The ASD-Plus Study:

Co-existing emotional and behavioural conditions in children with Autism Spectrum Disorder – frequency, severity and correlates from two large UK databases

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Abstract

Aim

The aims of this study were to investigate

1. The frequency, severity, associations and impact on the family of co-existing emotional and behavioural conditions in children with ASD
2. The role of unmet parent needs on the impact on families of children with ASD

Methods

The study used a cross-sectional observational design. Parents of children with ASD were recruited from two large ASD research databases. Standardised parent-report questionnaires were identified following a literature review, and used to measure co-existing conditions. A schedule was developed to estimate parents' unmet needs regarding their child's co-existing conditions and services received.

Main findings

80% of parents reported children had moderate-severe impairment in at least one co-morbid psychopathology; 42% reported moderate-severe impairment in three or more types of psychopathology. Co-morbid psychopathology was prevalent in children with ASD irrespective of age, gender, type of diagnosis, language level and school type. However, younger age, autism diagnosis and lower language ability significantly predicted internalizing problem behaviours. Age relationships with problems in sensory processing, eating, sleep and anxiety were found as hypothesised from the literature.

Co-morbid psychopathology, internalising and externalising problem behaviours, special schooling and having more than one child with ASD were associated with greater impact on families. Over two thirds of parents reported at least one unmet need; 41% had three or more unmet needs for support with co-existing emotional and behavioural conditions. The total unmet needs mediated the relationship between co-morbid psychopathology and the impact

on the family. The unmet needs mediated the impact on the family of externalising behaviours, but not of internalising behaviours.

Conclusions

Co-existing conditions are common in children with ASD and have significant impact on the family. Clinicians should look ‘beyond the diagnosis of ASD’ regarding what impacts families and whether additional support and services might ameliorate this impact.

Dedication

This thesis is dedicated to Regi, Reuben and Rhea Kurien.

Acknowledgements

The course of this thesis has been a tough, but exhilarating and beautiful journey. This journey would not have been possible without the help and support of many.

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Author’s declaration

I declare that the work in this thesis was carried out in accordance with the requirements of the University’s Regulations and Code of Practice for Research Degree Programmes for award of an academic degree and that the work described is my own work. I also declare that this thesis has not been submitted for any other academic award.

Signed…Beena……..

Date…25/2/2015……...

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Abbreviations

ABC – Aberrant Behaviour Checklist

ABPQ – Atypical Behaviour Pattern Questionnaire

AD – Anxiety Disorder

ADHD – Attention Deficit Hyperactivity Disorder

APSI – Autism Parenting Stress Index

AS – Asperger Syndrome

ASD – Autism Spectrum Disorders

ASD-CA – Autism Spectrum Disorders – Comorbidity for Adults

ASD-CC – Autism Spectrum Disorders – Comorbidity for Children

ASD-PBC – Autism Spectrum Disorders – Problem Behaviours in Children

ASD – UK – Autism Spectrum Database - UK

BASC – Behaviour Assessment System for Children

BISCUIT – 2 - Baby and Infant Screen for Children with aUtism Traits – 2

BPFAS – Behaviour Paediatric Feeding Assessment

CBCL - Child Behaviour Check List

CD – Conduct Disorder

CEBI – Children's Eating Behaviour Inventory

CSHQ – Children's Sleep Habits Questionnaire

CSI –Child Symptom Inventory

Dasl^{n}e - Database of children with ASD living in North East England

DD – Developmental Delay

DSM – Diagnostic and Statistical Manual of Mental Disorders

EQ-5D – Euro Quality of Life – 5D

GAD – Generalised Anxiety Disorder

GI – Gastro-intestinal

HFA – High Functioning Autism

IAN –Interactive Autism Network

ICD – International Classification of Diseases

ID – Intellectual Disability

IQ – Intelligence Quotient

Kiddie-SADS – Kiddie Schedule for Affective Disorders and Schizophrenia for School-age children

NAP-C – National Autism Plan for Children

NICE - National Institute for Health and Clinical Excellence

OCD - Obsessive compulsive Disorder

ODD – Oppositional Defiant Disorder

PBS – Paediatric Behaviour Scale

PCDI – Parent –child dysfunctional interaction

PD – Parental distress

PDD – Pervasive Developmental Disorder

PDD-NOS – Pervasive Developmental Disorder – Not otherwise specified

PSI – Parenting Stress Index

PSI-SF – Parenting Stress Index-Short Form

QoL – Quality of Life

QRS – Questionnaire on Resources and Stress

SAD – Separation Anxiety Disorder

SCAS-P – Spence Children's Anxiety Scale – Parent version

SDQ – Strengths and Difficulties Questionnaire

SEQ - Sensory Experience Questionnaire

SF – Short Form

SIB – Self-injurious behaviour

SNAP - Special Needs and Autism Project

SSP – Short Sensory Profile

STEP – Screening Tool of Feeding Problems

SP-Sensory Profile

SPA - Sensory Processing Assessment for Young Children

TD – Typically Developing

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1 Chapter 1. Introduction

"You get the diagnosis and you think it's going to be the magic key and then you open the cupboard and there's nothing in it"

- Parent of a child with ASD (Wallace, 2013)

1.1 Thesis overview

This thesis investigates co-existing emotional and behavioural conditions in children with Autism Spectrum Disorder (ASD) and explores the relationship between these conditions and impact on the family. The thesis also evaluates the role of services in the relationship between co-existing emotional and behavioural conditions and impact on the family. Since becoming a developmental paediatrician in 2005, I have believed clinicians need to move beyond 'the diagnoses' to look at co-existing conditions, which affect the everyday life of children and their families.

This introductory chapter describes the evolution of our understanding of ASD, co-existing emotional and behavioural conditions, impact on the family and the role of services before moving onto the structure of the thesis.

1.2 Historical background

There are anecdotal descriptions about autism peppered in history, some even dating back to ancient Mesopotamia (Gillberg, 2000). Paul Eugene Bleuler, a Swiss psychiatrist, first described the term autism in 1910 and he derived it from the Greek word, autos, meaning self (McGuinness, 2010).

In modern medicine, Hans Asperger, an Austrian paediatrician first described the term, 'autistic psychopathy' – combining autism (self) and psychopathy (personality) in a lecture in German in 1938 and subsequent in his publication in German in 1944 (Van Krevelen, 1971). Leo Kanner of John Hopkins University published the seminal paper in 1943 describing 'autistic disturbances of affective contact' in 11 children with 'extreme autism, obsessiveness, stereotypy and echolalia' (Kanner, 1943). Kanner later recognised autism as a unique syndrome with social and linguistic deficits and insistence on sameness (Kanner and Eisenberg, 1957).

1.3 Definition of ASD

The definition of ASD has evolved over time with our increasing understanding of its presentations, causative pathways and impact. Michael Rutter in a 1968 review article proposed three key features of autism – language development abnormalities, disturbance in interpersonal relationships and a variety of ritualistic and compulsive phenomena (Rutter, 1968). Lorna Wing and Judith Gould extended this further as a triad of impairments: 'absence or impairment of social interaction', 'absence or impairment of development of verbal or non-verbal language' and 'repetitive, stereotyped activities of any kind' (Wing and Gould, 1979).

For the purpose of this thesis, I have used definitions from the Diagnostic and Statistical Manual of Mental Disorders (4th ed.; DSM-4 TR; American Psychiatric Association (APA), 2000), a widely used nomenclature for the classification of mental disorders (APA, 2000). It defines autism or Autistic Disorders as (299.00): 'qualitative impairment in communication and social interaction with restricted repetitive and stereotyped patterns of behaviour, interests and activities; having onset prior to age 3 years'. Asperger's Disorder (299.80) is defined as 'qualitative impairment in social interaction along with restricted patterns of behaviour, interests and activities with no clinically significant general delay in language'.

The new criteria published in 2013 as DSM-5 aimed to simplify the diagnosis by including all children under the umbrella term of Autism Spectrum Disorder (APA, 2013). Instead of a triad, it concentrates on two areas – social communication and interaction, and restricted, repetitive patterns of behaviour and interests. Sensory difficulties are included for the first time under the diagnostic criteria of restricted, repetitive patterns of behaviour. The criteria also have severity grading of the two diagnostic variables as well as include other diagnoses such as intellectual impairment, language impairment and medical or genetic conditions.

1.4 Importance of ASD

There has been an exponential rise in the estimated prevalence of ASD in children over the last few decades: from 4.5 per 10,000 in the 1960s to 116 - 264 per 10,000 (Lotter, 1966; Baird et al., 2006; Baird, 2012). ASD is no longer

recognised solely as a childhood disorder; the third national survey of psychiatric morbidity in adults in England in 2007 showed a weighted prevalence of 9.8 per 1000 with 95% confidence interval, 3.0-16.5 (Brugha et al., 2011).

There is a substantial impact associated with ASD – financial, societal and on the family (Ganz, 2006). The experience of some families is summed up in a parent's reflection (Wallace, 2013):

"It controls every breath of your day, from the second you wake up to the second you go to sleep; you have to think about the preparation, the consequences, the rest of the family, in every decision you make concerning the person with autism"

The lifetime per capita incremental societal cost of autism is enormous: in the UK, for example, £1.5 million for a person with ASD and intellectual disability (ID) and £0.92 million for a person with ASD without ID, including both direct and indirect costs (Knapp *et al.*, 2009; Buescher *et al.*, 2014). The lifetime cost is $2.4 million for a person with ASD and ID in US (Ganz, 2006; Ganz, 2007; Buescher *et al.*, 2014). Specialist education, and health and social care along with parental productivity loss contributed the most to children's costs, while supported accommodation and lost individual productivity were the largest contributors to adults' costs (Ganz, 2006; Knapp *et al.*, 2009; Buescher *et al.*, 2014). The costs of supporting children and adults every year amount to £2.7 billion and £25 billion respectively in the UK (Knapp et al., 2009).

1.5 Aetiology of ASD

The aetiology of ASD is multifactorial as is the case for many other syndromes and disorders. The word "syndrome" was initially described as a series of totally different conditions presenting clinically as a cluster of symptoms through a final common pathway (Gillberg, 2000). A distinction between syndrome and disorder was made in DSM-IV; where syndrome refers to a group of symptoms presenting together characterising usually a single condition, and disorder a higher level and broader term referring to a cluster of symptoms (APA, 2000). Evaluating critically, the distinction might be related to the aetiology. Most of the syndromes have a definite aetiology. For example: Down syndrome and Turner syndrome have definite genetic aetiological basis; toxic shock syndrome has an

infectious aetiology. Disorders, while describing a collection of symptoms, have a broader aetiological basis. For example, neurodevelopmental disorders, developmental co-ordination disorder, autism spectrum disorder and attention deficit hyperactivity disorder all have multiple aetiological causative factors and pathways.

Both genetics and environmental factors are thought to contribute to the etiological neurobiology of autism; but our understanding of their interplay and individual impacts on the causative pathway is limited (Evans, 2013; Lai et al., 2014). Nevertheless, we have come a long way in our understanding of the central deficit in autism from a pure communication disorder to a neuro-cognitive/developmental disorder. However, researchers are still attempting to address some of the central questions about causative pathways (Evans, 2013).

1.6 Co-existing conditions in ASD

Children with ASD have multiple co-existing conditions, which can include a variety of conditions from comorbid health and mental health diagnoses, learning and intellectual disabilities to emotional and behavioural conditions (Close *et al.*, 2012; Skokauskas and Gallagher, 2012; Mannion and Leader, 2013b; Maskey *et al.*, 2013). Co-morbidity is defined as the occurrence of two or more forms of pathology in the same person or sharing of symptoms in a person across disorders (Matson and Nebel-Schwalm, 2007b; Gillberg, 2010). Many authors argue co-existence of disorders and co-morbidity is the rule rather than exception in developmental medicine (Gillberg and Billstedt, 2000; Sturm *et al.*, 2004; Hofvander *et al.*, 2009; Gillberg, 2010; Halleröd *et al.*, 2010; Goldstein *et al.*, 2011). Logically, this holds true for ASD as well. A recent review article has reiterated this relationship between different co-existing conditions and stressed on the future development of comorbid psychopathology research, analysing co-morbidities across different conditions including ASD (Matson and Williams, 2014).

Taking account of all the available evidence, the National Institute for Health and Care Excellence (NICE) (NICE, 2011) recognised the following co-existing conditions in children with ASD and can be used as a reference framework while evaluating these conditions:

- Mental and behavioural problems and disorders which include ADHD, anxiety disorders, mood disorders, etc.
- Neurodevelopmental disorders which include intellectual disability, motor co-ordination problems, etc.
- Medical or genetic problems which include epilepsy, chromosome disorders, etc.
- Functional problems and disorders which include sleep disturbances, feeding problems, etc.

The present study will concentrate on emotional and behavioural conditions that encompass the first and last categories: mental and behavioural problems and functional problems as described in the NICE guideline.

1.7 Co-existing emotional and behavioural conditions in ASD

Co-existing emotional and behavioural conditions (including hyperactivity, anxiety, temper tantrums, aggressive behaviour), occur more frequently in children with ASD compared with typically developing children (Bradley et al., 2004; Hess et al.). Our understanding of co-existing emotional and behavioural conditions in children with ASD is still evolving and there is limited literature evaluating these conditions holistically (Matson and Nebel-Schwalm, 2007b). This might be due to the following reasons:

- The complexity of diagnosis of ASD: ASD was until recently defined as comprising a number of related disorders in DSM-4 TR (APA, 2000). Most researchers have concentrated on autism or another subtype of ASD rather than evaluating the whole spectrum of ASD (Matson and Nebel-Schwalm, 2007b). The recent definition by DSM-5 unifying all 'autism conditions' under the umbrella term of ASD might help researchers to evaluate the whole spectrum as a group (APA, 2013).
- The presence of intellectual disability and communication difficulties in children with ASD makes it difficult to diagnose co-existing emotional and behavioural conditions (NICE, 2011; Volkmar *et al.*, 2014).
- The clinician might be prone to diagnostic overshadowing, where he/she fails to diagnose another co-existing emotional and behavioural conditions in the presence of a noticeable condition (Reiss et al., 1982).

Thus a clinician diagnosing ASD in a child might overlook another emotional and behavioural condition.

- Distinguishing co-existing problem behaviours from the core features of ASD can be challenging (Matson et al., 2008b).

In addition, we cannot ignore the debate that these co-existing emotional and behavioural conditions should be viewed as symptom clusters or 'epiphenomena' of ASD (Lainhart, 1999). However there is limited evidence to support this argument, as children with ASD have multiple different behavioural problems in addition to the core symptoms of autism (Matson and Nebel-Schwalm, 2007a). Over the past decade, researchers and clinicians have progressed towards a consensus of describing them as comorbid conditions or co-existing conditions alongside the core features of ASD (Matson and Nebel-Schwalm, 2007b; NICE, 2011; Kaat *et al.*, 2013; Mannion and Leader, 2013b; Volkmar *et al.*, 2014).

1.7.1 Dilemmas of diagnosing co-existing emotional and behavioural conditions in children with ASD

Before defining co-existing emotional and behavioural conditions, I would like to examine the dilemma of 'diagnosis' in clinical and research practice in child mental health. There is a stress on categorical diagnosis or the dichotomous distinction between 'disorder' and 'not disorder' in child psychiatry led by popular psychiatric classifications including DSM-5 and ICD-10 (WHO, 2008; APA, 2013). This has been criticised from the perspectives of co-existing mental health conditions in children and biological research. Children often have overlapping syndromes and cross-syndrome similarities making a categorical diagnosis of a 'disorder' difficult in these circumstances (Levy and Ebstein, 2009; Sonuga-Barke, 2009; Gillberg, 2010). For example: a child with Tuberous Sclerosis and ASD might have anxiety not fulfilling the diagnostic criteria or scoring above the indicative clinical cut-off of a standardised questionnaire. Though the child's anxiety symptoms might not fit the criteria, these symptoms can be debilitating for this particular child as well as the family.

The literature also supports the contention that clinically more children with ASD are impaired by psychiatric features than those who met symptom cut-off criteria (Kaat et al., 2013). To address some of these concerns, a dimensional

approach evaluating symptomatology as a continuum has been suggested and used in research practice (Sonuga-Barke, 1998; Nigg et al., 2005; Rutter, 2011). For example, analysing anxiety, the dimensional approach will evaluate symptoms on a continuous scale of one to ten rather than making a diagnosis of anxiety disorder above a cut-off of six. In addition, this dimensional approach also helps to monitor conditions such as anxiety that tend to be cyclical with remitting and relapsing phases. As a clinician, I use both categorical and dimensional approaches. While the categorical approach helps to reach a definite diagnosis, I move towards a dimensional approach to evaluate co-existing conditions for children with overlapping syndromes. The dimensional approach is also useful clinically to evaluate the progress of the child over time.

In the literature, different processes have been used to analyse and describe co-existing emotional and behavioural conditions in children with ASD: they range from single item questions looking at symptomatology (Mazurek et al., 2012) to diagnostic interviews and parent report questionnaires evaluating combinations of problem behaviours and psychiatric disorders (Huang et al., 2013; Kaat et al., 2013; Simonoff et al., 2013). Of late, studies have attempted to tease out these co-existing emotional and behavioural conditions into distinctive comorbid psychopathology apart from problem behaviours of children (Fodstad et al., 2010; Matson et al., 2010; Hattier et al., 2011; Mannion and Leader, 2013a; Mannion et al., 2013). Summarising, the complexity of defining and diagnosing co-existing conditions in children with ASD, has added to the already existing complexity of diagnosis of ASD in these children in both clinical and research practices.

1.7.2 Definition of co-existing emotional and behavioural conditions in children with ASD

For the purpose of this thesis, co-existing emotional and behavioural conditions are defined as co-existing behavioural conditions including co-morbid psychopathology and problem behaviours. Co-morbid psychopathology is defined as ‘occurrence of two or more psychopathologies in the same person’ (Matson and Nebel-Schwalm, 2007b). Problem behaviours or challenging behaviours are defined as ‘behaviours that are not socially acceptable, can physically harm someone, or affect education or living placement’ (Matson and Nebel-Schwalm, 2007a; Matson et al., 2010). Problem behaviours can be either

internalising or externalising (Matson and Nebel-Schwalm, 2007a). Internalising behaviours include 'fearful, inhibited or over controlled behaviours' and externalising behaviours include 'aggressive, antisocial or under-controlled behaviours' (Achenbach, 1991).

Beyond the operational diagnosis mentioned above, I will also explore other behaviours including sleep behaviour and feeding behaviour, classified under functional problems in the NICE guidelines, as well as sensory behaviour, during the course of this thesis (Lainhart, 1999; NICE, 2011; Maskey *et al.*, 2013).

This thesis will analyse co-existing emotional and behavioural conditions, henceforth known as co-existing conditions, and will not include neurodevelopmental disorders (for example, intellectual disabilities, motor co-ordination problems) and medical or genetic problems (for example, epilepsy and chromosome problems).

1.8 Prevalence of co-existing conditions in children with ASD

Depending upon the primary diagnosis (e.g.: autism, Asperger Syndrome, ASD or PDD-NOS) and the co-existing condition/s (e.g.: problem behaviour, anxiety, sleep behaviour) studied, the reported prevalence rates of co-existing conditions range from 24% to 84% in children and young people with ASD (Allik et al., 2006b; Brereton et al., 2006; Herring et al., 2006; Lecavalier, 2006; Davis III et al., 2010). Studies from the United Kingdom, Europe, Asia and the United States consistently report high prevalence of co-existing conditions in these children, but the methodological variations described above restrict the understanding of the true prevalence of these conditions (Simonoff *et al.*, 2008; Huang *et al.*, 2013; Kaat *et al.*, 2013; Mannion *et al.*, 2013; Maskey *et al.*, 2013). The prevalence of individual co-existing conditions is reviewed in detail in Chapter 3 (Literature Review).

1.9 Importance of co-existing conditions in children with ASD

Co-existing conditions can be overwhelming for the parent and child in everyday life as they impact day to day behaviour and add a significant burden to the care of a child with ASD (Allik et al., 2006a; Lecavalier et al., 2006; Manning et al., 2010). Co-existing conditions influence attainment of self-care skills, language

(Goldman et al., 2011) and social skills (Matson et al., 2010) and academic learning ability of the child (Pearson *et al.*, 2006a). These high rates of co-existing conditions persist into adulthood affecting the entire lifespan of individuals with ASD (Magiati et al., 2014).

In addition, co-existing conditions also contribute substantially to health care cost of children with ASD (Peacock et al., 2012). More importantly, it is shown that appropriate interventions for these conditions can improve the long term outcome of children with ASD (Matson and Nebel-Schwalm, 2007b). Compiling all this evidence, it is no surprise that national guidelines have recognised the importance of identifying co-existing conditions as early as at the time of diagnosis of the child to optimise the development potential of each child with ASD (NICE, 2011).

1.10 Impact on family of children with ASD

Both ASD as well as co-existing conditions impact families. Further sections analyse this impact of ASD and co-existing conditions individually.

1.10.1 Impact of ASD

The impact of ASD on the family, like the disorder itself, is pervasive and multifaceted (see a review by (Karst and van Hecke, 2012). Raising a child with ASD contributes to an increase in parental mental health concerns and parental stress (Davis and Carter, 2008; Ekas et al., 2010; De Andrés-García et al., 2012; Estes et al., 2013; Foody et al., 2014). Having a child with ASD results in a decrease in well-being and quality of life for parents (Allik et al., 2006a; Cappe et al., 2011; Werner and Shulman, 2013). Siblings of children with ASD are also affected; they can have higher levels of adjustment and mental health problems compared to their peers either as part of their own inherent Broad Autism Phenotype characteristics or as an extension of the impact of ASD in their own sibling/s (Benson and Karlof, 2008; Orsmond et al., 2009; Meyer et al., 2011).

The impact of ASD spreads beyond individuals affecting parental marriage and the family as a unit (Karst and van Hecke, 2012; Shtayermman, 2013). There are restrictions on employment opportunities for parents, and outings and holidays for families of children with ASD (Montes and Halterman, 2007). In

addition, there is usually a lifelong stress on parents as most people with ASD need some amount of care and support throughout their lives (Volkmar and Pauls, 2003; Volkmar et al., 2006), irrespective of the severity of autism and the age of diagnosis (Pottie and Ingram, 2008). This can precipitate crisis events throughout the lifespan, furthering the vulnerability of families of children with ASD (Weiss and Lunsky, 2011; White et al., 2012).

1.10.2 Impact of co-existing conditions

Co-existing conditions, along with the core autism symptoms of children with ASD, are shown to have an impact on the family (Davis and Carter, 2008; Silva and Schalock, 2012; Foody et al., 2014). They have a significant impact on the individual functioning of children with ASD (Patzold *et al.*, 1998; Sikora *et al.*, 2012b; Trembath *et al.*, 2012), as well as of that of their siblings and parents (Orsmond and Seltzer, 2007b; Orsmond *et al.*, 2009). Co-existing conditions also increase the rates of hospitalisation, medication and therapy of children with ASD (Frazier et al., 2001) and increase ASD related health care expenditure (Peacock et al., 2012).

Most studies show that multiple co-existing conditions and core symptoms of ASD have an additive impact on families (Davis and Carter, 2008; Phetrasuwan and Shandor Miles, 2009; van Steensel *et al.*, 2012a; Foody *et al.*, 2014). Parents reported that managing demanding behaviours and managing behaviours in public places were highest sources of parenting stress (Phetrasuwan and Shandor Miles, 2009). There are differences reported in parenting stress for mothers and fathers of children with ASD, where regulatory problems were associated with maternal stress and externalising behaviours with paternal stress (Davis and Carter, 2008). More recently, there is additional evidence showing problem behaviours as the more significant predictor for parenting stress in both groups of families of children with ASD and developmental delay (Estes et al., 2013). However, both socialisation deficits and behaviours such as oppositional behaviour were shown to be associated with parenting stress in a recent study of mothers of children with ASD (Foody *et al.*, 2014).

1.11 The role of services to meet unmet needs of families of children with ASD

It is difficult to understand and assess the role of services and the support they provide for families of children with disabilities (Quine, 1989). An assessment of any services should be in accordance with how they meet the needs of the user including the child and his/her family, in the case of child health services (Quine, 1989; Beresford, 1995). A needs assessment is usually done by measuring the perceived unmet need of families, so that appropriate services can be developed to support these unmet needs (Beresford, 1995). In a complex condition such as ASD, the assessment of perceived unmet needs can be used as a benchmark for the assessment of services; but the exploration of these needs in the available literature is limited (Brown et al., 2010; Brown et al., 2012). There is a dearth of literature analysing unmet parent needs regarding co-existing conditions of children with ASD. This thesis is the first attempt to describe and analyse these unmet parent needs. The detailed exploration of the theoretical basis of unmet parent needs is provided in section 2.3.2 (page 20).

1.12 Positioning of the present thesis in autism literature

It is clear from this brief literature summary that there are conflicting reports about the definition and frequency of co-existing conditions in children with ASD. There is emerging evidence of the impact of these conditions on the family of the child; but there is limited data to support the extent of this impact over and above that of ASD. It is no surprise then that the limitations of the available literature have prevented us from moving forward to defining the unmet needs of families regarding co-existing conditions.

This thesis aims to link the above concepts by understanding the role of co-existing conditions and their unmet needs on impact on the family by the following aims and objectives:

1.12.1 Research aims

The research aims of the project are:

- To analyse the frequency, severity, associations and impact on the family of co-existing emotional and behavioural conditions in children with ASD

- To investigate the role of unmet parent needs on the impact on families of children with ASD

1.12.2 Research objectives

The research aims will be delivered through the following objectives

- To review the existing literature to identify reliable and valid tools to assess the common co-existing conditions in children with ASD
- To describe the frequency, severity and correlates of common co-existing conditions in children with ASD using parent report questionnaires
- To describe the impact on the family of common co-existing conditions in children with ASD using a parent report standardised questionnaire
- To describe the perceived unmet needs of parents with children with ASD by a parent report questionnaire

1.13 Thesis structure

The thesis will consider the theoretical framework (chapter 2), before moving on to literature review (chapter 3), methodology (chapter 4), results (chapter 5), and discussion (chapter 6).

2 Chapter 2. Theoretical framework

This chapter deals with the theoretical frameworks used in the thesis. It begins with disability theories, moving on to theories around adaptation and the role of services, and finally looking at childhood disability research designs.

2.1 Disability theories

Though there is no single unifying theory of disability, the "International Classification of Functioning, Disability and Health" (ICF) model attempts to amalgamate the medical and social models of disability (WHO, 2001; Williams, 2001; Colver, 2005). The medical model considers the biological impairment as the starting point, extending support and help for the individual through therapeutic services (Barnes, 2003). The social model reflects on disability as being imposed on the individual due to the disabling barriers and processes in society (Oliver, 1990; Williams, 2001). A socio-medical model was also developed to integrate both the impact of biological impairment on the individual and the societal role in disability (Bury, 1997).

The World Health Organisation (WHO) initially presented a shared framework to define impairments, disability and handicap through its "International Classification of Impairments, Disabilities and Handicaps" (ICIDH) (Table 2.1) (WHO, 1980).

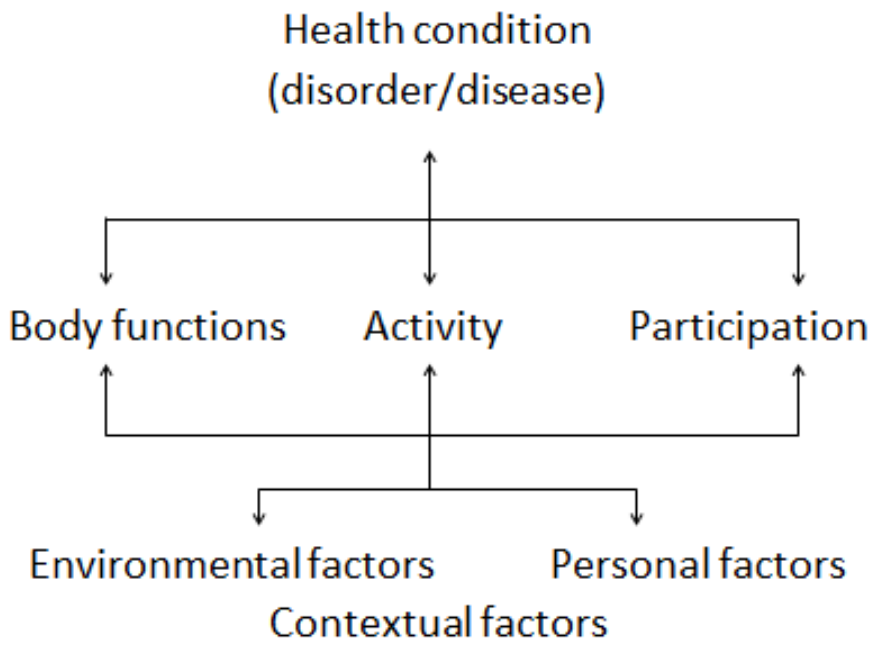
Table 1: Structure of International Classification of Impairments, Disabilities and Handicap

Impairment	Loss or abnormality of structure or function
Disability	Restriction of ability of the individual due to the impairment
Handicap	Disadvantage to the individual with disability in society

This framework was modified by WHO in its International Classification of Functioning, Disability and Health (ICF) model integrating the environmental or societal influences into a bio-psychosocial model (Figure 1) (WHO, 2001). Notably this model moved away from 'negative' handicap to 'positive' participation in society; from disability to activity or ability of the individual; and from 'negative' impairment to 'positive' body functions. Evaluating holistically, this model is relevant to all individuals with or without a disease/disorder:

participation in the society matters to every individual, not just people with disability. The ICF model strengthened the role of society/environment in shaping its active interaction with the individual enabling participation (Colver, 2005). The family as part of environmental factors contribute to improve body functions, activity and participation of the individual.

Figure 1: Adaptation of the ICF model (WHO, 2001)



2.2 Family and parental adjustment to disability

As shown in the ICF model, there is a dynamic exchange between the individual and environmental factors including the family. Children live in the context of families, their immediate nurturing shell, and affect family dynamics (Kazak, 1995). There is a continuous dynamic exchange between different members of the family; that is, disease and disability affect not just one individual but also the family triggering corresponding changes, adjustment and different ways of coping for each individual and the family as a whole. Different theoretical frameworks and models have been influential in our understanding of parental and family adjustment to paediatric chronic physical disorders and disability. All models explore the adaptation of an individual or the family to a stressor using available resources and coping strategies.

The Disability-Stress-Coping model explored the relationship between illness stressors as 'stress factors' and social support as 'resistance factors' affecting the outcome of familial adaptation and functioning (Wallander et al., 1989). This model was further developed with a mutual transaction among core factors (stress and resistance factors) including the processes of adaptation as a transactional stress and coping model (Thompson Jr et al., 1994; Wallander and Varni, 1998); an adapted model is presented in Figure 2.

Figure 2: Adaptation of conceptual model of parental adjustment to paediatric chronic physical disorders (Wallander and Varni, 1998)

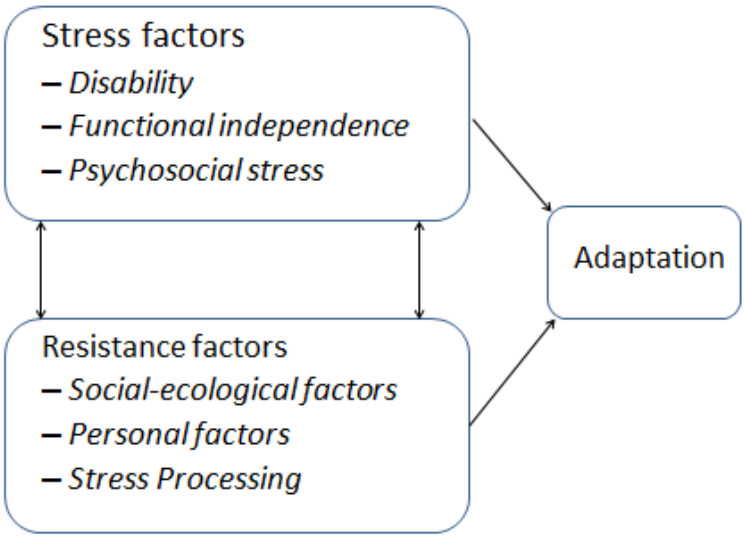
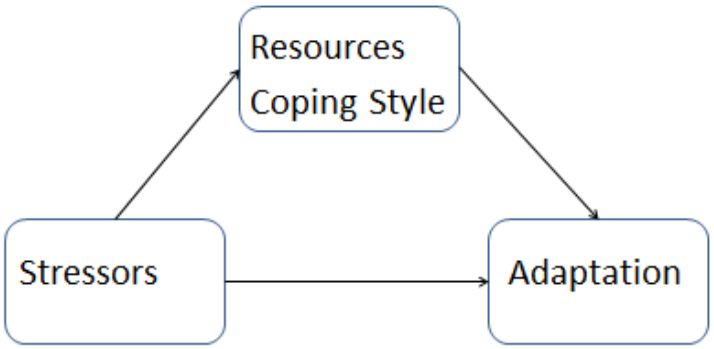


Figure 3: Adaptation of composite research model (McConachie, 1994)



The Disability-Stress-Coping model has been used in elaborating familial coping mechanisms in both families of children with Cerebral Palsy (Carona et al., 2013) and those of children with ASD (Altiere and Von Kluge, 2009).

The composite research model of stress and coping looked at four elements – stressors, resources, coping style and outcomes; where outcome including parental and family adaptation was influenced by the other three factors (McConachie, 1994). An adaptation of this model is presented in Figure 3; where resources and coping style mediate the effect of stressors on outcome. The formal sources of support, information and treatment through the services constituted one arm of the resources in this model. This model is adapted from the double ABCX family crisis model, which has three stages, a pre-crisis stage, crisis and a post-crisis stage (McCubbin, 1983). In the double ABCX model, 'a' as the present stressor interacts with family's resources ('b') and the family's perception of the stressor ('c') causing crisis 'x'. In the post-crisis stage, 'A' as the pile-up stressor interact with family's new resources ('B') and family's perception of the whole event of crisis ('C') resulting in the final adaptation of the family ('X').

