple tests was required.

Table 20: The associations between the family functioning measures (YP and parent report) when parent and young person variables were controlled for in partial correlations

Family functioning measure	**Controlled for Correlation coefficient (R^2)**					
	PHQ-9 (parent)	**GAD-7 (parent)**	**PSS (parent)**	**AQ (YP)**	**BDI (YP)**	**Anxiety (YP)**
Cohesion ratio	0.33 (10.89)^	0.32 (10.24)^	0.31 (9.61)	0.33 (10.89)	0.50 (25.00)^	0.37 (13.69)
Flexibility ratio	0.07 (0.49)^	0.07 (0.49)^	0.06 (0.36)	0.06 (0.36)	0.13 (1.69)^	0.21 (4.41)
FCS	0.39 (15.21)^	0.35 (12.25)^	0.41 (16.81)	0.26 (6.76)	0.28 (7.84)^	0.44 (19.36)
FSS	0.23 (5.29)^	0.27 (7.29)^	0.11 (1.21)^	0.20 (4.00)^	0.28 (7.84)^	0.27 (7.29)^

Partial correlations reported are Pearson's; ^ indicates Spearman's rho was used as a non-parametric alternative

To explore the fifth and final research question, hierarchical multiple linear regression models (Table 21) were used to explore the variables that predicted total family functioning[19] ratios. The first step of the parent-reported total family functioning ratio regression included full-time employment; no other demographic control variables were found to be significant predictors. PSS and GAD-7 scores were also significant predictors of total family functioning, with PSS scores accounting for the greatest variance. The final model accounted for 39% of the variation, and full-time employment was non-significant in the final model. The inclusion of PHQ-9 scores in a fourth step increased the adjusted R^2 to 0.40, but this change was non-significant (F=1.14, p=.29). The model met assumptions of normality and

[18] Regression, moderator, and mediator analyses were not used on the dyad sample as the sample size was too small and therefore underpowered to find significant effects.
[19] Total family functioning ratios were selected to subsume the cohesion and flexibility ratios into one family functioning regression model.

homoscedasticity based on visual inspection of a P-P plot and a scatterplot of the residuals (Appendix 8), and assumptions of multicollinearity based on acceptable VIF values (Johnston et al., 2018).

Table 21: The hierarchical multiple linear regression models predicting total family functioning ratios

Outcome variable	Predictor variables	β	t	R	Adjusted R^2	ΔR^2	VIF
Parent-reported total family functioning ratio (N=49)	*Step 1*			0.36	0.11	0.11**	
	Employment – full-time	0.36	2.67*				1.22
	Step 2			0.61	0.35	0.24***	1.25
	Employment – full-time	0.18	1.48				
	PSS	-0.53	-4.23***				
	Step 3			0.66	0.39	0.04*	1.28
	Employment – full-time	0.11	0.92				
	PSS	-0.44	-3.51**				
	GAD-7	-0.27	-2.10*				
Young person-reported total family functioning ratio	*Step 1*			0.34	0.09	0.09**	
	Age	-0.08	-0.68				1.63
	Non-student	-0.18	-1.81				1.17
	University education	-0.22	-1.94				1.45
	Step 2			0.50	0.22	0.13***	
	Age	-0.10	-0.89				1.59
	Non-student	-0.08	-0.83				1.18
	University education	-0.19	-1.78				1.45
	BDI	-0.38	-4.27***				1.05

*p<.05; **p<.01; ***p<.001

For the YP model, age, not being a student, and having a university-level education were significant predictors individually, and so were included in the first step of the model; together they resulted in a significant model, but each individual variable was non-significant. No other demographic variables were found to be significant predictors. BDI scores were also a significant predictor of total family functioning. In total, the model accounted for 22% of the variance. The inclusion of further variables was non-significant (AQ: F=0.19, p=.66; anxiety: F=0.05, p=.83; NSSI: F=0.28, p=.60). The model met assumptions for normality based on visual inspection of a P-P plot (Appendix 8) and assumptions of multicollinearity based on acceptable VIF values. A low level of heteroscedasticity was identified on visual

inspection of the scatterplot of residuals, which risks reducing precision and producing p-values smaller than they should be. As the model was significant at $p<.001$, this was not considered problematic enough to require further analysis.

In summary, parent total family functioning ratios were predicted by PSS and GAD-7 scores, accounting for 39% of the variation in scores with PSS accounting for the greatest variance. Full-time employment was also a significant predictor in the first stage of the model but was non-significant in the final model. YP-reported total family functioning ratios were predicted by BDI scores, accounting for 22% of the variance. Age, not being a student, and having a university-level education were also included as significant predictors in the first stage of the model but were non-significant in the final model.

2.5 Discussion

This is the first study to explore family functioning in families with an autistic child from the subjective perspective of *both* the parents and the autistic young people themselves (Greenlee et al., 2018; Milton, 2012; Walton & Tiede, 2020). The study also explored how parent and child mental health and child autism characteristics may explain differences in family functioning. The findings of this study are strengthened by the inclusion of a large sample of autistic young people, including a parent-child dyad subsample, allowing comparisons across and between families, and by the use of valid and reliable measures.

In this study both parents and young people reported overall balanced levels of family functioning, but low levels of family satisfaction. The child scores on all measures were lower than parent scores, with significant differences found on all scales for the full sample and lower satisfaction scores in the dyad subsample. No associations were found between the dyad family functioning scores in isolation or when controlling for autism or mental health. Young person anxiety and depression were significantly negatively associated with young

person-reported family functioning (except for anxiety and satisfaction), and young people who reported self-harm also reported significantly lower flexibility ratios, but there was no association with autism characteristics. Parent-reported flexibility was associated with all measures of parent mental health, in addition to cohesion with anxiety and stress, and communication with stress. No associations were found across the dyads. Total family functioning ratios were predicted by stress and anxiety for parents and by depression for young people.

On average both parents and autistic young people in this study reported balanced levels of family functioning. Autism characteristics were not associated with family functioning scores. These results should be reassuring for families, as they indicate that autism itself is not inherently associated with poorer family functioning despite the results of the previous meta-analysis (Section 1.4) that families with an autistic child report poorer family functioning than those with typically developing children. The FACES-IV measure was chosen due to its capacity for identifying dysfunctional as well as healthy family functioning. The finding of overall balanced functioning even on this measure can therefore help to challenge the caregiver burden narrative of a child's autism leading to negative outcomes for other family members (Turnbull et al., 2007). Indeed, the parent sample reported greater levels of cohesion than the normative sample, suggesting the experience of family functioning for parents of autistic children may be more nuanced across different dimensions. However, no demographic data are available for the normative sample. Given the emphasis on parent report in previous research, the normative sample may therefore not be a suitable comparison for the young person sample in this study, and the finding that the young person sample reported significantly lower scores should therefore be interpreted with caution. It is also possible that the normative sample includes data from clinical samples reporting poorer family functioning than the general population (Walton & Tiede, 2020).

Nonetheless, these data demonstrate the novel finding that on average both parents and, critically, autistic young people themselves report overall balanced family functioning.

Both parents and young people reported low levels of family satisfaction, which is the degree to which family members feel happy and fulfilled by each other. The normative sample for family satisfaction reported by parents follows a U curve across the family life cycle, and is lowest when children are in late adolescence (Olson et al., 1989). This sample captures families with adolescent children, which may explain the low levels of satisfaction reported by this sample. However further research is needed to assess whether this U curve replicates in families with an autistic child, who may not follow a typical life cycle trajectory. The discrepancy between balanced levels of family functioning in the sample yet low levels of family satisfaction suggests that family satisfaction may be better explained by other variables not measured in the FACES-IV questionnaire package. Given that family satisfaction accounts for nearly half of the variance in an individual's quality of life (Olson et al., 1989), this finding warrants further research. Factors beyond the individual and family system levels may be more relevant for family satisfaction in families with an autistic child. For example, families with autistic children experience inequalities around physical health, mental health, employment and education, which has only been exacerbated by the recent covid-19 pandemic (National Autistic Society, 2020). This indicates that further research and interventions may need to be targeted at the level of the mesosystems, exosystems and macrosystems (Section 1.2.2), consistent with a community psychology approach.

Young people reported lower scores on all family functioning measures in the full sample and lower levels of family satisfaction in the dyad sample compared to the parent samples. The dyad sample reported better family functioning and mental health than the wider young person sample, which may explain the differential findings between young adult and parent scores in the samples. It is highly plausible that this effect is causal, with families

with poorer functioning less likely to jointly participate in research on families, which could be linked to both family functioning itself or external family stressors and barriers. However, the findings in both the full sample and dyad subsample that young people report lower family functioning are consistent, suggesting this is a valid effect that should be further explored.

The correlations between parent and young person reported family functioning are trivial to medium in size, but not statistically significant. The consistency of the correlation direction suggests the lack of statistical significance may be due to the small sample size. However, given both informants are reporting on the same construct, the weak correlations demonstrate that each individual's subjective experience of their family's functioning is very different. This is consistent with research in other populations, where correlations between parent and child reports are of a similar magnitude (Olson, 2010; Palmer et al., 2016). This emphasises the importance of using multiple family informants on family functioning in both research and clinical settings. Given the small dyad sample size and their tendency to be younger and better functioning, it cannot be assumed, however, that the results from the dyad sample generalise to families with poorer functioning.