2.2.1 Individual coping and adaptation

The composite research model of stress and coping can be used as the model to understand individual adaptation as well as that of the family. An individual faced with a stressful event uses available resources and personal coping strategies to adapt to the stressor. For individuals, two broad coping strategies are described (Benson, 2010; Cappe *et al.*, 2011). The first strategy which is described as 'approach coping' strategy including problem solving ability and ability to seek support can help parents to cope positively. The second strategy of 'avoidance coping' predominantly uses emotion-focused strategies and results in negative coping. Parental self-efficacy, defined as 'individual's self-appraisal of ability to effectively parent' is an important positive process as well as outcome of this coping process (Coleman and Karraker, 1998). The positive coping mechanisms result in parental well-being and parental self-efficacy while negative coping strategies increase parental stress eventually leading to mental health problems including depression (Benson, 2010; Werner and Shulman, 2013). Parental resilience, defined as parents' ability to cope with adverse events, is shown to buffer the effects of parenting stress (Meadan *et al.*, 2010).

Parental personality characteristics can influence both resilience as well as coping mechanisms and can vary from individual to individual (Bitsika *et al.*, 2013).

2.2.2 Family coping and adaptation

In addition to individuals, the family copes with the stressor as a unit. Family resilience, which is equivalent to the resilience of an individual, defines "the positive behavioural patterns and functional competence, individuals and the family unit demonstrate under stressful or adverse circumstances, which determine the family's ability to recover by maintaining its integrity as a unit while insuring, and where necessary restoring, the well-being of family members and the family unit as a whole" (McCubbin, 1996). The family resilience is intricately linked to family hardiness, an equivalent of self-efficacy. The hardiness is defined as the family perception of its control over life events, stressors and outcome and the confidence that they can endure potential stressors and challenges (McCubbin, 1983; McCubbin, 1996; Weiss *et al.*, 2013). The family hardiness and resilience can positively impact the outcome of any stressor and act as a mediator between stressors and outcomes (Greeff and Van Der Walt, 2010; Kapp and Brown, 2011). In addition the family hardiness can enhance positive coping mechanisms of the parents. Supporting family members who understood child's needs can improve parental resilience and buffer the impact on the family (Bitsika *et al.*, 2013).

Families are diverse and complex not only in their constitution, but also in their response to stressors and challenges (Ellis *et al.*, 2002; Meadan *et al.*, 2010). The differences in the internal dynamicity of a single-parent family from that of a double-parent family make its responses and coping strategies to stressors different. The family also cannot be defined as a single congruous entity as it can be either defined as 'an entity where all family members live under one roof' to 'all descendants of an ancestor' (Weiss, 1991). However, the thread of coping to stressors run though all families irrespective of their diverse and complex compositions, albeit with varying degrees.

To summarise, the different research models recognise the presence of ongoing stressors in the family of a child with chronic disability; the resilience or resistance factors which protect the family; the coping styles of individuals

including parents and the final adaptation of the family which will again feedback into the model as a stressor or strength.

2.3 The role of services for families of children with ASD

The service system including clinical, educational and social support services are designed to support children and their families (Armbruster *et al.*, 1997). Children with ASD use a higher number of services (averaging between 6-7 services) compared to children without disabilities (averaging 1.75 services) (Kohler, 1999; Thomas et al., 2007; Brown et al., 2010). These high service needs may be related to both the complexity of ASD (Kohler, 1999) as well as the frequent occurrence of co-existing conditions (Gillberg and Billstedt, 2000). For example, families of children with feeding problems might use the services of a health visitor or a community nurse to improve feeding practices, a dietician to improve nutrition intake and a paediatrician or a speech and language therapist to rule out any associated physical causes. It should be noted that a family uses these services multiple times for initial screening, diagnostic evaluation, possible interventions/suggestions and monitoring.

The composite research model of stress and coping directed the role of services (as part of resources) as one of the elements affecting family and parental adaptation (McConachie, 1994). The services encompassing clinical, educational and social support services, can support children with ASD not only to improve their social communication skills but also to address their frequent co-existing conditions (Kohler, 1999; Gillberg, 2000; Thomas *et al.*, 2007; Brown *et al.*, 2010). In addition, the services should be able to support the needs of and empower parents of children with ASD (Brown *et al.*, 2010; Brown *et al.*, 2011). The need for the services to offer such a holistic standard of care has been recognised in the NICE guidelines, where there are recommendations to enable and strengthen existing child health and mental health services and enable parental participation (NICE, 2013). Specifically parental participation in formal services can not only be advantageous to services to understand parental needs but also to parents themselves in enhancing their own positive coping strategies (Bourke-Taylor *et al.*, 2010).

In addition to formal services, the perception of social support can also contribute to the final adaptation (Carona *et al.*, 2013). The informal social

support, defined as 'the availability of significant others to provide adequate help, care or company' (Sarason *et al.*, 1983) includes in addition to family members, social support workers, non-governmental organisation support and other parents as well as accessibility to respite care. This social support is central to parents and families in their coping and final adaptation to the stressor. For example, appropriate respite care is shown to alleviate stress, enhance positive coping skills and improve marital quality (Harper *et al.*, 2013). Similarly increased community and social support is also associated with improved coping strategies (Hall, 2012; Paynter *et al.*, 2013). Both formal services and informal social support can augment positive coping mechanisms of individuals and the family unit.

How do we analyse the role of services? To begin with, we cannot quantify the amount of services received as a measure of services. There is a recognisable gap between service need and service utilisation for child mental health service, where only one-third of children who need this service receive it (Armbruster *et al.*, 1997). Different barriers that contribute to this gap include access to services, financial circumstances, parental perceptions and poorly co-ordinated clinical, educational and social support components of services (Armbruster *et al.*, 1997; Brown *et al.*, 2012).

In addition, there are problems in defining the standards by which services have to be assessed (Quine, 1989). There is agreement that the standards of assessment should tally with the needs of the end-user; that is the family and the child in case of children with ASD (Beresford, 1995). But, do we know enough about the needs of families of children with ASD? The emerging evidence suggests that there is a gap in our understanding of the specific needs of families who have children with ASD (Brown *et al.*, 2010; Brown *et al.*, 2012).

2.3.1 Parental needs

A child's impairment as well as disability can result in a substantial range of needs (Beresford, 1995). A perceived need is defined as "an individual's judgement of the discrepancy between actual states or conditions and what is normative, desired or valued from a help seeker's and not a help giver's perspective" (Dunst, 1988). The perceived needs of parents of a child with disability include: early diagnosis and support, information about the condition

and the services, opportunity to discuss the child's progress regularly, help with developing the child's communication and developing new skills, emergency services for times of difficulty and help with child-minding care (Quine, 1989). Parents of children with ASD report similar needs and stress the need for information about services, for information about managing problem behaviours and the need for co-ordination of services including respite services (Ellis *et al.*, 2002; Cassidy *et al.*, 2008; Wallace, 2013). It is also important to highlight that needs of children with ASD and their families can change over time as children develop through their childhood and adolescence, as ASD is a developmental disorder (Brown *et al.*, 2012).

2.3.2 Parental unmet needs to assess services

An assessment of services should be in accordance with how they meet the needs of the user including the child and his/her family (Quine, 1989; Beresford, 1995). This evaluation through the eyes of the parents can give a clear picture of what parents consider important for their children and what they lack (Quine, 1989). The assessment of perceived unmet needs is particularly useful to analyse ASD services, as families of children with ASD have differing needs through different stages of life (Beresford, 1995; Brown et al., 2010). This also assesses the process factors of access to care including delays in getting the diagnosis and waiting times to access the support services, in addition to the outcomes (Krauss et al., 2003).

A recent article analysing unmet needs of families of school-aged children with ASD in Canada identified age-appropriate social activities for the child, information about relevant services and continuous service support as the three common unmet parent needs (Brown et al., 2012). A recent parent UK survey found that parents reported the support for managing co-existing conditions including problem behaviours as an unmet parent need (Wallace, 2013). In summary, it is suggested that unmet parent needs can be used as a surrogate to measure the role of services in meeting the needs of the service users.

2.4 Study designs in Disability research

Epidemiological study designs are the most appropriate processes to identify the frequency of individuals with disability in a population: "To make people count, we have to count people right" (WHO/UNESCAP, 2008).

2.4.1 Epidemiological study designs in disability research

Population censuses, surveys, and registration data collection are used in epidemiological study designs (WHO/UNESCAP, 2008). Though population censuses are ideal to calculate the true prevalence of disability, they are expensive and time-consuming (UN, 2001). Sample surveys can be used to collect data, but will still require a large enough sample to effectively look at different disabilities (UN, 2001).

Registration data collection can effectively prepare sampling frames for specific disabilities for both service and research purposes; they can be a one-time data collection at the time of diagnosis or intermittent or continuous data pooling (UN, 2001; WHO/UNESCAP, 2008). A good disability register or database can help in service planning, individual patient care, clinical audit and epidemiological research (Hutchison, 2001). There are two main types of disability registers based on their utilisation: health registers and research registers (McConachie et al., 2009; Ogdie et al., 2012). Health registers are government national or regional registration databases of service users of different health conditions while research databases are voluntary registers intended only for research (Colver, 2003; McConachie et al., 2009). Health registers can be useful mainly for patient care and public health surveillance, but can be expensive to initiate and maintain (Newton, 2002). In addition, these health registers have information about service users only, not about the whole population of individuals with a particular condition. They can be of use in countries where standardised services are available with an empowered population with high service use for a health condition.

Health service registers have both strengths and weaknesses. A well maintained health service register can aid in comprehensive epidemiological research including incidence, risk factors and evolution of health conditions (Newton, 2002). The biggest disadvantage is the cost of resources involved in initiating and maintaining registers. Coding, accuracy and privacy of data are crucial in maintaining the register, but can be resource intensive in terms of human resources, time and finances (McConachie *et al.*, 2009). Researchers and database co-ordinators also have to go the extra mile to ensure enrolment of the ‘hard to hear’ population including minorities. A comprehensive register

can accurately analyse the population it studies, but generalizability to the wider population needs to be done with caution (Colver, 2003).

Similarly, research databases or registers can be expensive to initiate and maintain but can help to conduct high quality research by bringing together large number of individuals with a particular health condition (Newton, 2002; Colver, 2003; McConachie *et al.*, 2009). The research databases, having a voluntary enrolment, will have only individuals who enrolled out of their own interest and not the whole population of individuals with a specific health condition. Though this can be disadvantageous from an epidemiological point of view, it can be fruitful to have motivated individuals willing to participate in research, which in turn can improve response rates, participation and results of research (Newton, 2002). Research databases can help a researcher to select the sample population for the study appropriately; be it a large sample for prevalence studies or a small sample with strict inclusion criteria for an intervention study. Recruiting through a research database enables the researcher to reach the target sample without the difficulties of recruiting directly from a large population. Consequently, these databases can overall improve research infrastructure and increase research capacity of a condition (McConachie et al., 2009). There are research databases on different childhood disorders exploring epidemiology and outcomes: for example: Cerebral Palsy (Colver, 2003; Colver, 2006) and Diabetes Mellitus (Awa et al., 2013; Bechtold et al., 2014).

2.5 Study designs in autism research

Over the last decade, to understand the frequency of co-existing conditions in children with ASD, the majority of the studies have looked at surveys of clinic samples (Kaat et al., 2013), community samples (Dominick et al., 2007; Mannion et al., 2013; Simonoff et al., 2013) and combined clinic and community samples (Hurtig et al., 2009; Thorson and Matson, 2012). Apart from the population referenced studies (Simonoff *et al.*, 2008; Maskey *et al.*, 2013; Simonoff *et al.*, 2013), these studies fail to capture the true prevalence or frequency of co-existing conditions in children with ASD.

Epidemiological study designs, which can define prevalence more accurately, are also used in ASD: large surveillance health and treatment registries (Levy et

al., 2010; Mazurek et al., 2013) and population-representative cohorts (Simonoff *et al.*, 2008; Maskey *et al.*, 2013; Simonoff *et al.*, 2013). Usually a single item question is used to identify a problem in large surveillance records limiting the understanding of severity of the problem (WHO/UNESCAP, 2008; Levy et al., 2010; Mazurek and Engelhardt, 2013). Population representative research databases can attempt to bridge this gap by encouraging both prevalence studies and detailed analysis of a condition in a cost-effective and efficient process (McConachie *et al.*, 2009; Maskey *et al.*, 2013).

There are databases for autism research based in different countries – examples include the Database of children with ASD living in North East England (Dasl^{n}e) and the Autism Spectrum Database–UK (ASD-UK) based in the UK; the Autism Treatment Network (ATN), the Interactive Autism Network (IAN) and the National Database for Autism Research (NDAR) based in the USA, the Autism Western Australia (Autismwa) and the National Epidemiologic Database for the Study of Autism in Canada (NEDSAC) based in Australia and Canada respectively (McConachie et al., 2009; Lee et al., 2010; Hall et al., 2012; Ouellette-Kuntz et al., 2012; Warnell, 2013). Databases can support researchers with recruitment to different studies and analyses: epidemiological studies (Levy *et al.*, 2010; Maskey *et al.*, 2013; Mazurek *et al.*, 2013); phenotype-genotype correlation studies (Lee et al., 2010); risk factor studies (Mazurek and Engelhardt, 2013); and intervention studies (Maskey *et al.*, 2014).

To summarise, this section highlights the utility of population referenced ASD databases to conduct a range of studies in children with ASD including frequency and severity of co-existing conditions as planned in this study project.

2.6 Chapter summary

This chapter has analysed briefly the evolution of the ICF model of disability moving away from handicap towards participation; and different models of stress, coping and adaptation for parents and family to disability. It also briefly analysed the role of services and unmet parent needs in these models and finally the value and convenience of using databases for the present project.

3 Chapter 3. Literature Review

This chapter presents a review of the available literature to address the objective to identify valid and reliable measures to assess common co-existing conditions of children with ASD (refer to section 1.12.2 – page 12). The review proposes to briefly analyse the prevalence and predictors of co-existing emotional and behavioural conditions and their impact on family as well as the collective and individual measures to assess these conditions and their impact. The literature review covers the following topics:

- Co-existing emotional and behavioural conditions in children with ASD and the measures available
- Specific emotional and behavioural conditions in children with ASD such as anxiety and hyperactivity and their measures
- The impact of co-existing emotional and behavioural conditions on the family and the role of services on the impact on family.

3.1 Literature Search Methodology

The literature search utilised Scopus electronic database (all years – present date) and Ovid Medline® (1996 – present date). The search was conducted using standardised terms (see below and Appendix 2) and was sequentially performed on the databases with subsequent updates and alerts. The search included using standardised search terms in ‘title-abstract-keyword’ in Scopus and ‘title’ in Ovid Medline®. The initial searches were conducted between October 2012 and January 2013 with subsequent monthly updates until date. Specific behaviours such as anxiety, hyperactivity, sleep, feeding and sensory behaviours were not adequately covered in measures assessing co-existing conditions thus necessitating a dedicated literature review for these specific behaviours and their measures. A lot of the studies included in these reviews are recent highlighting that the study of co-existing conditions of children with ASD is an emerging topic. This thesis attempts a novel direction of evaluating the impact of total co-existing conditions on family and the role of unmet parent needs in this relationship.

3.1.1 Inclusion criteria for literature review

From the literature search, articles for the initial literature screening were selected if they fulfilled the following criteria

- Children with ASD aged between 2-18 years of age or their parents
- Study analysed the particular co-existing condition of children with ASD. The definition for total or specific co-existing condition is provided for each search category below.
- Full article available or translated to English

After initial screening, articles were further assessed for the type of study, the number of participants and for the type of measure used to assess the co-existing condition, so that relevant studies could be included for detailed review. All population-representative community based studies and longitudinal follow-up studies were included. Other community-based studies and combined or clinic-based studies were included for this detailed review if they fulfilled the inclusion criteria of

- More than 50 subjects (children with ASD or their parents)
- A parent report questionnaire or a parent report item or parent report interview was used to assess the co-existing condition

3.2 Co-existing conditions in children with ASD

Co-existing conditions in children with ASD as per definitions in section 1.7.2 page 7 include both comorbid psychopathology and problem behaviour. For the purpose of the review, emotional and behavioural conditions will be defined as:

- Parent report of behavioural disorder/behavioural problems or
- Fulfils the DSM-IV criteria for a behavioural/emotional disorder or
- Scores above the cut-off scores for behavioural/emotional disorder on a standardised measure.

The search term used was behavio*. The results of the literature search are summarised in

Table 2 and Table 3.

Table 2 : Results of literature search about co-existing conditions on Scopus (all years – October 2012)

Search term number	Search terms used	Search results
1.	Autis* in 'title-abstract-keyword'	33,050
2.	Behavio* in 'title-abstract-keyword'	2,835,119
3.	Search term number 1 and 2 combined	13,888
4.	Search term number 3 and 'questionnaire or tool or measure' combined	5011

Table 3 : Results of literature search about co-existing conditions on Ovid-medline (1996 – October 2012)

Search term number	Search terms used	Search results
1.	Autistic disorder.mp or autistic disorder in title	10,338
2.	Behavio*.mp in title	580,844
3.	Search term number 1 and 2 and 'questionnaire.mp or tool.mp or measure.mp' combined	395

184 articles were identified relevant after initial screening. These articles were assessed for the type of study, the number of participants and whether parent reporting was used to analyse co-existing conditions. The articles fulfilling the inclusion criteria of relevant studies including a sample size of more than 50 subjects with parent reporting of symptoms were analysed in detail, the summary of which is provided in Table 4.

3.2.1 Methodological considerations

Literature uses different terminologies to describe co-existing emotional and behavioural conditions in children with ASD: refer to sections 1.7.1 (page 6) and 1.7.2 (page 7) for detailed definitions, discussions and dilemmas. I attempt to bring the different terminologies used into a common platform for the purpose of this review; but at times revert back to the original authors' vocabulary. For

example, I aimed to use co-existing conditions to describe the total co-existing emotional and behavioural conditions throughout the literature review; but for clarity also use terms such as co-morbid psychiatric disorders in line with authors' vocabulary.

Sampling of most of these studies comes from community, clinic or a combined population; some are from population representative cohorts (Simonoff *et al.*, 2008; Maskey *et al.*, 2013; Simonoff *et al.*, 2013). There are large-sample studies reported from health and treatment registries, but these studies often use a single question for surveillance purposes (Levy et al., 2010; Mazurek et al., 2013). Some of the earlier studies evaluated specific subgroups of ASD such as Asperger syndrome (AS), Pervasive Developmental Disorders-Not Otherwise Specified (PDD-NOS); but of late studies have started analysing the broader ASD group (Kaat et al., 2013; Mannion and Leader, 2013a; Simonoff et al., 2013).

3.2.2 Results of literature review

The summary of relevant studies considered for this literature review is provided in Table 4. This table gives information about the main research question, sampling frame, age of children, type of diagnosis, main findings and short critique of each article regarding the type of study and the measure used. Subsequent sections on prevalence, predictors and measures of co-existing conditions utilise this literature review table as well as recent published literature reviews (Matson and Nebel-Schwalm, 2007b; Mannion and Leader, 2013b).

Table 4 : Summary of important studies evaluating total co-existing conditions in children with ASD (Reverse chronological order)

Author (date)	Research question	Sampling (N = number of participants)	Age	Tools used for co-existing conditions	Main findings	Brief critique
Mannion (2013)	Analysis of predictors of psychopathology, gastro-intestinal problems and epilepsy in ASD	Community recruitment (schools, parent groups) N = 89 ASD	3-16 years	ASD-CC (ASD-Comorbid for Children)	Behaviours classified as moderate and severe impairment respectively in Tantrum behaviour – 23.6% moderate and10.1% severe Repetitive behaviour – 27% and 7.9% Worry/depressed – 19.1% and 2.2% Avoidant behaviour – 21.3% and 13.5% Under-eating – 16.9% and 11.2% Conduct – 10.1% and 13.5% Overeating -12.4% and 9% GI symptomatology significant predictor for co-morbid psychopathology (R^2=.309). Other factors considered age, gender, ID, anxiety disorder, ADHD and sleep problems. GI symptomatology predicted tantrum behaviour (R^2=.074), worry/depressed (R^2=.266), avoidant behaviour (R^2=.194) and	Community sample: evaluating co-morbid psychopathology: ASD specific tool

					conduct behaviour (R^2=.221).	
Simonoff (2013)	Longitudinal analysis of persistence of psychiatric problems in ASD and predictors	Population representative Special Needs and Autism Project (SNAP) Cohort N = 81 ASD	12 year followed to 16 year	Strengths and Difficulties Questionnaire (SDQ)	No significant difference in psychiatric problems over time: 48.9% at 12 years; 44% at 16 years Lower IQ predicted higher hyperactivity and total scores Lower social class, poorer maternal mental health, family based deprivation predicted higher emotional problems	1st longitudinal study: Population representative: used ASD cut offs for SDQ
Kaat (2013)	Relation of psychiatric symptom-induced impairment to mental health in children with ASD	Referral developmental disability clinic – consecutive referrals N = 115 ASD	6-12 Years	Child and Adolescent Symptom Inventory DSM-4 referenced rating scales	Parents report: 81% - impaired socially or academically by ≥ 1 psychiatric disorder Most common – ADHD (67%), Anxiety Disorder (47%) and ODD (35%) More youth impaired by psychiatric symptoms than met symptom cut off criteria	Clinic sample
Huang (2013)	Examine effects of autistic behaviour and individual emotional and behavioural	Recruited from 5 hospitals and 2 clinics N = 52 autism	3-12 years	SDQ	ASD children at high risk for problems of hyperactivity/Inattention – 55.8%; peer problems – 51.9% and pro-social behaviour – 65.4% Stress scores of mild/moderate and severe emotional and behavioural problems higher	Clinic sample

	problem on parenting stress				than that with no problems.	
Mannion (2013)	Co-morbid psychopathology, gastrointestinal symptoms, sleep problems and epilepsy in ASD	Community recruitment (schools, parent groups) N = 89 ASD	3-16 years	ASD-CC (ASD-Comorbid for Children)	46.1% ≥ 1 co-morbid conditions (78.7% if ID included) ADHD – 18%; anxiety -15.7% Mean of each subscale shows no impairment	Community sample: ASD specific tool
Mazurek (2013)	Relationship between videogame use and problem behaviour in ASD	Recruited from IAN (Interactive Autism Network) N = 169 boys ASD	8-18 years	Vanderbilt Attention Deficit/Hyperactivity Disorder Parent Rating Scale	No relationship between hours of videogame use and problems of inattention, hyperactivity and ODD In multivariate analysis, problem videogame playing test score predicted inattention (other factors – age, hours, videogame genre). ODD was predicted by problem videogame playing test score and role-playing genre.	Database sample: only boys
Maskey (2013)	Predictors and frequency of co-existing emotional and behavioural conditions in children with ASD	Population representative database N = 863 ASD	2-18 years	Ten item parent report questionnaire	52.6% - ≥ 4 co-existing emotional and behavioural problems frequently Most common problems: Eating – 58% Sensory reactions – 57% Lower age, speech of less than sentences and special school attendance - significant predictors of behavioural problems (other factors considered – type of autism, gender)	Population representative

O'Donn-ell (2012)	Relation between sensory problems & problem behaviour	Early intervention group – retrospective; N =75 ASD	3 years	Aberrant Behaviour Checklist (ABC)	Significant relationship between sensory problems and problem behaviour score	Early childhood retrospective study
Mandy (2012)	Sex differences in children with ASD	Specialist clinic sample N=325 ASD	3-18 years	SDQ	Girls – less repetitive stereotyped behaviour, when controlled for social and communication impairment Boys – More externalising and social problems	Clinic sample
Mazurek (2012)	Prevalence and correlates of physical aggression in ASD	Autism Treatment Network registry N=1584 ASD	2-17 years	Single item aggression question, CSHQ, SSP	Aggression – 53% - highest prevalence in young children Aggression associated with SIB, sleep problems, sensory problems, GI problems, communication and social functioning. Multivariate model – SIB, sleep and sensory problems strongly associated with aggression	Single item on a large database sample
Skokau-skas (2012)	Patterns of comorbid psychiatric problems in children with ASD and their parents compared to controls and their parents	Community recruitment N = 67 ASD (Age and IQ matched controls– 67)	6-18 years	CBCL	ASD Children – either internalising or externalising problems. Controls – more externalising problems In ASD: ADHD – 44.78%; Anxiety – 46.2% 22.4% of fathers and 23.8% of mothers of ASD – possible psychopathology (17.9% and 23.9% respectively for fathers and mothers of controls) Parents' distress associated with neither externalising and internalising problems in children with ASD	Community sample

Tseng (2011)	Relationship between emotional and behavioural problems and sensory problems	6 centres' samples N = 67 autism; 45 typically developing (TD)	48-84 month (mts)	CBCL – Chinese version	73.1% ≥ 1 syndrome scale score in CBCL 96% of (ASD & internalising behaviour) and 82% of (ASD & externalising) children had at least 1 atypical sensory behaviour; Linear regression analysis: male and sensory avoidance predicted internalising and sensory sensitivity predicted externalising.	Multi-centre study from Taiwan
Gjevik (2011)	DSM-IV disorders in ASD	Special school population N = 71 ASD	6 – 17.9 years	Kiddie SADS interview (Schedule for Affective Disorders and Schizophrenia for School-Age children	72% - at least 1 co-morbid condition 30% - at least 2 conditions Anxiety – 41%; ADHD – 31% OCD-older children ODD/Conduct – PDD-NOS	School sample
Pandolfi (2011)	Psychometric evaluation of CBCL in ASD	Combined samples – ASD clinic samples + Down syndrome prevalence sample N = 122	6-18 years	CBCL	Results compared between ASD with emotional and behavioural disorders and ASD alone. Children with emotional and behavioural disorders had higher mean scores on CBCL. The subscales showed good sensitivity but low specificity for detecting co-occurring conditions	2 different samples combined

		autism				
Hattier (2011)	Occurrence of challenging behaviours in ASD and controls	Children enrolled in early intervention programme N =633 ASD (1498 – controls)	17 – 37 mts	BISCUIT – 3 (Baby and Infant Screen for Children with aUtism Traits –3)	ASD – higher frequency in all problem behaviours. 66.8% ≥ 2 problem behaviours 70% ≥ 1 aggressive/destructive behaviour; 47% ≥ 1stereotypy behaviour ; 30% ≥ 1 self-injurious behaviour	Clinic sample of toddlers and young children: ASD specific tool: comparison with controls
Goldm-an (2011)	Relationship between sleep and problem behaviours	Autism Treatment Network registry N = 1784 ASD	2-18 years	Parental concerns questionn aire (PCQ)	Poor sleepers had higher problem behaviours on all subscales Poor sleepers 20% higher to have SIB and 10% higher to have mood swings, aggression and compulsive behaviour 3/4ths of poor sleepers– problems with attention span and social interaction Younger children with poor sleep – more problems with language, aggression, hyperactivity and poor eating habits than older children with poor sleep	Large database sample
Fodstad (2010)	To investigate emergence and aetiology of comorbid conditions in ASD	Early intervention centre N=109 ASD (160 controls)	12-39 mts	BISCUIT – 2 (Baby and Infant Screen for	Co-morbid behaviour emerged at toddler stage (more symptoms for ASD children than controls) and increased as age increased.	Clinic sample: ASD specific tool: comparison with controls

				Children with aUtism Traits –2)		
Levy (2010)	Frequency of co-occurring non-ASD diagnosis and symptoms in ASD	Population representative surveillance health registry cohort N = 2568 ASD	8 years	Surveillance registry data collection method	83% ≥ 1 non-ASD developmental disorder 10% ≥ 1 psychiatric diagnosis 16% ≥ 1 neurological diagnosis 4% - genetic diagnosis	Large health database sample: single item surveillance question
Gau (2010)	Behavioural problems and parenting style in children with autism and their siblings in Taiwan	Samples from 2 medical centres N = 151 autism	3-12 years	CBCL	Children with autism had greater severity in all symptoms of CBCL compared to siblings More attention problems, social problems, thought problems and withdrawal in ASD compared to controls	Clinic sample in Taiwan
Matson (2010)	Interaction between verbal communication skills, social skills and challenging behaviour	Community recruitment from schools, support groups N=109 ASD	3-16 years	ASD-PBC (ASD – Problem Behaviour – Children)	Challenging behaviour correlated with both communication skills and total social skills. Both challenging behaviour and communication skills influenced social skills. Children who had low challenging behaviour, minimal impact of verbal communication skills on social skills. In children with high challenging behaviour, better communication skills predicted worst social skills compared to children with poor communication skills.	Community sample: ASD specific tool

Hess (2010)	Frequency of comorbid psychiatric disorders in children compared to controls	Community recruitment N = 65 ASD (control – 72)	14-16 years	ASD-CC and combined checklist from DSM-4TR and ICD-10	Mean number of comorbid psychiatric disorders higher in ASD children compared to typically developing peers. Mean scores of all subscales higher in ASD than in peers. Factors of worry/depressed, avoidant behaviour, over-eating, under-eating and repetitive behaviour contributed significantly to the difference between groups. Factors of conduct and tantrum behaviour did not contribute significantly to difference.	Small community sample: ASD specific tool: comparison with controls
Joshi (2010)	Examine patterns of psychiatric comorbidity	Referral academic psychopharmacology program N = 217 ASD (non-ASD – 217	3-17 years	Kiddie-SADS	ASD group had higher number of comorbidities compared to non-ASD group. Higher rates of language disorders, multiple anxiety disorders and encopresis in ASD group.	Clinic sample: comparison with controls
Mattila (2010)	Prevalence and types of co-morbid psychiatric disorders in Asperger/High functioning Autism	Combined community and clinic based sample N=50 AS/HFA	9-16 years	Kiddie-SADS	84% ≥ 1 co-morbid lifetime diagnosis Most common – behavioural disorders (44%), anxiety disorders (42%) and tic disorders (26%) Current problems more common in primary school age than in secondary school age Level of functioning in everyday life decreased with the number of co-morbid psychiatric disorders	Combined sample: only AS/HFA
Witwer (2010)	Effect of IQ and language skills	Clinic + community	6-17 years	Nisonger Child	Lower IQ had fewer reported problems on ADHD and GAD	Combined sample

	on behaviour	N = 61 ASD		Behaviour Rating Form	IQ < 70 – sub-syndromal for GAD; IQ ≥ 70 reported more worries OCD symptom not impacted by IQ Non-verbal individuals – sub-syndromal for ODD	
Matson (2010)	Progression of challenging behaviour in ASD	Variety of school and clinic settings N=167 ASD	3-14 years	ASD-PBC	No difference between young children, children and young adolescents on each item of problem behaviour	Combined sample: ASD specific tool
Hartley (2009)	Explore sex difference with behaviour problems in toddlers with ASD	Tertiary autism clinic N = 199 ASD	1.5-3.9 years	CBCL	No difference in total behaviour problems (CBCL composite scores) between boys and girls. Girls - more sleep problems and anxious/depressed affect.	Clinic sample
Matson (2008)	Validity of ASD-CC (ASD-Comorbid for Children)	Variety of school and clinic settings N = 177 ASD	2-17 years	ASD-CC and Behaviour Assessment System for Children-2 (BASC-2)	7 factor structure – internal consistency of each factor between .70 and .86. Strong convergence between ASD-CC score and BASC-2 clinical subscale (r=.66). Construct validity acceptable for each subscale except avoidant behaviour subscale	Combined sample: ASD specific tool validation

Matson (2008)	Reliability of ASD-CC (ASD-Comorbid for Children)	Variety of school and clinic settings N = 113 ASD	2-16 years	ASD-CC	High internal consistency, moderate test-retest and interrater reliability	Combined sample: ASD specific tool reliability
Matson (2008)	Reliability of ASD-BPC (ASD-Problem Behaviours for Children)	Variety of school and clinic settings N=110 ASD (controls-108)	2-16 years	ASD-PBC	High internal consistency, moderate test-retest and interrater reliability	Combined sample: ASD specific tool reliability
Gadow (2008)	Examine mental health risk/protective factors for DSM-IV symptoms in children with ASD and their contribution to functioning	Referral Developmental clinic – consecutive samples N = 238 ASD	6-12 years	Child Symptom Inventory - 4	Combination of multiple risk factors contributed to the variance in severity of psychiatric symptom. Family history of psychiatric illness predicted most psychiatric symptoms except ADHD and specific phobia. Psychotropic medication use predicted ADHD, ODD; pregnancy complications predicted SAD and specific phobia.	Clinic sample
Simonoff (2008)	Identify rates and types of psychiatric co-morbidity in ASD	Population representative Special Needs and Autism Project (SNAP) Cohort N=112 ASD	10-14 years	Parent interview – DSM 4 based diagnosis ; SDQ	70% - 1 condition 41% ≥ 2 conditions Anxiety disorder – 41.8% ADHD – 28.3% No predictor identified	Population representative sample
Xue (2008)	Clinical co-occurrences in	Retrospective chart review	2-18 years	Clinical informatio	52%-sleep disorders 51% - food intolerance	Retrospective chart review

	ASD	of autism clinic sample N = 160 ASD		n form with history, DSM-IV criteria	Mood disorder – 26% Aggressive/self-injurious – 32% Most common aggression toward mother or younger sibling Most common SIB – child biting his or her own hands or forearms AS–more psychiatric disorders; PDD-NOS – more medical disorders	
Hartley (2008)	Prevalence and risk factors of maladaptive behaviour	Autism clinic sample N = 169 autism	1.5 – 5.8 years	CBCL	One-third of children – clinically significant maladaptive behaviour 29.6% - internalising problems; 27.2% - externalising problems Most common – withdrawn (70.4%), attention problems (38.5%) and aggression (22.5%) Multivariate – non-verbal cognition predictor for externalising problems; adaptive behaviour predictor for internalising problems Boys – higher sleep problems Age - +ve correlation with somatic complaints and withdrawal syndrome scales Non-verbal cognition, expressive language and adaptive behaviour - –ve correlation with CBCL attention, aggression and anxious/depressed Autism severity not correlated with any scales	Clinic sample

De Bruin (2007)	Prevalence of co-morbid psychiatric disorders in PDD-NOS	Outpatient clinic sample – consecutive referrals N=94 PDD-NOS	6-12 years	DISC-IV-P Interview	80.9% ≥ 1 and 54.3% ≥ 2 co-morbid psychiatric disorders 61.7% - co-morbid disruptive behaviour disorder and 55.3% - anxiety disorder Children with more social communication deficits – higher chance of co-morbid disorder	Clinic sample: only PDD - NOS
Dominick (2007)	Atypical behaviour in ASD compared to that of HLI	N = 54 ASD (control - 39 Having Language Impairment) Part of another community project	4 year 2 mt - 14 year 2 mt	Atypical Behaviour Patterns Questionnaire (ABPQ)	Concurrent validation with ADI-R and VABS interviews done 98% ASD - 1 atypical behaviour; majority – 2-3 problems Atypical eating, self-injurious behaviour and temper tantrums significantly more in ASD (not abnormal sleep and aggression) Aggression related to both verbal and non-verbal cognition, language measures and autistic symptoms; SIB related to expressive language Low non-verbal IQ and low expressive language predict more atypical behaviour	Community sample: autism specific tool validation
Leyfer (2006)	Development of Autism Co-morbidity Interview – present and lifetime version (ACI –PL)	Community sample N=109 Idiopathic Autism	5-17 years	Autism Co-morbidity Interview	Most common lifetime diagnosis – specific phobia (44%), OCD (37%) and ADHD (31%) Median and mode number of co-morbid diagnoses per child - 3	Community sample: development of autism tool: only idiopathic autism

Le Cavalier (2006)	Examine prevalence of specific behaviour problems and assess the impact of subject characteristics	State evaluation project sample N = 487 PDD	3-21 years	Nisonger Child Behaviour Rating Form	High rates of behaviour and emotional problems Age directly correlated with anxious/insecure subscale Lower adaptive skills associated with less pro-social behaviour, symptoms of anxiety and higher other behavioural problems.	Community sample: only PDD
Le Cavalier (2006)	Correlates of caregiver stress	Community sample N= 293 ASD	9 years	Nisonger Child Behaviour Rating Form	Behavioural problems stable over 1-year period; Parenting stress and change in stress predicted behavioural problems	Community sample evaluating behaviour and parent stress

ABC – Aberrant Behaviour Checklist, ABPQ – Atypical Behaviour Patterns Questionnaire, ASD-CC - ASD-Comorbid for Children, ASD-PBC – ASD-Problem Behaviour Children, BISCUIT – Baby and Infant Screen for Children with aUtism Traits , CBCL – Child Behaviour Check List, CSHQ – Children's Sleep Habits Questionnaire, Kiddie SADS interview - Schedule for Affective Disorders and Schizophrenia for School-Age children, SDQ - Strengths and Difficulties Questionnaire, SSP – Short Sensory Profile

3.2.3 Prevalence of co-existing conditions in children with ASD

Reviews have reported that children with ASD had increased rates of co-existing conditions in comparison with typically developing children and these high rates persisted throughout the lifespan (Matson and Nebel-Schwalm, 2007b; Mannion and Leader, 2013b). There are conflicting reports about the frequency of co-existing conditions in children with ASD. A surveillance report of eight-year-olds with ASD showed 10% having at least one psychiatric diagnosis, but this was based on health surveillance parent-reports of definite diagnosis (Levy et al., 2010). Other studies reported high frequency of co-existing conditions in children with ASD: the median number being three per child with 70% of children with ASD having one condition and more than half having four or more frequent conditions (Leyfer *et al.*, 2006; de Bruin *et al.*, 2007; Simonoff *et al.*, 2008; Hess *et al.*, 2010; Joshi *et al.*, 2010; Levy *et al.*, 2010; Mattila *et al.*, 2010; Gjevik *et al.*, 2011; Maskey *et al.*, 2013). There are concurring reports of high frequency of co-existing conditions from non-western countries (Gau *et al.*, 2010) as well as population representative samples in the UK (Simonoff *et al.*, 2008; Maskey *et al.*, 2013). The common co-existing conditions include eating problems, sensory reactions, anxiety disorder, ADHD and ODD (Leyfer *et al.*, 2006; Mattila *et al.*, 2010; Huang *et al.*, 2013; Kaat *et al.*, 2013; Mannion and Leader, 2013a; Maskey *et al.*, 2013).