Young person depression and anxiety were consistently negatively associated with young person-reported family functioning. In the regression models, young person depression predicted their reported total family functioning ratio. These results are consistent with the research in the general population linking poorer family functioning with poor mental health, suggesting the bidirectional associations replicate in autistic young people and their families. It is also consistent with previous findings that the association between autism characteristics and family functioning can be accounted for by child internalising disorders (Green et al., 2016). Extensive literature from the typically developing population has linked self-harm to family functioning, but few significant differences was found in this sample. These findings

may be due to the small sample size, as well as the use of the binary yes/no screening question which cannot distinguish different levels of frequency or severity of self-harm. Prior research has found that lower levels of flexibility are associated with frequency and duration of self-harm in typically developing adolescents (Halstead et al., 2014), although it should be noted that the mean score for the self-harm group in this sample would still be interpreted as balanced flexibility. Overall, these results suggest that child depression and anxiety may be a key target for intervention, both at an individual level and a family level. It is therefore essential that clinicians are equipped with the skills to for making clinical adaptations to therapies for autistic young people, with a need for training and ongoing supervision (Cooper et al., 2018). However, the regression model only explained 22% of the variance in family functioning total ratios, indicating there are other important mechanisms that need to be elucidated.

Parent-reported mental health was negatively associated with parent-reported family functioning, although findings were less consistent across all measures. In the regression model, parent stress and anxiety predicted the parent-reported ratio, although the model only accounted for 39% of the variance. PSS scores were significantly higher than a Norwegian normative sample (mean=31, t=15.3700, df=1143, p<.0001; Nærde & Hukkelberg, 2020), consistent with previous research that parents of autistic children experience higher levels of stress (Hayes & Watson, 2013). Depression was a non-significant predictor in the regression model, in contrast with previous research that has linked parental depression to family functioning (Baker et al., 2011; Jellett et al., 2015; Walton & Tiede, 2020). These results suggest that targeting parental stress and anxiety may be a key intervention focus for families. Stress can be defined as a relationship between the person and their environment that is appraised as exceeding coping resources (Lazarus & Folkman, 1984). Interventions targeting stress may therefore take the form of individual intervention targeting those appraisals or

coping resources, but also may involve support at the level of the meso-system from services to decrease stressors or provide additional coping resources. Overall, these results suggest that the meta-analytic findings of poorer family functioning in families of autistic children (Section 1.4) may be partially explained by individuals within these families experiencing poorer mental health than other families. This interpretation highlights a significant clinical need to address the mental health of both autistic people and their families, with a notable gap of rigorous research on systemic family therapies in this population (Walton & Tiede, 2020).

The partial correlations used in this study suggest that the difference in informant family functioning scores is not well explained by autism characteristics or mental health variables, so further exploratory research involving parent-child dyads exploring this discrepancy is warranted. Very few significant associations were found across dyads, suggesting that poor mental health affects an informant's subjective experience of their family functioning but not that of other family members. This is consistent with Yorke et al.'s (2018) suggestion that mental health and family functioning outcomes are inflated through shared rater effects, and findings that autistic child internalising disorders are not associated with parent-reported family functioning (Nuske et al., 2018). Mental health should therefore be assessed in clinical settings and controlled for in future research. Given the interdependency of family functioning and mental health treatment outcomes (Dardas, 2019; Feeny et al., 2009; Jongerden & Bogels, 2015; Keeton et al., 2013), further understanding of these associations has the potential to identify treatment pathways for improving family functioning by targeting an individual's mental health and/or improving mental health by targeting family functioning. Research that can allow causal and directional mechanisms to be elucidated is therefore needed. Larger dyad samples would also allow cross-dyad variables to be included in the family functioning models.

2.5.1 Strengths and Limitations

The FACES-IV measure used is a well validated and psychometrically robust measure (Alderfer et al., 2008; Olson, 2011). Family functioning measures have previously been criticised for not capturing the extremes of family dysfunction (Glidden, 2012), which the FACES-IV is able to capture within one overall ratio of balanced to unbalanced functioning. However, the FACES-IV, like other measures of the same construct, has been developed with neurotypical populations and therefore the model may not validly apply to families with autistic children (Turnbull et al., 2007). The FACES-IV package of questionnaires generally demonstrated acceptable internal consistency as measured by Cronbach's alpha in this sample. The parent-reported enmeshment subscale, however, fell below the 0.7 threshold, suggesting some items may be poorly inter-related or the subscale may be heterogenous in its dimensions in this population. Families with an autistic child may communicate and display cohesion in different ways to typically developing families (Greenlee et al., 2018). Higher parent-reported enmeshment is not associated with decreased parent happiness in families with an autistic child, in contrast to typically developing families (Walton & Tiede, 2020). Parents of autistic children who score highly on enmeshment also use more positive coping strategies than those with other cohesion styles, suggesting enmeshment may be an adaptive and healthy strategy for these families (Altiere & von Kluge, 2009). The finding that the enmeshment scale may lack internal consistency in this parent sample suggests the meaning, measurement and clinical implications of cohesion and enmeshment may differ in families of autistic children compared to typically developing families, and this warrants further research. The measurement of depression in autistic young people also requires further research; the BDI-II used in this study has previously been identified as the best available measure, but is still not particularly strong (Gotham et al., 2015).

This study focused on autistic individuals without a co-occurring diagnosis of ID, and these clear exclusion criteria meant that autism-specific factors could be explored beyond general neurodevelopmental factors. However, neither autism diagnosis nor lack of ID diagnosis were validated through assessments or accessing health records. Feedback from participants suggested many were unsure about the distinction between autism and learning disabilities. The results, therefore, may be subject to exposure bias.

This study was cross-sectional, and it is therefore not possible to determine causality or directionality of the associations identified. The reliance on convenience sampling risks introducing bias into the sample, and the significantly better family functioning found in the dyad sample also suggests recruitment bias in this subsample. Recruitment through schools was limited by the closures and disruption of the Covid-19 pandemic, meaning the overall smaller sample size was underpowered to find statistical significance at small effect sizes raising the likelihood of type II errors. The use of the FACES-IV measure provided rich clinical data but resulted in multiple tests being performed for each research question. Correction for multiple tests was also therefore required, which can be considered overly conservative.

The parent sample was predominantly mothers, in keeping with past research (Greenlee et al., 2018; Hayes & Watson, 2013), and the views of fathers are therefore not represented by this research. Given the discrepancy between maternal and paternal differences in family functioning found in the previous meta-analysis (Section 1.4.4), it cannot be assumed that the findings in a predominantly maternal sample here apply to fathers. The overall sample was predominantly white/Caucasian, with high levels of education and an above average socioeconomic status, which also limits the generalisation of the results to diverse populations. Recruitment for future samples should therefore explicitly aim to recruit fathers and more diverse groups of participants, and consideration should be

given to the recruitment strategy to facilitate this. The number of dyads recruited was also lower than hoped for, so consideration of how to facilitate and incentivise whole-family participation in research should also be considered, holding in mind the bias this may introduce as discussed above.

The data collection period for this study was during the Covid-19 pandemic. In the UK this included periods of significant disruption and restriction to family life, including intermittent lockdowns where schools were closed, and families were restricted from leaving the house for non-essential reasons. While family shielding was a non-significant variable in the regression analyses, the reported impact of Covid-19 on participants was variable, with over half of parents reporting that it had affected their own, their child's and their family's wellbeing in a negative way. This trend towards a negative impact but variability between participants is consistent with research on the mental health of autistic adults (Bundy et al., 2022) and on family relations in the general population (Soejima, 2021). A scoping review of the impact on the mental health and wellbeing of caregivers of autistic children identified an increase in demands, stress and mental health symptoms during lockdown measures (Lee et al., 2021). In the general population, a review of family resilience noted the importance of healthy relationships and communication, highlighting the importance of family functioning during this time (Gayatri & Irawaty, 2021). It is therefore likely that overall (but not consistently) participants in this study have experienced poorer mental health and family functioning due to the pandemic, and it remains to be seen whether these findings continue to be applicable to the 'living with covid' era.

2.5.2 Conclusions

In summary, this study was the first to explore family functioning from the subjective experience of both autistic young people and their parents, and to consider the potential association between mental health, autism, and family functioning reports. The findings

suggest the informant's mental health (child depression, and parent anxiety and stress) predict their family functioning reports, while autism characteristics themselves do not. Taken together with the findings of the previous meta-analysis (Section 1.4), these results suggest poorer family functioning in families with autistic children may be partially mediated by their poorer mental health rather than autism itself. This highlights a significant clinical need for mental health support for some (but by no means all) autistic young people and their families, as well as a role for support from services within the meso-system. The discrepancy between parent and child reports of family functioning also highlights a need for researchers and clinicians to include multiple perspectives from family members within their work.