Evaluating literature about problem behaviours, high rates of behaviour problems ranging from 24% to 73.9% are reported in children and young people with ASD (Brereton *et al.*, 2006; Herring *et al.*, 2006; Lecavalier, 2006; Dominick *et al.*, 2007; Hartley *et al.*, 2008; Gau *et al.*, 2010; Hattier *et al.*, 2011). The problem behaviours include internalising (including anxiety/depression, withdrawal, and somatic complaints) and externalising behavioural problems (including attention problems, aggressive behaviour, and rule-breaking actions) (McFarlane J, 2003; Skokauskas and Gallagher, 2012). .

There is only a limited literature evaluating progression of co-existing conditions in children with ASD. Population representative studies in adolescents with ASD have shown that the high frequency of comorbid psychiatric problems persisted over time (Simonoff et al., 2008; Simonoff et al., 2013). In contrast a pattern of increase over time was shown in an early childhood study, which showed co-

morbid conditions emerging at toddler stage and increasing with growing age (Fodstad et al., 2010). There was no longitudinal study evaluating the progression of problem behaviours. In a cross-sectional study, each problem behaviour was shown to be persistent in young children, children and young adolescents (Matson et al., 2010).

3.2.4 Predictors of co-existing conditions in children with ASD

Reviews have identified that factors such as age, gender and intellectual disability, which affect comorbid psychopathology in the general population, have been studied in children with ASD (Gadow *et al.*, 2008; Witwer and Lecavalier, 2010; Kaat *et al.*, 2013; Mannion and Leader, 2013a; Mannion and Leader, 2013b; Mannion *et al.*, 2013; Maskey *et al.*, 2013; Simonoff *et al.*, 2013).

There are studies evaluating the influence of age on co-existing conditions. Age did not influence psychiatric problems in a longitudinal analysis of psychiatric problems in adolescents with ASD (Simonoff et al., 2013). Age was again not a significant predictor for co-morbid psychopathology in a recent community based study of children and adolescents by Mannion and Leader, 2013 (Mannion and Leader, 2013a). Matson et al., 2010 evaluated a combined clinic and community sample of children and adolescents and reported no difference between young children, children and young adolescents on each item of problem behaviour (Matson et al., 2010). There are however contradicting findings, showing lower age predicting more behavioural problems (Mattila *et al.*, 2010; Maskey *et al.*, 2013) and aggression (Mazurek et al., 2013); while higher age has been reported to correlate with anxious/insecure state (Lecavalier, 2006). Of note among these studies is the population representative study by Maskey and colleagues which reported lower age as a significant predictor for the frequency of co-existing conditions (Maskey *et al.*, 2013).

Boys are reported to have more repetitive behaviour, externalising and social problems, while girls more sleep problems and anxious/depressed affect (Hartley et al., 2008; Hartley and Sikora, 2009; Mandy et al., 2012; Werling and Geschwind, 2013). Hartley and Sikora explored gender differences in a clinic sample of toddlers and reported no difference in total problem behaviour

between boys and girls, but more sleep problems and anxious/depressed affect in girls (Hartley and Sikora, 2009). Population representative studies have described minimal influence of gender in rates of co-existing conditions in children and adolescents with ASD (Simonoff *et al.*, 2008; Maskey *et al.*, 2013; Simonoff *et al.*, 2013).

Evaluating the influence of cognition on co-existing conditions, there are conflicting reports. In a recent study by Mannion and Leader on a community sample of children and adolescents with ASD, intellectual disability did not predict co-morbid psychopathology (Mannion and Leader, 2013a). Initial evaluation around 12 years of age of the population representative Special Needs and Autism Project (SNAP) cohort revealed no significant predictors, including IQ for psychiatric co-morbidity (Simonoff et al., 2008). However, the longitudinal follow-up study of this cohort at 16 years of age showed that lower IQ at 12 years of age predicted total difficulties and hyperactivity scores on SDQ at 16 years of age (Simonoff et al., 2013). Higher rates of co-existing conditions were reported in children with lower IQ (Amr et al., 2012), lower expressive language (Hartley *et al.*, 2008; Maskey *et al.*, 2013) and higher social communication deficits (de Bruin et al., 2007).

Xue and colleagues in a retrospective chart review of children and adolescents with ASD identified that children with Asperger syndrome (AS) had more psychiatric disorders and children with PDD-NOS had more medical disorders (Xue et al., 2008). Tonge and colleagues in a clinic sample of children and adolescents with AS and HFA reported that AS had higher levels of psychopathology than High Functioning Autism (HFA) (Tonge et al., 1999). However, population representative studies have shown no significant contribution of either autism severity or type of autism to the frequency of co-existing conditions (Simonoff *et al.*, 2008; Maskey *et al.*, 2013; Simonoff *et al.*, 2013).

3.2.5 Measures of co-existing conditions in children with ASD

Co-existing conditions can be measured using parent report questionnaires, parent interviews, child/adolescent self-report questionnaires, child/adolescent interviews and direct observations of the individual in different settings. Refer to

section 1.7.2 (page 7) for discussion about dilemmas of diagnosing co-existing conditions in children with ASD.

A recent review by Mannion and Leader reported the lack of valid and reliable diagnostic instruments to measure co-existing conditions in children with ASD (Mannion and Leader, 2013b). Studies have predominantly used parent report questionnaires developed for the general population and for other specific groups such as intellectual disability (ID).

Parent report questionnaires to measure behavioural problems in the general population such as the Child Behaviour Check List (CBCL) (Achenbach, 1991), the Behaviour Assessment System for Children-2 (BASC-2) (Reynolds, 2004), the Child Symptom Inventory-4(CSI-4) (Gadow, 2002) and the Strengths and Difficulties Questionnaire (SDQ) (Goodman, 2001) are reliable tools used extensively for both clinical and research purposes. These questionnaires developed for the general population have been used in children with ASD (Brinkley *et al.*, 2007; Gadow *et al.*, 2009; Pandolfi *et al.*, 2012; Skokauskas and Gallagher, 2012). The challenge of identifying and discriminating co-existing conditions from core features of ASD becomes magnified with the use of these measures meant for the general population (Matson et al., 2008b). Psychometric evaluation of the CBCL in ASD through an archival data analysis showed good sensitivity of subscales but low specificity for detecting co-occurring conditions in children with ASD (Pandolfi et al., 2012). Similarly, evaluation of the CBCL in another community study of children with autistic disorder showed higher relative scores on subscales of attention, social problems and thought problems and relative low scores on subscale of somatic complaints (Bölte et al., 1999). A recent study evaluating the longitudinal progression of psychiatric disorders in the population representative SNAP cohort used ASD-specific cut-offs for the SDQ (Simonoff et al., 2013). This study reported that the general population cut-offs identified a larger proportion of individuals as affected compared to ASD-specific cut-offs, with the exception of ADHD.

Some measures have been developed specifically for children with ID, for example the Aberrant Behaviour Checklist (ABC) (Aman et al., 1985) and the Nisonger Child Behaviour Rating Form (NCBRF) (Aman et al., 1996). Factor

analysis of the ABC in children and youth with ASD revealed a moderate fit for a five factor structure; but there was discrepancy in this structure for a subset of individuals with high self-injury (Brinkley et al., 2007).

Of late, measures have been developed specifically to address the inherent defects noted with the more generic measures, and effectively measure co-existing conditions in the ASD population (Matson et al., 2009b). The Autism Spectrum Disorder Comorbidity Child Version (ASD-CC) (Matson and Wilkins, 2008) evaluates co-morbid psychopathology in children with ASD and has moderately good inter-rater (κ=.46) and test-retest reliability (κ=.51) (Matson and Wilkins, 2008; Thorson and Matson, 2012). It has good internal consistency (α=.91). The subscales of the ASD-CC except avoidant behaviour were shown to have acceptable construct validity by correlation with subscales of the BASC-2 (Matson et al., 2009b). Matson has included the Autism Spectrum Disorder Problem Behaviour Child Version (ASD-PBC) (Matson et al., 2008a) in the same assessment battery for children with ASD to evaluate problem behaviours. It has acceptable reliability and validity (Matson et al., 2008a; Mahan and Matson, 2011; Beighley et al., 2013). Both ASD-CC and ASD-PBC have been used in other community and clinic based studies (Davis III et al., 2010; Davis Iii et al., 2011b; Mannion et al., 2013; Rieske et al., 2013). Appendix 1 provides detailed information on the psychometric properties including strengths and weaknesses of the measures considered for this study.

3.2.6 Summary of literature review on co-existing conditions

Children with ASD have a high frequency of co-existing conditions compared to their peers. There are conflicting reports about the prevalence and predictors of co-existing conditions in children with ASD. There is a lack of appropriate instruments to measure these co-existing conditions in children with ASD. Of late, there are attempts to develop ASD-specific tools and to adapt other behavioural assessment tools for children with ASD.

3.3 Anxiety in children with ASD

Anxiety is a significant problem for individuals with ASD (Rodgers et al., 2012). For the purpose of this thesis, anxiety will be defined as: parent report of anxiety disorder or fulfils the DSM-IV criteria for an anxiety disorder or scores above the cut-off scores for anxiety on a standardised measure of anxiety(van Steensel et

al., 2011). The search term used was 'anxiety'. The results of the literature search are summarised in Appendix 2. 104 articles were identified after initial screening. The inclusion criteria of 3.1.1 (sample size above 50 and parent reporting of symptoms) were used to identify relevant articles for detailed review.

3.3.1 Results of literature review

Table 5 summarises the relevant studies of anxiety in children with ASD considered for this study. Van Steensel's meta-analysis of anxiety disorders in ASD (van Steensel et al., 2011) and Grondhuis's review article on assessment of anxiety in ASD (Grondhuis and Aman, 2012) were particularly helpful.

3.3.2 Prevalence and predictors of anxiety

Van Steensel and colleagues in a meta-analysis reported a frequency rate of 39.6% of co-morbid anxiety disorders in children with ASD (van Steensel et al., 2011) compared to 5% in the general population (King and Ollendick, 1997). The most frequent subtype identified across studies was 'specific phobia (29.8%), followed by OCD (17.4%) and social anxiety disorder (16.6%)'.

Subsequent studies have reported a frequency rate ranging from 15.7% to 58% (Skokauskas and Gallagher, 2012; Kaat *et al.*, 2013; Mannion *et al.*, 2013; Maskey *et al.*, 2013). One study of a community recruited sample of ASD based on parent report of co-occurring conditions reported a rate of 15% (Mannion et al., 2013). Another study of a population representative sample of ASD based on parent report of behavioural concerns found prevalence rate of 42% (Maskey *et al.*, 2013).

The moderator analyses done in the recent meta-analysis reported age as significant moderator for anxiety; higher age was found to correspond to higher prevalence rates in anxiety in general and generalised anxiety disorder (GAD) and lower age corresponded to higher rates in obsessive compulsive disorder (OCD) and separation anxiety disorder (van Steensel et al., 2011). Evaluation of anxiety symptoms across the lifespan in a mixed sample study reported that anxiety rose from toddlerhood to childhood, then decreased to early adulthood and again increased in late adulthood (Davis Iii et al., 2011a). A recent community based study of 3-16 year olds (Mannion and Leader, 2013a) and

another using ASD specific tool in 2-14 year olds (Davis Iii et al., 2011b) reported age as not being a significant predictor for anxiety. Both these studies however had comparatively smaller sample sizes of less than 100 children with ASD.

Table 5 : Summary of relevant studies evaluating anxiety in children with ASD (Reverse chronological order)

Author (date)	Research question	Sampling (N = number of participants)	Age	Tools used for anxiety	Main findings	Brief critique
Lidstone (2014)	Relationship between repetitive behaviour, anxiety and sensory features in ASD	2 different study databases N =120 ASD	2-17 years	Spence Children's Anxiety Scale- Parent version (SCAS-P)	Anxiety related to repetitive behaviour not age; anxious children had more sensory sensitivity and sensation avoidance than non-anxious; Relation between anxiety and insistence on sameness mediated by sensory avoidance and sensory sensitivity	Relation between anxiety, sensory problems and repetitive behaviour
Mannion (2013)	Analysis of predictors of psychopathology, gastrointestinal problems and epilepsy in ASD	Community recruitment (schools, parent groups) N = 89	3-16 years	Parent report of diagnosis	Gender (being male) and co-morbid psychopathology (especially worry/depressed subscale and avoidant behaviour subscale) predicted anxiety disorder (R^2=.897). Other factors considered age, ID, epilepsy, ADHD, sleep and gastrointestinal problems.	Worry/depressed subscale in ASD-CC is another measure of anxiety

Rieske (2013)	Explore effect of autism symptomatology on anxiety N = 2366 including atypically developing, autistic and PDD-NOS	Early intervention group sample	17-36 months	BISCUIT – 2 (Baby and Infant Screen for Children with aUtism Traits – 2)	Autism symptomatology accounted for 50% of variance in total anxiety scores	Infant and toddler group study: ASD specific tool: comparison with controls
Kaat (2013)	Relation of psychiatric symptom-induced impairment to mental health in children with ASD	Referral developmental disability clinic – consecutive referrals N = 115 ASD	6-12 years	Child and Adolescent Symptom Inventory DSM-4 referenced rating scales	Parent report: Anxiety Disorder - 47%	Clinic sample
Mannion (2013)	Co-morbid psychopathology, gastrointestinal symptoms, sleep problems and epilepsy in ASD	Community recruitment (schools, parent groups) N = 89	3-16 years	Parent report	Anxiety – 15.7%	Community sample: ASD specific tool
Maskey (2013)	Predictors and frequency of co-existing emotional and behavioural conditions in children with ASD	Population representative database N = 863 ASD	2-18 years	Single item parent report	Anxiety – 42%	Population representative ; single item

Davis (2012)	Understand effect of communication on anxiety in 3 groups of infant and toddlers: autistic disorder, PDD-NOS and controls (atypically developing)	Early intervention group sample N = 107 Autistic N=110 PDD-NOS N=518 controls	15-36 months	BISCUIT – 2	Anxiety more in autistic than PDD-NOS Age (older age), type of diagnosis and communication skills (not gender) predicted anxiety. Anxiety increased as communication (both receptive and expressive) increased for both autistic and PDD-NOS; not in controls	Only infant and toddler sample: ASD specific tool: comparison with controls
Skokauskas (2012)	Patterns of comorbid psychiatric problems in children with ASD and their parents compared to controls and their parents	Community recruitment N = 67 ASD (Control – 67)	6-18 years	CBCL (Child Behaviour Check List)	ASD Children – Anxiety – 46.2%	Community sample
Davis (2011)	Relationship between anxiety, communication skills and autism	Combined sample N = 33autistic N=33 PDD-NOS N=TD (typically developing)	2-14 years	ASD-CC (Autism Spectrum Disorders – Comorbidity for Children)	Boys more anxious; age not significant Anxiety decreased as communication deficits increased for autism diagnosis. For children with PDD-NOS, anxiety increased as communication deficits increased	Combined sample: ASD specific tool: comparison with controls
Davis (2011)	Anxiety symptoms across lifespan	Multiple methods; combined sample	18 months - 65	BISCUIT-II, ASD-CC, ASD-CA (Autism	Anxiety rose from toddlerhood to childhood; decreased from childhood to early adulthood; increased again from early	1st lifespan study of anxiety; but not

		N = 131ASD	years	Spectrum Disorders-Comorbidity for Adults)	adulthood to older adulthood	longitudinal
Davis (2010)	Compare anxiety between 3 groups of toddlers: autistic disorder, PDD-NOS and controls (atypically developing)	Early intervention group sample N = 159 Autistic N=154 PDD-NOS N=200 controls	17-37 months	BISCUIT-II	More severe anxiety symptoms in autistic > PDD-NOS > controls	ASD specific tool: comparison with controls
Hess (2010)	Frequency of comorbid psychiatric disorders in children compared to controls	Community recruitment N = 65 ASD (control – 72)	14-16 years	ASD-CC and combined checklist from DSM-4TR and ICD-10	Mean of worry/depressed subscale higher in ASD than in peers: contributed significantly to the difference between groups	Small community sample: ASD specific tool: comparison with controls
Witwer (2010)	Effect of IQ and language skills on behaviour	Clinic + community N = 61 ASD	6-17 years	Nisonger Child Behaviour Rating Form	IQ < 70 – sub-syndromal for GAD; IQ ≥ 70 reported more worries	Combined sample

ASD-CC - ASD-Comorbid for Children, ASD-CA – ASD – Comorbidity for Adults, ASD-PBC – ASD-Problem Behaviour Children, BISCUIT – Baby and Infant Screen for Children with aUtism Traits , CBCL – Child Behaviour Check List, SCAS – Spence Children's Anxiety Scale

Similar to other behavioural problems, anxiety also can be affected by gender differences. A recent study by Mannion and Leader in children with ASD reported more anxiety symptoms in boys; however, the sample size was less than 100 and the diagnosis was by a single item parent report (Mannion and Leader, 2013a). A study of children with ASD using an ASD specific tool found similar finding, but the sample size was again less than 100 (Davis Iii et al., 2011b). This difference between genders was not found in a study of toddlers using an ASD specific tool (Davis et al., 2012).

The meta-analysis (van Steensel et al., 2011) reported that lower mean IQ was associated with higher frequency rates of anxiety in general and social anxiety disorder; even when multiple moderators were included in the model. Higher mean IQ was associated with higher rates of OCD and separation anxiety disorder, but not when multiple moderators were included.

As per the meta-analysis (van Steensel et al., 2011), ASD subtype was found to be significant for anxiety in general and all anxiety subtypes. This analysis reported that anxiety was more likely to be reported in PDD-NOS followed by autistic disorder and AS. Davis evaluated toddlers with ASD using an ASD specific tool and reported more severe anxiety symptoms in autistic children than in PDD-NOS (Davis III et al., 2010). Another study of toddlers, including ASD and atypically developing controls, produced a similar finding, where autism symptomatology accounted for 50% of variance in total anxiety scores (Rieske et al., 2013).

3.3.3 Measures of anxiety

A recent review highlighted the unique challenges presented by assessment methods of anxiety in children with ASD: overlapping anxiety symptoms with presentations of autism and the inability of individuals with communication and cognitive deficits to convey their anxiety accurately (Grondhuis and Aman, 2012). The review evaluated both measures analysing anxiety in general and those investigating different subtypes of anxiety. Both the Autism Spectrum Disorders – Comorbidity for Children (ASD-CC) and the Baby and Infant Screen for Children with aUtism Traits – 2 (BISCUIT – 2) were identified as parent report measures empirically derived for ASD with acceptable psychometric

properties. But both these measures investigated anxiety in general without evaluating specific subtypes. Two popular parent report measures used in the general population to analyse different subtypes of anxiety, the Spence Children's Anxiety Scale (SCAS) (Spence et al., 2003) and the Screen for Child Anxiety-Related Emotional Disorder (SCARED) (Wren et al., 2007) were lacking in validation for ASD. Subsequent to the initial review, the SCARED was shown to have acceptable psychometric properties in a clinic-based sample of high functioning ASD, though the parental cut-offs had to be raised in the ASD group to improve specificity (Van Steensel et al., 2013). Appendix 1 summarises the measures considered for this study including their psychometric properties.

3.3.4 Summary of literature review about anxiety in children with ASD

Children with ASD have a high frequency of anxiety disorders, with an average prevalence rate of 40%. The moderators evaluated in literature include age, IQ, and gender. There is ongoing research to validate measures analysing subtypes of anxiety for the population with ASD.

3.4 Hyperactivity in children with ASD

The diagnosis of Attention Deficit Hyperactivity Disorder (ADHD) was excluded in the presence of ASD as per both DSM-IV TR and ICD-10 (Taurines et al., 2012; Matson et al., 2013). The recent DSM-V has rectified this difficulty and ASD and ADHD are not considered mutually exclusive anymore (APA, 2013).

For the purpose of this thesis, hyperactivity will be defined as: parent report of hyperactivity or fulfils the DSM-IV criteria for ADHD or scores above the cut-off scores for hyperactivity/inattention/ADHD on a standardised measure. The search term used was 'hyperactivity'. The results of the literature search are summarised in Appendix 2. 72 articles were identified after the initial screening in addition to the previously analysed literature. The inclusion criteria of 3.1.1 (sample size above 50 and parent reporting of symptoms) were used to identify relevant articles for detailed review.

3.4.1 Results of literature search

Table 6 summarises the relevant studies evaluated in the literature review. There have been some recent review articles evaluating ADHD in children with ASD (Taurines et al., 2012; Matson et al., 2013; Mannion and Leader, 2014a).

3.4.2 Prevalence and predictors for ADHD

A recent review reported the prevalence of Attention Deficit/Hyperactivity Disorder (ADHD) in the ASD population to be between 20 to 70% (Matson et al., 2013). The inattentive type has been reported as the most common subtype (Leyfer et al., 2006; Gjevik et al., 2011). The review by Mannion and Leader identified many correlates for children with ASD and ADHD including autism severity, adaptive functioning, social impairment and quality of life (Mannion and Leader, 2014a). In a large network based sample of 3066 children with ASD, the combined ASD and ADHD group had lower adaptive functioning and poorer health related quality of life compared to the group with ASD alone (Sikora et al., 2013). In a recent study of a comparatively smaller sample, children with both ASD and ADHD had lower cognitive functioning, higher social impairment, more stereotypic and repetitive behaviour and impaired adaptive functioning (Rao and Landa, 2014). This study also reported more boys in the combined ASD and ADHD group than in the ASD only group; age, ethnicity and socio-economic state had no associations. The first longitudinal study of a population-representative sample reported that lower IQ and adaptive functioning at 12 years predicted higher hyperactivity at 16 years (Simonoff et al., 2013).

Table 6 : Summary of important studies evaluating hyperactivity in children with ASD (Reverse chronological order)

Author (date)	Research question	Sampling (N=number of participants)	Age	Tools used for ADHD	Main findings	Brief critique
Rao (2014)	Association between behaviour, ADHD and ASD	Combined sample from community and clinics N = 62 ASD	4-8 years	BASC-2: hyperactive and inattention subscales	29% of ASD had ADHD: 33% hyperactive, 33% attention problems and 33% combined More males in ASD+ ADHD group than ASD only group (other factors considered: age, ethnicity, SES) ASD+ADHD had lower cognitive functioning, lower adaptive skills, more social skills impairment and more stereotypic and repetitive behaviour	Combined sample
Steijn (2014)	Relationship between ASD, ADHD, depression symptoms and stress in parents	Recruited from family genetic studies N = 174 ASD	2-20 years	Conner's Long Version Rating Scales Revised	Parents have more stress with ASD+ADHD Parental depression more when child has ASD or ASD+ADHD Paternal ASD, maternal ADHD – increased parental stress	Effect of parental factors on parental stress

Mannion (2013)	Analysis of predictors of psychopathology, gastrointestinal problems and epilepsy in ASD	Community recruitment (schools, parent groups) N = 89 ASD	3-16 years	Parent report	No predictors for ADHD (factors considered age, gender, ID, comorbid psychopathology, sleep problems and gastrointestinal problems)	Community sample, ASD specific tool; smaller sample size
Simonoff (2013)	Longitudinal analysis of persistence of psychiatric problems in ASD and predictors	Population representative Special Needs and Autism Project (SNAP) Cohort N = 81 ASD	12 followed to 16 years	Strengths and Difficulties Questionnaire (SDQ)	ASD specific cut-offs lower than population specific cut-offs for hyperactivity on SDQ. Using ASD specific cut-offs 23.3% hyperactive at 12 years and 10.6% at 16 years. Lower IQ and adaptive functioning at 12 years predicted higher hyperactivity at 16 year	1st longitudinal study: Population representative: used ASD cut offs of SDQ
Kaat (2013)	Relation of psychiatric symptom-induced impairment to mental health in children with ASD	Referral developmental disability clinic – N = 115 ASD	6-12 years	Child and Adolescent Symptom Inventory	Most common – ADHD (67%)	Clinic sample
Huang (2013)	Examine effects of autistic behaviour and individual emotional and behavioural problem on parenting stress	Recruited from 5 hospitals and 2 clinics N = 52 Autism	3-12 years	SDQ	ASD children at high risk for problems of hyperactivity/Inattention – 55.8%;	Clinic sample

Mannion (2013)	Co-morbid psychopathology, gastrointestinal symptoms, sleep problems and epilepsy in ASD	Community recruitment (schools, parent groups) N = 89	3-16 years	Parent report	ADHD -18%	Community sample; ASD specific cut-off: one item parent report
Sikora (2013)	Frequency of ADHD in ASD	Recruited from ATN (Autism Treatment Network) N = 3066 ASD	2-17.9 years	CBCL – 2 scales: attention problem and attention deficit hyperactivity problem	40% - 1 elevated T score 19% - both T scores elevated ADHP scale: 2-5 years age group lower % than 6-11 and 12-17 years groups Age did not affect combined both scales elevated ASD+ADHD group had lower adaptive score and poorer health related quality of life	Large network based sample
Maskey (2013)	Predictors and frequency of co-existing emotional and behavioural conditions in children with ASD	Population representative database N = 863 ASD	2-18 years	Parent report	Parent report of ADHD: 9-17%; frequent hyperactivity – 42%	Population representative; one item about hyperactivity
Mandy (2012)	Sex differences in children with ASD	Specialist clinic sample N=325 ASD	3-18 years	SDQ	Parent report – girls and boys similar hyperactivity Teacher report – boys more hyperactive at school	Clinic sample
Skokauskas (2012)	Patterns of comorbid psychiatric problems in children with ASD compared to controls	Community recruitment N = 67 ASD (Control – 67)	6-18 years	CBCL	ADHD – 44.78%: borderline clinically significant ADHD difficulties – 47%	Community sample

Gjevik (2011)	DSM-IV disorders in ASD	Special school population N = 71 ASD	6 – 17.9 years	Kiddie SADS interview (Schedule for Affective Disorders and Schizophrenia for School-Age children	ADHD – 31% ODD/Conduct – PDD-NOS	School sample
Antshel (2011)	Effect of ADHD on social skills group intervention treatment efficacy in children with ASD	Community N = 83 ASD	7 – 12 years	Parent report of ADHD	ADHD affected the improvement in social skills of children with ASD	ADHD affects intervention outcome
Simonoff (2008)	Identify rates and types of psychiatric co-morbidity in ASD	Population representative SNAP Cohort N=112 ASD	10-14 years	Parent interview – DSM 4 based diagnosis	ADHD – 28.3%	Population representative sample

BASC – Behaviour Assessment System for Children, CBCL – Child Behaviour Check List, Kiddie SADS interview - Schedule for Affective Disorders and Schizophrenia for School-Age children, SDQ - Strengths and Difficulties Questionnaire

3.4.3 Measures of ADHD

Matson and his colleagues in their review evaluated different measures to assess ADHD in children with ASD and reported that methods are in the process of being developed (Matson et al., 2013). The authors suggested use of several measures in combination to assess ADHD to improve diagnosis. The review by Matson and colleagues evaluated a new measure being developed for ASD and ADHD: the Multi-dimensional Scale for Pervasive Developmental Disorders and Attention – Deficit/Hyperactivity Disorder (MSPA) (Funabiki et al., 2011). This measure has 14 domains covering ASD, ADHD, Developmental Co-ordination Disorder, sensory, sleep disturbance, learning and language development; but has to be rated by a trained professional. The measure can be of use in clinical practice as it provides a profile of strengths and difficulties of each child.

Among parent report measures, studies have used subscales of behavioural measures such as the CBCL, the BASC-2 and the SDQ (Simonoff *et al.*, 2008; Mandy *et al.*, 2012; Huang *et al.*, 2013; Sikora *et al.*, 2013; Simonoff *et al.*, 2013; Rao and Landa, 2014). General population based cut-offs are not ideal for the ASD population. In the population-representative longitudinal study of SNAP cohort, ASD specific cut-offs for hyperactivity in the SDQ were lower than the normal population-specific cut-offs (Simonoff et al., 2013). Other measures are yet to undergo a similar validation for the ASD population. Conners' Rating Scales–third edition is a parent/teacher/self-report measure to obtain information about inattention, hyperactivity and oppositional behaviour (Conners, 1989). This measure has been used in correlational and intervention studies in populations with ASD (Posey *et al.*, 2006; Gjevik *et al.*, 2011; Van Steijn *et al.*, 2012). Appendix 1 provides a summary of measures considered for this project.

3.4.4 Summary of literature review about ADHD in children with ASD

The recent DSM-V criteria revised the previous standards of not diagnosing ADHD in the presence of ASD. The prevalence of ADHD in ASD was reported to be between 20-70%. Specific measures to assess ADHD in ASD are in the process of being developed.

3.5 Sleep problems in children with ASD

Sleep problems are recognised as common co-existing conditions in children with ASD and are shown to affect health related quality of life in children and parental well-being including parental mental health (Lopez-Wagner et al., 2008; Hodge et al., 2013; Delahaye et al., 2014). For the purpose of this thesis, sleep problems were defined as: parent report of sleep problems or sleep problems detected in parent-report sleep diary or sleep observations or scores above the cut-off scores for sleep problems on a standardised measure. The search term used was 'sleep'. The results of the literature search are summarised in Appendix 2. 117 articles were identified after the initial literature screening in addition to the previously analysed literature. The inclusion criteria of 3.1.1 (sample size above 50 and parent reporting of symptoms) were used to identify relevant articles for detailed review.

3.5.1 Results of literature search

Table 7 summarises the relevant articles evaluated in the literature review. Review articles have evaluated sleep problems extensively in children with ASD (Cortesi et al., 2010; Hollway and Aman, 2011; Spruyt and Gozal, 2011; Hodge et al., 2012; Mannion and Leader, 2013c).

3.5.2 Prevalence and predictors of sleep problems

The different reviews reported high prevalence of sleep problems in children with ASD (Cortesi et al., 2010; Hollway and Aman, 2011; Hodge et al., 2012; Mannion and Leader, 2013c). The frequency rates are reported to be between 33%-80.9% by different studies (Dominick *et al.*, 2007; Goldman *et al.*, 2011; Goldman *et al.*, 2012; Hodge *et al.*, 2013; Mannion and Leader, 2013a; Maskey *et al.*, 2013). A population-based study reported the frequency of sleep problems as 39.3% in ASD, compared to 3.6% in typically developing children; children with ASD had ten times more insomnia than their peers (Sivertsen et al., 2012). Delayed sleep initiation, frequent night waking and reduced sleep duration are the most common problems. In a recent longitudinal evaluation of sleep patterns in children aged 6 months to 11 years of age, there was no difference in sleep duration in infancy, but children with ASD had lesser night time sleep duration from 30 months of age onward compared to peers (Humphreys et al., 2014).