2.6 References for the Empirical Study

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CHAPTER 3 – PRESS RELEASES

3.1 Families of children with neurodevelopmental conditions report that they function together less well than families with neurotypical children

Families of autistic children or children with intellectual disability report that their family functioning is poorer compared to families with neurotypical children, according to a review bringing together twenty-four different studies from across the globe. This finding indicates we need to know more about why autism and intellectual disabilities are linked to poorer outcomes for families. It also highlights that these families may need greater support from services.

Autism and intellectual disability are lifelong conditions that mean someone's brain works differently to other people. Autistic people experience social relationships differently to others and tend to have a smaller range of interests, while people with intellectual disability can find it harder to learn new things. Autism and intellectual disability are different medical diagnoses, but in practice they often co-occur in the same person and there is a lot of overlap between them, including in how they impact the whole family. Previous reviews have found that parents and siblings in these families experience poorer mental health and individual functioning, but no one has considered the experience of the whole family. We were particularly interested in family functioning, which is about what a family's overall environment is like, including their relationships with each other. Families that function well have clear roles, open and straight-forward communication, and regulate their emotions well together. We know that family functioning is important because families with good functioning tend to have happier, healthier family members.

This review involved searching online databases for any studies that had asked neurodiverse/neurodivergent families and neurotypical families to fill out a questionnaire about their family functioning. The results of each study were combined – a total of 24 studies and 14169 participants! The overall size of the difference was small, but small differences over time can have big effects on families. The result was also significant enough that we can be sure the findings are not just due to chance.

We also found that in families with an autistic child compared to a neurotypical child, mothers reported poorer family functioning than fathers did. This is important, as mothers tend to fill out questionnaires in research, and we know they can also experience poor mental health which might affect how they experience their family life. We are therefore recommending that more fathers need to get involved in research, and we also need to make sure we are asking neurodivergent children and young people what they think about their families. We also need further research on the link between family functioning and mental health for neurodiverse/neurodivergent families.

The studies were all checked to see if any of them were overly influential or too different from the result, and this led to two studies being taken out of the analysis. One of these was because different family members (the child or the parent) filled out the questionnaire; this shows us how important it is to ask different family members about their views as their individual perspectives can be quite different. Another study was taken out because it used an adapted questionnaire, and this tells us how important it is to use questionnaires that have been well tested by researchers.

Each paper was rated on how well the study was conducted, so high quality studies had more of an effect on the overall result. We also checked to see if it was likely that any studies were missing (as studies finding big results are more likely to be published than studies finding small results), and this was not found to be a problem.

3.2 Mental health, but not autism itself, is linked to how autistic young people and their parents feel about how their families

Autistic young people and their families say their families work less well together (known as family functioning) when they are experiencing poor mental health, according to a new study. The research also found that there was no link between the young people's autism characteristics and how they said their families were functioning together. This finding challenges a commonly held view that autism itself affects families in a bad way, and instead shifts the focus to ensuring autistic young people and their families have access to timely and tailored mental health support to help both the individuals and their families.

Autism means someone's brain works in a different way from a very young age, and over 1 in 50 people in England are autistic. Autistic young people and their families are more likely than non-autistic (or neurotypical) people to experience poor mental health – in fact, 70-80% of autistic young people experience mental health problems, such as depression and anxiety. Their parents also experience higher levels of depression, anxiety, and stress. It's therefore important for researchers to find out more about mental health in families with an autistic child so services can provide better support for them.

Autism affects the person themselves, as well as their relationships with other people. ‘Family functioning’ is about how well a family works together, including their emotional bonding (cohesion), their family roles and rules (flexibility), how well they can talk to each other (communication) and how happy and fulfilled by each other they are (satisfaction).

We know that family functioning is linked to young people’s mental health, including how difficulties start and keep going, as well as how they respond to treatments like therapy. The researchers previously did a review of studies of family functioning in families with an autistic child, and found that overall, these families say their family functioning is poorer than families with non-autistic children. We wanted to find out why this is the case. Previous researchers have tended to ask parents (usually mothers) about their views on how well their families are functioning. This was the first study to ask autistic young people themselves and their parents to tell us about how their families are doing, and to see how this linked to their autism and their mental health.

We asked 115 autistic young people aged 14-15 and 50 parents of autistic young people about their family functioning in an online survey. Twenty-five of these young people and parents took part together in family dyads so we could understand different perspectives from the same families. We also asked autistic young people about their autism characteristics and their experience of depression, anxiety, and self-harm, and we asked parents about their experience of depression, anxiety, and stress. We used statistics to explore how participants’ answers to different questionnaires were linked.

We found that, on average, autistic young people and their parents say their families function in a balanced or healthy way, but that they are not very satisfied with their family life. This is

important as how we feel about our families is a large part of how good we feel our lives are overall. This might be because of the stage of life these families were at (as adolescence is known to be a tough time for many families!), but we need to find out more about how to help families feel more satisfied.

We found that autistic young people tend to say their family functioning is less healthy than parents say. In our young person and parent dyads, there was a low level of agreement between different family members. This shows how important it is for researchers and clinicians to ask everyone in the family about their functioning, as individuals experience their family life in different ways.

We found that high levels of depression predicted young people saying their families had poor functioning, and that high levels of stress and anxiety predicted the same for their parents. Neither autism nor self-harm were linked to family functioning. This suggests that support for depression, anxiety and stress is key for families – both in terms of individual and family therapies, but it also means families need support to reduce the number of stressful things they are experiencing.

Surprising, parents in this study said they experienced higher levels of very intense emotional bonding (called enmeshment) compared to other people from previous studies who have filled out the same questionnaire. We know that families of autistic children may experience emotional closeness differently to (rather than worse than) families with non-autistic children, and their answers in this study also suggest we need to know more about emotional closeness in families with an autistic child.

It is hoped that this study will prompt other researchers to ask more dyads of autistic young people and their parents to take part in research in the future. It is also hoped that the value of individual mental health support for individuals and families will be recognised, and that future research will explore how families could be supported together by services.

This review and study were funded by Higher Education England as part of the lead researcher's clinical psychology training for the NHS.

Appendix 1: Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) checklist

Supplementary Table 1: The PRISMA checklist, from Moher et al. (2009)

Section/topic	#	Checklist item	Reported on page #
TITLE			
Title	1	Identify the report as a systematic review, meta-analysis, or both.	1
ABSTRACT			
Structured summary	2	Provide a structured summary including, as applicable: background; objectives; data sources; study eligibility criteria, participants, and interventions; study appraisal and synthesis methods; results; limitations; conclusions and implications of key findings; systematic review registration number.	1
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of what is already known.	2-8
Objectives	4	Provide an explicit statement of questions being addressed with reference to participants, interventions, comparisons, outcomes, and study design (PICOS).	8-9
METHODS			
Protocol and registration	5	Indicate if a review protocol exists, if and where it can be accessed (e.g., Web address), and, if available, provide registration information including registration number.	9
Eligibility criteria	6	Specify study characteristics (e.g., PICOS, length of follow-up) and report characteristics (e.g., years considered, language, publication status) used as criteria for eligibility, giving rationale.	9-11
Information sources	7	Describe all information sources (e.g., databases with dates of coverage, contact with study authors to identify additional studies) in the search and date last searched.	9-13
Search	8	Present full electronic search strategy for at least one database, including any limits used, such that it could be repeated.	9-10
Study selection	9	State the process for selecting studies (i.e., screening, eligibility, included in systematic review, and, if applicable, included in the meta-analysis).	9-13
Data collection process	10	Describe method of data extraction from reports (e.g., piloted forms, independently, in duplicate) and any processes for obtaining and confirming data from investigators.	13
Data items	11	List and define all variables for which data were sought (e.g., PICOS, funding sources) and any assumptions and simplifications made.	13-14
Risk of bias in individual studies	12	Describe methods used for assessing risk of bias of individual studies (including specification of whether this was done at the study or outcome level), and how this information is to be used in any data synthesis.	14-18
Summary measures	13	State the principal summary measures (e.g., risk ratio, difference in means).	13
Synthesis of results	14	Describe the methods of handling data and combining results of studies, if done, including measures of consistency (e.g., I2) for each meta-analysis.	18-21

Risk of bias across studies	15	Specify any assessment of risk of bias that may affect the cumulative evidence (e.g., publication bias, selective reporting within studies).	14-18
Additional analyses	16	Describe methods of additional analyses (e.g., sensitivity or subgroup analyses, meta-regression), if done, indicating which were pre-specified.	36
RESULTS			
Study selection	17	Give numbers of studies screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally with a flow diagram.	10-13
Study characteristics	18	For each study, present characteristics for which data were extracted (e.g., study size, PICOS, follow-up period) and provide the citations.	22-27
Risk of bias within studies	19	Present data on risk of bias of each study and, if available, any outcome level assessment (see item 12).	14-18
Results of individual studies	20	For all outcomes considered (benefits or harms), present, for each study: (a) simple summary data for each intervention group (b) effect estimates and confidence intervals, ideally with a forest plot.	21-34
Synthesis of results	21	Present results of each meta-analysis done, including confidence intervals and measures of consistency.	21-36
Risk of bias across studies	22	Present results of any assessment of risk of bias across studies (see Item 15).	14-18
Additional analyses	23	Give results of additional analyses, if done (e.g., sensitivity or subgroup analyses, meta-regression [see Item 16]).	36-38
DISCUSSION			
Summary of evidence	24	Summarize the main findings including the strength of evidence for each main outcome; consider their relevance to key groups (e.g., healthcare providers, users, and policy makers).	38-45
Limitations	25	Discuss limitations at study and outcome level (e.g., risk of bias), and at review-level (e.g., incomplete retrieval of identified research, reporting bias).	38-45
Conclusions	26	Provide a general interpretation of the results in the context of other evidence, and implications for future research.	38-45
FUNDING			
Funding	27	Describe sources of funding for the systematic review and other support (e.g., supply of data); role of funders for the systematic review.	121

Appendix 2: Further detail on the inclusion and exclusion criteria, including measures that were included or excluded

The differing terminology for ID between countries and changes in this terminology over time required full-text screening to identify whether a sample could be included. A frequent example of this was use of the term ‘learning disabilities’ to refer to what would be called specific learning difficulties (e.g., dyslexia) in the UK. Poorly defined samples were also excluded, such as children “at risk for learning disabilities” identified through a record of poor academic achievement in reading or writing (Emam & Abu-Serei, 2014) or children with a “mental disability” (Javadian, 2011). Other neurodevelopmental conditions and mental health disorders were excluded as comparison groups, due to potential for comorbidity and phenotypic overlap (Dewey, 2018; Licari et al., 2019; Morris-Rosendahl & Crocq, 2020) resulting in confounding of groups.

Family functioning measures were also ill-defined, as highlighted in other reviews in this area (Turnbull et al., 2007). Decisions for inclusion were based on a combination of the paper author’s description of their use, measures identified in the Alderfer et al. (2008) review, and through review of the measure by the researcher. A list of included or excluded measures and rationales for exclusion are given below in supplementary tables 1 and 2. A common reason for exclusion was a measure being situated at the level of an individual, family sub-system, or wider mesosystem. Other constructs, such as family quality of life, family empowerment and family life satisfaction, were also excluded due to previous research considering them as separate constructs to family functioning.

Supplementary Table 2: The family functioning measures that were included in the meta-analysis

Measure	Measure details	Interpretation
Bespoke resilience score (Schneider et al., 2019)	Based on the Walsh (2003) theory of family resilience – three domains of family belief systems, organisational patterns, communication/problem-solving	Higher scores = better functioning
Family Adaptability and Cohesion Evaluation Scale (FACES-III, FACES-IV, Chinese version of FACES-III)	Based on the Circumplex model of family functioning (D. H. Olson, 1990) FACES-IV – six scales measuring balanced and unbalanced levels of cohesion, flexibility, and communication FACES-III – two scales measuring adaptation and cohesion	Balanced subscales selected Cohesion subscale selected if combined subscale scores unavailable Higher scores = better functioning
Family Assessment Measure (FAM-III)	Based on the Process Model of Family Functioning (Skinner et al., 2000) 3 forms – general (whole family), dyadic, self-rating 6 subscales – task accomplishment, communication/affective expression, role performance, affective involvement, control, values, and norms	General (whole family) form selected Higher scores = worse functioning
Family Environment Scale (FES)	Social-environmental characteristics (Moos & Moos, 2009) 3 forms – real, ideal, expectation 10 subscales over 3 conceptual domains – relationships, personal growth or goal orientation, and system maintenance	Real form selected Cohesion subscale of the relationships domain selected Higher scores = better functioning

Measure	Measure details	Interpretation
Family Orientated Personal Evaluation Scale (F-COPES)	Based on the Resiliency Model of Family Adjustment and Adaptation (McCubbin & McCubbin, 1996) 5 factors – acquiring social support, reframing, seeking spiritual support, mobilising to acquire and accept help, and passive appraisal	Higher scores = better functioning
Family Relations Scale (FARS)	Family systems and family pathology (Höök & Cederblad, 1992) 5 subscales – attribution, joint interest, isolation, chaos, intrusion	Higher scores = worse functioning
McMaster Family Assessment Device (FAD)	Based on the McMaster Model of Family Functioning (Epstein et al., 1983) 7 scales – general functioning, affective involvement, affective responsiveness, behavioural control, communication, problem solving and roles	General functioning subscale used where whole measure unavailable Higher scores = worse functioning
Pediatric Quality of Life Family Impact Module: The Family Functioning Scale (PedsQL^{TM})	Health-related quality of life (Varni et al., 2004) 2 domains - family daily activities and family relationships	Higher scores = better functioning
Systemic Clinical Outcome and Routine Evaluation (SCORE-28)	Systemic family therapy outcome (Cahill et al., 2010) 3 factors – family strengths, difficulties, and communication	Higher scores = worse functioning

Supplementary Table 3: The family functioning measures that were excluded at full-text screening

Measure	Rationale for exclusion
The Beach Centre Family Quality of Life Scale (Hoffman et al., 2006) Family Quality of Life Survey (Brown et al., 2006) Family Quality of Life People Survey (Verdugo et al., 2005) KINDL (Ravens-Sieberer & Bullinger, 2000) The Family Member Well-being Index (H. I. McCubbin & Patterson, 1996) Family Empowerment Scale (Koren et al., 1992) The Kansas Family Life Satisfaction Scale (Schumm et al., 1986) Family Adjustment Measure (Daire et al., 2014) Family Support Scale (Dunst et al., 2007)	Measures mesosystem interactions beyond the family microsystem. Considered different constructs within the literature: Family quality of life (R. I. Brown & Brown, 2014; Summers et al., 2005; Turnbull et al., 2007) Family empowerment (Cunningham et al., 1999) Family life satisfaction (Szcześniak & Tułecka, 2020)
Parenting and Family Adjustment Scales (Sanders & Morawska, 2010) The Life Management Survey (Scorgie et al., 1997)	Primarily measures parent-level functioning
Family Impact Questionnaire (Donenberg & Baker, 1993) The Questionnaire on Resources and Stress (Holroyd, 1974) Impact on Family Scale (Stein & Riessman, 1980) Family Life Impairment Scale (Mian et al., 2018) Family Management Measure (Knafl et al., 2012)	Measures the impact of the child on the family system rather than the family microsystem as a whole
Family Stress and Coping Interview (Nachshen et al., 2003)	Measures stressors

Measure	Rationale for exclusion
The Family Relations Test (Anthony & Bene, 1957)	Primarily measure dyadic or triadic relationships within the family microsystem
The Kvebaek Family Sculpture Technique (Cromwell et al., 1980)	
The Lausanne Trilogue Play (Corboz-Warnery et al., 1993)	
Family Function section of the 2007 National Survey of Children's Health (*Data Resource Center for Child and Adolescent Health.*, 2007)	
Family Network Method (Widmer et al., 2013)	
Bespoke variables –e.g. family status, ill treatment, adoption, 'unfavourable' family situation as judged by a psychologist, challenged living arrangements, severe family illness, family problem-solving discussions, intellectual disability in other family members, child deaths, social class, disharmony/disruption, changes in the household, domestic violence, disturbance to daily life, discrimination, concerns about the future, child legitimacy, long-term separations, emotional support, educational stimulation	Not primarily measuring a psychological construct of functioning within the family microsystem

Appendix 3: Letter from ethics committee providing permission to proceed with the research

From: Research Support Services
Sent: 01 September 2020 15:50
Subject: Application for Ethical Review ERN_20-0819

Dear X,

Re: "Wellbeing in Autistic Young People and their Families"
Application for Ethical Review ERN_20-0819

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

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

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

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

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

Kind regards

Research Ethics Manager
Research Support Group

Appendix 4: The promotional materials, information sheets, and consent forms

Are you an autistic young person or a parent/caregiver of an autistic young person?

We invite you to take part in a new autism research project. Our research is interested in the wellbeing of autistic young people between the ages of 14-25 years. This project will focus on autistic young people who *do not* have a co-occurring diagnosis of intellectual/learning disability. We are also interested in family well-being and so also invite their parents/caregivers to participate. For both the autistic young people taking part and their parent/caregiver, participating in this study entails completing a short survey in either an online or postal format. Participants will be reimbursed for their participation at the rate of £10 per hour.

If you are interested in taking part or would like more information, please contact

Wellbeing in Autistic Young People and their Families

Parent/Caregiver Information Sheet

Please read this information carefully before deciding whether you/your child want to take part in the study. If you have any questions about the research, or need a verbal explanation of the study, please contact… When you are happy that you have all the information required to decide whether to participate in the study, please inform the research team. A member of the research team with then send you a link to, or post, the relevant consent forms, and survey. Please ensure you complete the consents forms before participating in the survey.

What is the research about and who can take part?

Adolescence and young adulthood are a time of huge physical, social, and emotional change for young people. Autistic young people likely have a very different experience of this age compared to their neurotypical peers, however very little is known about autistic young people, and in particular their well-being and mental health through these years. Therefore, this study aims to learn more about the wellbeing of autistic young people, in order to develop our understanding of autism across this age period. We are also interested in family well-being as experienced by the parents/caregivers of autistic young people.