Table 7: Summary of relevant studies evaluating sleep problems in children with ASD (Reverse chronological order)

Author (date)	Research question	Sampling (N = number of participants)	Age	Tools used for sleep	Main findings	Brief critique
Delahaye (2014)	Relationship between sleep problems in ASD and health related quality of life (HRQoL) of parents	Clinic sample N = 86 ASD	4-12 years	Children's Sleep Habits Questionnaire (CSHQ)	Negative relationship between sleep disturbance and HRQoL (total, physical and psychosocial) of parents Sleep duration and sleep anxiety negatively correlated with HRQoL	Clinic sample
Humphreys (2014)	Longitudinal sleep patterns in children with ASD	Clinic sample N = 73 ASD	6 months – 11 years	Sleep data questionnaire	No difference in sleep duration in infancy; lesser night time sleep duration between 30 months – 11 years age in ASD. Frequent night time wakings from 30 months of age;	Longitudinal study of sleep in ASD
Mannion (2013)	Analysis of predictors of psychopathology, gastrointestinal problems and epilepsy in ASD	Community recruitment (schools, parent groups) N = 89 ASD	3-16 years	CSHQ	80.9% - Sleep problems Avoidant behaviour, under-eating and GI symptom inventory total score predicted sleep problems (R^2=.259) specially sleep anxiety (R^2=.138), parasomnias ((R^2=.132) and daytime sleepiness (R^2=.187)	Community sample

Hodge (2013)	Relationship between maternal stress and children's sleep in ASD	From research dataset N = 90 ASD and 90 TD controls	4-12 years	CSHQ	More sleep problems in ASD than TD Maternal sleep and maternal stress mediated the relationship between children's sleep and mother's mental health for mothers of both ASD and TD	Comparison with TD
Hollway (2013)	Correlates and risks for sleep problems for children in Autism Treatment Network	Autism Treatment Network Database N = 1583 ASD	2- 17 years	CSHQ	Anxiety, autism symptom severity, sensory sensitivities (under-responsive/sensory seeking, auditory filtering and taste/smell sensitivity subscales) and GI problems related with sleep problems: IQ positively predicted sleep anxiety: Age and communication negatively predicted total sleep problem: AS more vulnerable. Total R^2 = .201	Large database study
Kozlowski (2012)	Sleep and feeding problems in toddlers with ASD	Research database N =506 autistic N = 522 PDD-NOS N= 719 atypical development	17 – 37 months	BISCUIT-part 2	Children with autistic disorder significantly more sleep and feeding problems than PDD-NOS. PDD-NOS more problems than atypically developing group. No effect of age and gender	Toddler group studied
Maskey (2013)	Predictors and frequency of co-existing emotional and behavioural conditions	Population representative database N = 863 ASD	2-18 years	Parent report	Frequent sleep problems – 45%	Single item report: population representative sample

Goldman (2012)	Variations in parental sleep concerns with age	Autism Treatment Network sample N = 1859 ASD	3-18 years	CSHQ	Total sleep problems significant across all age groups Adolescents, older children – delayed sleep onset, shorter sleep duration and daytime sleepiness Younger children – bedtime resistance, sleep anxiety, parasomnias, night wakings	Large database sample analysing effect of age on sleep problems
Mazurek (2012)	Prevalence and correlates of physical aggression in ASD with sleep	Autism Treatment Network registry N=1584 ASD	2-17 years	CSHQ	Aggression associated sleep problems Multivariate model –sleep problems strongly associated with aggression	Single item on a large database sample
Goldman (2011)	Relationship between sleep and problem behaviours	Autism Treatment Network registry N = 1784 ASD	2-18 years	CSHQ	Poor sleepers had higher problem behaviours on all subscales Poor sleepers 20% higher to have SIB and 10% higher to have mood swings, aggression and compulsive behaviour ¾ths of poor sleepers– problems with attention span and social interaction 1 unit increase in parasomnia, 20% increase in anxiety, sensory issues, aggression, hyperactivity, attention, mood swings and SIB.	Large database sample
Hartley (2009)	Explore sex difference with behaviour problems in toddlers with ASD	Tertiary autism clinic N = 199 ASD	1.5-3.9 years	CBCL subscale of sleep problems	Girls – more sleep problems	Clinic sample: toddler sample

Mayes (2009)	Variables related to sleep problems in ASD	Retrospective analysis of clinical data N = 477 Autism	1-15 years	10 sleep items on the Paediatric Behaviour Scale (PBS)	Most common sleep problems – difficulty falling asleep and restlessness during sleep Sleep problems not related to age, IQ, SES, gender, learning ability and neuropsychological functioning Autism severity and co-existing conditions increased sleep problems Strongest predictors – autism severity, hyperactivity, aggression and mood variability	
Xue (2008)	Clinical co-occurrences in ASD	Retrospective chart review of autism clinic sample = 160 ASD	2-18 years	Parent information form	52%-sleep disorders	Retrospective chart review
Hartley (2008)	Prevalence and risk factors of maladaptive behaviour	Autism clinic sample N = 169 Autism	1.5 – 5.8 years	CBCL subscale of sleep problems	Boys – higher sleep problems	Clinic sample
Giannotti (2008)	Sleep, epilepsy and EEG in developmentally regressed children	Clinic sample N = 104 autism (70 regressed, 34 regressed) and 162 TD	30 months – 8 years	CSHQ	Regressed group – more circadian rhythm disorders – higher bedtime resistance, sleep onset delay, higher night-wakings and lower sleep duration. Regression – increased sleep disorders Lower IQ - more sleep disorders	Clinic sample; studied group with regression

Dominick (2007)	Frequency and inter-relationship of atypical behaviour (eating, sleeping, self-injurious behaviour, aggression and temper tantrums) in ASD and HLI	Community voluntary recruitment N = 67 ASD 39 HLI (Having Language Impairment)	4 year 2 month – 14 year 2 months	Atypical Behaviour Patterns Questionnaire (ABPQ)	ASD Sleep problems – 63.6%: 50% of sleep problems – initial and middle insomnia; terminal insomnia least frequent 80% of problems – constant Began – year 1 or 2 – persistent in 60%	Community sample

ABPQ – Atypical Behaviour Patterns Questionnaire, BISCUIT – Baby and Infant Screen for Children with aUtism Traits , CBCL – Child Behaviour Check List, CSHQ – Children's Sleep Habits Questionnaire, PBS – Paediatric Behaviour Scale

Reviews reported that studies evaluating predictors of sleep problems reported mixed results (Cortesi et al., 2010; Hollway and Aman, 2011). Age was shown to have a negative correlation with total sleep problems in some studies (Giannotti et al., 2008; Hollway et al., 2013), while others reported sleep problems being present across all age groups (Goldman et al., 2012). A large treatment network based study reported that adolescents and older children had more delayed sleep onset, shorter sleep duration and daytime sleepiness; while younger children had more bedtime resistance, sleep anxiety, parasomnias and night-waking (Goldman et al., 2012). Another recent article evaluating a large number of children with ASD from the same treatment network reported anxiety, sensory sensitivities, autism severity and gastrointestinal problems were related to sleep problems (Hollway et al., 2013).

Reviews have reported that autism severity had a direct relationship with severity of sleep problems (Hollway and Aman, 2011; Mannion and Leader, 2013c). There are reports of autism severity influencing sleep problems (Mayes and Calhoun, 2009; Hollway et al., 2013) and lower sleep duration predicting autism severity (Schreck et al., 2004). A population based study reported autism severity as the strongest predictor for sleep problems (Sivertsen et al., 2012). A recent review also highlighted the possibility of a bidirectional relationship between sleep and co-morbid psychopathology (Mannion and Leader, 2013c).

3.5.3 Measures of sleep problems

Reviews have analysed both objective and subjective measures of sleep problems in children with ASD (Bauer and Blunden, 2008; Spruyt and Gozal, 2011; Hodge et al., 2012). The parent-report subjective measures were noted to be valid for screening but were inconsistent in estimating different sleep variables including sleep duration and night-waking with the exception of sleep onset latency (Bauer and Blunden, 2008; Hodge et al., 2012). The subjective measures analysed included sleep diaries, single item responses and multi-item parent report measures (Dominick *et al.*, 2007; Giannotti *et al.*, 2008; Mayes and Calhoun, 2009; Goldman *et al.*, 2011; Kozlowski *et al.*, 2012a; Hodge *et al.*, 2013; Mannion and Leader, 2013a; Mannion and Leader, 2013c).

The Children's Sleep Habits Questionnaire (CSHQ) (Owens et al., 2000) was recognised as widely used in children with ASD. Comparing the CSHQ with objective measures such as actigraphy and polysomnography, parents' reports were objectively confirmed for sleep latency, but not for other subscales of the CSHQ (Hodge et al., 2012). All measures considered for this study are summarised in Appendix 1.

3.5.4 Summary of literature review of sleep problems in children with ASD

Sleep problems present as a common co-existing condition in children with ASD. Studies have reported different findings regarding predictors for sleep problems. Subjective measures to assess sleep problems have drawbacks when compared to objective measures.

3.6 Feeding problems in children with ASD

For the purpose of this thesis, feeding problems were defined as: parent report of feeding or eating problems or significant scores on a standardised measure for feeding/eating problems. The search term used was 'feeding'. The results of the literature search are summarised in Appendix 2. 68 articles were identified after the initial screening in addition to the previously analysed literature. The inclusion criteria of 3.1.1 (sample size above 50 and parent reporting of symptoms) were used to identify relevant articles for detailed review.

3.6.1 Results of literature search

Table 8 summarises the relevant studies evaluated in the literature review. There are some reviews assessing feeding/eating problems in children with ASD (Nadon et al., 2008; Cermak et al., 2010; Kral et al., 2013).

3.6.2 Prevalence and predictors of feeding problems

The reviews recognised that study of eating behaviours in children was an emerging field of research (Kral et al., 2013). A population representative database-based study reported the frequency of eating problems as 58%, but this was based on a single item (Maskey *et al.*, 2013). Another community study reported higher prevalence of 76.4%, with preference for particular foods in 58% and restricted range of foods in 63% (Dominick et al., 2007). Food selectivity has been reported as the major feeding problem from other studies as well (Bandini *et al.*, 2010; Bicer and Alsaffar, 2013). Other studies analysing

the broader gastrointestinal symptoms of children with ASD reported frequencies between 24% - 46.5% (Ming *et al.*, 2007; Mazurek *et al.*, 2012; Chandler *et al.*, 2013).

A community study of ASD stated that eating problems began in the first year of life and persisted to childhood (Dominick et al., 2007). Age and gender did not have any effect on reported rates in another study of toddlers with autistic disorders and PDD-NOS (Kozlowski *et al.*, 2012a). Neither autism severity nor the type of autism predicted feeding problems in other studies (Schreck and Williams, 2006; Matson *et al.*, 2009a). From the limited literature available, it appears that the usual variables of age, gender, IQ and autism severity had limited association with feeding problems. Reviews are moving towards the possibility of associations between sensory sensitivity and feeding problems including food selectivity (Cermak et al., 2010; Kral et al., 2013).

3.6.3 Measures of feeding problems

A review published in 2008 assessing measures of feeding problems in pervasive developmental disorders reported that none of the measurement tools assessed all factors that might influence eating in these children (Nadon et al., 2008). There are reviews and studies analysing gastro-intestinal (GI) problems of children with ASD; however, the measures developed to address GI symptomatology do not cover feeding/eating problems including food refusals and limited food repertoire (Mannion and Leader, 2014b).

Table 8: Summary of relevant studies evaluating feeding problems in children with ASD (Reverse chronological order)

Author (date)	Research question	Sampling (N=number of participants)	Age	Tools used for feeding	Main findings	Critique
Meral (2014)	Screening tool of feeding problems (STEP) in Turkey	School sample N = 360	9.4 years	STEP, BAMBI (Brief Autism Mealtime Behaviour Inventory)	Good internal consistency, split half reliability and criterion related validity; acceptable goodness of fit	Turkish adaptation of STEP
Mazurek (2013)	Anxiety, sensory over-responsivity and GI problems in ASD	Autism Treatment Network N = 2973	2-17 years	Gastrointestinal (GI) symptom inventory	24% -at least 1 GI problem; commonest – constipation, abdominal pain, bloating, diarrhoea. Each GI problem associated with high sensory-over-responsivity and anxiety. Both sensory issue and anxiety predicted GI problem	Large database sample
Chandler (2013)	GI symptoms in ASD children in SNAP cohort	Population-derived N=132ASD; 81 special needs; 82 TD	10-14 years	GI symptom questionnaire	46.5% ASD at least 1 lifetime GI symptom; 29.2% -special needs and 21.8% TD ASD more current constipation and soiling than others No association between GI problems and IQ, ASD severity, ASD regression or limited diet	Population representative

Mannion (2013)	Analysis of predictors of psychopathology, gastrointestinal problems and epilepsy in ASD	Community recruitment (schools, parent groups) N = 89	3-16 years	GI symptom inventory	Intellectual disability and Sleep problems predicted GI problems (R^2=.403) (other factors considered – age, gender, co-morbid conditions, comorbid psychopathology)	Community sample
Bicer (2013)	Feeding problems in Turkish children with ASD	4 autism centres in Istanbul N = 164 ASD	4-18 years	3-day food record; feeding assessment surveys	Major feeding problem – food selectivity; 58.5% - obese/over-weight and 11% - thin and severely thin based on Body Mass Index; Intake of calcium, zinc, vitamin B6 and folate deficient; salt intake in all children high; cholesterol intake in normal, overweight and obese – high	Study from Turkey
Mannion (2013)	Co-morbid psychopathology, gastrointestinal symptoms, sleep problems and epilepsy in ASD	Community recruitment (schools, parent groups) N = 89	3-16 years	Gastrointestinal (GI) symptom inventory	79.3% ≥ 1 GI problem Most common – abdominal pain (51.7%), constipation (49.4%) and diarrhoea (45.9%)	Community sample; evaluated all GI symptoms
Maskey (2013)	Predictors and frequency of co-existing emotional and behavioural conditions in ASD	Population representative database N = 863 ASD	2-18 years	Single item parent response	Eating – 58% - most common problem	Population representative; single item

Kozlowski (2012)	Sleep and feeding problems in toddlers with ASD	Research database N =506 autistic N = 522 PDD-NOS N= 719atypical development	17 – 37 months	BISCUIT-part 2	Children with autistic disorder significantly more sleep and feeding problems than PDD-NOS. PDD-NOS more problems than atypically developing group. No effect of age and gender	Toddler group studied
Bandini (2010)	Food selectivity in ASD	Community + research database N = 53 ASD; 58 TD	3-11 years	Modified food frequency questionnaire and 3-day food record	ASD – food refusal 41.7% of food compared to TD (18.9%); more limited food variety 19 vs 22.5 foods in TD Limited food variety group had inadequate intake of nutrients	Evaluated food refusal
Matson (2009)	Relationship between feeding problems and core features of autism	Combined sample N = 72 Autism; 40 PDD-NOS; 53 atypical development; 114 TD	3-16 years	Items and subscales of ASD-CC	Autism and PDD-NOS similar feeding problems; but higher feeding problems than with atypically and typically developing children	Combined sample
Ming (2008)	Clinical co-occurrences in ASD	Autism clinic sample; retrospective chart review N = 160 ASD	2-18 years	Clinical information form with history	51%-long term food intolerance 59% - GI dysfunction Food intolerance and sleep dysfunction associated with GI dysfunctions	Retrospective clinic sample

Lukens (2008)	Development of BAMBI (Brief Autism Mealtime Behaviour Inventory)	Community sample N = 68 ASD; 40 TD	3-11 years	BAMBI Behaviour Paediatric Assessment (BPFAS)	Good internal consistency; high test-retest reliability, 3 factor structure; strong construct and criterion validity	Autism specific measure
Dominick (2007)	Frequency and inter-relationship of atypical behaviour (eating, sleeping, self-injurious behaviour, aggression and temper tantrums) in ASD and HLI (Having Language Impairment)	Community voluntary recruitment N = 67 ASD 39 HLI	4 year 2 month – 14 year 2 months	Atypical Behaviour Patterns Questionnaire (ABPQ)	ASD Eating problems – 76.4% significantly higher in ASD than HLI Of eating problems: Preference for particular foods – 58%; 30% on texture, 14% on colour, 16% on taste Restricted range of foods – 63% Problems begin 1st year of life – persist to childhood Eating problems related to temper tantrums	Community sample
Schreck (2006)	Food preferences and food selectivity in ASD	Database sample N = 138 ASD	4-12 years	Children's Eating Behaviour Inventory (CEBI); food preference inventory	ASD-limited food variety; not related to food texture; 72% -restricted food; 57% -food refusal; refusals related to food presentation; family food preferences not autism severity predicted child food preferences	Database sample; not ASD-specific tool

ABPQ – Atypical Behaviour Patterns Questionnaire, ASD-CC - ASD-Comorbid for Children, ASD-PBC – ASD-Problem Behaviour Children, BAMBI – Brief Autism Mealtime Behaviour Inventory, BISCUIT – Baby and Infant Screen for Children with aUtism Traits , BPFAS – Behaviour Paediatric Feeding Assessment, CEBI – Children's Eating Behaviour Inventory, CEBI – Children's Eating Behaviour Inventory, HLI – Having Language Impairment, STEP – Screening Tool of Feeding Problems

The measures assessed for this thesis are included in Appendix 1. The Brief Autism Mealtime Behaviour Inventory (BAMBI) (Lukens and Linscheid, 2008) was developed specifically to address feeding problems in children with ASD. It has a three-factor structure covering limited food variety, food refusal and features of autism. The BAMBI was shown to correlate with objective feeding assessment in a small-sample study of ASD children aged 3-8 years of age (Sharp et al., 2013).

3.6.4 Summary of literature review of feeding problems in children with ASD

Feeding and eating problems constitute another common co-existing condition in children with ASD. There is only a limited literature evaluating feeding problems; however available literature points to restricted influence of common demographic and autism specific variables barring sensory sensitivities on feeding problems. Autism specific measures to evaluate feeding problems are being developed.

3.7 Sensory problems in children with ASD

Sensory problems including sensory processing and sensory integration problems are common in children with ASD; the recent DSM-V has included these in the diagnostic criteria for ASD (APA, 2013). Dunn in her seminal article described a model of sensory processing having different sensory styles based on neurological thresholds for stimulation and individual self- regulation strategies (Dunn, 1997). The neurological thresholds can be either high or low and self-regulation strategies can be active or passive. Crossing these factors, four sensory processing styles are described.

- Low registration: This sensory processing style has high neurological threshold for stimulation and passive self-regulation strategy. Individuals with this style can be described as disconnected from the environment
- Sensory seeking: This sensory processing style has high neurological threshold for stimulation and active self-regulation strategy. Individuals with this style seek out sensations.
- Sensory avoiding: This sensory processing style has low neurological threshold for stimulation and active self-regulation strategy. Individuals with this style tend to avoid sensory inputs and remain withdrawn

- Sensory sensitivity: This sensory processing style has low neurological threshold for stimulation and passive self-regulation strategy. Individuals with this style tend to be distracted by sensory inputs, but do not avoid them.

Using Dunn's model, Miller and colleagues suggested a classification for sensory modulation disorders (Miller *et al.*, 2007). The classification suggested three distinct sensory modulation disorders: sensory over-responsivity, sensory under-responsivity and sensory sensitivity. Individuals with sensory over-responsivity respond to sensations faster; while individuals with sensory under-responsivity do not respond or disregard sensations. Individuals with sensory seeking seek out sensation.

For the purpose of this thesis, sensory problems or atypical sensory behaviours were defined as: parent report of sensory problems or significant scores on a standardised measure for sensory problems. The search term used was 'sensory'. The results of literature search are summarised in Appendix 2. 95 articles were identified after the initial screening of the literature in addition to the previously analysed literature. The inclusion criteria of 3.1.1 (sample size above 50 and parent reporting of symptoms) were used to identify relevant articles for detailed review.

3.7.1 Results of literature search

Table 9 summarises the relevant studies evaluated in the literature review after using initial screening and subsequent inclusion criteria and published after the meta-analysis by Ben-Sasson (Ben-Sasson et al., 2009).

Table 9: Summary of relevant studies evaluating sensory problems in children with ASD (Reverse chronological order)

Author (date)	Research question	Sampling (N = number of participants)	Age	Tools used for sensory problems	Main findings	Brief critique
Lidstone (2014)	Relationship between repetitive behaviour, anxiety and sensory features in ASD	2 different study databases N =120 ASD	2-17 years	Sensory Profile (SP)	Age not related to total score or quadrants of atypical sensory behaviour except negatively related to sensory seeking; Relation between anxiety and insistence on sameness mediated by sensory avoidance and sensory sensitivity	Relationship between anxiety, sensory problems and repetitive behaviour
Hollway (2013)	Correlates and risks for sleep problems for children in Autism Treatment Network	Autism Treatment Network (ATN) Database N = 1583 ASD	2- 17 years	Short Sensory Profile (SSP)	Sensory problems (under-responsive/sensory seeking, auditory filtering and taste/smell sensitivity subscales) related with sleep problems	Large database study
Mazurek (2013)	Prevalence and correlates of physical aggression in ASD	ATN N=1584 ASD	2-17 years	SSP	Multivariate model –Sensory problems strongly associated with aggression	Large database sample
O'Donnell (2013)	Relation beween sensory problems, adaptive behaviour and problem	Early intervention group – retrospective	3 years	SSP	75% - atypical sensory scores Significant relationship between sensory problems and problem behaviour score; no relationship between sensory	Early childhood retrospective study

	behaviour	study; N =75 ASD			problems and cognition or adaptive behaviour	
Maskey (2013)	Predictors and frequency of co-existing emotional and behavioural conditions	Population representative database N = 863 ASD	2-18 years	Parent report	Sensory problems – 57%	Population representative; single item
Mazurek (2012)	Anxiety, sensory over-responsivity and GI problems in ASD	ATN N = 2973	2-17 years	SSP	Sensory over-responsivity or sensory sensitivity predicted GI problem	Large database sample
Brock (2012)	Temperament and sensory features in ASD	Sampling not mentioned N= 54 ASD; 33 atypical development	3-7 years	SP, Sensory Experience Questionnaire (SEQ) ;Sensory Processing Assessment for Young Children (SPA)	Sensory hypo-responsiveness or low registration associated with slowness to adapt, low reactivity, low distractibility in temperament in ASD. Increased sensory features across hyper-responsiveness, hypo-responsiveness and sensory seeking patterns – associated with increased withdrawal and more negative mood	Sampling details not clear
De la Marche (2012)	Sensory processing in adolescents with ASD	Study database N = 80 ASD; 56 siblings; 33 TD	11-18 years	Adolescents SP (self-report)	ASD differed from controls in sensation seeking and sensation avoidance. Siblings showed reduced sensation seeking – possible intermediate phenotype	Adolescent study
Tseng (2011)	Relationship between emotional and behavioural problems and sensory	6 centres' samples N = 67 autism; 45	48-84 months	SP – Chinese version	Sensory pattern abnormal in all quadrants; low registration most common 96% of (ASD+internalising behaviour)	Study from Taiwan; multicentric

	problems	TD			and 82% of (ASD+externalising) – at least 1 atpical sensory behaviour;	
Goldman (2011)	Relationship between sleep and problem behaviours	ATN N = 1784 ASD	2-17 years	SSP	1 unit increase in parasomnia - 20% increase in sensory issues	Large database sample
Lane (2010)	Relation between sensory processing and adaptive behaviour in ASD	Early intervention programme N = 54 autism	33-115 months	SSP	Sensory subtypes predicted communication competence and maladaptive behaviour	Early childhood study
Watson (2010)	Relationship between sensory patterns and social communication	Combined recruitment N =72 autism; 44 developmentaldelay	Mean (SD) – 52 months (16)	SP, SEQ, SPA	Hypo-responsiveness positively associated with social communication symptom severity in both groups; hyper-responsiveness not associated; Sensory seeking positively correlated with communication severity for autism, reverse for atypically developing	Evaluating different sensory patterns

SEQ – Sensory Experience Questionnaire, SSP – Short Sensory Profile, SP – Sensory Profile, SPA – Sensory Processing Assessment for young children

3.7.2 Prevalence and predictors of sensory problems

In a meta-analysis, the authors reported that individuals with ASD had a significant higher frequency of sensory symptoms compared to peers (Ben-Sasson et al., 2009). The meta-analysis used the classification proposed by Miller and colleagues in analysing sensory modulation disorders (Miller *et al.*, 2007). The mean effect sizes across studies were high and significant with under-responsivity being the greatest area of difference (Ben-Sasson *et al.*, 2009).

A retrospective chart review of three year-olds with ASD reported a frequency rate of 75% for sensory problems (O'Donnell et al., 2012); while the rate was reported as 57% from a population-representative study (Maskey *et al.*, 2013). Other studies also have reported similar high frequency of sensory problems or atypical sensory behaviours in children with ASD (Tseng *et al.*, 2011; De La Marche *et al.*, 2012). Under-responsivity was the commonest type reported in a multi-centre study in Taiwan similar to the findings in the meta-analysis (Tseng et al., 2011). Another study of adolescents with ASD reported that sensation seeking and sensation avoidance differed the maximum between children with ASD and controls (De La Marche et al., 2012).

In the meta-analysis, for total sensory, sensory over-responsivity and sensory seeking scores, the effect size rose from studies of 0-3 year-olds to studies of 3-6 year-olds to 6-9 year-olds and then decreased in studies of above 9 year-olds (Ben-Sasson et al., 2009). For under-responsivity, the highest effect size was for 6-9 year-olds indicating under-responsivity being highest in this age group of ASD. The meta-analysis also suggested that individuals with autism had higher sensory seeking than those with non-autism pervasive developmental disorder; but not under and over-responsivity.

Evaluating subsequent studies, a recent study of children with ASD reported that age did not correlate to sensory profile score or sub-domain score, but negatively correlated to sensory seeking behaviour (Lidstone et al., 2014). Studies from the large Autism Treatment Network (ATN) database reported that atypical sensory behaviours were associated with and predicted sleep problems, gastrointestinal problems and aggression (Mazurek et al., 2012; Hollway et al., 2013; Mazurek et al., 2013). Atypical sensory behaviours are

reported to be associated with temperament including negative mood and withdrawal (Brock et al., 2012) as well as problem behaviours in children with ASD (Lane et al., 2010; Tseng et al., 2011; O'Donnell et al., 2012). Reviews of other co-existing conditions such as anxiety and feeding problems have acknowledged that atypical sensory behaviours affect these conditions (Cermak et al., 2010; van Steensel et al., 2011).

3.7.3 Measures of sensory problems

The meta-analysis in 2009 reported that 79% of studies used a version of the Sensory Profile (Ben-Sasson et al., 2009). The Sensory Profile (Dunn and Westman, 1997) comprises the full profile, the short sensory profile, the infant/toddler sensory profile and the adolescent self-report sensory profile. The factor structure of this measure corresponds to over-responsivity, under-responsivity and sensory seeking types of sensory modulation disorders. Most of the subsequent studies after meta-analysis also utilised one of the sensory profile measure (Lane *et al.*, 2010; Goldman *et al.*, 2011; Watson *et al.*, 2011; Brock *et al.*, 2012; Mazurek *et al.*, 2012; Mazurek *et al.*, 2013; Lidstone *et al.*, 2014). Appendix 1 provides a summary of measures considered for this project.

3.7.4 Summary of literature review of sensory problems in children with ASD

Sensory problems or atypical sensory behaviours are a common co-existing condition in individuals with ASD and are now incorporated into the recent DSM-V diagnostic criteria for ASD. These problems are associated with and predict other co-existing conditions in children with ASD. The most common measure used in individuals with ASD belongs to the sensory profile group including the Sensory Profile and the Short Sensory Profile.

3.8 Impact on the family of children with ASD

Both the core features of ASD and co-existing conditions have a considerable impact on individuals with ASD and their families: refer to 1.10 (page 9). Both ASD specific behaviours and behaviours related to co-existing conditions can act as stressors in the double ABCX and the composite research models of coping and adaptation: refer to 2.2 (page 14).

‘Impact on the family’ studies included any studies of parents who had children of 2-18 years with ASD covering impact on family or quality of life (QoL) of

family or parent stress. The search item was ‘impact on the family’ or ‘quality of life’. 153 articles were identified after the initial screening of the literature in addition to the previously analysed literature. For the subsequent inclusion criteria to identify relevant articles for detailed review, the number of subjects was reduced to 40 to include more studies for consideration. The results of literature search are summarised in Appendix 2.

3.8.1 Results of literature search

Table 10 summarises the relevant studies evaluated in the literature review published after 2010, the earlier of the two recent reviews (Meadan *et al.*, 2010; Karst and van Hecke, 2012). Most of the studies covered one or all outcome adaptations of parents, siblings or the family as a unit.

3.8.2 Prevalence and predictors of impact on family

Recent reviews have analysed stress and support for families of children with ASD (Meadan et al., 2010; Karst and van Hecke, 2012). The reviewers evaluated stress within three subsystems of family – marital subsystem, parental subsystem and sibling subsystem and how all these subsystems interrelated with each other and with the individual having ASD. The reviews reported high stress for parents with mothers reporting more impact than fathers. There was impact on parent well-being and mental and physical health affecting the parent-child relationship. There was only limited literature evaluating the marital subsystem; but the available evidence pointed to low family harmony (Karst and van Hecke, 2012). Subsequent studies have confirmed the review findings of parent stress being higher among parents of children with ASD compared to either those of children with developmental delay or those of typically developing children (Dabrowska and Pisula, 2010; Ingersoll and Hambrick, 2011; Silva and Schalock, 2012; Estes *et al.*, 2013; Hodge *et al.*, 2013; Huang *et al.*, 2013). The parenting stress can affect parental health including endocrine control systems (Foody et al., 2014) and parental mental health (Skokauskas and Gallagher, 2012; Shtayermman, 2013).