We are looking for autistic young people aged between 14-25 years to participate in this research. Additionally, we are also asking you as their parent/caregiver to participate. This is necessary for young people under 16 but is optional for those over 16. You have been approached about taking part in the study because a professional (E.g., Educational or support staff) or yourself has indicated that this research might be applicable for your child/yourself. Approximately 250 autistic young people and their parents/caregivers will be participating in this study.

What will participation involve?

Your child with complete the young person section of the survey, and if participating you will complete the parent/caregiver section.

Researchers will send you/your child a link to complete an online survey, however if preferred a hard copy can be sent in the post. The young person survey should take approximately 30 minutes to complete. The survey will primarily focus on their autism and their wellbeing, including questions on mental health, mood and self-harm. Participants are not required to answer any questions they feel uncomfortable about and can take breaks from the survey as/when needed. Whilst some questions may be of a personal and sensitive nature, we do not anticipate that they will cause any distress to participants; however, your child may wish to talk about it with you afterwards.

We are also interested in family well-being and are therefore asking parents/caregivers to participate in a separate section of the study. The parent/caregiver survey take approximately 30 minutes to complete. Again, you will not be required to answer any questions that make you uncomfortable. If your child is under 16, you will both need to participate. For parents/caregivers of young people over 16, you do have to participate

in the survey for your child to participate in the study, however it would be greatly appreciated if you could.

Are there any potential risks of participating?

There is a very small chance that answering questions about themselves may cause some distress for your child. However, past research tells us that any distress is usually short-lived, and people feel the benefits of contributing to research outweigh this. The survey has been designed to minimise any distress but we understand that everyone's experiences are different. Therefore, your child will be made aware prior to participating that they do not have to answer all questions and that they can take a break/withdraw at any point during the survey. There will be no consequences for not fully completing the survey or missing out any questions. Additionally, upon completion of the questionnaire, details will be provided of organisations/services which might be able to offer support to both you and your child. You and your child will also be able to contact the research team using the contact details provided. If your child tells us anything that suggests they are at high or imminent risk of harm, we have safeguarding policies in place that mean this information may be passed on to you or another appropriate person.

What are the benefits of participating?

Participants will be reimbursed for their participation at the rate of £10 per hour. Upon study completion, we will provide all participants with a short summary of the study and its findings. Additionally, this study aims to help us to find out more about the lives of autistic people and the difficulties they face. By taking part in this study, you and your child might help us to improve the lives of autistic people and their families in the future.

Where will data be stored?

All data collected will be kept in locked or password protected storage at the University of Birmingham. All information gathered about you and your child will be stored separately from any information that would allow someone to identify you or your child (this is known as personal identifying information, E.g., full names, address, contact details). Your personal identifying information will be stored in a locked space at the University of Birmingham and only members of our research team will have access to it. We will only be able to trace the information we have collected about you and your child back to you using a special reference number which we will store in a password protected database held at the University of Birmingham. Only members of our research team will have access to that database. Personal identifying information will be treated as strictly confidential and handled in accordance with the provisions of the General Data Protection Regulation 2018 (GDPR) and the Data Protection Act 2018.

What will happen to the data afterwards?

Ten years after the end of the study we will destroy all data collected during the study. At the end of the study, any personal details will be destroyed unless you tell us otherwise. This means that we would no longer be able to trace the results of your/your child's questionnaires back to you.

Can I withdraw my child/myself from the study?

Even after consent has been granted, you are free to request for both you and your child to be withdrawn from the study without providing us with a reason. Upon request, we will also destroy any data we have collected on you/your child. There will be no consequences for withdrawing, and you will still receive any agreed feedback. You will have up to 1 month after participation to withdraw. After this time, your personal information will no longer be linked to your data, meaning we will no longer be able to trace your assessment results/interview back and withdraw you from the study.

What happens next?

After reading all the information, carefully considering it, and asking any questions you want answered, please contact the research team using the contact details provided if you/your child are interested in participating in the study. We will then either send a link to an online survey or send a hard copy in the post. The first section of the survey will be consent forms. You need to consent on behalf of your child if they are under 16 years, and yourself if participating in the parent/caregiver portion, to take part in the study. You cannot participate until consent has been obtained.

Where can I get more information from?

If you have any questions about the current study, or would like any more information please contact a member of the research team:

Wellbeing in Autistic Young People and their Families

Young Person Information Sheet

Please read this information carefully before deciding whether you want to take part in the study. If you have any questions about the research, or need a verbal explanation of the study, please contact… When you are happy that you have all the information you need to decide whether or not to take part in the study, please inform the research team. A member of the research team with then send you a link to, or post, the relevant consent forms and survey. Please ensure you complete the consents forms before participating in the survey.

What is a research study?

A research study is a way to find out new information about people or things. Research is a good way of finding out new or better ways to help people.

What is the research about and why am I being asked to take part in this study?

Adolescence and young adulthood are a time of huge physical, social and emotional change for young people. Autistic young people likely have a very different experience of this age compared to their neurotypical peers, however very little is known about autistic young people, and in particular their well-being and mental health through these years. Therefore, this study aims to learn more about the wellbeing of autistic young people, in order to develop our understanding of autism across this age period. We are also interested in family well-being as experienced by the parents/caregivers of autistic young people.

To research this we are we are looking for autistic young people aged 14-25 years to take part in this research. Additionally, we are also asking their parents/caregivers to participate. If you are under 16, your parent/caregiver will need to take part. If you are over 16 your parent/caregiver does not have to take part for you to participate, but we would really appreciate it if they did. We are asking you to be in the study because either you, your parent/caregiver, or a professional (E.g., Educational or support staff) who knows you has told us that you might fit the description of the people we want to take part. About 250 autistic young people and their parents/caregivers will be in this study.

What will I have to do if I want to take part in the study?

A member of the research team will send you a link to take part in the online survey, however if preferred a hard copy can be sent through the post. The survey should take approximately 30 minutes to complete. The survey will primarily focus on your autism and well-being, including questions about your feelings, mood and self-harm. You do not have to answer any questions that might make you uncomfortable and you can take breaks from the survey as/when needed. Whilst some questions may be of a personal and sensitive nature we do not anticipate that they will upset you; however, you may wish to talk about it with a parent, caregiver or significant adult afterwards.

We are also interested in family well-being and relationships, and are therefore asking parents/caregivers to participate in a separate section of the study.

What are the risks of taking part in the study?
There is a very small chance that answering questions about yourself, your autism and your feelings may cause you to feel slightly upset. However other research participants say this is usually short-lived and that overall they are glad to take part in research. The survey has been designed to minimise any distress but we understand that everyone's experiences are different. Therefore, you do not have to answer all questions and can take a break/stop at any point during the survey. There will be no consequences for not fully completing the survey or missing out any questions. Additionally, upon completion of the survey, details will be provided about various organisations you may wish to go to for help. You will also be able to contact the research team using the contact details provided. If you tell us anything that makes us think you might be at high or immediate risk of hurting yourself or someone else, we may need to do something to keep you safe. We would try to speak to you first about what you want to happen in this situation, however it might mean we need to tell a parent/caregiver.

Will I benefit from taking part in the study?
Participants will be paid for their participation at the rate of £10 per hour Upon study completion, we will provide all participants with a short summary of the study and its findings. Additionally, this study will help us to find out more about the lives of autistic people and the difficulties that people they may face. By taking part in this study, you might help us to improve the lives of autistic people the future.
Do my parents/caregivers know about this study?

If you are under 16 years of age, we will need to provide them with information about the study and get permission from them for you to take part. You can talk this over with them before you decide whether you want to be involved. Additionally, we are asking parents/caregivers to also complete their own questionnaires., so we will likely contact them regarding that part of the study. However, if you are over 16, you can take part without asking your parent/caregiver and it is your choice whether they complete a survey themselves.

How will the information collected about me be stored?

The data collected will be kept in locked or password protected storage at the University of Birmingham. All information gathered about you will be stored separately from any information that would allow someone to know it was you taking part in the research (this is known as personal identifying information, E.g., your full name, your address, your contact details). This information will be stored in a locked space at the University of Birmingham and only members of our research team will be able to look at it. We will only be able to trace the information we have collected about you back to you using a special reference number which we will store in a password protected database held at the University of Birmingham. Only members of our research team will have access to that database. Personal identifying information will be treated as strictly confidential and handled in accordance with the law (General Data Protection Regulation 2018 (GDPR) and the Data Protection Act 2018).

What will happen to this information after the study?
Ten years after the end of the study we will destroy all the information collected during the research.

At the end of the study, any personal details will be destroyed unless you tell us otherwise. This means that we would no longer be able to trace the results of your questionnaires back to you.

Do I have to take part and what if I decide later that I no longer want to take part?

You do not have to be in the study and no one will be upset if you don't want to. You do not even have to give us a reason for not taking part.

Even after you have agreed to take part, you can request to be withdrawn from the study and for your research data to be destroyed. You will have up to 1 month after taking part to tell us you want to withdraw from the study. There will be no consequences to you or your family for withdrawing, and you will still receive any agreed feedback. After this, your personal information will no longer be linked to any information collected about you during the study. This means that we would no longer be able to trace any information collected back to you and withdraw you from the study.

What happens next?

After reading all the information and asking any questions you want answered, please contact the research team using the contact details on this information sheet if you want to take part in the study. We will then need to take your consent or have a parent/caregiver consent for you to be part of the study. Consent just means you telling us that you understand the study and what we are asking you to do as part of it, and that you agree to take part. You cannot take part until you have consented.

Where can I get more information from?

If you have any questions about the current study, or would like any more information please contact a member of the research team:

CONSENT FORM A (Consent provided by parents/caregivers on behalf of participants below 16 years of age)

Title of Project: Wellbeing in Autistic Young People and their Families

1. I have read and understood the information sheet (Version/date) provided for this study explaining what is expected of my child. I have asked any questions I wanted to ask and had these answered satisfactorily. ☐
2. I understand that my child's participation in the study is voluntary and that I am free to withdraw my child at any time without giving any reason. This will not impact any feedback I receive. ☐
3. I understand that relevant official bodies may access the data obtained in this study to check the quality of the research. I give permission for these individuals to have access to my child's records for these purposes. ☐
4. I understand that my child is being asked to take part in a survey, as described in the information sheet, that will include questions about themselves, their autism and their wellbeing and mental health, and am happy for this to happen. ☐
5. I understand that my child's data will be securely stored at the University of Birmingham in accordance with University policies and the provisions of the General Data Protection Regulation 2018 (GDPR) and the Data Protection Act 2018. ☐
6. I understand that my child's data will be kept for a maximum of 10 years, in line with the University of Birmingham policies, and after which it will be destroyed. ☐
7. As a parent or legal caregiver, I agree for my child to take part in the above study. ☐

The next question is optional and will not affect your participation in the research study.

9. I agree to being contacted regarding future studies taking place within the team that may be relevant to myself or my child. ☐

CONSENT FORM B (Parent and Young Person Participants aged over 16 years)

Title of Project: Wellbeing in Autistic Young People and their Families

1. I have read and understood the information sheet (Version/date) provided for this study explaining what is expected of me. I have asked any questions I wanted to ask and had these answered satisfactorily. ☐
2. I understand that my participation in the study is voluntary and that I am free to withdraw at any time without giving any reason. This will not impact any feedback I receive. ☐
3. I understand that relevant official bodies may access the information collected in this study to check the quality of the research. I give permission for these people to have access to my records for these purposes. ☐
4. I understand I am asked to take part in a survey, as described in the information sheet, that will include questions about myself, my autism and my wellbeing and mental health, and am happy for this to happen. ☐
5. I understand that my data will be securely stored at the University of Birmingham in accordance with University policies and the provisions of the General Data Protection Regulation 2018 (GDPR) and the Data Protection Act 2018.
6. I understand that my information will be kept for a maximum of 10 years, in line with the University of Birmingham policies, and after which it will be destroyed. ☐
7. I agree to take part in the above study. ☐

The next question is optional and will not affect your participation in the research study

8. I agree to being contacted regarding future studying taking place within the team that may be relevant to me. ☐

ASSENT FORM (Participants aged below 16 years)

Title of Project: Wellbeing in Autistic Young People and their Families

1. I have read, or had read to me, the study information and I understand what the research is about and what is expected of me. I have been able to ask all the questions I want to and am happy with how they have been answered ☐
2. I know I do not have to take part in this study, that I can leave at any time without a reason, and that I will not be in trouble if I do leave. ☐
3. I know that people within the research team may need to look at my information sometimes, and I am okay with this. ☐
4. I know I am being asked to take part in a survey that will ask questions about myself and I am happy to do this. ☐
5. I know that my information will be securely stored at the University of Birmingham in accordance with University policies and the law. ☐
6. I understand that my information will be kept for up to 10 years, in line with the University of Birmingham policies, and after which it will be destroyed. ☐
7. I agree to take part in the above study. ☐

The next question is optional and will not affect you taking part in the study.

8. There is a chance that during or after the study, the researchers might find other questions needing answers that require future studies. I would like to hear about these future studies. This does not mean that I must take part in a new study, just that the researchers will let me know about it. ☐

Appendix 5: The demographic questionnaires

Background Questionnaire (Young Person participant on behalf of themselves)

Please complete the following questions about yourself.

1. **Date of Birth**: ___/___/_____ **Age**:_____
2. **Gender** (please tick appropriate):
   - ☐ Female (including transgender female)
   - ☐ Male (including transgender men)
   - ☐ Prefer not to say
   - ☐ Prefer to self-describe as (please specify E.g. non-binary, gender-fluid, agender, Etc.,…) _____)
3. **Do you identify as transgender or have a transgender history?**
   - ☐ Yes
   - ☐ No
   - ☐ Prefer not to say
4. **Ethnicity**
   - ☐ White/Caucasian
   - ☐ Asian/Asian British
   - ☐ Black/African/Caribbean/Black British
   - ☐ Mixed/Multiple ethnic groups
   - ☐ Other (please specify _____)
5. **Which of the following have you been diagnosed with?** (please tick appropriate):
   - ☐ Autism Spectrum Disorder
   - ☐ Autism Spectrum Condition
   - ☐ Autism
   - ☐ Asperger Syndrome
   - ☐ Pervasive Developmental Disorder
   - ☐ Pervasive Developmental Disorder Not Otherwise Specified
   - ☐ Pathological Demand Avoidance
   - ☐ Other (please specify_____)
6. **Age of autism diagnosis** (Year and months): _____
7. **Who did you receive a diagnosis of autism from?** (Please tick appropriate)
   - ☐ Pediatrician
   - ☐ Psychiatrist
   - ☐ Clinical Psychologist
   - ☐ Speech and Language Therapist
   - ☐ Specialist autism team
   - ☐ Other (please specify_____)

8. **Have you ever received a diagnosis of any of the following?** (Please tick appropriate)
   - ☐ Learning disability
   - ☐ Intellectual disability
   - ☐ Global developmental delay
   - ☐ Unsure/prefer not to say.
9. **Which of the best describes your employment status?** (Please tick appropriate)
   - ☐ Employed full-time
   - ☐ Employed part-time
   - ☐ Self-employed
   - ☐ Unemployed
   - ☐ Student (school/college/university)
   - ☐ Prefer not to say
   - ☐ Other (please specify _____
10. **Which of the following do you/did you attend?**
   - ☐ Mainstream middle/high/secondary school
   - ☐ Special Educational Needs (SEN) unit in a mainstream school
   - ☐ Special Educational Needs (SEN) school
   - ☐ Home-school
   - ☐ Other (please specify _____
11. **What is your highest level of education?**
   - ☐ No qualifications
   - ☐ GCSE's (or equivalent)
   - ☐ A-Levels (or college equivalent)
   - ☐ Undergraduate degree (or equivalent)
   - ☐ Postgraduate degree (or equivalent)
   - ☐ Unsure/Prefer not to say
   - ☐ Other (please specify) _____
12. **What is your relationship status?** (Please tick appropriate)
   - ☐ Single
   - ☐ Married
   - ☐ Cohabiting
   - ☐ Divorced
   - ☐ Widowed
   - ☐ Prefer not to say.

13. **What is your residential status?** (Please tick appropriate)
    - ☐ Living with parents
    - ☐ Living with another caregiver who is not a parent
    - ☐ Living in a supported living facility
    - ☐ Living with friends
    - ☐ Living with a partner or spouse
    - ☐ Living in shared accommodation (not with friends)
    - ☐ Living on own.
14. **Is your household income:** (Please tick appropriate)
    - ☐ Above the UK median (> £29,400)
    - ☐ Below the UK median (< £29,400)
    - ☐ Unsure/Prefer not to say
15. **Do you have any physical or mental health diagnoses?** (Please tick appropriate)
    - ☐ Yes (Please specify all)
    - ☐ No
    - ☐ Prefer not to say
16. **Does anyone else in your immediate family (i.e., a parent or sibling) have a diagnosis of autism?** (Please tick appropriate)
    - ☐ Yes (Please specify all)
    - ☐ No
    - ☐ Prefer not to say
17. **Does anyone in your immediate family (i.e., a parent or sibling) have any physical or mental health diagnoses?** (Please tick appropriate)
    - ☐ Yes (Please specify all)
    - ☐ No
    - ☐ Prefer not to say

Background Questionnaire (Parent participant)

Please complete the following questions about yourself.