Table 10: Summary of relevant studies evaluating Impact of Family of children with ASD (Reverse chronological order)

Author (date)	Research question	Sampling (sample size)	Age	Tools used for impact/ QoL	Main findings	Critique
Steijn (2014)	Relationship between ASD, ADHD, depression symptoms and stress in parents	Recruited from family genetic studies N = 174 ASD	2-20 years	Parenting Stress Index (PSI)	Parents have more stress with ASD+ADHD Parental depression more when child has ASD or ASD+ADHD Paternal ASD, maternal ADHD – increased parental stress	Effect of parental factors on parental stress
Foody (2014)	Health outcomes in mothers of ASD	Community sample N = 74 ASD	2-14 years	Parenting Stress Index (PSI)	High levels of parenting stress & anxiety; dysregulation of hypothalamo-pituitary-adrenal axis. Quantity of unmet service needs, sleep problems, socialisation deficits, adaptive behaviour, coping strategies of self-blame & behavioural disengagement predicted parental distress	Community sample; study of maternal health
McStay (2013)	Parenting stress in ASD	School sample N = 150 ASD (able children)	6.4-18.8 years	PSI	Hyperactivity not autism severity, age of child, verbal ability or perceived child quality of life, predicted parenting stress	Child behaviour predicted parenting stress

Estes (2013)	Parent related stress in ASD	Study dample N = 46 ASD; 25 developmental delay (DD); 25 TD	18-30 months	Brief Symptom Inventory and Questionnaire on Resources & Stress (QRS)	Parents of ASD higher stress than DD and TD; Child behaviour problems not daily living skills predictor for parenting stress	Toddler study
Huang (2013)	Examine effects of autistic behaviour and individual emotional and behavioural problem on parenting stress	Recruited from 5 hospitals and 2 clinics N = 52 Autism	3-12 years	Parenting Stress Index – Short Form (PSI-SF)	3 domains of parental stress – 28.8% parental distress (PD), 21.2% parent-child dysfunctional interaction (PCDI) and 40.4% difficult child (DC). DC and PD scores higher. Parent of mild/moderate autistic behaviour perceived lower parenting stress than those of children with no or severe autistic behaviour. Stress scores of mild/moderate and severe emotional and behavioural problems higher than that with no problems. DC parent stress model– 32.9% contributed by conduct problems; PCDI stress model– mild to moderate autistic behaviour + pro-social behaviour contributed 40%; PD model – no variables contributing	Clinic sample
Hodge (2013)	Relationship between maternal stress and children's	From research dataset N = 90 ASD	4-12 years	PSI	More sleep problems in ASD than TD Maternal sleep and maternal stress mediated the relationship between children's sleep and mother's mental health for	Comparison with TD

	sleep in ASD	and 90 TD controls			mothers of both ASD and TD	
Harper (2013)	Respite care and parental stress	Community & school recruitment; N = 101 parents of ASD	Not mentioned	Dyadic Adjustment Scale; Hassles and Uplift Scale	Number of hours of respite care improved marital quality mediated by perceived daily stresses.	Community sample
Paynter (2013)	Factors affecting family adaptation	Intervention centre sample N = 43 ASD	2.5 – 6 years	Impact on Family (IOF)	Challenging behaviour, pile up demands, internal and external resources, active avoidant coping strategies predicted IOF (ASD severity not affecting)	Early intervention sample; double ABCX model
Shtayermann (2013)	Parental stress and marital satisfaction	Interactive Autism Network database N = 253 ASD	<18 years	Perceived stress scale	Lower levels of marital satisfaction; 13% parents –depressive disorder; 15% - GAD; Higher stress associated with higher depression and anxiety	Database sample
Peters-Scheffer (2012)	Child factors affecting parenting stress	Research study sample N = 104 ASD	2-9 years	PSI every 6 months for 2 years	Maternal stress stable over time; emotionally reactive behaviour, withdrawn behaviour & attention problems; not behavioural inflexibility nor social problems predicted parental stress. No relation to developmental age/IQ, receptive/expressive language, adaptive behaviour, severity/type of ASD	Community study
Van Steensel (2012)	Anxiety and QoL	Community centres N = 115 ASD;	Mean -11 years	Euro Quality of Life – 5D	ASD-like behaviour and higher anxiety severity negatively predicted QoL in both groups	Community study

		122 anxiety		(EQ-5D)		
Hall (2012)	Community support services and coping in ASD	Community enrolment N = 38	2-21 years	Family Crisis Oriented Personal Evaluation Scale	Community social services support predicted family coping better than CARS (Childhood Autism Rating Scale) score	Small community sample
Skokauskas (2012)	comorbid psychiatric problems and distress	Community recruitment N = 67 ASD (Control – 67)	6-18 years	Brief Symptom Inventory	Parents' symptoms of psychological distress associated with neither externalising and internalising problems in children with ASD	Community sample
Kheir (2012)	Quality of life of caregivers of children with ASD in Qatar	Clinic sample N = 56 ASD; 42 TD	3-17 years	SF-36 (Short Form -36)	No significant difference between QoL between two groups; caregivers of ASD rated their health as poor	Evaluated Health related QoL
Silva (2012)	Autism Parenting Stress Index (APSI): Initial psychometrics	Early intervention group N = 107 ASD; 28 atypically developing; 139TD	24-72 months	Autism Parenting Stress Index (APSI)	Mean parenting stress for autism group 4 times higher than that of TD and double that of other atypically developing children; 3 factor structures identified – each relating to core deficits, co-morbid behavioural symptoms and co-morbid physical symptoms	Specific tool for autism – separate for core and co-morbid.
Ingersoll (2011)	Relationship of parent and child characters to parent stress	Interactive Autism Network Database N = 149 ASD	< 18 years	PSI – Short Form (PSI-SF)	Child symptom severity and parent Broad Autism Phenotype (BAP) positively correlated with parent stress; Relationship between BAP and stress partially mediated by maladaptive coping & social support.	Study analysing Broad Autism Phenotype of parents

Brown (2011)	Parents' unmet needs in ASD	Research database N = 97 ASD	6-13 year	IOF	Impact of family +vely correlated with unmet need	Low participation (20.2%)
Cappe (2011)	Quality of Life: key variable to consider in adaption of parents	Combined sample N = 160 ASD	Different ages	Appraisal of Life Events; Coping Checklist; Quality of Social support; Quality of Life Scale	Parents who viewed their experience as threat– lowest QoL, quality of daily, family and social life; parents who viewed their experience as challenge – better relation with child & greater self-fulfilment; Parents with emotion-focused coping strategies – felt nervous and distressed at work; parents with problem-solving coping strategies – better relationships with child; Parental stress +vely correlated with poorer QoL and emotion-focused coping strategies; Lesser available social support associated with more emotion-focused coping strategies	Combined sample study exploring parental experience, coping skills and outcome of QoL
Khanna (2011)	Psychometric properties of Caregiver Strain Questionnaire (CGSQ) in ASD	Community sample N = 304 ASD	2-18 years	Short Form – 12 (SF-12)	Modified model good convergent validity and internal consistency; reasonable fit	Evaluates health related QoL
Manning (2010)	Double ABCX model in racially diverse families	Study sample N = 195 Autism	6-12year	Family Environment Scale	Behaviour problems and reframing coping strategy significantly predicted family outcome (not family income, life stress, autism severity, informal social support, spiritual support, family & friends, subjective social statue) – total model 28% of variance	Double ABCX model

Dabrowska (2010)	Stress in parents of Downs and ASD	Early intervention centre/ kindergartens N = 51 autism; 54 Downs & 57 TD	2-6 years	Coping inventory; QRS	Higher level of stress in parents of children with ASD; mothers had higher stress. Emotion-oriented coping and level of education positively predicted stress in ASD not income and number of children. Child's diagnosis affected total stress, dependency and management, limits on family opportunities; life span care; family disharmony; terminal illness stress; physical limitations and personal burden (no difference in financial stress and lack of personal reward)	Preschool sample

APSI – Autism Parenting Stress Index, EQ-5D – Euro Quality of Life – 5D, IOF – Impact on Family, PSI - Parenting Stress Index, PSI – SF – Parenting Stress Index – Short Form, QRS – Questionnaire on Resources and Stress, SF – Short Form

To understand different predictors for the impact on the family, the detailed reviews are evaluated with reference to models of coping and adaptation. The initial stressors identified included the ASD specific core symptoms of communication and social deficits as well as worry about the future: see reviews (Meadan et al., 2010; Karst and van Hecke, 2012). The reviews also highlighted the emergence of children's co-existing emotional and behavioural problems as ongoing stressors in the literature. Evaluating subsequent literature, a community-based study of children with ASD supported the findings of the review that the predictors of parent distress included socialisation deficits, sleep problems and adaptive behaviour of children as well as coping strategies of self-blame, behavioural disengagement and quantity of unmet service needs of families (Foody et al., 2014). A longitudinal community-based study of mothers of children with ASD moved beyond this knowledge level, to report that not developmental age/IQ, receptive/expressive language or adaptive behaviour; but attention problems and emotionally reactive behaviour predicted maternal stress (Peters-Scheffer et al., 2012). There are concurring reports from other studies as well highlighting the impact of co-existing conditions including ADHD (Steijn *et al.*, 2014), sleep disturbances (Delahaye *et al.*, 2014)

Regarding adaptation to the stressors, both family and child characteristics influence the final outcome of family adaptation, with different support systems and coping mechanisms acting as mediators. There is also the emergence of evidence of bidirectional relationships between different components of the model (Karst and van Hecke, 2012). The support systems including social, formal and informal service support positively augment family and parent coping mechanisms (Hall, 2012; Harper *et al.*, 2013). Among coping mechanisms, both approach-oriented and problem-focused coping were shown to have a more positive outcome compared to avoidance-oriented or emotion-focused coping strategies (Dabrowska and Pisula, 2010; Manning *et al.*, 2010; Cappe *et al.*, 2011; Paynter *et al.*, 2013). Families with a variety of coping strategies were reported to experience lower levels of stress (Cappe *et al.*, 2011).

There are conflicting reports to the impact of co-existing conditions on families above and beyond that of autism. A community-based study reported that parents' symptoms of psychological distress associated with neither externalising nor internalising problems in children with ASD (Skokauskas and

Gallagher, 2012). Another clinic-based study of families of children with autism stressed the impact of autistic behaviour on parenting stress (Huang et al., 2013). Of late, studies have reported that not autism severity nor type of autism of the child, but challenging behaviour, sleep problems, anxiety and hyperactivity of children affected the final impact on the family significantly (Hall, 2012; Peters-Scheffer *et al.*, 2012; van Steensel *et al.*, 2012a; McStay *et al.*, 2013; Paynter *et al.*, 2013; Delahaye *et al.*, 2014; Foody *et al.*, 2014).

In addition to the factors discussed above, parental ASD and ADHD characteristics were also reported to impact family adaptation (Ingersoll and Hambrick, 2011; Steijn *et al.*, 2014). Evaluating the role of services impacting the final family/parent adaptation, the quantity of unmet service needs was shown to predict parent distress in a community study of mothers of children with ASD (Foody et al., 2014). Both internal and external resources and pile-up stressors were reported to impact family in a clinic-based sample of parents of pre-schoolers with ASD (Paynter et al., 2013). A community-based study of families of children and young people with ASD reported that the community social service support predicted family coping (Hall, 2012). These articles reinforced the role of services in family coping and family adaptation as proposed in the research models outlined earlier: refer to section 2.2 (page 14).

3.8.3 Measures to assess impact on the family

Studies have used a range of measures to assess parent/family adaptation. As expected all these measures were subjective and parent-reported. The tool used in most of the studies was a version of the Parent Stress Index (PSI) and dealt with parent adaptation (Peters-Scheffer *et al.*, 2012; Hodge *et al.*, 2013; Huang *et al.*, 2013; McStay *et al.*, 2013; Foody *et al.*, 2014; Steijn *et al.*, 2014). The Impact on Family has been used to assess family adaptation as a whole in families of children with ASD (Brown et al., 2011; Paynter et al., 2013). Appendix 1 summarises the measures considered for this project.

3.8.4 Summary of literature review of Impact on family

All studies and reviews agreed on the high impact of ASD on families/parents. Autism-specific symptoms and co-existing conditions act as ongoing stressors for the families, but there are conflicting reports of their significance to parent

stress and the impact on family. Support including that of services helps in promoting family coping and adaptation.

3.9 Unmet parent needs in families of children with ASD

Measuring unmet parent needs can be used to evaluate services: refer to section 2.3.2 (page 20) for details. For the purpose of this thesis, unmet needs were defined as: parent report of unmet parental or service needs. The search term used was 'needs'. The results of the literature search are summarised in Appendix 2. After initial screening, 26 articles were identified in addition to the literature evaluated earlier. For the subsequent inclusion criteria to identify relevant articles for detailed review, the number of subjects was reduced to 30 to include more studies for consideration.

3.9.1 Results of literature search

Table 11 summarises the relevant studies evaluated in the literature review. Brown and colleagues in their review recognised the ambiguity of the definition of needs in children with ASD (Brown et al., 2010). The authors proposed a conceptual framework where parent's perceptions of unmet needs are influenced by both service use and child and family characteristics.

3.9.2 Prevalence and predictors of unmet parent needs

A review of unmet parent needs highlighted the limited literature evaluating them in families of children with ASD (Brown et al., 2010). It is important to stress that studies about parental needs are relevant as parent and professional opinions do not match always (Dillenburger et al., 2010). In the review, the need for information, followed by the co-ordination of services and the need for proper communication were reported as prominent unmet needs (Brown et al., 2010).

Table 11: Summary of important studies evaluating parent needs in families of children with ASD (Reverse chronological order)

Author (date)	Research question	Sampling (sample size)	Age	Tools used for needs	Main findings	Critique
Molteni (2014)	Parental experiences of children with ASD in Italy	No profit organisation sample N = 31 families	4-10 years	Interviews & thematic analysis	Parents need support during diagnosis and for future planning to avoid crisis	Thematic analysis of experiences
Foody (2014)	Health outcomes in mothers of ASD	Community sample N = 74 ASD	2-14 years	Parent report	Quantity of unmet service needs predicted parental distress	Community sample; study of maternal health
Paynter (2013)	Factors affecting family adaptation	Intervention centre sample N = 43 ASD	2.5 – 6 year	Revised Social Readjustment Scale	Pile up of demands, internal and external resources predicted IOF	Early intervention sample; double ABCX model
Brown (2012)	Unmet needs of families of children with ASD	Research database N = 101	6-13 years	Family needs questionnaire	Common – 78.2% -social activities for the child; 77.2% -information about services; 74.3% - continuous service provision	Low participation (20.2%)
Brown (2011)	Parents' unmet needs in ASD	Research database N = 97 ASD	6-13 year	Family needs questionnaire	Functional independence of children –vely correlated with unmet need; impact of family +vely correlated with unmet need;	Low participation (20.2%)

Dillenburger (2010)	Parental and professional views about needs	Combined clinic & community N = 100 ASD	1-16 years	Parent and professional needs questionnaire	Parent & professional opinions do not match always; Parents – services are not efficient in meeting needs	Study from Ireland: combined sample
Papageorgiou (2010)	Parental needs of children with ASD in Greece	Parent support groups N = 299 ASD	Not mentioned	Open ended questionnaire	Common needs – communication, behavioural and practical problems Parent education not gender affected needs perception; Parents of girls perceived more support for self-help skills, while of boys social problems; parents of younger children – communication, while of older children behaviour, self help skills and occupation	Study from Greece
Keenan (2010)	Parental experiences at diagnosis & forward planning	Combined clinic & community N = 100 ASD	1-16 years	Parent and professional needs questionnaire	Diagnostic & planning processes stressful for parents; long time for diagnosis & limited participation for planning major concerns	Study from Ireland; combined sample
Kogan (2008)	Health care experiences of families of children with ASD in US	National survey N = 2088 ASD	3-17 years	Survey	Special health care needs (other emotional, developmental or behavioural problems) increased unmet needs of parents of children with ASD. Caused significant financial and employment burden.	Large survey
Bromley (2004)	Mothers supporting children with ASD: satisfaction with services	National Autism Society Database N = 68 ASD	2-18 years	Interviews	Challenging behaviours of children associated with unmet needs and high psychological stress of mothers	Database based study

A recent study exploring needs of parents of children with ASD through semi-structured interview reported that parents needed support during diagnosis as well as for future planning to avoid crisis (Molteni and Maggiolini, 2014). In a research database-based study of children with ASD, the common unmet needs included those for social activities for the child, information about services and continuous service provision (Brown et al., 2012). Another combined sample study from Ireland reported lengthy waiting time for diagnosis and limited family participation for planning as major concerns (Keenan et al., 2010).

Parental and child factors were shown to influence parental perception of unmet needs in a study of children with ASD in Greece using an open ended questionnaire (Papageorgiou and Kalyva, 2010). A research database-based study reported that lower functional independence of children and impact on family correlated with high unmet parent needs (Brown et al., 2011). There are other studies also showing the influence of child and parent characteristics on parental perception of unmet needs (Bromley *et al.*, 2004; Kogan *et al.*, 2008; Papageorgiou and Kalyva, 2010; Brown *et al.*, 2011; Foody *et al.*, 2014).

3.9.3 Measures to assess unmet parent needs

Open ended questionnaires (Papageorgiou and Kalyva, 2010) and semi-structured interviews (Molteni and Maggiolini, 2014) were utilised in literature to assess the unmet parent needs. The Family Needs Questionnaire (Brown et al., 2011; Brown et al., 2012) and the Parent Needs Questionnaire (Dillenburger et al., 2010; Keenan et al., 2010) were also used. A measure to evaluate the unmet needs regarding co-existing conditions in children with ASD, and thus families' needs for support from service providers, was not identified from this literature review.

3.9.4 Summary of review of unmet needs

Recent reviews and articles have attempted to describe and define the unmet parent needs in families of children with ASD. Family and child characteristics ahave been shown to influence the unmet needs in the limited literature available.

3.10 Summation of all literature reviews

Compilation of evidence from widely varying methodologies of literature was a challenge with primary diagnosis varying from broad ASD to specific subtypes; definition of some of the conditions including co-existing conditions, sleep problems and unmet needs being in the process of being described; different sampling frames studied; and the specific measures to assess these co-existing conditions in children with ASD being in the process of development. Though there is a consensus of high frequency of co-existing conditions in children with ASD, there are conflicting reports about their predictors. Almost all of the studies that evaluated the influence of co-existing conditions on familial adaptation have used measures developed for the general population.

The present project proposes to bridge this gap in the literature to evaluate the effect of co-existing conditions as measured by appropriate tools on familial adaptation. The project aims to include specific measures for the ASD population in the study and the best alternative option, if none available. Drawing from the background information elicited in the literature review, the hypotheses are enumerated as follows.

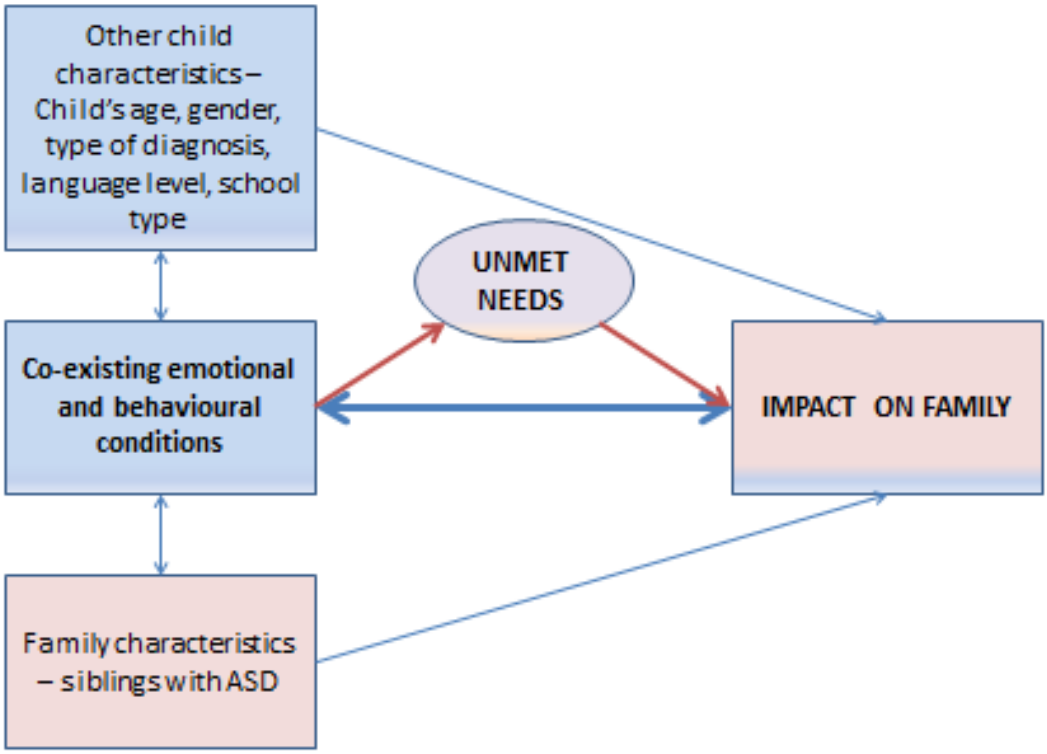
3.11 Research Hypotheses

1. There is no difference in the prevalence of co-existing conditions including total co-morbid psychopathology and total problem behaviour of children with ASD in different groups of ASD diagnosis, age, gender, type of school and language level.
2. Individual types of co-morbid psychopathology will differ between children according to age group and ASD diagnosis such that:
    a. Total anxiety scores will be higher in the older age group and children with Asperger Syndrome.
    b. Feeding issues such as under eating, limited food variety and food refusal will be more common in younger children.

c. Total sleep problems will be seen across both older and younger age groups.
d. Younger children will have higher atypical sensory behaviour and sensory seeking behaviour scores compared to older children.
e. Atypical sensory behaviour will have direct correlation with comorbid psychopathology and problem behaviours.

3. Irrespective of the type of diagnosis, language level, school type, gender and age of the child, total co-morbid psychopathology and total problem behaviours will impact directly on the family.
4. Externalising behaviours will have more impact on family than internalising behaviours.
5. Regarding unmet parent needs, it is hypothesised that parents who have met more professionals will report they received more support for co-existing conditions. Similarly parents who have received more informal support will also report more support for these conditions.
6. Parental perception of unmet need will have a mediational effect on the relationship between coexisting conditions and impact on family (Figure 4).

Figure 4: Model showing mediational effect on unmet needs on relationship between co-existing emotional and behavioural conditions and impact on family



Blue colour – Child characteristics, pink colour – family characteristics

4 Chapter 4. Methodology

4.1 Introduction

This section describes the methodology for the project. It includes the design of the study, participants including the databases from which they are drawn, measures, procedures and analysis.

4.2 Design of the study

The design is a cross-sectional observational study. Standardised parent-report questionnaires were used to collect information about co-existing conditions and the impact on the family.

4.3 Research participants

The research participants were recruited from two ASD research databases.

4.3.1 Databases

The Newcastle University ASD research team coordinate two databases holding information about families who have children with ASD. Parents consent for data to be held about their child and to be contacted about research projects (through separate existing ethical approvals).

4.3.1.1 Dasl^{n}e (Database of children with ASD living in the North East of England)

Dasl^{n}e (the Database of children with ASD living in the North East of England) is a regional database of families with children with ASD in the North East of England covering six districts including Gateshead, Newcastle, North Tyneside, Northumberland, South Tyneside and Sunderland. This database has a population sampling frame, and is representative of the overall childhood ASD population in the North East of England (McConachie *et al.*, 2009). It now includes more than 1400 families. For recruitment, parents of newly diagnosed children with ASD are approached usually by their local clinical team. After informed consent, they complete the short Dasl^{n}e questionnaire and nominate a professional to complete the professional questionnaire.

4.3.1.2 Autism Spectrum Database – UK (ASD-UK)

The ASD-UK is a national family research database of children with ASD and covers the rest of the UK (areas outside the North East of England). ASD-UK includes more than 1200 families recruited through paediatricians and child development teams across the UK. Professionals inform parents of children with ASD about the database and provide an information sheet that explains the database. Parents contact the research team through the contact form provided. ASD-UK collects data about the same child characteristics as Dasl^{n}e. Analysis shows ASD-UK is likely to be representative of children with ASD in the UK (Warnell, 2013).

At enrolment, both databases use identical ten-item parent questionnaire to collect information from parents about co-existing conditions in children with ASD (Appendix 3).

4.3.2 Recruitment of Participant Parents

4.3.2.1 Inclusion Criteria

Parents of children with ASD enrolled in either database with the following inclusion criteria: parents of children aged 2-18 years of age; who gave informed consent to participate in this project.

There was no exclusion criterion.

4.3.2.2 Sample Size calculation for participant parents

In published studies the prevalence of co-existing emotional and behavioural conditions reported ranges from 22% to more than 90% (Matson and Nebel-Schwalm, 2007b; Hollway and Aman, 2011; van Steensel *et al.*, 2011; Kozlowski *et al.*, 2012a; Mannion *et al.*, 2013; Maskey *et al.*, 2013). Taking the average prevalence as 50%, the required sample size was 384 using the following formula:

$$n = \frac{t^2 \times p(1-p)}{m^2}$$

n = required sample size

t = confidence level at 95% (standard value of 1.96)

p = estimated prevalence from previous studies
m = margin of error at 5% (standard value of 0.05)

4.4 Measures

We used the ten-item parent database questionnaire, completed by parents at enrolment, to obtain baseline information about co-existing conditions (Appendix 3).

4.4.1 Ten-item parent database questionnaire

At the time of enrolment to the databases, parents give detailed information about their child's characteristics. This information includes the frequency of 10 common co-existing conditions (sleep problems, hyper periods, toileting problems, temper tantrums, aggression to other people, injury to self, reluctance to separate from parent, anxiety, selective about eating, sensory reactions). For the majority of families, the information is less than 2 years old or recently updated. For Dasl^{n}e, immediately before this study, a request for an update of co-existing conditions was sent to all parents whose child was enrolled in Dasl^{n}e before January 2011 and aged 2-18 years, by post or e-mail by the database management. This questionnaire is provided in Appendix 3 and is referred to hereafter as the 'screening questionnaire'.

4.4.2 Selection of tools for this study

A systematic review of available literature was carried out to enable selection of appropriate parent questionnaire tools for common co-existing conditions in children with ASD based on conditions identified on the screening questionnaires. Tools were selected to investigate behaviour, hyperactivity, anxiety, feeding, sleep, and sensory difficulties based on the following criteria:

- Domains measured by the tool
- Age range
- Format of the tool and the type of respondents
- Use in children with ASD
- Psychometric properties including validity and reliability

- Acceptance of tool as a measure beyond the developing research group

The summary of tools considered is enclosed in Appendix 1, with detailed information on reliability and validity evidence for each tool.

4.4.3 Tools used in the study

The following standardised tools were selected for the study

4.4.3.1 The Autism Spectrum Disorder-Comorbidity Child Version (ASD-CC) (Matson and Wilkins, 2008; Matson *et al.*, 2009b)

This tool was used for all children aged 2-18 years of age to identify their behavioural co-morbidity including problem behaviours. The tool is validated for 2-16 years of age, but was used up to 18 years of age in our study for continuity of analysis. The ASD-CC evaluates co-morbid psychopathology and is part of the same assessment battery that has the Autism Spectrum Disorder-Problem Behaviour Child Version (ASD-PBC). The ASD-CC has 39 items and seven factors – tantrum behaviour, repetitive behaviour, worry/depressed, avoidant behaviour, under-eating, conduct, over-eating. It has the following options for each item: 0 – 'not a problem or impairment'; 1 – 'mild problem or impairment' and 2 – 'severe problem or impairment'. Parents could mark X if an item did not apply to their child.

The ASD-CC has moderately good inter-rater (κ=.46) and test-retest reliability (κ=.51) (Matson and Wilkins, 2008; Thorson and Matson, 2012). It has good internal consistency (α=.91). The subscales of the ASD-CC except avoidant behaviour were shown to have acceptable construct validity by correlation with subscales of the BASC-2 (Matson et al., 2009b). The possible minimum and maximum scores for ASD-CC are 0 and 78 respectively.

4.4.3.2 The Autism Spectrum Disorder-Problem Behaviour Child Version (ASD-PBC)

The ASD-PBC is a measure developed for assessment of problem behaviours of children with ASD. It is validated for 2-16 years of age, but was used up to 18 years of age for the continuity of analysis. The ASD-PBC has 18 items and two factors: internalising and externalising. It has the following options for each item: 0 – 'not a problem or impairment'; 1 – 'mild problem or impairment' and 2

– ‘severe problem or impairment’. It has acceptable reliability and validity (Matson et al., 2008a; Mahan and Matson, 2011; Beighley et al., 2013). The possible minimum and maximum scores for ASD-PBC are 0 and 36 respectively.

4.4.3.3 The Impact on Family Scale (Stein and Riessman, 1980; Stein and Jessop, 2003)

This tool was used to assess the parents’ perceptions about impact on family and has 15 questions and one factor. The possible minimum and maximum scores are 15 and 60 respectively. Parents can rate each item on a four point Likert Scale as 1 – ‘strongly agree’; 2 – ‘agree’; 3 – ‘disagree’; or 4 – ‘strongly disagree’ with the items reverse scored as necessary. The scale has acceptable reliability and validity (Stein and Riessman, 1980; Stein and Jessop, 2003).

4.4.3.4 The Conner’s 3 ADHD index

This tool was used for children aged 2-18 years of age, whose parents reported concerns regarding ‘hyper’ periods in the screening questionnaire. It contains 10 questions to evaluate hyperactivity and inattention. Parents rate on a Likert scale: 0 – not true at all (never, seldom); 1 – just a little true (occasionally); 2 – pretty much true (often, quite a bit); and 3 – very much true (very often, very frequently). The Likert scale was converted to raw score based on instructions for each item. The possible minimum and maximum scores are 0 and 30 respectively.

4.4.3.5 The Spence Children’s Anxiety Scale – Parent version (SCAS-P) (Spence, 1998; Nauta *et al.,* 2004)

This tool was used for children whose parents reported concerns regarding anxiety or reluctance to separate from parent in the screening questionnaire. The school version was used for all children aged 2-18 years of age for continuity of analysis. The SCAS-P is appropriate for describing anxiety in children who use words; the covering instructions therefore explained that parents should not complete this where they considered the questions inappropriate for their child. The SCAS-P has 38 items and response to each item is on a Likert scale of never, sometimes; often and always. It has six subdomains – panic attack and agoraphobia, separation anxiety, physical injury

fears, social phobia, obsessive compulsive disorder and generalised anxiety disorder.

4.4.3.6 The Short Sensory Profile (SSP) (Dunn, 1994; Ermer and Dunn, 1998)

The SSP is validated for children up to 10 years of age. This tool was used for children aged 2-18 years of age, whose parents reported concerns regarding sensory behaviours in the screening questionnaire; for continuity of analysis. The SSP has been widely used in children with ASD and has 38 items with Likert response of 'Always', 'Frequently', 'Occasionally', 'Seldom' and 'Never'. It has seven sub-domains – tactile sensitivity, taste/smell sensitivity, movement sensitivity, seeks sensation/under-responsivity, auditory filtering, low energy/weak and visual/auditory sensitivity.

The subdomain of sensory sensitivity is shown to overlap with sensory under-responsivity according to scoring instructions of this scale. The sensory under-responsivity sub-domain is reported to be more commonly affected in children with ASD (Ben-Sasson *et al.*, 2009). But analysing individual items in the sensory under-responsivity/seeks sensation sub-domain of the SSP, most of the items characterise sensory seeking behaviour; thus this thesis will analyse the sensory seeking/under-responsivity sub-domain and will describe it as sensory seeking.

4.4.3.7 The Children's Sleep Habits Questionnaire (CSHQ) (Owens *et al.,* 2000)

The Children's Sleep Habits Questionnaire is validated for 2.5-10 years of age and had been used in other studies in older children (Goldman *et al.*, 2012). This tool was used for children, aged 2-18 years of age, whose parents reported concerns regarding sleep in the screening questionnaire, for continuity of analysis. It has 33 items and sub-domains of bedtime resistance, sleep onset delay, sleep duration, sleep anxiety, night wakings, parasomnias, daytime sleepiness and sleep disordered breathing. Parents rate items on a three-point Likert scale based on the last week: rarely – never or one time a week; sometimes – 2-4 times a week; or usually – 5 or more times a week. The second column of questions is to determine whether that item is a problem for parent/caregiver with responses 'Yes', 'No' or 'N/A'.

4.4.3.8 The Brief Autism Mealtime Behaviour Inventory (BAMBI) (Lukens and Linscheid, 2008)

The BAMBI is validated up to 11 years of age. For continuity of analysis, this tool was used for children, aged 2- 18 years of age, whose parents reported concerns regarding eating in the screening questionnaire. Parents can rate behaviour based on the last six months a five point Likert scale: 1 – never/rarely; 2 – seldom; 3 – occasionally; 4 – often; or 5 – at almost every meal. The second part of each item enquires whether that is a problem for the parent/caregiver with the options of 'Yes' and 'No'. It has 18 items and three factors – limited food variety, food refusal and features of autism.

4.4.4 The Needs and services questionnaire

A self-developed measure was utilised to estimate the met and unmet needs of parents regarding co-existing conditions in children with ASD. We used three previous surveys to develop a parent report questionnaire to assess these needs of parents and their experience of the formal and informal services and support available; previous surveys included the national survey of parents caring for a severely disabled child (Beresford, 1995), the evaluation of stress and coping in families (Quine, 1989) and a Newcastle based survey of behavioural problems in children with neuromuscular disease (Darke *et al.*, 2006). The Needs and services questionnaire, in particular the layout and response format, were piloted with parents of children with ASD and typically developing children and their suggestions incorporated. The final version was seen and approved by parents and had five questions (Appendix 4).

The first question explored the areas where parents wanted support and advice over the last two years for co-existing conditions: feeding/eating problems, sleep problems, anxiety, hyperactivity, other behavioural problems and sensory problems. Further four questions maintained this format of co-existing conditions to understand the support and unmet parent needs for individual condition.

The second question explored the level of support obtained from professionals and non-professionals for each individual condition. The professionals enumerated under the options ranged from clinical professionals (paediatrician, psychologists, etc.) to education professionals (teachers, special support

assistant, etc.). Parents were encouraged to tick all professionals, whom they had met for support for a condition. The number of professionals met by parents for a condition was calculated by adding the number of positive responses for that individual condition. The options under non-professional help included voluntary organisations and other parents, and a positive response of support from either of them was counted as non-professional help.

The third question explored the level of support as perceived by parents for individual co-existing condition. They chose one of the five options for each condition namely: 'got enough support', 'got support, but wanted more', 'have not got support, but did not want it', 'have not got support, but wanted it' and 'no support was needed'. The third question explored whether the support provided by professionals was useful or not for individual co-existing condition. The final question explored whether parents still require support for that co-existing condition or not. Parents who reported partial/no support for each co-existing condition, when support was needed, were considered to have unmet need for support. Parents who did not need support and who got enough support when support was needed were considered to have met needs: refer to Appendix 6 for detailed scoring procedures.

4.5 Research methodology

4.5.1 Ethical approval and approval from databases

The project was given favourable opinion on 11th March, 2013 by The Black Country NRES Committee, West Midlands (Appendix 5). The project was submitted to the database research committees and permission to recruit families through the confidential databases was obtained. The Northumberland Tyne and Wear NHS Foundation Trust acted as the sponsor for the study (Appendix 5).

4.5.2 Parent survey

Parents were contacted by post and informed consent for this project was returned in a return-addressed stamped envelope. Appropriate parent-report tools were presented as two packs.

A. Pack 1

All parents in both the databases who fulfilled the inclusion criteria were sent Pack 1 along with the cover letter, information sheet, instructions for completion and a return-addressed stamped envelope. Parents who joined the databases more than 2 years ago, and who did not respond to the recent update on co-existing conditions, received another prompt regarding the update along with Pack 1. Pack 1 contained ASD-CC and ASD-PBC, the Impact on Family and the Needs and services questionnaire.

B. Pack 2

Pack 2 was individualised based on parental report of the problem that occurred frequently (data from the screening questionnaire). If a parent reported that their child had frequent problems with 'hyper' periods, anxieties (including reluctance to separate from one parent), eating problems, sleep problems or sensory reactions, corresponding questionnaire/s were included in Pack 2. In the event of a parent marking more than one co-existing condition as frequent on the screening questionnaire, Pack 2 contained all the relevant standardised questionnaires and the parent was given the choice of answering at least one of them, based on their current priority and perceptions. They were requested to complete all the questionnaires, if they wished. These instructions were given on the instruction sheet for Pack 2. Pack 2 had some or all of the following measures:

1. The Conner's 3 ADHD index
2. The Spence Children's Anxiety Scale – Parent version (SCAS-P) (Spence, 1998; Nauta *et al.*, 2004)
3. The Short Sensory Profile (SSP) (Dunn, 1994; Ermer and Dunn, 1998)
4. The Children's Sleep Habits Questionnaire (CSHQ) (Owens *et al.*, 2000)
5. The Brief Autism Mealtime Behaviour Inventory (BAMBI) (Lukens and Linscheid, 2008)

Some parents did not receive Pack 2, if the frequent concerns they reported in the screening questionnaire included only temper tantrums, aggression to other people or injury to self. Questions about these areas were included in the ASD-CC and ASD-PBC in Pack 1. Parents who did not report any co-existing condition received only Pack 1.

Our preparatory work showed that each questionnaire would take about 5 minutes to complete. After reading the cover letter, information sheet and instructions, Pack 1 could be completed in 15-20 minutes. It took around 20 minutes to complete Pack 2 when it included all 5 questionnaires. The sample packs were weighed to get the postage right, so that parents did not have to pay excess postage. A reminder was sent to non-responder parents after 2 weeks by post or e-mail.

4.6 Data entry of quantitative data

The quantitative data were entered into IBM SPSS statistics software version 20. Data entry errors were analysed by re-checking around 5% of entries of questionnaires and assessing the distributions and frequencies of each entered item. Missing value analysis was performed for all questionnaires using IBM SPSS software. Patterns were analysed to see whether the missing value was happening at random. Missing value corrections for each questionnaire are described in detail in Appendix 6. Any individual questionnaire with more than 20% of items missing was excluded from the analysis. If the questionnaire had subscales and any subscale had more than 20% missing, the whole questionnaire was excluded. If a questionnaire with only one factor (no subscales) had less than 20% missing, the mean of the whole questionnaire replaced the missing value. If a questionnaire had less than 20% missing from the whole questionnaire and less than 20% missing from its subscales, the respondent's mean of each subscale replaced the missing value.

4.7 Statistical Analysis of quantitative data

4.7.1 Descriptive Analysis

The basic demographic and ASD specific variables were imported from the databases. The demographic variables included gender and age. ASD specific

variables included the type of diagnosis, the type of school attended, the language level and whether siblings were also diagnosed with ASD. The details of variables are provided below and details of their coding in Appendix 7:

- Age was analysed as both continuous and categorical (younger than and including 8 years of age as the younger age group and above 8 years as the older age group).
- Gender was analysed as male and female
- Type of diagnosis of ASD: There were three categories of diagnosis of ASD – Autism, ASD and Asperger syndrome. For mediation analysis, two levels of diagnosis were used – Autism and diagnosis other than Autism.
- Type of school: The school types were categorised into special schools and schooling other than special schools.
- Language level: The children were categorised into two groups – those who spoke fluent sentences and those who did not.

Descriptive analyses were performed on all questionnaire scores. The distribution of the scores was evaluated visually by histogram and box-plot. Skew and kurtosis were evaluated for the distribution to understand normality. If the value of skew divided by standard error is less than 3.29, the distribution was considered normal (Pallant, 2010). Parametric tests were used for normal distribution and non-parametric tests for non-normal distribution. When parametric tests were used, effect sizes were reported as Cohen's d wherever applicable, if the p was less than or equal to .05. When non-parametric tests such as Mann-Whitney U test were used, effect sizes were reported as r wherever applicable, if the p was less than or equal to .05.

4.7.1.1 Variables derived from questionnaires

4.7.1.1.1 The Impact on the Family

The impact on the family was calculated as the total score on the Revised Impact on Family measure (Stein and Jessop, 2003).

4.7.1.1.2 Co-existing Conditions

Co-existing conditions included both co-existing psychopathology and problem behaviour. Co-existing psychopathology was calculated from the ASD-CC and problem behaviour from the ASD-PBC. Regarding co-existing psychopathology, total scores and subscale scores were both used. These scores were compared with results from the parent study analysing 639 children (Thorson and Matson, 2012) and an Irish study evaluating 89 children (Mannion *et al.*, 2013). The ASD-CC subscale score also was analysed as categorical variable where the score was divided into no/minimal impairment/problem and moderate to severe impairment/problem based on cut-off scores published (Thorson and Matson, 2012).

4.7.1.1.3 Unmet parent needs

The unmet parent needs were calculated by addition of unmet needs scores from six domains of sleep, feeding, anxiety, hyperactivity, other behavioural issues and sensory issues from the Needs and services questionnaire. The unmet needs were compared to the objective use of formal and informal support as reported by parents.

4.7.1.1.4 The Conner's 3 ADHD Index

The total score was calculated by adding the scores of ten items on the scale. Using the probability score table available with the measure, the total score was used to predict the probability of classification of ADHD.

4.7.1.1.5 The Spence Children's Anxiety Scale-Parent version

The total score was calculated from the whole measure as per the recommendations available on the website (http://www.scaswebsite.com/). A possible clinical cut-off is indicated by a total score of 37 for boys and 44 for girls (http://www.scaswebsite.com/).

4.7.1.1.6 The Short Sensory Profile

The total score and the sensory seeking sub-scale score were calculated according to the instructions available in the measure. These scores were compared to the norms on the measure to indicate three levels of performance 'typical', 'probable difference' and 'definite difference'.

4.7.1.1.7 The Children's Sleep Habits Questionnaire (CSHQ)

The total CSHQ was calculated according to the instructions available in the measure and total score above 41 is reported to be a cut-off for identification of probable sleep problems (Owens *et al.*, 2000).

4.7.1.1.8 The Brief Autism Mealtime Behaviour Inventory (BAMBI)

The total BAMBI score and subscale scores for limited food variety and food refusal were calculated according to the instructions available in the measure. The factor 'limited food variety' included items reflecting limited variety of food consumed by the child. The factor 'food refusal' included five items related to both food refusal and disruptive mealtime behaviour (Lukens and Linscheid, 2008). There are no cut-off scores for the total score or subscale scores.

Curve estimation was performed to evaluate the relationship between continuous independent and dependent variables. For categorical independent variables such as gender, type of diagnosis, type of school, language level and sibling involvement, student t test was used for analysis.

4.7.2 Hypothesis testing

For hypothesis 1, outcome variables were co-morbid psychopathology (ASD-CC) and problem behaviour (ASD-PBC). Independent student's t-test was used to evaluate differences across variables with two levels - gender, age, type of school and language level (eg: gender – male and female) and one-way ANOVA for variables with three levels (eg: type of diagnosis – Autism, ASD and Asperger). If the distribution was not normal, corresponding non-parametric tests were used. Post-hoc analysis was performed when more than two levels of independent variable was present with a significant result on the initial evaluation. The relationship between age and the ASD-CC subscales was also explored using independent samples student's t-test to understand the evolution of these conditions across younger (≤ 8 years) and older (> 8 years) age groups.

If there were significant results on the uni-variate analysis, further multivariate regression analysis was performed to understand the factors affecting co-morbid psychopathology and total problem behaviours. In multiple regression, age was evaluated as a continuous variable and diagnosis was evaluated as

two levels; children with a diagnosis of autism and those without a diagnosis of autism. For the variable of diagnosis, ASD and Asperger syndrome were recoded as a diagnosis of 'ASD, but not autism'.

For hypothesis 2, data from SCAS-P were used to calculate the total anxiety score. The feeding problems were evaluated using under-eating subscale of the ASD-CC and food refusal and limited food variety factors of the BAMBI scale. Information about sleep problems was gathered from the CSHQ scale. The total atypical sensory behaviour and the sensory seeking subscale score were gathered from the SSP scale. The analysis plan was similar to that for hypothesis 1. To explore the relationship between atypical sensory behaviour and co-morbid psychopathology/problem behaviours, correlational analysis were performed between the Short Sensory Profile (SSP) score and subscale scores of the ASD-CC/ASD-PBC.

For hypothesis 3, the impact on the family was calculated from the Impact on Family (IOF) scale. Preliminary analyses were performed evaluating the following:

- The normality distribution of criterion variable (IOF score)
- The association between independent variables and criterion variable was analysed. Pearson correlations were used when both variables were continuous with normal distribution of the criterion variable. When independent variables were categorical, independent samples student t-test and Analysis of Variance (ANOVA) were performed with categorical variable with two and three levels respectively. For continuous variables, curve estimation was used in addition to affirm the linear relationship.

After the preliminary analyses, a hierarchical regression analysis was conducted with IOF as the criterion variable, demographic variables (age and gender) as independent variables on step 1, ASD specific variables (type of diagnosis, language level, type of school and whether siblings had a diagnosis of autism) as independent variables on step 2, number of moderate-severely impairing co-morbid psychopathology on step 3 and problem behaviours on step 4. The percentage of variance explained at each step and the change of

variance was analysed. Collinearity diagnostics to check for multi-collinearity and residual plots to check for error variance assumptions were performed.

For hypothesis 4, a similar hierarchical regression was conducted with the subscales of ASDCC on step 3 of the model to evaluate the subscale with the maximum impact.

For hypothesis 5, the unmet needs for each co-existing condition and the total unmet needs were calculated from the needs and services questionnaire. The information about the number of professionals involved with the family and whether they received informal support was also obtained from the needs and services questionnaire. After descriptive analysis, the number of professionals met for each co-existing condition was compared to the level of support perceived (enough support, partial support and no support) using an Independent Samples Kruskal-Wallis test. To understand whether the involvement of non-professionals resulted in higher percentage of parents reporting fully met 'enough support', further analysis was performed using cross-tabulation and chi-square tests.

For hypothesis 6, mediation analysis was performed. To demonstrate the relationship between total unmet needs and co-existing conditions, a regression model with total unmet needs as dependent variable with co-existing conditions and demographic and ASD specific variables as independent variables was performed. If this model held well along with hypothesis 3, further mediational analysis was performed using Hayes's model and templates on SPSS (http://www.afhayes.com/public/templates.pdf).

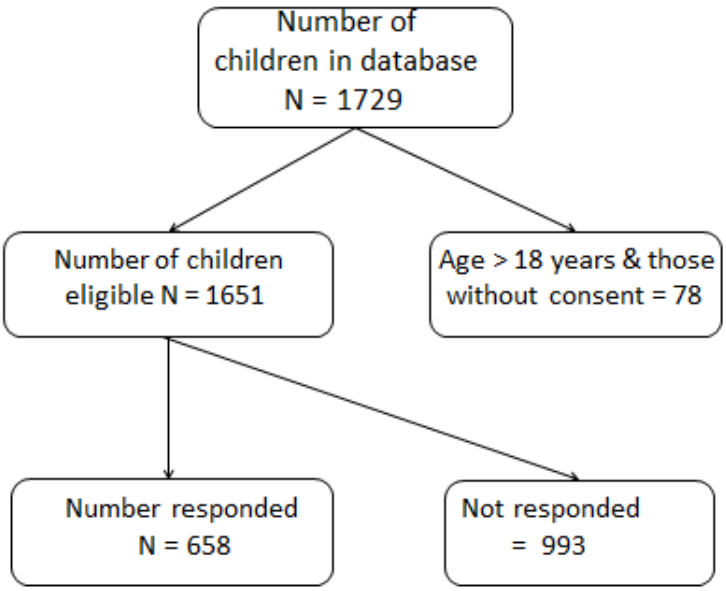
5 Chapter 5. Results

This chapter initially presents a comparison of the characteristics of study responders and non-responders, moving onto internal consistency and descriptive analyses of each questionnaire, and then to the analyses which address the hypotheses of the study.

5.1 Participant parents

The total number of children in both databases at the time of recruitment was 1729 and 1651 children fulfilled the inclusion criteria to participate in this study. 658/1651 eligible children from both databases participated in the study (39.9%) (Figure 5).

Figure 5: Flowchart of study participants



The details of different phases of sending and the response rates are enclosed in Appendix 8. Table 12 compares the characteristics of responders and non-responders to the study with the whole group of eligible children in the databases. The characteristics evaluated include age, gender, type of diagnosis, language level, type of school, siblings with ASD and the mean number of frequent co-existing emotional and behavioural conditions (frequent defined as behaviour is present three or more times a week) as reported by parents on the screening questionnaire within the ASD databases.

Table 12 shows no significant differences between the characteristics of responders and non-responders with the exception of age. The children of non-responders were slightly older in age (10.92 for non-responders vs. 9.71 for responders); however, the standard deviations were comparable.

Table 12: Comparison of characteristics between responders, non-responders and the whole group

	Whole group n = 1651	Non-responders n = 993	Responders n =658	Comparison: responders vs. non-responders
Age Mean (SD)	10.44 (3.9)	10.92 (3.9)	9.71 (3.8)	p = .000[a]
Gender n (%)	1380 (83.7%)	838 (84.4%)	542 (82.7%)	p = .376[b]
Type of diagnosis n (%) Autism ASD Asperger	 354 (21.4%) 1000 (60.6%) 297 (18.0%)	 210 (21.1%) 610 (61.3%) 175 (17.6%)	 144 (22.0%) 390 (59.5%) 122 (18.6%)	p = .748[b]
Type of school n (%) Preschool Mainstream school Special school	 174 (10.5%) 944 (57.2%) 533 (32.2%)	 98 (9.8%) 562 (56.5%) 335 (33.7%)	 76 (11.6%) 382 (58.2%) 198 (30.2%)	p = .243[b]
Language level Sentences n (%)	900 (54.5%)	529 (53.2%)	371 (56.6%)	p = .189[b]
Siblings with ASD n (%)	175 (10.6%)	109 (11.0%)	66 (10.1%)	p = .624[b]
Number of frequent problems Mean (SD)	3.89 (2.7)	3.90 (2.7)	3.88 (2.6)	p = .883[a]

[a] – Student t-test, [b] – Chi-square test

5.2 Background information for the study sample

5.2.1 Demographic and ASD specific variables

The demographic and ASD specific variables for the children whose parents participated in the study were obtained from the databases (Table 13). Most children had a diagnosis of ASD (59.4%). More than 80% of children in each diagnostic group were male. The mean age at diagnosis was 4.2 years for

children with Autism; and 5.2 years and 7.4 years for children with ASD and Asperger syndrome respectively. The Asperger syndrome group had a higher percentage (88.6%) of children in the older age range of above eight years of age.

Table 13: Baseline characteristics of children in the study (n=658)

	Autism	ASD	Asperger
Number of children n(%)	144 (21.9%)	391 (59.4%)	123 (18.7%)
Gender[1] - Male n (%) - Female	117 (81.3%) 27 (18.8%)	319 (81.8%) 71 (18.2%)	108 (87.8%) 15 (12.2%)
Mean age at diagnosis (years)[2]	4.2	5.2	7.4
Age bands at project n (%)			
≤ 8 years of age	60 (41.7%)	176 (45%)	14 (11.4%)
>8 years of age	84 (58.3%)	215 (55%)	109 (88.6%)

[1] – Information missing for 1
[2]– Information about age of diagnosis missing for 81

5.2.2 Proportion of children with frequent co-existing emotional and behavioural conditions

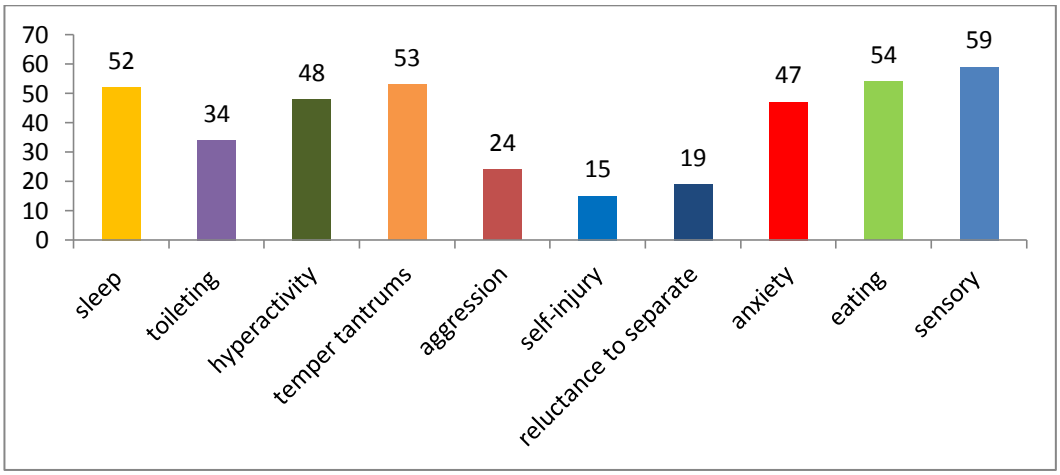
Table 14 shows the rate of frequent co-existing emotional and behavioural conditions as reported by the parent in the screening questionnaire (occurring more than three times per week) for the study population. Almost 90% of children were reported to have at least one co-existing emotional and behavioural condition. 55% of children were reported to have four or more co-existing conditions.

Table 14: Proportion of children with frequent co-existing condition/s (more than three times a week) (n=658)

Number of problems	0	1	2	3	4	5	6	7	8	9	10
n (%) children	76 (11.6)	66 (10)	80 (12.2)	80 (12.2)	86 (13.1)	87 (13.2)	65 (9.9)	60 (9.1)	34 (5.2)	16 (2.4)	8 (1.2)

Figure 6 shows that the frequently reported conditions in study children most commonly included sensory problems (59%), eating problems (54%), temper tantrums (53%) and sleep problems (52%).

Figure 6: Percentage of children with individual frequent emotional and behavioural conditions (n=658)



5.3 Results for the measures included in Pack 1

5.3.1 Response rate and exclusions for Pack 1

All parents received the measures included in Pack 1. The details of the number of questionnaires sent out to all database families, the responses received, the number of questionnaires excluded (>20% of scale/subscales missing) and the final number included in analyses are given in Table 15.

Table 15: Response rate and exclusions for the questionnaires included in Pack 1

Questionnaire	Sent	Received (% response)	Excluded (>20% missing)	Included in analyses
ASD-CC	1651	658 (39.9%)	19	639
ASD-PBC	1651	658 (39.9%)	2	656
Needs and services questionnaire	1651	651 (39.4%)	102	549
Impact on Family	1651	658 (39.9%)	1	657

Eight parents returned the unmet needs and services questionnaire without filling any details and 37 parents did not turn the page over to fill in details. This resulted in the questionnaire being excluded for these families as the unmet

needs section was on page two of the questionnaire. However, the parents included in this analysis did not differ from those who were not included, in the rates of frequent co-existing emotional and behavioural conditions on independent student t-test (3.87 vs 3.84 respectively; t=-.222, p =.822)

5.3.2 Co-existing/co-morbid psychopathology (ASD-CC) in the study population

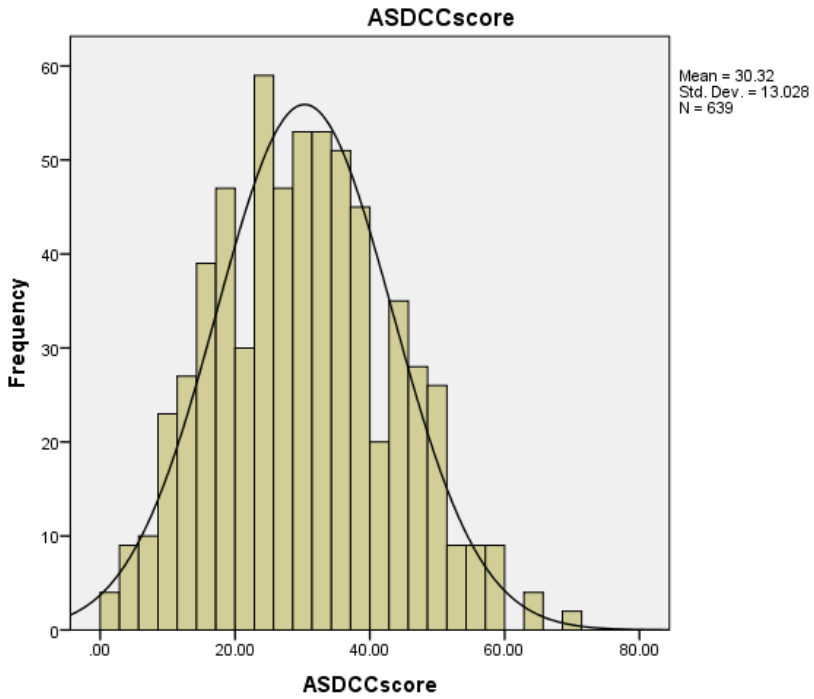
The ASD-CC is designed to detect co-morbid psychopathology in children with ASD and has seven factors (Matson *et al.*, 2009b). The Cronbach's alpha for internal consistency for the total measure in the present study sample was good at α .920. The Cronbach's alpha for the subscales ranged from .722 to .854. The mean scores of each subscale are provided in Table 16 to enable comparison with the other studies which have used the measure (Thorson and Matson, 2012; Mannion *et al.*, 2013); their data are also shown in Table 5. The mean scores in this study are comparable to those by Mannion et al., 2013 (p values ranged from .07 to .96). The mean scores of tantrum behaviour (p=.00), repetitive behaviour (p=.02), conduct (p=.00) and over-eating (p=.04) were higher than those published by Thorson and Matson, 2012. However, the standard deviations of all the subscales were comparable. The ASD-CC scale had a normal distribution (Figure 7).

Table 16: Mean of subscale scores for the ASD-CC (SD)

	Present study (n= 639) Mean (SD)	Thorson and Matson, 2012* (n=639) Mean (SD)	Mannion et al., 2013** (n=89) Mean (SD)
Tantrum Behaviour	9.36 (4.8)	6.70 (4.2)	8.66 (4.2)
Repetitive Behaviour	6.72 (3.9)	4.91 (3.7)	7.00 (3.7)
Worry/depressed	3.51 (2.6)	2.69 (2.8)	3.08 (2.7)
Avoidant Behaviour	6.22 (3.1)	4.23 (2.9)	5.63 (2.9)
Under-eating	0.97 (1.6)	0.82 (1.5)	1.01 (1.7)
Conduct	2.10 (2.1)	1.49 (1.8)	1.81 (1.9)
Over-eating	1.52 (1.8)	1.20 (1.6)	1.35 (1.9)

*- Combined clinic, community and school sample covering the United States of America and southern Canada, Age: 2-17 years; **- Community recruitment sample from schools and parent groups, Age 3-16 years

Figure 7: Distribution of ASD-CC scores (Mean = 30.32; Median = 30; Skew =.212; Kurtosis =-.392)



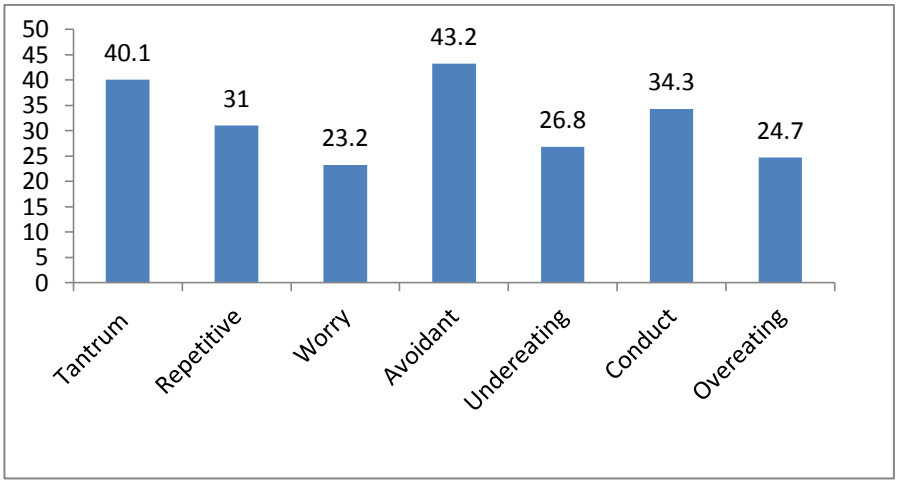
The ASD-CC subscales were split into no/minimal impairment and moderate to severe impairment according to the published cut-offs (Thorson and Matson, 2012). The frequency of the different levels of impairment in each subscale is shown in Table 17; parentheses provide the raw value cut-offs for each subscale. The majority of children had no impairment in each subscale. The report of moderate impairment ranged from 14.7% (under-eating) to 25.9% (avoidant behaviour). Severe impairment was most frequently reported for tantrum behaviour (17.6%), and the least frequently for worry (3.4%).

Figure 8 provides combined moderate-severe impairment for each subscale. The highest levels of moderate-severe impairment were reported for avoidant behaviour (43.2%) and tantrum behaviour (40.1%) and the lowest level was reported for worry (23.2%).

Table 17: The proportion of children with different levels of impairment for each ASD-CC subscale (n=639)

	Level of impairment	Number	Percentage
Tantrum	No impairment (0-10)	383	59.9%
	Moderate impairment (11-14)	144	22.6%
	Severe impairment (≥15)	112	17.6%
Repetitive	No impairment (0-8)	441	69%
	Moderate impairment (9-12)	141	22%
	Severe impairment (≥13)	57	9%
Worry/depressed	No impairment (0-5)	491	76.8%
	Moderate impairment (6-8)	126	19.8%
	Severe impairment (≥9)	22	3.4%
Avoidant	No impairment (0-6)	363	56.8%
	Moderate impairment (7-9)	166	25.9%
	Severe impairment (≥10)	110	17.3%
Under-eating	No impairment (0-1)	467	73.2%
	Moderate impairment (2-3)	94	14.7%
	Severe impairment (≥4)	77	12.1%
Conduct	No impairment (0-2)	420	65.7%
	Moderate impairment (3-4)	135	19.2%
	Severe impairment (≥5)	84	13.1%
Over-eating	No impairment (0-2)	481	75.3%
	Moderate impairment (3-4)	101	15.8%
	Severe impairment (≥5)	57	8.9%

Figure 8: The proportion of children with moderate to severe impairment in each subscale of ASD-CC (%) (n=639)



Considering total moderate to severe impairment across all subscales, 80% of families reported their child to have moderate to severe impairment in at least

one co-morbid psychopathology; 42% reported moderate to severe impairment in three or more subscales (Table 18).

Table 18: Proportion of children with moderate to severe impairments on ASD-CC subscales representing different co-morbid psychopathologies (n=639)

Number of subscales	0	1	2	3	4	5	6	7
n children (%)	130 (20.3)	129 (20.2)	112 (17.5)	106 (16.6)	86 (13.5)	48 (7.5)	24 (3.8)	4 (0.6)

5.3.3 Problem behaviour (ASD-PBC) in children with ASD

The ASD-PBC measuring problem behaviour had good internal consistency in this study, with Cronbach's alpha .877 for the total. The externalising subscale had Cronbach's alpha of .876 and the internalising subscale .790. Figure 9 and Table 19 provide descriptive information for the total score and subscale scores of internalising and externalising behaviours. The distribution is positively skewed.

Figure 9: Distribution of total problem behaviours as measured by ASD-PBC (Mean = 9.47; Median = 8; Skew =.866; Kurtosis =.365)

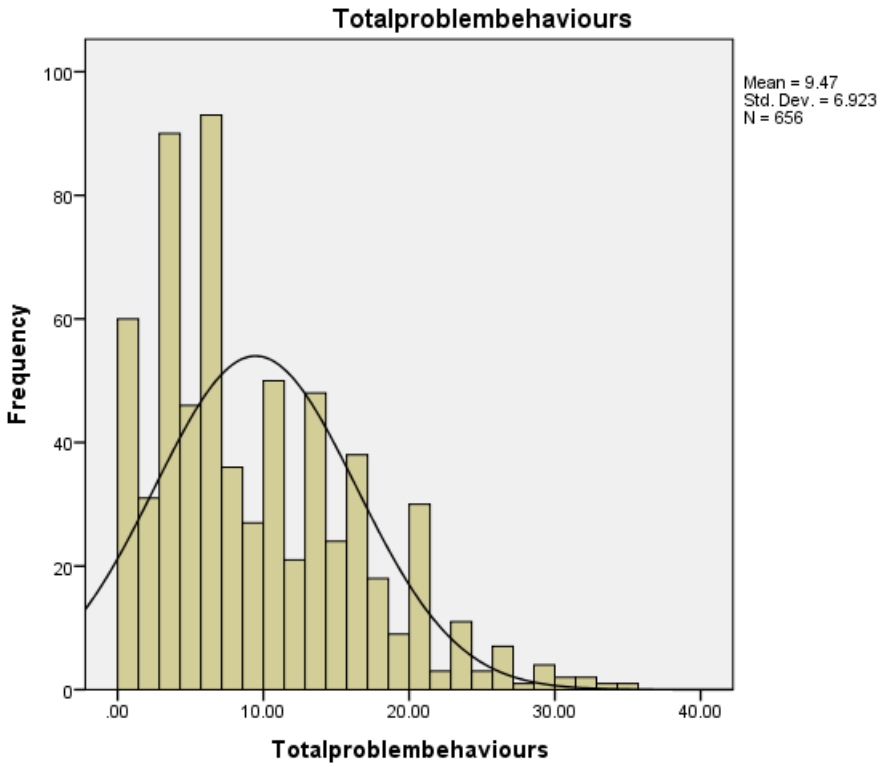


Table 19: Descriptive information for total problem behaviour, externalising and internalising behaviour scores (n=656)

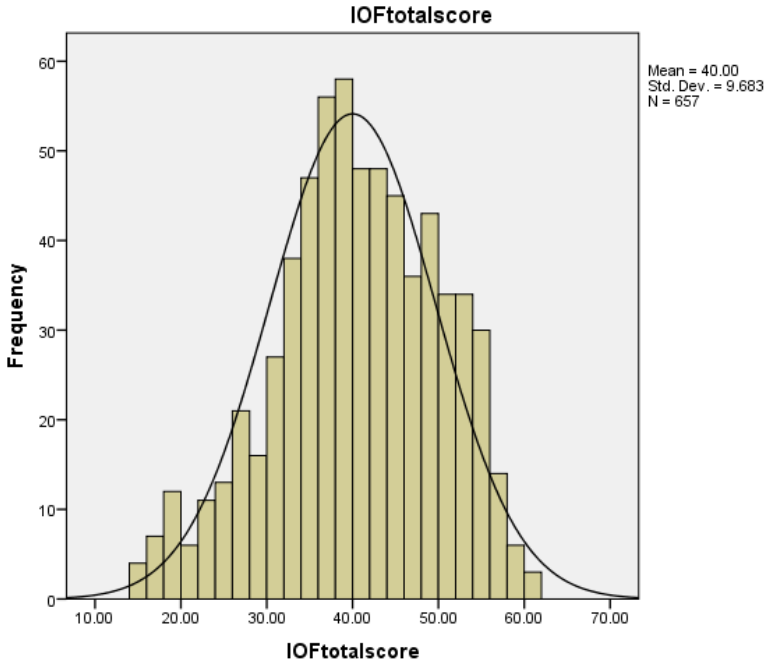
	Total problem behaviour score (n=656)	Externalising behaviour score	Internalising behaviour score
Mean	9.47	4.60	4.88
Median	8.00	4.00	4.00
Standard Deviation	6.9	3.9	3.9
Range	0-35	0-14	0-21

The mean total problem behaviour score and externalising score in the present study were higher than those reported in a sample of 219 children diagnosed with ASD based on DSM-V criteria (8.41 and 3.40 respectively; p=.00 for both). The mean internalising behaviour score was comparable (5.01; p =.18) (Beighley *et al.*, 2013). The standard deviations for both externalising and internalising behaviours were comparable (3.2 and 3.7 respectively) with the present study.

5.3.4 Impact on the family

The Impact on Family measure had a Cronbach's alpha of .91 in this study. Figure 10 shows that the total scores had a normal distribution.

Figure 10: Distribution of Impact on Family (IOF) total scores (Mean = 40; Median = 40; Skew = -.301; Kurtosis =-.368)



5.4 Results for the questionnaires included in Pack 2

Pack 2 data collection was tailored based on parents' report of frequent co-existing emotional and behavioural conditions. The questionnaires included in this pack were the Spence Children's Anxiety Scale – Parent version (SCAS-P) for anxiety, the Short Sensory Profile (SSP) for sensory symptoms, the Children's Sleep Habits Questionnaire (CSHQ) for sleep problems, the Brief Autism Mealtime Behaviours Inventory (BAMBI) for feeding problems and the Conner's 3 ADHD Index for hyperactivity.

Table 20 provides the number of questionnaires sent, the responses received including exclusions for each questionnaire in Pack 2.

Table 20: Response rate and exclusions for the questionnaires included in Pack 2

Questionnaire	Number sent	Number received (% response)	Excluded (>20% missing)	Number included in analyses
Spence Children's Anxiety Scale – Parent version	665	291 (43.7%)	74	217
Short Sensory Profile	778	353 (45.3%)	34	319
Children's Sleep Habits Questionnaire	684	293 (42.8%)	84	209
Brief Autism Mealtime Behaviour Inventory	711	308 (43.3%)	80	228
Conner's 3ADHD Index	628	280 (44.6%)	16	264

The Conner's 3 ADHD Index and the SCAS-P had non-normal distributions. The internal consistency of these measures and their descriptive statistics are provided in Table 21. The median value for the Conner's 3 ADHD index in this study is 26. The total raw score of 20 corresponds to 99% probability of a classification of ADHD, according to the manual's probability score table. The median score of 26 corresponds to a diagnosis of ADHD, which is as expected as all these forms were completed by parents who reported concerns of hyperactivity in the screening questionnaire. The median value of 37 for the SCAS-P total score is at the 'indicative clinical cut-off' for anxiety in boys with ASD (http://www.scaswebsite.com/).

Table 21: Internal consistency and descriptive statistics of SCAS-P and Conner's 3ADHD Index

	Internal consistency (Cronbach's alpha)	Median totalscore	Range
Conner's 3 ADHD Index	.824	26	6-30
Spence Children's Anxiety Scale – Parent version	.916	37	2-106

The SSP, the CSHQ and the BAMBI scores had normal distributions. The internal consistency of these measures and their descriptive statistics are provided in Table 22. The mean values for both total SSP score and sensory seeking score in our study, as would be expected, indicated a 'definite difference' in reference to the scoring instructions of this measure. The mean value of CSHQ total score was higher than the cut-off for identification of probable sleep problems (Owens *et al.*, 2000).

Table 22: Internal consistency and descriptive statistics of SSP, CSHQ and BAMBI

	Internal consistency (Cronbach's alpha)	Mean score (SD)
Short Sensory Profile	.897	108.96 (23.7)
Sensory seeking score	.812	17.98 (6.5)
Children's Sleep Habits Questionnaire	.553	56.87 (9.6)
Brief Autism Mealtime Behaviour Inventory	.627	50.95 (12.3)
Limited food variety	.722	28.25 (6.2)
Food refusal	.832	10.87 (4.9)

5.5 Summary of descriptive analyses of the initial screening and standardised questionnaires

The parents who participated in this study were representative of the families included in the databases from which they were recruited across all variables, with the exception of a small difference in average age. Parents reported high rates of frequent co-existing emotional and behavioural conditions for their children with ASD. Almost 90% of children were reported to have at least one co-existing emotional or behavioural condition. 80% of families reported their child to have moderate to severe impairment in at least one co-morbid psychopathology; 42% reported moderate to severe impairment in three or

more conditions. The descriptive evaluation of measures included in Pack 2 revealed high levels of difficulty for each co-existing emotional and behavioural condition – these results concur with the parents' report of frequent concerns in similar problem behaviour in the screening questionnaire.

5.6 Testing Hypothesis 1

Hypothesis 1: There is no difference in the prevalence of co-existing conditions including total co-morbid psychopathology and total problem behaviour of children with ASD in different groups of ASD diagnosis, age, gender, type of school and language level.

5.6.1 Total co-morbid psychopathology

Parametric tests were used to test this hypothesis with total co-morbid psychopathology (ASD-CC), as the total score had a normal distribution. The variables considered included gender, age, type of ASD diagnosis, language level and type of school.

Independent samples t-tests were conducted with the study sample to test the following characteristics on total co-morbid psychopathology score: gender, age (<8 years vs >8 years), language level (full sentences vs less than full sentences), type of schooling attended (special school vs no special school); the results are provided in Table 23. Gender, age, language level and type of school were not significantly associated with total co-morbid psychopathology score.