1. **What is your age?**

   _____

2. **What is your gender?**
   - ☐ Female
   - ☐ Male
   - ☐ Prefer not to say
   - ☐ Other (please specify)
3. **What is your ethnicity?**
   - ☐ White/Caucasian
   - ☐ Asian/Asian British
   - ☐ Black/African/Caribbean/Black British
   - ☐ Mixed/Multiple ethnic groups
   - ☐ Prefer not to say
   - ☐ Other (please specify)
4. **What is your relationship to the young person involved in this study?**
   - ☐ Parent
   - ☐ Stepparent
   - ☐ Grandparent
   - ☐ Adopted parent
   - ☐ Foster parent
   - ☐ Other (please specify) _____
5. **Do you live in the same household as the young person involved in this study?**
   - ☐ Yes – all of the time
   - ☐ Yes – some of the time
   - ☐ No
6. **What is your relationship status?**
   - ☐ Single
   - ☐ Married
   - ☐ Cohabiting
   - ☐ In a relationship but not cohabiting
   - ☐ Divorced
   - ☐ Widowed
   - ☐ Prefer not to say
   - ☐ Other (please specify)
7. **What is your highest level of education?**
   - ☐ No qualifications
   - ☐ GCSE’s / O-Levels (or equivalent)
   - ☐ College / A-Levels (or equivalent)
   - ☐ Undergraduate degree (or equivalent)
   - ☐ Postgraduate degree (or equivalent)
   - ☐ Prefer not to say
   - ☐ Other (please specify)

8. **What is your employment status?**
   - ☐ Employed full-time
   - ☐ Employed part-time
   - ☐ Self-employed
   - ☐ Not employed
   - ☐ Retired
   - ☐ Student (school/college/university)
   - ☐ Prefer not to say
   - ☐ Other (please specify)
9. **Has your employment status changed due to the Covid-19 pandemic?**
   - ☐ Yes
   - ☐ No

If yes, what was your employment status before the Covid-19 pandemic?

10. **What is your household income?**
    - ☐ Above the UK median (more than £29,400)
    - ☐ Below the UK median (less than £29,400)
    - ☐ Prefer not to say
11. **Has your household income changed due to the Covid-19 pandemic?**
    - ☐ Yes
    - ☐ No

If yes, what was your household income before the Covid-19 pandemic?

12. **Do you have any physical health conditions?**
    - ☐ Yes (please specify)
    - ☐ No
    - ☐ Prefer not to say
13. **Do you have any mental health conditions?**
    - ☐ Yes (please specify)
    - ☐ No
    - ☐ Prefer not to say
14. **Do you have any developmental conditions?**
    - ☐ An autism spectrum condition
    - ☐ A learning / intellectual disability
    - ☐ Prefer not to say
    - ☐ Other (please specify)

_____

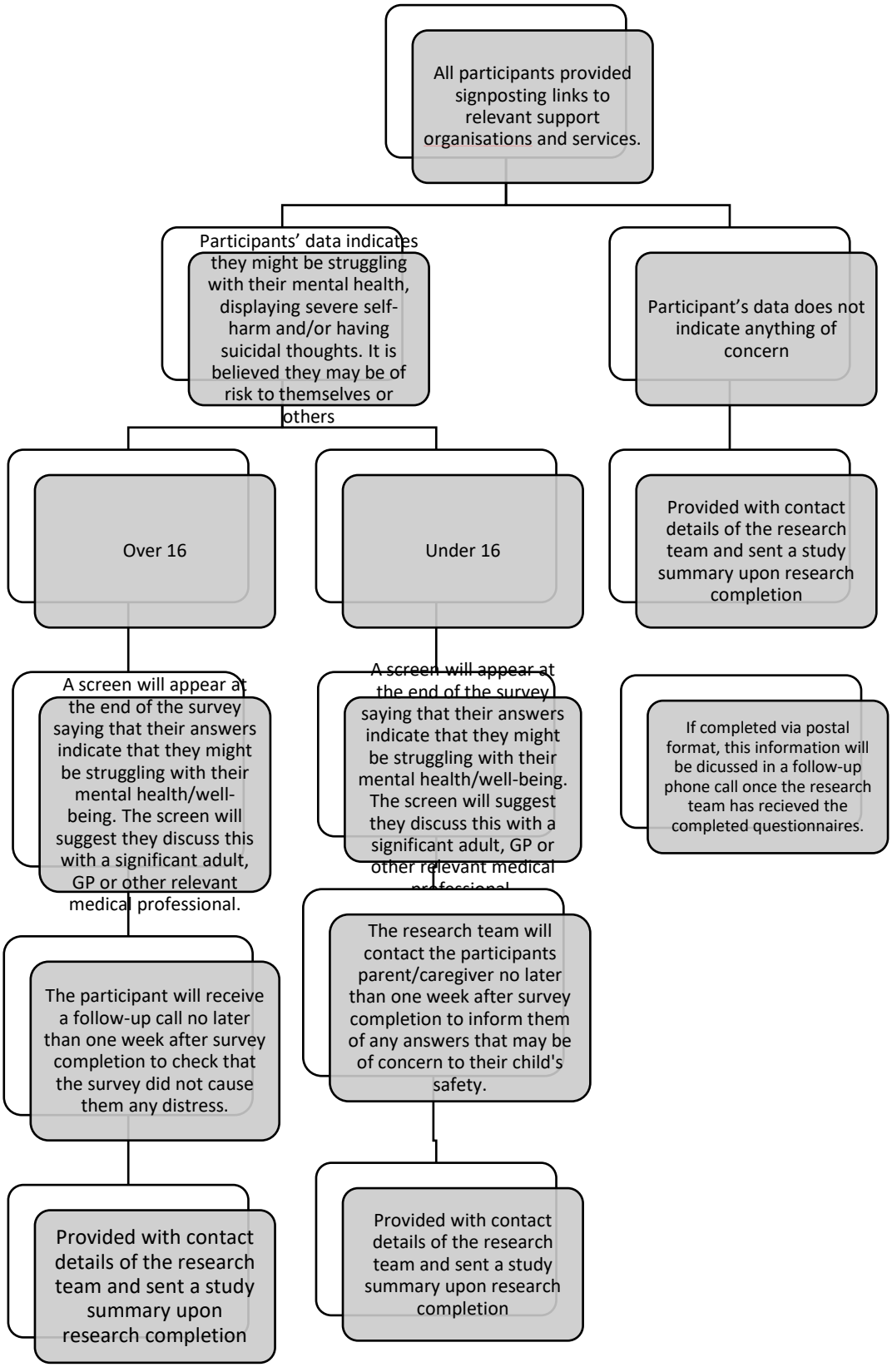
Please complete the following questions about the young person involved in the study.

15. **At what age did the young person receive their diagnosis of an autism spectrum condition?**

_____

16. **Does the young person have a diagnosis of a learning or intellectual disability?**
    - ☐ Yes
    - ☐ No
    - ☐ Prefer not to say
17. **Has your family received any support from services around the young person's autism spectrum condition?**
    - ☐ Yes (please specify)
    - ☐ No
    - ☐ Prefer not to say

Appendix 6: The risk-management protocol



Appendix 7: The tests of normality and Cronbach's alpha for the FACES-IV

Supplementary Table: The Shapiro-Wilk tests of normality for all measures

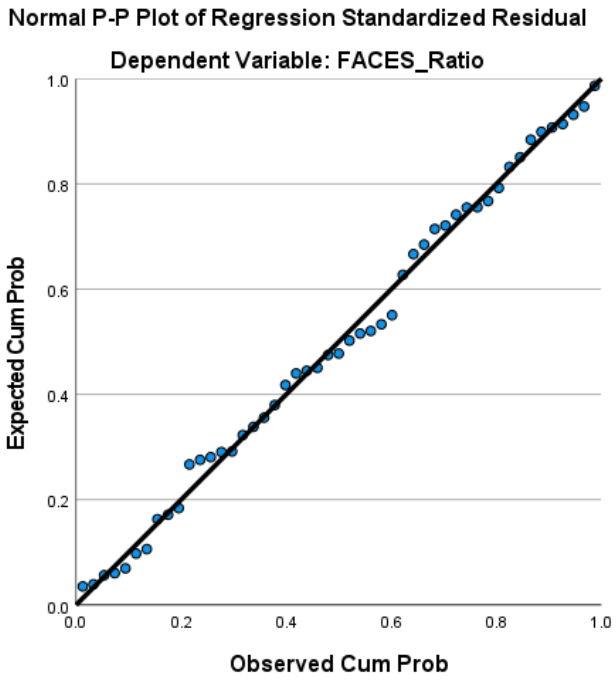
Measure		Shapiro-Wilk			Shapiro-Wilk		
		Statistic	**df**	**Sig**	**Statistic**	**df**	**Sig**
		Whole parent sample (N=50)			*Dyad parent sample (N=25)*		
FACES-IV	Cohesion Raw Score	0.94	50	.03*	-	-	-
	Flexibility Raw Score	0.93	50	<.01*	-	-	-
	Disengaged Raw Score	0.92	50	<.01*	-	-	-
	Enmeshed Raw Score	0.96	50	007	-	-	-
	Rigid Raw Score	0.98	50	.48	-	-	-
	Chaotic Raw Score	0.91	50	<.01*	-	-	-
	Cohesion Ratio	0.96	50	.11	0.96	25	.36
	Flexibility Ratio	0.98	50	.56	0.96	25	.38
	Combined Ratio	0.97	50	.25	0.93	25	.08
FCS Total Score		0.95	50	.03*	0.94	25	.11
FSS Total Score		0.97	50	.22	0.86	25	<.01*
PHQ-9 Total Score		0.90	49	<.001*	0.91	25	.04*
GAD-7 Total Score		0.92	50	<.01*	0.91	25	.03*
PSS Total Score		0.98	49	.64	0.94	25	.17
		Whole young person sample (N=115)			*Dyad young person sample (N=25)*		
FACES-IV	Cohesion Raw Score	0.94	115	<.001*	-	-	-
	Flexibility Raw Score	0.97	115	.02*	-	-	-
	Disengaged Raw Score	0.97	115	.02*	-	-	-
	Enmeshed Raw Score	0.96	115	<.01*	-	-	-
	Rigid Raw Score	0.96	115	<.01*	-	-	-
	Chaotic Raw Score	0.99	115	.27	-	-	-
	Cohesion Ratio	0.93	115	<.001*	0.97	25	.71
	Flexibility Ratio	0.93	115	<.001*	0.96	25	.34
	Combined Ratio	0.95	115	<.001*	0.95	25	.28
FCS Total Score		0.98	115	.06	0.94	25	.14
FSS Total Score		0.97	115	.02*	0.94	25	.17
AQ Total Score		0.97	110	.02*	0.97	24	.58
BDI Total Score		0.96	112	<.001*	0.86	25	<.01*
ASA-A Total Score (age 16-25)		0.98	103	.05*	-	-	-
ASC-ASD Total Score (age 14-15)		0.71	7	<0.01*	-	-	-
ASA-A/ASC-ASD Standardised Scores		0.98	110	0.17	0.953	24	0.31