Table 23: Associations between child characteristics and total co-morbid psychopathology (ASD-CC) (n=639)

Child characteristic	Mean ASD-CC total score (SD)	t-value	p-value
Gender	male 30.20(13.1)	.518	.604
	Female 30.91(12.9)		
Age	≤8 years 29.50(12.8)	-1.434	.152
	>8 years 30.98(13.2)		
Language level	full sentences 30.8(13.4)	-1.094	.275
	less than full sentences 29.67(12.5)		
Type of school	special school 31.7(13.1)	-1.776	.076

	not attending special school 29.7(13.0)		

A one-way between-subjects ANOVA was conducted to compare the effect of type of diagnosis (ASD vs autism vs AS) on total co-morbid psychopathology score. There was no significant difference in total ASD-CC score between the different diagnostic groups; (F (2,636) = 1.992, p=.137). As hypothesised, no significant differences were found in total co-morbid psychopathology score according to age, gender, language level, type of school and type of diagnosis. Further multiple regression analysis was not done as uni-variate analysis did not reveal any significant predictors.

The relationship between age and the ASD-CC subscales was explored using student's t-test for subscales where there was normal distribution and Mann-Whitney U test for subscales with non-normal distribution; the results are provided in Table 24. Younger children were reported to have more repetitive behaviour and under-eating, while older children had more worry, conduct problems and over-eating.

Table 24: Associations between age and ASD-CC subscales

ASD-CC subscale	Mean subscale score in younger and older age groups (SD)	t-value/ U-value	p-value
Tantrum[a]	Younger 9.64 (4.9)	1.412	.159
	Older 9.11 (4.7)		
Repetitive[a]	Younger 7.29 (3.9)	3.552	.000
	Older 6.20 (3.9)		
Worry[a]	Younger 2.63 (2.4)	-8.378	.000
	Older 4.28 (2.6)		
Avoidant[a]	Younger 5.96 (2.9)	-1.926	.055
	Older 6.42 (3.1)		
Under-eating[b]	Younger 1.17 (1.7)	40464.5	.002
	Older .81 (1.5)		
Conduct[b]	Younger 1.81 (1.9)	44845.5	.003
	Older 2.36 (2.2)		
Over-eating[b]	Younger 1.14 (1.6)	47060	.002
	Older 1.85 (1.8)		

[a] – Independent samples student's t-test [b] – Mann-Whitney U test

5.6.1.1 Total number of moderate-severely impairing types of co-morbid psychopathology

This section analyses hypothesis 1 for the frequency of moderate-severely impairing co-morbid psychopathology. The frequency was calculated by adding the total number of moderate-severely impairing co-morbid psychopathologies. As this distribution was non-normal (Table 18), non-parametric tests were used. The independent variables used were the same child characteristics.

Independent samples Mann-Whitney U tests were used when the independent variable had two levels to compare the distribution and median across groups and the results are summarised in Table 25. No differences were found in the frequency of moderate-severely impairing co-morbid psychopathology based on gender, age, language level or type of school; see Table 25.

Table 25: Associations between child characteristics and the total number of moderate-severely impairing co-morbid psychopathology (n=639)

Variable	Results – median number (range)	U value	p-value
Gender	male 2 (0-7)	28054	.653
	Female 2 (0-6)		
Age	≤8 years 2 (0-6)	47591.513	.204
	>8 years 2 (0-7)		
Language level	full sentences 2 (0-7)	47242.522	.240
	less than full sentences 2 (0-6)		
Type of school	special school 2 (0-7)	38485.540	.060
	not attending special school 2 (0-7)		

The median number (range) of moderate-severely impairing co-morbid psychopathology of children with Autism, ASD and Asperger syndrome was 2 (0-7) for each group; the distribution was similar across each groups in an Independent Samples Kruskal-Wallis Test (H = 1.296; p=.523). Further multiple regression analysis was not done as uni-variate analysis did not reveal any

significant differences. Total co-morbid psychopathology and the number of moderate-severely impairing co-morbid psychopathology were equally prevalent in all groups of children with ASD irrespective of age, gender, type of diagnosis and language level. However, there were differences in some particular types of psychopathology by age.

5.6.2 Problem behaviour in children with ASD

As problem behaviours, measured by the ASD-PBC had a non-normal distribution, the Independent Samples Mann-Whitney U test was used to evaluate the distribution and medians with two independent groups and the Independent Samples Kruskal-Wallis test with more than two independent groups.

The relationship between independent variables and total problem behaviours is explored in Table 26, internalising behaviours in Table 27 and externalising behaviours in Table 28.

Considering total problem behaviours (Table 26), younger children had significantly higher reported problem behaviours than older children, effect size r =.18. Children who spoke in full sentences had significantly lower total problem behaviours than those who spoke in less than full sentences, r = .23. Children who attended special schools had significantly higher total problem behaviours than those who did not attend special schools, r =.11.

Table 26: Associations between child characteristics and total problem behaviours (ASD-PBC) (n=656)

Child characteristic	Total problem behaviours median (range)	U-value	p-value
Gender	male 8 (0-35)	30083.542	.859
	female 8 (0-31)		
Age	≤8 years 10 (0-35)	42320	.000
	>8 years 7 (0-27)		
Language level	full sentences 7 (0-34)	38654.511	.000
	less than full sentences 10 (0-35)		
Type of school	special school 10 (0-35)	38917.526	.004
	not attending special school 7 (0-34)		

Younger children had higher internalising behaviours than older children, effect size r =.26 (Table 27). Children who spoke in full sentences had lower internalising behaviours than those who spoke in less than full sentences, r =.37. Children who attended special schools had higher internalising behaviours than those who did not attend special schools, r =.16.

Table 27: Associations between child characteristics and internalising behaviours (n=656)

Variable	Internalising behaviours median (range)	U-value	p-value
Gender	male 4 (0-21)	30687.5	.979
	female 4 (0-18)		
Age	≤8 years 5 (0-21)	37759	.000
	>8 years 3 (0-17)		
Language level	full sentences 3 (0-21)	30540	.000
	less than full sentences 6 (0-21)		
Type of school	special school 5 (0-21)	36650	. 000
	not attending special school 4 (0-14)		

The total externalising behaviour did not significantly differ based on gender, age, language level or type of school (Table 28).

Table 28: Associations between child characteristics and externalising behaviours (n=656)

Variable	Externalising behaviours median (range)	U-value	p-value
Gender	male 4 (0-14)	29975.5	.812
	female 4 (0-14)		
Age	≤8 years 4 (0-14)	49582.528	.117
	>8 years 3.5 (0-14)		
Language level	full sentences 3 (0-14)	49473	.161
	less than full sentences 4 (0-14)		
Type of school	special school 4 (0-14)	42290	.169
	not attending special school 3 (0-14)		

In summary, child characteristics of age, language level and type of school had significant associations with total problem behaviours. Younger children, those with less language and those attending special schools were reported to have more difficulties. Internalising behaviours also had similar association with age, language level and type of school. No significant differences were found between the groups for externalising behaviours.

A main effect of diagnostic group was found for the median total problem behaviour score (H = 31.266, p=.000) on the Independent Samples Kruskal-Wallis Test. Further analyses between groups were carried out with Independent Samples Mann-Whitney U test. Total problem behaviours were found to be higher in children with Autism (median 11) than in children with ASD (median 7); U =21589, z=-4.001, p=.000, r=.16; and in children with Autism than in children with Asperger syndrome (median 6); U = 5393, z=-5.443, p=.000, r=.21. There was also a significant difference between the total problem behaviours of children with ASD and children with Asperger syndrome; U =19950, z=-2.819, p=.005, r=.11.

A multiple regression analysis evaluated whether any independent variable predicted total problem behaviours (Table 29). The R^2 of the model was .095, implying the predictor variables together accounted for only a small proportion of the variance (9.5%). The type of diagnosis of autism, lower age and lower language level were significant independent predictors. Total problem behaviours were more frequently seen in younger children and those with lower language ability and with a diagnosis of Autism.

Table 29: Multiple regression analysis predicting frequency of total problem behaviours (n=656)

Variable	Coefficient	Standard error	95% CI for B	p value
Gender	.004	.687	-1.283, 1.416	.923
Age (years)	-.174	.075	-.466, -.170	.000
Type of school	.079	.617	-.012, 2.410	.052
ASD diagnosis	.134	.652	.963, 3.523	.001
Language level	-.110	.612	-2.743, -.338	.012

5.7 Hypothesis 2

Individual types of co-morbid psychopathology will differ between children according to age group and ASD diagnosis such that:

a. Total anxiety scores will be higher in the older age group and children with Asperger Syndrome.
b. Feeding issues such as under eating, limited food variety and food refusal will be more common in younger children.
c. Total sleep problems will be seen across both older and younger age groups.
d. Younger children will have higher atypical sensory behaviour and sensory seeking behaviour scores compared to older children.
e. Atypical sensory behaviour will have direct correlation with comorbid psychopathology and problem behaviours.

5.7.1 Anxiety

Anxiety was measured using the Spence Children's Anxiety Scale – Parent version (SCAS-P). Non-parametric tests were used for the total anxiety score, as the distribution was non-normal.

Using a Mann-Whitney U test, older children had significantly higher total anxiety scores than younger children (median 38 vs. 33 respectively and ranges 2-106 vs 8-78 respectively) ($U = 4497$, $z=-2.474$, $p = .013$, $r=.17$).

To evaluate the impact of diagnosis on the total anxiety scores Independent Samples Kruskal-Wallis test was performed. For the total anxiety score, there was a significant main effect of diagnostic group ($H = 5.96$, $p=.05$). Further post-hoc analyses with Independent Samples Mann-Whitney U tests revealed that for total anxiety score, children with Asperger syndrome had higher scores than children with ASD (median 44.5 vs. 35 respectively) ($U = 2085$, $z=-2.356$, p

=.018, r=.16) and higher scores than children with Autism (median 35) (U = 459, z=-1.937, p =.053, r=.13). The descriptive values are provided in Table 30.

Table 30: Median (range) of total anxiety score for children with different types of ASD diagnoses (n=217)

	Asperger Syndrome Median (range) (n=38)	ASD Median (range) (n=146)	Autism Median (SD) (n=33)
Total Anxiety Score	44 (13-98)	35 (2-106)	35 (4-70)

To summarise the results on anxiety, children older than eight years of age had higher total anxiety than younger children. Children with Asperger syndrome had higher total anxiety scores than children with ASD. These findings were as hypothesised.

5.7.2 Feeding problems

Feeding problems were measured in two ways; first using the under-eating subscale of the ASD-CC and second using the factors of the Brief Autism Mealtime Behaviour Inventory (BAMBI). For the under-eating subscale, both the total score and the rates of moderate and severe impairments were used.

14.7% of 639 parents reported moderate impairment and 12.1% reported severe impairment in the subscale of under-eating of ASD-CC (Table 17) (page 117). To compare the total under-eating score between younger (≤ 8 years) and older (>8 years) children, Independent Samples Mann-Whitney U test was used, as the distribution was non-normal. The under-eating mean (SD) total score was significantly higher in younger children (mean 1.17 (1.7)) than in the older children (mean .81 (1.5)); U = 47060, z=-3.142, p=.002, r=.12.

The factors scores of the BAMBI (limited food variety and food refusal) were used to investigate under-eating further. As these scores had normal distributions, independent students t-test was used to compare between younger (≤ 8 years) and older (>8 years) children. For limited food variety, younger children had similar mean scores to older children (28.58 vs 27.8 respectively; t=.948, p =.344). Food refusal scores were significantly higher in younger than older children (12.86(5) vs 8.7(3.8) respectively; t=7.068, p=.000,

Cohen's d = .94). Summarising about feeding problems, younger children had higher under-eating and food refusal compared to older children, as hypothesised, while there was no difference between younger and older age groups regarding limited food variety.

5.7.3 Sleep problems

Sleep problems were measured using the Children's Sleep Habits Questionnaire (CSHQ). As the total score had normal distribution, independent students t-test was used to compare between younger (≤8 years) and older (>8 years) children.

Younger children had a similar total mean score (57.11) to older children (56.66); t=.339, p=.735. Both younger and older children had similar total sleep problem scores.

5.7.4 Sensory problems

Sensory problems were measured using the Short Sensory Profile (SSP). As the total score had a normal distribution, independent students t-test was used to compare between younger (≤8 years) and older (>8 years) children.

In the Short Sensory Profile, lower scores indicate higher atypical sensory behaviour. The mean total sensory score was in the 'definite difference' range for both younger and older children.

There was no difference between younger and older children in total atypical sensory behaviour (mean (SD) 110.28 (24.0) vs 107.51 (23.4) respectively; t=-3.526, p=.300). However, younger children had significantly more sensory seeking behaviours than older children (mean (SD) 16.87 (6.1) vs 19.3 (6.7) respectively; t=1.037, p=.000, Cohen's d=.38). The total atypical sensory behaviour persisted across both younger and older age groups, whilst sensory seeking behaviour was more common in the younger age group than in older children.

5.7.5 Relationship between atypical sensory behaviour and co-existing conditions

It is hypothesised that the atypical sensory behaviour will have direct correlation with comorbid psychopathology and problem behaviours.

The following table evaluates the Pearson-correlations between total SSP score and subscales of co-morbid psychopathology and problem behaviours (Table 31). All correlations in Table 31 are significant correlations between individual co-morbid psychopathology/problem behaviour and sensory behaviour. The strongest correlation was with avoidant behaviour (Pearson's correlation coefficient -.514) and worry (Pearson's correlation coefficient -.466). Other variables which had moderate correlations with atypical sensory behaviour were atypical behaviour and tantrum behaviour. Under-eating, over-eating, conduct, externalising and internalising problems had weak but significant correlations with atypical sensory behaviour. The atypical sensory behaviour correlated with both co-morbid psychopathology and problem behaviours.

Table 31: Correlation coefficients of the SSP score with co-morbid psychopathology and problem behaviour

	Correlation coefficient	Significance
Over-eating	-.122	.030
Conduct	-.268	.000
Under-eating	-.228	.000
Avoidant	-.514	.000
Worry	-.466	.000
Repetitive behaviour	-.419	.000
Tantrum	-.339	.000
Externalising behaviour	-.228	.000
Internalising behaviour	-.206	.000

5.8 Hypothesis 3

Irrespective of the type of diagnosis, language level, school type, gender and age of the child, total co-morbid psychopathology and total problem behaviours will impact directly on the family.

5.8.1 Associations between child characteristics and the Impact on family

The impact on the family was calculated from the Impact on Family (IOF) scale. The score had a normal distribution (Figure 10, page 119). Curve estimation

analysis of all continuous variables such as age, co-morbid psychopathology and problem behaviours revealed that they had linear relationships with the dependent variable of the Impact on Family (IOF) (data not shown). These variables except age had significant correlations with the IOF score, Table 32.

Table 32: Correlation analyses between continuous variables and the IOF score

Variables	Pearson's correlation coefficient	p-value
Age	-.050	.201
Number of moderate-severely impairing co-morbid psychopathology	.462	.000
Over-eating subscale of ASD-CC	.213	.000
Conduct subscale of ASD-CC	.238	.000
Under-eating subscale of ASD-CC	.194	.000
Avoidant subscale of ASD-CC	.438	.000
Worry subscale of ASD-CC	.230	.000
Repetitive subscale of ASD-CC	.414	.000
Tantrum subscale of ASD-CC	.559	.000
Externalising behaviour	.503	.000
Internalising behaviour	.469	.000

The categorical independent variables were gender, type of diagnosis, language level, type of school and sibling being diagnosed with ASD. On Independent Samples Student t-test, there was no difference between boys and girls regarding the impact on the family (mean (SD) 41.62 (9.5) vs 39.67 (9.7) respectively; t=1.957, p=.051). Families who had more than one child with ASD reported more impact than those who had only one child with ASD (mean (SD) 44.59 (8.7) vs 39.49 (9.6) respectively; t=4.107, p=.000). Children who spoke in less than fluent sentences were reported to have more impact on the family than those who spoke fluent sentences (mean (SD) 41.53 (9.3) vs 38.82 (9.9) respectively; t=3.60, p=.000). Children who attended special schools were reported to have more impact on the family than those not attending special schools (mean (SD) 42.87 (9.2) vs 38.76 (9.6) respectively; t=5.089, p=.000).

On ANOVA, there was a significant main effect of diagnostic group (F = 9.389, p=.000) on the impact on the family. The descriptive statistics are provided in Table 33.

Table 33: Mean (SD) of total IOF score for children with different types of ASD diagnoses (n=657)

	Autism Mean (SD) n=142	ASD Mean (SD) n=392	Asperger syndrome Mean (SD) n=123
IOF score	42.99 (8.2)	39.42 (9.73)	38.41 (10.38)

5.8.2 Hierarchical regression analysing predictors for the impact on family

Two hierarchical regression models were built with the impact on family as the criterion variable – level 1 had demographic variables (age and gender); level 2 included ASD specific variables (type of ASD diagnosis, type of school, language level and sibling being diagnosed with ASD); level 3 included co-morbid psychopathologies and level 4 included problem behaviours (internalising and externalising behaviours). The first model used individual co-morbid psychopathology at level 3 (eg: avoidant behaviour, under-eating) while the second model used the number of moderate-severely impairing co-morbid psychopathology at level 3.

Table 34 provides the summary of the first model and gives R^2, SE (Standard Error), change in R^2, change in F and significance of change in F. Table 35 provides details about the significant predictors of each model level. The final model explained 42% of variance of the impact on family; siblings with ASD (p=.046), special schooling (p=.001), avoidant behaviour (p=.000), tantrum behaviour (p = .000) and internalising behaviour (p=.000) were the significant predictors.

Table 34: Model summary of hierarchical regression analysing predictors for impact on the family

Model	R	R^2	Adjusted R^2	SE	Change in R^2	Change in F	df 1	df2	Sig. F change
1	.093	.009	.006	9.684	.009	2.763	2	627	.064
2	.291	.085	.076	9.335	.076	12.932	4	623	.000
3	.642	.412	.399	7.526	.327	48.935	7	616	.000
4	.659	.434	.421	7.392	.023	12.224	2	614	.000

Predictors for each model
1 – age, gender
2 - age, gender, ASD diagnosis, siblings with ASD, type of school, language level

3 - age, gender, ASD diagnosis, siblings with ASD, type of school, language level, avoidant, over-eating, conduct, under-eating, repetitive, worry, tantrum
4 - age, gender, ASD diagnosis, siblings with ASD, type of school, language level, avoidant, over-eating, conduct, under-eating, repetitive, worry, tantrum, externalising, internalising

Table 35: Significant predictors for each level of hierarchical regression model of impact on the family

Model	Variable	Coefficient	Standard error	P value
1	Gender	-.085	1.02	.033
2	Sibling with ASD	.132	1.262	.001
	Type of ASD diagnosis	.112	.948	.006
	Type of school	.167	.891	.000
3	Sibling with ASD	.065	1.03	.041
	Type of school	.117	.734	.001
	Avoidant behaviour	.222	.128	.000
	Tantrum	.425	.091	.000
4	Sibling with ASD	.062	1.013	.046
	Type of school	.113	.722	.001
	Avoidant behaviour	.216	.126	.000
	Tantrum	.332	.113	.000
	Internalising behaviour	.197	.123	.000

The second hierarchical regression analysis was performed with the frequency of moderate-severely impairing co-morbid psychopathology at level 3 with all other levels remaining the same. This model was considered to exclude the overlap between items on the subscales of co-morbid psychopathology and problem behaviours. All the subscales of ASD-CC had significant correlations ranging from .138 to .776 (p=.000) with externalising and internalising behaviours with the exception of the worry subscale.

Table 36: Model summary of hierarchical regression analysing predictors of IOF using frequency of moderate-severely impairing co-morbid psychopathology

Model	R	R^2	Adjusted R^2	SE	Change in R^2	Change in F	df 1	df2	Sig. F change
1	.095	.009	.006	9.671	.009	2.835	2	627	.059
2	.291	.085	.076	9.324	.076	12.857	4	623	.000

3	.529	.280	.272	8.277	.195	168.267	1	616	.000
4	.606	.367	.358	7.772	.087	42.700	2	614	.000

Predictors for each model
1 –age, gender
2 - age, gender, ASD diagnosis, siblings with ASD, type of school, language level
3 -age, gender, ASD diagnosis, siblings with ASD, type of school, language level, number of moderate-severely impairing co-morbid psychopathology
4 - age, gender, ASD diagnosis, siblings with ASD, type of school, language level, number of moderate-severely impairing co-morbid psychopathology, externalising, internalising.

The second model explained 36% variance of the impact on family with sibling(s) with ASD (p=.024), special schooling (p=.003), number of moderate-severely impairing co-morbid psychopathology (p=.000), externalising (p=.000) and internalising behaviours (p=.000) being significant predictors (Table 36 and Table 37).

In both models, the type of diagnosis of ASD did not predict the impact on the family; both co-morbid psychopathology and problem behaviours did have significant impact on family. The frequency of moderate-severely impairing co-morbid psychopathology, externalising behaviour and internalising behaviour predicted the impact on the family (Table 37). This is consistent with the hypothesis that co-morbid psychopathology and problem behaviours will impact directly on the family.

Table 37: Significant predictors for each level of hierarchical regression model of IOF model 2

Model	Variable	Coefficient	Standard error	P value
1	Gender	-.085	1.020	.033
2	Sibling with ASD	.132	1.262	.001
	Type of ASD diagnosis	.112	.948	.006
	Type of school	.167	.891	.000
3	Sibling with ASD	.081	1.127	.019
	Type of ASD diagnosis	.108	.841	.003
	Language level	-.081	.785	.043
	Type of school	.123	.795	.001
	Number of moderate-severely impairing co-morbid psychopathology	.447	.190	.000
4	Sibling with ASD	.074	1.059	.024

	Type of school	.104	.749	.003
	Number of moderate-severely impairing co-morbid psychopathology	.215	.227	.000
	Externalising behaviour	.247	.108	.000
	Internalising behaviour	.199	.110	.000

5.9 Hypothesis 4

It is hypothesised that externalising behaviours will have more impact on the family than internalising behaviours.

The subscales of ASD-CC included under-eating, over-eating, tantrum behaviour, avoidant behaviour, worry and repetitive behaviour. From the uni-variate analysis described in Table 32, the highest correlation with the IOF score was found for tantrum behaviour (r=.559) and avoidant behaviour (r=.438). These two co-morbid psychopathology subscales remained significant predictors for the impact on family in the first hierarchical regression model (Table 35).

Among problem behaviours, both externalising (r=.503) and internalising behaviours (r=.469) had strong positive correlations with the IOF score (Table 32). Both these behaviours remained significant predictors for the impact on family in the second hierarchical regression model of IOF (Table 37).

To summarise, the predictors for impact on the family of children with ASD, the frequency of moderate-severely impairing co-morbid psychopathology and problem behaviours were both significant predictors alongside siblings being diagnosed with ASD and type of schooling attended. Tantrum and avoidant behaviours were the co-morbid psychopathologies that had the greatest impact, while both internalising and externalising behaviours impacted families.

5.10 Hypothesis 5

Regarding unmet parent needs, it is hypothesised that parents who have met more professionals will report they received more support for co-existing conditions. Similarly parents who have received more informal support will also report more support for these conditions.

The unmet needs for each co-existing condition and the total unmet needs were calculated from the Needs and services questionnaire and included parents who reported no support was needed. The parents who reported partial/no support for each co-existing condition, when support was needed, were considered to have unmet need for support. Parents who did not need support and who got enough support when support was needed were considered to have met needs. The information about the number of professionals involved with the family and whether they received informal support was also obtained from the Needs and services questionnaire.

5.10.1 Parents' report of level of support and unmet needs

Parents who indicated they needed support reported differing levels of support ranging from 'enough support', 'partial support' to 'no support' for each co-existing emotional and behavioural condition (Table 38). The range of parents who reported that 'no support was needed/wanted' varied from 50.1% (feeding problems) to 24.8% (other behaviour problems). When support was needed, the highest proportion of parents reported 'enough support' for sleep problems (25.1%) and the highest proportion of parents reported 'no support' for other behaviour problems (14.9%).

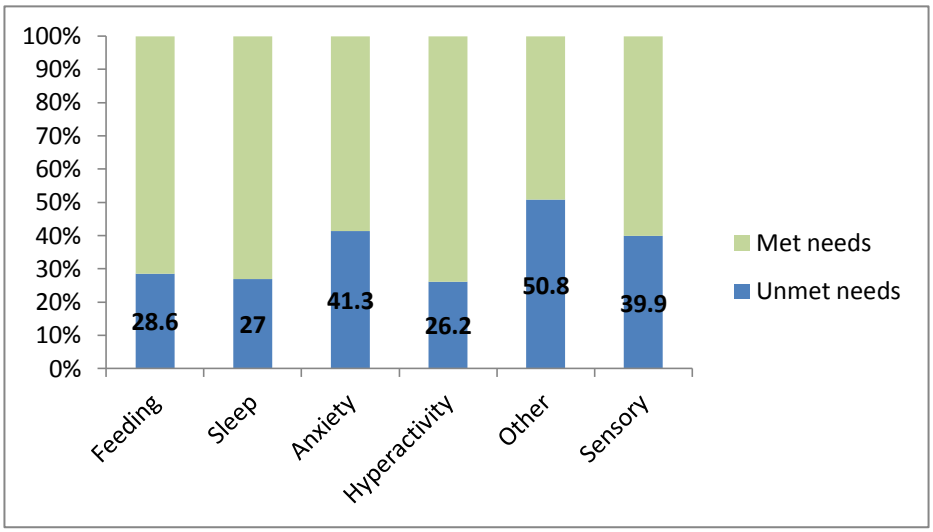
Table 38: Level of support reported for each co-existing condition (n=549)

Level of support	Enough support	Partial support	No support	No support needed/wanted
Feeding problem n(%)	117 21.3%	98 17.9%	59 10.7%	275 50.1%
Sleep problem n(%)	138 25.1%	101 18.4%	47 8.6%	263 47.9%
Anxiety n(%)	109 19.9%	154 18.1%	73 13.3%	213 38.8%
Hyperactivity n(%)	66 12%	96 17.5%	48 8.7%	339 61.7%
Other behaviour problems n(%)	134 24.4%	197 35.9%	82 14.9%	136 24.8%
Sensory problems n(%)	131 23.9%	162 29.5%	57 10.4%	199 36.2%

The highest level of unmet need was reported for other behavioural conditions (50.8%), anxiety (41.3%) and sensory problems (39.9%) (

Figure 11). Parents reported 28.6% unmet needs for feeding problems; 27% unmet needs for sleep problems and 26.2% unmet needs for hyperactivity.

Figure 11: Proportion of parents with unmet and met needs for individual co-existing condition



Regarding all co-existing conditions, 70% of families had at least one unmet need (Table 39). Forty one percent of families had three or more unmet needs for support regarding co-existing emotional and behavioural conditions.

Table 39: Rates and percentages of total unmet needs of families (n=549)

Number of unmet needs	0	1	2	3	4	5	6
Number of parents (%)	169 (30.8%)	72 (13.1%)	82 (14.9%)	76 (13.8%)	70 (12.8%)	50 (9.1%)	30 (5.5%)

5.10.2 Role of professionals

The next evaluation examines the role of professionals in providing support for each co-existing emotional and behavioural condition. Table 40 provides information about the average number of professionals met for each condition compared with parents' perceived level of support.

As expected, parents who did not need/want support met fewer number of professionals compared to parents who needed support. Considering parents who needed support, parents who indicated they received no support met fewer professionals for each co-existing condition, compared with parents who

reported partial and enough support. For example, considering sensory problems, parents reported seeing on average the following number of professionals: parents who reported no support 2 professionals, partial support 3.52 professionals and enough support 2.95 professionals. However, parents who reported they had partial support actually met more professionals compared to those who reported they received enough support for each co-existing condition. For example, considering feeding problems, families who reported enough support met on average 2.46 professionals while those reporting only partial support met on average 2.90 professionals.

To evaluate whether there was a significant difference in the number of professionals met and the support reported when parents needed support, further analysis excluded families who did not want/need support. Non-parametric Kruskal - Wallis test was performed to determine whether there was a significant difference between groups who got full support, partial support and no support regarding the number of professionals they met; the results are provided in Table 40. There was a significant difference in the number of professionals met for sleep problems, anxiety, hyperactivity, other behavioural problems and sensory problems. For all the co-existing conditions in bold in Table 40, parents who reported partial support met more professionals than those who reported enough support.

Table 40: Average number of professionals (SD) met by parents for each condition compared with parent's perceived level of support (n = 549)

Level of support	Enough support	Partial support	No support	No support needed/wanted	Kruskal-Wallis test result analysing enough support vs partial support vs no support (p value)
Feeding problem Mean(SD)	2.46 (1.7)	2.90 (2.2)	2.32 (2.1)	0.17 (0.6)	3.392 (.183)
Sleep problem Mean(SD)	2.09 (1.6)	2.57 (1.9)	1.98 (2.3)	0.22 (1)	8.044 (.018)
Anxiety Mean(SD)	3.19 (2)	3.66 (2.2)	2.70 (2.2)	0.23 (0.9)	10.367 (.006)
Hyperactivity Mean(SD)	2.62 (2.1)	3.44 (2.2)	1.89 (1.9)	0.10 (0.5)	18.506 (.000)
Other behaviour problems Mean(SD)	3.53 (2.3)	4.22 (2.3)	3.33 (2.4)	0.52 (1.2)	12.366 (.002)
Sensory	2.95 (2)	3.52 (2.3)	2.00 (1.9)	0.26 (0.8)	22.596 (.000)

problems Mean(SD)					

5.10.3 The role of support from non-professionals

It is hypothesised that parents who have received support from informal sources will report more support for managing their children's co-existing conditions. The information about the involvement of non-professionals was obtained from the Needs and services questionnaire. A third of families who needed/wanted support for a co-existing condition reported support from non-professionals (Table 41).

Table 41: Proportion of parents requiring support for co-existing condition having contact with non-professionals

	Feeding problem n = 274	Sleep problem n = 286	Anxiety n = 336	Hyperactivity n = 210	Other behaviour problems n = 413	Sensory problems n = 350
n parents (%)	92 (33.6%)	95 (33.2%)	114 (33.9%)	67 (31.9%)	157 (38.1%)	126 (36.0%)

To understand whether the involvement of non-professionals resulted in a higher percentage of parents reporting 'enough support' overall, further analysis was performed using cross-tabulation and chi-square tests and the results presented in Table 42. Families who sought additional help from non-professional informal sources were less likely to report 'enough support' for each co-existing condition, compared to families who did not seek help from informal sources. For example, considering feeding problems, 41.8% of families who had non-professional help reported adequate support, compared to 43.2% of families who did not have non-professional help (difference not statistically significant). Statistically significant differences were found for 'other behavioural problems' and sensory problems; this suggests that despite having more informal support for these conditions, parents continued to report lower levels of 'enough support' for 'other behavioural problems' and sensory problems.

Table 42: Proportion of parents reporting enough support from families who received non-professional help and those who did not receive it, for each co-existing condition

	Non-professional help	No non-professional help	Chi-square value (p)
Feeding n= 274	38 (41.8%)	79 (43.2%)	0.041 (.98)
Sleep n= 286	43 (46.2%)	95 (49.2%)	0.658 (.720)
Anxiety n= 336	28 (24.6%)	81 (36.5%)	5.394 (.067)
Hyperactivity n = 210	15 (22.4%)	51 (35.7%)	3.823 (.148)
Other behavioural problems n= 413	37 (23.6%)	97 (37.9%)	8.828 (0.012)
Sensory n = 350	40 (31.7%)	91 (40.6%)	8.248 (.016)

5.11 Hypothesis 6

Parent perceptions of unmet need will have a mediating effect on the relationship between coexisting conditions and impact on family.

There were two pre-requisites to proceed with this hypothesis

1. Validity of hypothesis 3. To recap, the results supported hypothesis 3 that co-morbid psychopathology and problem behaviours will impact directly on the family.
2. The relationship between unmet needs and other variables, which will be demonstrated in the subsequent regression analysis.

5.11.1 Hierarchical regression analysing predictors of unmet needs

A regression analysis was performed with unmet needs as the criterion variable. Level 1 had demographic variables; level 2 ASD specific variables; level 3 frequency of moderate-severely impairing co-morbid psychopathology and level 4 problem behaviours. The model summary is provided in Table 43.

The ASD specific variables did not significantly contribute to the model while demographic variables, number of moderate-severely impairing co-morbid psychopathology and problem behaviours contributed to the model (R^2 - .201).

The significant predictors for each level are provided in Table 44. Younger age (p=.012), the number of moderate-severely impairing co-morbid psychopathology (p=.000) and externalising behaviour (p=.000) were significant predictors of total unmet needs.

Table 43: Model summary of hierarchical regression analysing predictors of unmet needs

Model	R	R^2	Adjusted R^2	SE	Change in R^2	Change in F	df 1	df2	Sig. F change
1	.132	.017	.014	1.911	.017	4.566	2	517	.011
2	.173	.030	.019	1.906	.013	1.664	4	513	.157
3	.400	.160	.149	1.775	.130	79.354	1	512	.000
4	.448	.201	.187	1.735	.041	12.946	2	510	.000

Predictors for model
1 –age, gender
2 - age, gender, ASD diagnosis, siblings with ASD, type of school, language level
3 - age, gender, ASD diagnosis, siblings with ASD, type of school, language level, number of moderate-severely impairing co-morbid psychopathology
4 - age, gender, ASD diagnosis, siblings with ASD, type of school, language level, number of moderate-severely impairing co-morbid psychopathology, externalising, internalising.

Table 44: Significant predictors for each level of hierarchical regression model of total unmet needs

Model	Variable	Coefficient	Standard error	P value
1	Age	-.130	.022	.003
2	Age	-.151	.024	.002
	Sibling with ASD	.091	.287	.041
3	Age	-.155	.023	.001
	Number of moderate-severely impairing co-morbid psychopathology	.366	.045	.000
4	Age	-.113	.023	.012
	Number of moderate-severely impairing co-morbid psychopathology	.212	.055	.000
	Externalising behaviour	.208	.027	.000

5.11.2 Mediation analysis

In order to determine the impact of parent reported unmet needs on the relationship between the presence of co-existing conditions and impact on the family a series of mediation analyses were undertaken using Hayes' SPSS templates (http://www.afhayes.com/public/templates.pdf). Mediation analyses enable the evaluation of the direct effects of variable X (co-existing conditions) on Y (impact on the family) and the indirect effect of X on Y through the mediator M (unmet needs). Co-existing conditions constituted three different variables - number of moderate-severely impairing co-morbid psychopathology, externalising behaviour and internalising behaviour and thus three different mediation analyses were performed.