*Significant p-value with p=.05 as threshold

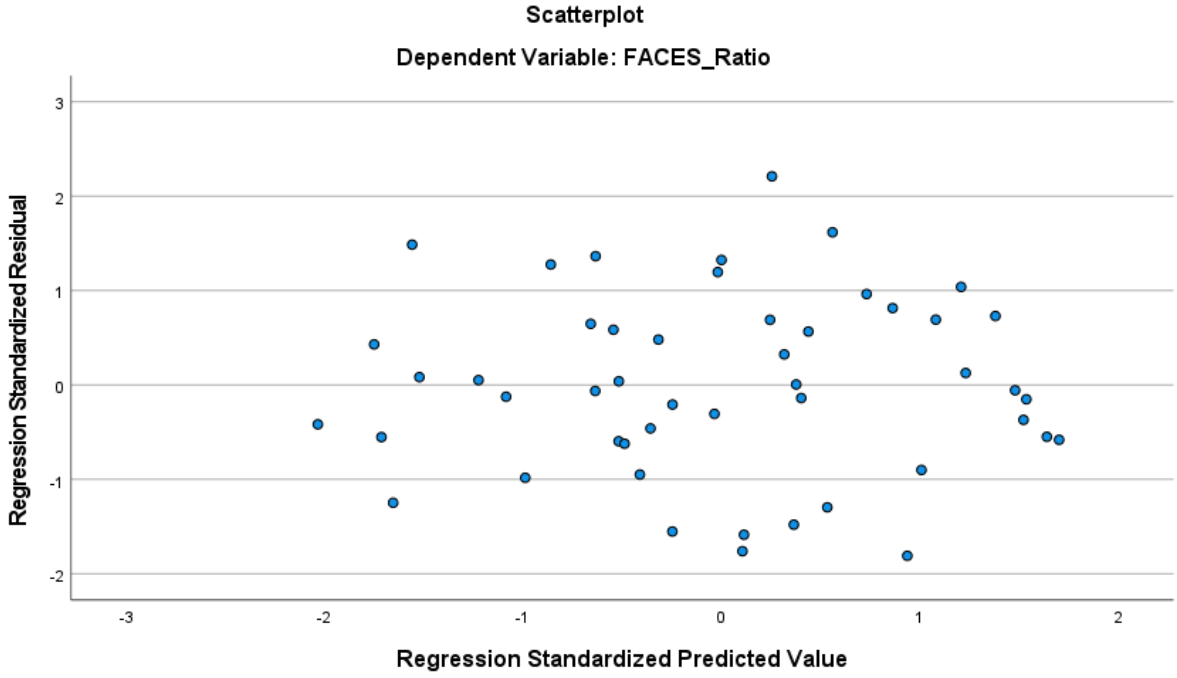
Supplementary table: The Cronbach's alpha for the FACES-IV package

Subscale	Parent Sample	Young Adult Sample
Balanced Cohesion	0.73	0.90
Balanced Flexibility	0.71	0.75
Disengaged	0.77	0.77
Enmeshed	0.64	0.74
Rigid	0.79	0.73
Chaotic	0.82	0.78
FCS	0.83	0.90
FSS	0.89	0.89

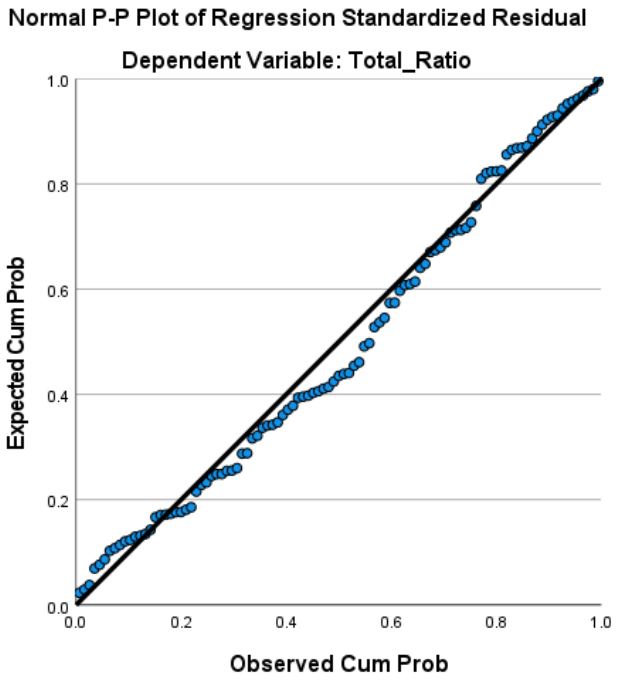
Appendix 8: Testing the Assumptions of the Regression Lines



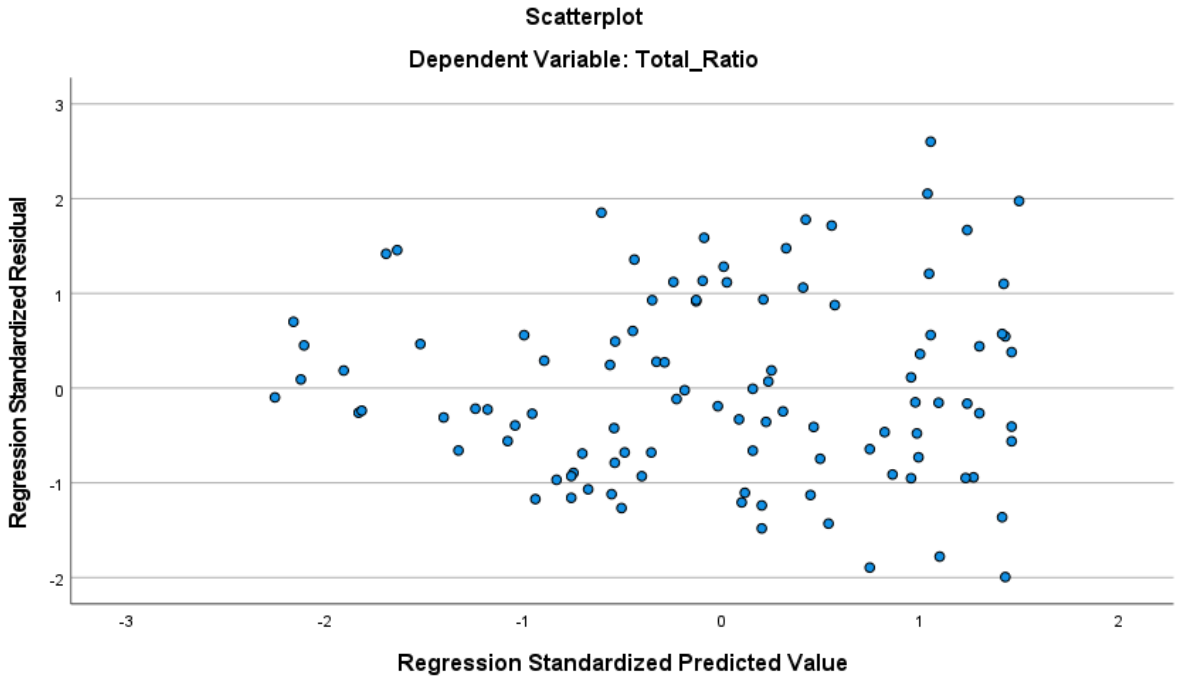
Supplementary figure 1: The normal P-P plot of regression standardised residuals for the parent-reported total family functioning ratio regression model



Supplementary figure 2: The scatterplot of the regression standardised residuals against the predicted values for the parent-reported total family functioning regression model



Supplementary figure 3: The normal P-P plot of regression standardised residuals for the young person-reported total family functioning ratio regression model



Supplementary figure 4: The scatterplot of the regression standardised residuals against the predicted values for the parent-reported total family functioning regression model