In the first mediation analysis, the variables added to the analysis were age, gender, the type of diagnosis, siblings with ASD, language level, the type of school, the number of moderate-severely impairing co-morbid psychopathology and externalising and internalising behaviours. The number of moderate-severely impairing co-morbid psychopathology was the X variable with all the other variables added as control variables. The Impact on Family (IOF) score was the Y variable and the total unmet needs the M mediator. Similarly subsequent analyses were performed with externalising and internalising behaviours as X criterion variable respectively.

Figure 12: Pictorial representation of the first mediation model with the number of moderate-severely impairing co-morbid psychopathology as X variable, the IOF score as the criterion variable and the total unmet needs as the mediator

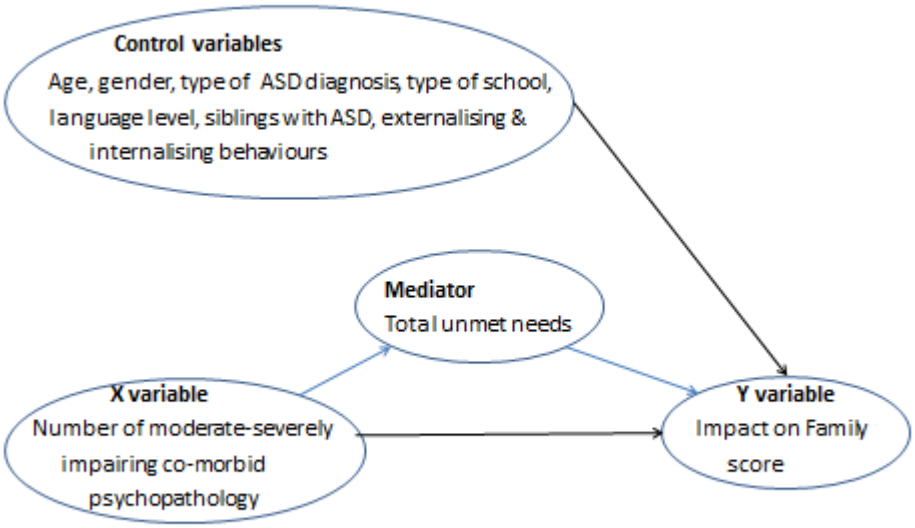


Table 45 presents data on the direct effect of the three separate X variables on the Y variable (Impact of family score) and the indirect effect or mediation effect through the mediator – total unmet needs. 95% Confidence Interval (CI) and SE are provided for both direct and indirect effects. There is a significant effect if the 95% CI does not include 0.

The direct effect of number of moderate-severely impairing co-morbid psychopathology on impact on family was .921 with SE of .245; 95% confidence interval being .439 and 1.402. The indirect effect of number of moderate-severely impairing co-morbid psychopathology was .191 (SE-.061); 95% confidence interval being .095 and .351. This indirect effect or mediation was through unmet needs. As 95% confidence interval of mediation did not include 0, unmet needs significantly mediated the effect of the frequency of moderate-severely impairing co-morbid psychopathology on impact on the family.

Similarly total unmet needs mediated the relationship between externalising behaviour and the impact on family score, whereas total unmet needs did not mediate the relationship between internalising behaviour and the impact on

family score as the 95% CI for this analysis included 0. The raw data for these three analyses are provided in Appendix 9.

Table 45: Results of mediation analyses of the total unmet needs on the Impact on family score

X variable	Direct effect			Indirect effect		
	Effect	SE	95% CI	Effect	SE	95% CI
Number of moderate-severely impairing co-morbid psychopathology	.921	.245	(.439;1.402)	.191	.061	(.095; .351)
Externalising behaviour	.581	.12	(.346;.817)	.09	.033	(.041; .176)
Internalising behaviour	.440	.121	(.203; .678)	.035	.03	(-.018; .105)

In summary, the total unmet parent needs (a surrogate marker for the role of services) mediated the impact on family of both co-morbid psychopathology and externalising problem behaviours. This may imply that appropriate services can ameliorate the impact on family of co-morbid psychopathology and externalising problem behaviours of children with ASD.

6 Chapter 6. Discussion

This chapter discusses the results of this thesis in relation to objectives of the project and the available literature. The probable mechanisms fundamental to these findings about co-existing conditions in children with ASD, and their implications for researchers, clinicians, families and policy makers are discussed. The chapter presents initially the representativeness and summary of the results before moving onto discussions around co-existing conditions, their impact on families and the role of services. The discussions around these concepts will move from the results of this study to corroborating and contradicting evidence in the literature before exploring possible explanatory reasons for the results.

The overarching objectives of this project included evaluating the presence and persistence of co-existing conditions across all age groups, types of ASD and ability levels; how they impact on families irrespective of these characteristics; and the role of services in alleviating this impact.

The study has unique strengths. The participants were recruited from two large ASD databases of Dasl^{n}e and ASD-UK. The Dasl^{n}e is shown to be a valid and representative database for the North-east of England (McConachie *et al.*, 2009). The ASD-UK database has child characteristics similar to that of Dasl^{n}e, highlighting that this database is also likely to be representative of UK children with ASD (Warnell, 2013).

There was no significant difference between the characteristics of responders to the study and non-responder children and families, with the exception of age; the children of non-responders were slightly older. Families have been enrolled in the Dasl^{n}e database since 2003 and the response rate from families enrolled in the early years was lower than that from those enrolled more recently. It is possible that the address for some families enrolled earlier might not be accurate or updated, underpinning some poorer response from families of older children.

The present study had a large sample of more than 600 participants and also used standardised questionnaires to assess co-existing conditions of children

with ASD and their impact on the family. It further explored the role of services through parent report of unmet needs for support in managing these co-existing conditions of their child.

The active participation and the support of parents have helped this study to produce some clinically notable results. Co-morbid psychopathology was equally prevalent across different groups of age, gender, type of ASD diagnosis, language level and type of school, highlighting the need to clinically screen and monitor these conditions across different groups. Problem behaviours were associated with lower age, lower levels of language ability and a diagnosis of autism. Children older than eight years of age had higher anxiety than younger children. Younger children had higher levels of under-eating and food refusal than older children, while age was not associated with limited food variety. Age was again not associated with total sleep problems and total atypical sensory behaviour; while sensory seeking behaviour was more common in the younger age group than in older children.

Both co-morbid psychopathology and problem behaviours significantly impacted the family irrespective of different child characteristics including the type of ASD diagnosis. This finding strengthens the need to identify and address co-existing conditions of children with ASD as soon as they are in contact with services. The evaluation of services revealed that services mediated to some extent the impact of co-morbid psychopathology and externalising behaviours on families; this finding highlights the role of services to support families in managing co-existing conditions in addition to the communication and learning needs of children with ASD and their families.

6.1 Co-existing conditions

Under this section, the frequency of co-existing conditions in children with ASD is explored initially along with the possible reasons for their high frequency followed by analyses of predictors of the two components of co-existing conditions, that is, co-morbid psychopathology and problem behaviours. Towards the end of the section, a critical scrutiny of the plan of this project to analyse co-morbid psychopathology and problem behaviours independently is undertaken.

6.1.1 Frequency of co-existing conditions in children with ASD

The literature review suggested that children with ASD had a high frequency of co-existing conditions compared to their peers (Huang *et al.*, 2013; Kaat *et al.*, 2013; Mannion *et al.*, 2013; Maskey *et al.*, 2013; Simonoff *et al.*, 2013). The present study substantially adds evidence to this literature in that it utilised a screening questionnaire and an ASD-specific questionnaire to assess these conditions, and it has a large sample of over 600 from two large ASD-databases in UK.

At least one co-existing condition was reported for the majority of children (90%) in the screening questionnaire and this result was similar to a previous report from the Dasl^{n}e database (Maskey *et al.*, 2013). This corroborates with the majority of available evidence in the literature with more reports emerging of late (Leyfer *et al.*, 2006; Simonoff *et al.*, 2008; Amr *et al.*, 2012; Huang *et al.*, 2013; Kaat *et al.*, 2013; Mannion *et al.*, 2013; Simonoff *et al.*, 2013). .

This study evaluated co-existing emotional and behavioural conditions as having two components of co-morbid psychopathology and problem behaviour. In existing literature, these two components are usually evaluated together, whereas this study separates these components for evaluation.

The frequency of moderate-severely impairing co-morbid psychopathology was estimated using the cut-offs published for the scale used (Thorson and Matson, 2012). In addition to the total co-morbid psychopathology score, this frequency score can help to identify the burden of co-morbidity. Eighty percent of the parents reported at least one moderate-severely impairing co-morbid psychopathology (Page 115). Evaluating individual co-morbid psychopathology, the highest frequency of moderate-severe impairment was reported for avoidant behaviour (43.2%) and tantrum behaviour (40.1%) in the present study. Both avoidant behaviour (34.8%) and tantrum behaviour (33.7%), in addition to repetitive behaviour (34.9%) were reported to have the highest level of moderate-severe impairment in another community study of 89 children with ASD (Mannion and Leader, 2013a).

6.1.2 Why are co-existing conditions highly frequent in children with ASD?

The possible reasons for the high frequency of co-existing conditions in children with ASD can be one or many of the underlying and associated factors of ASD. That is, the conditions may be linked by sharing either common aetiological or neurophysiological pathways or arise as the secondary deficits of the primary condition of ASD.

6.1.2.1 Shared aetiological basis

The first possibility is that ASD is one of the manifestations of an injury to early brain growth. The aetiology of this early brain injury could be genetic, in-utero factors, prematurity, neonatal, infantile and early childhood intrinsic (inherent to the child) and extrinsic (external to the child) factors. These causes, depending on their timing, affect neurogenesis, astrocyte proliferation, dendrite growth and synapse formation altering the brain structure and function (Kolb *et al.*, 2011). The recovery from this early brain injury might be a continuum between plasticity and vulnerability of the early brain (Anderson *et al.*, 2011) and exhibits a wide range of cognitive and behavioural outcomes, irrespective of the motor manifestations (Armstrong-Wells *et al.*, 2010). This early brain injury has been considered as one of the probable aetiologies for ASD (Trottier *et al.*, 1999; Thirtamara Rajamani *et al.*, 2013). An injury at this early stage of brain development is unlikely to produce only the core social communication deficit, but can also affect attention, sensory-motor integration, sleep co-ordination and other behaviours. This might explain the high frequency of co-existing conditions seen along with ASD.

6.1.2.2 Shared neurophysiological basis

Another possibility is a shared neurochemical or neurotransmitter deficit irrespective of the aetiology and causative pathways. In a recent review, consistent alterations of Gamma-aminobutyric acid (GABA) and serotonin levels were reported in ASD (Lai *et al.*, 2014). In ADHD, in addition to the conventional dopaminergic and noradrenergic pathways, serotonin pathways and its relative levels are garnering attention (Blum *et al.*, 2008). For anxiety, serotonin is not only implicated for aetiology but also used for treatment modalities (Schinka *et al.*, 2004). Both GABA and serotonin play a crucial role in the sleep-wake cycle

and are associated with sleep problems (Saper *et al.*, 2005). Despite a limited understanding of the detailed neurochemical basis of these conditions, we cannot overlook the possibility of neurochemical deficits linking ASD and other co-existing conditions and can be a fruitful direction of future research.

6.1.2.3 Multiformity and overlap of symptoms

Another explanation usually proposed to address the co-occurrence of ADHD and ASD is multiformity, that is, the presence of one disorder increases the risk of the other (Rhee *et al.*, 2008; Taurines *et al.*, 2012). This might be conceptually linked to both shared aetiological and neurophysiological bases discussed earlier or this relationship might be due to other unknown factors. Recent reviews have concurred that research in both ASD and ADHD suggest 'common neurodevelopmental pathways, overlapping symptoms and co-occurring disorders' (Taurines *et al.*, 2012; Matson *et al.*, 2013; Mannion and Leader, 2014a).

Commonly, symptoms of ASD can be seen with other conditions. For example, ASD can co-exist with genetic syndromes such as Down, Cri du chat, Angelman and Fragile X syndromes (Moss *et al.*, 2013a; Moss *et al.*, 2013b; Oliver *et al.*, 2013). There can be also overlap of symptoms across different conditions. Self-injurious behaviour can be seen not only in children with ASD, but also in children with intellectual disability and those with genetic syndromes (Richards *et al.*, 2012; Petty *et al.*, 2014). Repetitive behaviours, a hallmark of diagnosis of ASD, can be seen in typically developing children as well as in children with other developmental disorders including intellectual disability (Honey *et al.*, 2007). Children with anxiety disorders have more autism spectrum traits and children with ASD have more anxiety than typically developing children (van Steensel *et al.*, 2011; van Steensel *et al.*, 2012b). Such overlap in symptoms across different conditions can increase the rate of diagnosis of a co-existing condition.

6.1.2.4 Co-existing conditions as secondary deficits of ASD

Another possibility is that ASD is a separate clinical entity with its own individual and specific aetiology and pathogenesis, and co-existing conditions are associated as its secondary deficits. These secondary deficits can arise

downstream after the initial social communication deficit due to various other compounding factors including, but not limited to, immediate family environment, learning milieu and societal interactions. There is evidence that parenting stress, which in turn predicts the occurrence of behaviour problems (Lecavalier *et al.*, 2006), can be improved by appropriate services and support for parents (Harper *et al.*, 2013; Foody *et al.*, 2014). Further prospective research is needed to evaluate whether appropriate advice about co-existing conditions at the time of diagnosis itself will lead to lower frequency of problem behaviours in children with ASD.

6.1.3 Predictors of co-existing conditions

The understanding of predictors for co-existing conditions in children with ASD can not only aid autism researchers to unravel their causative pathway but also alert professionals to clinically evaluate a child with additional risk factors more carefully. This thesis evaluates co-existing conditions having two components of co-morbid psychopathology and problem behaviours. The distribution and predictors of these two components have differed in this study and are thus presented with separate discussions.

6.1.3.1 Predictors of co-morbid psychopathology

In the present study, age, gender, type of ASD diagnosis, type of schooling and language level did not influence either total co-morbid psychopathology or the frequency of moderate-severely impairing co-morbid psychopathology. This lack of influence of child characteristics on co-morbid psychopathology and psychiatric problems of children with ASD has been reported from other studies.

Another population representative study of children with ASD in South Thames, London as part of the Special Needs and Autism Project (SNAP) also did not find any strong predictor for psychiatric problems based on parent interviews (Simonoff *et al.*, 2008). Simonoff and colleagues critically analysed this lack of 'risk factor-disorder association' to propose explanations which included a possibility of co-existing conditions in ASD being conceptually and aetiologically different from similar conditions in other child groups (Simonoff *et al.*, 2013). A recent report of a community study from Ireland also reported that age, gender

and intellectual disability of children did not predict co-morbid psychopathology as measured with ASD-CC (Mannion and Leader, 2013a).

However there are contradictory reports as well. A population representative study of the Dasl^{n}e database reported lower age, speech of less than sentences and special school attendance as significant predictors of co-existing conditions in children with ASD (Maskey *et al.*, 2013). Analysing this study further, the ten-item questionnaire used for data collection aimed to capture problem behaviours as reported by parents. Another clinic-based study reported that children with co-morbid psychiatric disorder were associated with lower intelligence, but this was on a sample size of only 60 children with ASD (Amr *et al.*, 2012).

Can there be any underlying reason for the lack of significant predictors for total co-morbid psychopathology, as reported by this study? Can these reasons be related to the probable shared neurobiological basis of ASD and co-morbid psychopathology? The discussions in 6.1.2.1 and 6.1.2.2 explored the probability of shared neuroanatomical and neurochemical links respectively between ASD and its co-existing conditions. Can the common shared neurobiological basis be the driver for the co-existing conditions irrespective of child-specific characteristics? The possible mechanisms are discussed below under subheadings of neurostructural and neurochemical bases.

6.1.3.1.1 Neurostructural reasons

There is an understanding that the basic social communication deficit, which manifests during early childhood due to a variety of antenatal, perinatal and postnatal causes, persists in individuals with ASD over lifespan, albeit in varying degrees (Rutter, 2011). We do not understand fully the reasons for this persistence, but may be related to the stable brain effect caused by an early brain injury. It is possible that an early brain injury, caused by either genetic or environmental factors, causes an irreversible change in either at the macro or micro structure of the brain with manifestations differing over time. The available evidence points to atypical neural connectivity at the micro level resulting in an atypical generalised early brain growth macroscopically in the late infancy for children with ASD (Silver and Rapin, 2012; Lai *et al.*, 2014). The manifestations

of this early brain effect can vary depending on the developmental level of the individual and dynamic interacting factors; but the underlying brain effect remains stable probably reflecting in the stable total co-morbid psychopathology as well as the stable autism behaviour.

6.1.3.1.2 Neurochemical reasons

Irrespective of the aetiology, a shared neurochemical or neurotransmitter deficit between ASD and its associated co-existing conditions might explain the lack of predictors for co-existing conditions. It might be possible that this shared basis drives the association of ASD and its co-existing conditions irrespective of other known child-specific characteristics (Simonoff *et al.*, 2013). Our understanding of the roles of neurochemicals and neurotransmitters in mental health conditions including ASD is evolving, but the exact state of their inter-linkage and roles in causative pathway awaits clarification (Lai *et al.*, 2014). There are many promising signs in the right direction. There is evidence emerging of the role of neurochemicals and neurotransmitters, especially serotonin, in a wide variety of mental health conditions including ASD, ADHD, anxiety and sleep problems (Schinka *et al.*, 2004; Saper *et al.*, 2005; Blum *et al.*, 2008; Lai *et al.*, 2014). Animal studies and advanced neuroimaging studies can further help to unravel this mystery around the role of neurochemicals in ASD and its co-existing conditions in the future (Chugani, 2012; Miller *et al.*, 2013).

The first longitudinal study of co-existing conditions in adolescents with ASD from the SNAP cohort also reported the persistence and stability of psychiatric problems in this population (Simonoff *et al.*, 2013). Having evaluated the possible reasons for this persistence of co-existing conditions, further research is needed to understand and elucidate the neurobiological links between ASD and its co-existing conditions to take this field forward.

6.1.3.2 Predictors of problem behaviours

The distribution and predictors of problem behaviours have differed from those of co-morbid psychopathology in this cross-sectional study. While co-morbid psychopathology remained stable across age, gender, type of diagnosis and language level, in contrast lower age, lower levels of language ability and the diagnosis of autism predicted problem behaviours. Younger children were

reported to have more problem behaviours and internalising problems than older children in this study and there are concurring reports in the literature (Mattila *et al.*, 2010; Maskey *et al.*, 2013; Mazurek *et al.*, 2013). Another study from the Dasl^{n}e database also reported that younger children with ASD had higher co-existing emotional and behavioural conditions using the ten item questionnaire to screen problem behaviours (Maskey *et al.*, 2013). A parent interview based study of children with Asperger syndrome also reported higher current problems in primary school age than in secondary school age (Mattila *et al.*, 2010). However, another cross-sectional study using the same measure as the present study, the Autism Spectrum Disorders – Problem Behaviours in Children (ASD-PBC), reported that problem behaviours were not different between young children, children and adolescents (Matson *et al.*, 2010). That cross-sectional study had used different age cohorts of 3-6 years, 7-10 years and above 10 years and had a sample size of 167 children with ASD.

Gender did not have any effect on total problem behaviour, internalising or externalising problems. Other studies also have reported no difference in problem behaviours between males and females with ASD using Child Behaviour Check List (CBCL) (Hartley and Sikora, 2009), Strength and Difficulties Questionnaire (Simonoff *et al.*, 2008) and ASD-PBC (Kozlowski *et al.*, 2012b). This is in possible contradiction to an early meta-analytical review of children with intellectual disability where males were found to have more challenging behaviours than girls (McClintock *et al.*, 2003).

Children with lower verbal ability had higher total problem behaviours and internalising behaviours. This result concurs with the report from another Dasl^{n}e database study of children with ASD where lower levels of language ability predicted frequent behaviour problems (Maskey *et al.*, 2013). A clinic-based study of maladaptive behaviour using CBCL in children with autism also concurred that non-verbal cognition and expressive language negatively predicted attention, aggression and anxious/depressed subscales of CBCL (Hartley *et al.*, 2008).

The diagnostic grouping, as a proxy for severity of autism had an effect on total problem behaviour, internalising and externalising behaviours. These

behaviours were reported higher in children with Autism than with those with Asperger syndrome. Another study of 94 children with PDD-NOS using clinic based interviews reported that children with more social communication deficits had a higher chance of a co-morbid disorder (de Bruin *et al.*, 2007). Aggressive behaviour was found to correlate with autistic symptoms in another community based study of children with ASD (Dominick *et al.*, 2007). Contradictory reports have come from a clinic-based study where the autism severity did not predict maladaptive behaviour using CBCL, but this study included 169 children with only a diagnosis of autism (Hartley *et al.*, 2008).

In the multiple regression, age, type of diagnosis and language level were found to be significant predictors for problem behaviours in our study. Lower age and lower language level might mean lower ability and lower adaptation skills. The probable factors affecting problem behaviours are discussed below.

6.1.3.2.1 Developmental unfolding

ASD is a developmental disorder (defined as a disorder affecting developmental patterns of children). Human development is a continuous process from conception to maturity; but the rates of acquisition of skills vary from individual to individual (Illingworth, 1983). This developmental unfolding of skills is a natural phenomenon and involves acquisition of adaptation skills as the child grows older (Ikiugu, 2007). The adaptation or coping skills of an individual, defined as the ability to interact and cope with adverse environmental circumstances, can improve with age (Illingworth, 1983). These adaptation skills are moderated by interactions with the environment mainly the family and the society (Eccles, 1999). Good adaptation skills can help an individual to cope with adverse environment and events. There is a possibility that this bidirectional interaction between individual's adaptation skills and the environment can influence individual's behaviours including problem behaviours.

Along with adaptation skills, problem behaviours also unfold along different timelines. Problem behaviours are seen in other populations in addition to children with ASD; commonly in those with intellectual disability (Yu *et al.*, 2006). In children with intellectual disability, verbal learning ability is inversely

related to problem behaviours (Prior *et al.*, 1999; Yu *et al.*, 2006) and echoes findings in our study of children with ASD. There are additional concurring reports of lower age (Maskey *et al.*, 2013), and lower levels of language skill and non-verbal intelligence (Dominick *et al.*, 2007; Hartley *et al.*, 2008) predicting behaviour problems in children with ASD.

In addition to problem behaviours, individual co-morbid psychopathology also can evolve over time. Considering one example from the present study, older children had more worry compared to younger children. This is in concurrence with a recent meta-analysis that reported that higher prevalence of anxiety disorder was found with older age (van Steensel *et al.*, 2011) and also with an intervention centre based study which reported age and communication were directly related to anxiety (Davis *et al.*, 2012). There are contradictory reports where lack of relationship between age and anxiety is reported, but the sample sizes of these studies were 89 and 120 respectively (Mannion and Leader, 2013a; Lidstone *et al.*, 2014). It is possible that anxiety evolves over time as the child grows older and communicates better, due to the developmental unfolding of this co-existing condition over time. In addition to this inherent nature of a condition, environmental interactions can also play a role.

6.1.3.2.2 Environmental interactions

Environmental interactions modulate the manifestations of behaviours including positive and problem behaviours in typically developing children and those with neurodevelopmental disorders (DiLalla *et al.*, 2009; Nikolas *et al.*, 2010). Environmental interactions are interactions the individual has with his/her immediate family environment, educational/vocational environment and the society at large. It is possible that though the underlying neurobiology in children with ASD may be stable, manifestations of behaviour are influenced by these environmental interactions. For example, parent and peer behaviour are shown to interact with genotype to shape a child's aggression during interactive peer play at five years of age (DiLalla *et al.*, 2009). There is emerging consensus that both genes and environment act synergistically to affect even basic human behaviour, debunking 'gene versus environment' debates (Varki *et al.*, 2008).

What I propose further is again amalgamating the synergistic influence of different factors on human behaviour rather than analysing one reason versus the other. The present study showed a lack of influence of child-characteristics on the total co-morbid psychopathology, but factors like lower age and lower verbal ability, possibly related to lower adaptation skills, predicted problem behaviours of children with ASD. The factors and the probable reasons they influence/do not influence co-existing conditions as discussed above are not mutually exclusive of each other, rather they can be synergistic. It is possible that all these three concepts of neurobiology, developmental unfolding and dynamic environmental interactions are linked together. The underlying brain effect, of an early brain injury, be it brain microstructural or neurochemical, persists over time, but individual manifestations differ based on developmental unfolding and dynamic environmental interactions. This hypothesis might explain the differing distributions and predictors of co-morbid psychopathology and problem behaviours as seen in this study.

The synergistic effect of underlying neurobiology, developmental unfolding and dynamic environmental interactions is not just true for ASD, but also for other neurodevelopmental disorders. As discussed earlier, it is very unlikely that an early brain injury resulting in a stable brain effect will only affect social communication, but usually results in multiple neurodevelopmental conditions (Gillberg, 2000; Lichtenstein *et al.*, 2010). Multiple neurodevelopmental disorders (MNDD) are a group of conditions that include a variety of neurodevelopmental disorders including ASD, ADHD, developmental co-ordination disorder, sensory integration disorders, etc. (Gillberg, 2000; Gillberg and Billstedt, 2000; Coghill and Banaschewski, 2009; Gillberg, 2010; Gillberg *et al.*, 2013; Matson *et al.*, 2013). They can share common neurobiology including genetics, epigenetics (intracellular modification of genetic matter in cells) and neurochemical basis, but manifest in varying degrees of social communication problems, hyperactivity and inattention, motor co-ordination problems, specific learning disabilities, etc. (Gillberg, 2010; Rangasamy *et al.*, 2013). The discovery of the Fox-1 gene defect responsible for MNDD across different disorders recently, might have been one of the scientific breakthroughs needed to cement this concept (Hammock and Levitt, 2011). As a clinician, I meet

children with MNDD on a regular basis and understand the need to evaluate the overall development of the child in addition to the specific concern parents bring the child with. Child health and mental health research might also need to 'zoom out' of single conditions such as ASD and evaluate this diverse and broad spectrum of MNDD to understand the neurobiology, causative pathway and downstream developmental unfolding of behavioural manifestations of these conditions.

6.1.4 Co-morbid psychopathology and problem behaviours

Having evaluated the predictors of co-morbid psychopathology and problem behaviours in the previous sections, this section analyses the logic of separating problem behaviours from co-morbid psychopathology. Conventionally both co-morbid psychopathology and problem behaviours are combined together in both the DSM and ICD diagnostic criteria, as well as in measurement tools such as the Child Behaviour Checklist (CBCL) and the Behaviour Assessment System for Children (BASC) (Achenbach, 1991; Reynolds, 2004). There is indeed overlap between problem behaviours and co-morbid psychopathology. For example, breaking rules is often considered problem behaviour, but can be part of a conduct disorder; confrontational behaviour is another problem behaviour that is a component of oppositional defiant disorder. Conventionally such problem behaviours attain significance if they fulfil the diagnostic criteria or score above the cut-off for a specific diagnostic condition. Most of the diagnostic criteria require behaviours to be manifest in at least more than one setting (for example, school and home).

Evaluating problem behaviours and co-morbid psychopathology separately adds a new dimension. As discussed, some problem behaviours might be isolated and not fulfil the criteria for a diagnosis of conduct disorder or other co-morbid psychopathology, yet significantly impair learning, self-care skills and day to day life of the individual. For example, behaviour of breaking rules may happen in only certain circumstances or as isolated incidents making a diagnosis of conduct disorder difficult. Still even one isolated incident can impair learning and skill acquisition of the individual and affect the family significantly. It will be helpful to separate problem behaviours from co-morbid

psychopathology to collectively analyse all behaviours which are detrimental to learning, skill acquisition and care, which may not fulfil a cut-off or diagnostic criteria.

It is unlikely that co-morbid psychopathology and problem behaviours will have a different aetiological or neurochemical basis. The likely explanation is that though underlying co-morbid psychopathology remains stable across different groups, problem behaviours are possibly manifested more by individuals with poorer adaptation skills such as those with lower age and lower verbal ability. Environmental interactions also might play a role in the expression of these problem behaviours as described above. Considering all these factors, it is no surprise that the ASD Assessment Battery for Children analyses co-morbid psychopathology and problem behaviours as distinct diagnostic issues (Matson, 2009).

The present study has shown a clear distinction in the distribution and predictors of co-morbid psychopathology and problem behaviours as two components of co-existing conditions. However, the concept of co-morbid psychopathology being distinct from problem behaviours needs to be justified across different study settings and populations in the future. Further evidence is needed to support the hypothesis that environment plays a role in the manifestation of problem behaviours; thus more remediable by modifying the environment. For clinicians as well, this finding is notable as it stresses the importance to evaluate and provide treatment and intervention for both co-morbid psychopathology and problem behaviours.

6.2 The impact of co-existing conditions on family

In this section, the impact of co-existing conditions on families of children with ASD is explored. This section will initially discuss the results described in this thesis regarding the impact on the family in relation to evidence in the literature, before moving on to explore the possible reasons for this impact.

This thesis has shown that the frequency of moderate-severely impairing co-morbid psychopathology and both internalising and externalising problem behaviours predicted the impact on the family, irrespective of the type of

diagnosis of ASD (page 133). Having another child with ASD and the child attending special school were other significant predictors. This finding of co-existing conditions impacting on families of children with ASD is notable that the diagnostic category of ASD did not impact on the family in the final model.

There is more recent evidence added to the already existing literature regarding the impact of co-existing conditions on the family. Not only can co-existing conditions cause social and academic impairments in the child (Kaat *et al.*, 2013) but also parenting stress (Lecavalier *et al.*, 2006; Meadan *et al.*, 2010; Karst and van Hecke, 2012; Van Steijn *et al.*, 2012; Estes *et al.*, 2013; Huang *et al.*, 2013; McStay *et al.*, 2013). There has been evidence for more than a decade that parents of children with autism have more stress, poorer mental and emotional health, more worries about the future and less quality of life compared to parents of typically developing children (Konstantareas and Homatidis, 1989; Montes and Halterman, 2007; Cassidy *et al.*, 2008; Shu, 2009; Dabrowska and Pisula, 2010).

There have been more attempts to tease out the contribution of co-existing conditions in addition to the core behavioural issues of children with ASD, on impact on the family in recent years. For example, Konstantareas and Homatidis, as early as 1989 reported that the best predictor for parent stress was child's self-injurious behaviour in a population-representative group of 44 children with autism (Konstantareas and Homatidis, 1989). However, there were contradictory reports as well. A community study of 67 children with ASD reported that neither externalising nor internalising behaviour problems were associated with parent stress (Skokauskas and Gallagher, 2012). Subsequent literature strengthened the role of co-existing conditions including behaviour problems impacting families (Lecavalier *et al.*, 2006; Cassidy *et al.*, 2008; Davis and Carter, 2008; Manning *et al.*, 2010; Ingersoll and Hambrick, 2011; Silva and Schalock, 2012; van Steensel *et al.*, 2012a). Recent studies have re-stressed the significance of co-existing conditions including behaviour problems and hyperactivity and not autism severity impacting families of children with ASD (Estes *et al.*, 2013; McStay *et al.*, 2013). This is in concurrence with the findings of this thesis that co-existing conditions including co-morbid psychopathology

and problem behaviours, and not the diagnostic grouping impacted on families of children with ASD.

All these studies have used either clinical interviews (Konstantareas and Homatidis, 1989; Cassidy *et al.*, 2008; van Steensel *et al.*, 2012a) or parent-report questionnaires (Van Steijn *et al.*, 2012; Estes *et al.*, 2013; Huang *et al.*, 2013; Foody *et al.*, 2014) or a combination of both (van Steensel *et al.*, 2012a) to understand co-existing conditions. The parent report questionnaires used included Strength and Difficulties Questionnaire (Huang *et al.*, 2013), Conner's Parent Rating Scales (Van Steijn *et al.*, 2012; Foody *et al.*, 2014), Brief Symptom Inventory (Estes *et al.*, 2013) and Vineland Adaptive Behaviour Scales (Foody *et al.*, 2014); none of them was developed nor adapted for the ASD population. This study adds value to the existing literature in that it used ASD specific questionnaires to assess co-morbid psychopathology and problem behaviours.

When analysing individual types of co-morbid psychopathology, this thesis has shown that avoidant and tantrum behaviours were found the strongest predictors impacting on the family (page 136). Though there are studies recognising higher rates of avoidant and tantrum behaviours in children with ASD (Konst *et al.*, 2013; Tureck *et al.*, 2014), this is the first study to report their impact on families. There are reports of tantrum behaviour affecting parents in other conditions such as Prader-Willi syndrome (Dykens *et al.*, 2011; Tunnicliffe *et al.*, 2014). There are other reports of individual conditions impacting the family of children with ASD above and beyond autism severity, including hyperactivity (McStay *et al.*, 2013) and conduct problems (Lecavalier *et al.*, 2006). Having evaluated how results of the present study confirm and add new evidence to the impact of co-existing conditions on families of children with ASD, the next section evaluates the reasons for this impact.

6.2.1 Why do co-existing conditions impact on the family?

This thesis has shown that co-existing conditions of children with ASD impacted on the family. The family unit consists of individuals, who function independently as well as together as a dynamic unit. Under this section, the impact of co-existing conditions on individuals (individuals with ASD, siblings and parents)

and the family unit as a group is explored. The dynamic nature of interactions within a family means that what impacts upon one individual may have repercussions on other individuals as well as the whole family, thus necessitating an evaluation of the effects on individuals initially.

6.2.1.1 Impact on the child with ASD

Co-existing conditions such as sleep and feeding problems affect the day to day functioning of the child; feeding problems can affect the child's nutrition and their long term health (Cermak *et al.*, 2010). Other behaviours such as hyperactivity and problem behaviours can affect children academically, and their attainments of future life skills (Pearson *et al.*, 2006b; Kaat *et al.*, 2013). Co-existing conditions can also affect the development of language and social skills (Matson *et al.*, 2010; Goldman *et al.*, 2011). The impairments related to these co-existing conditions limit a variety of abilities for the individual starting from attainment of self-care skills to vocation to independent living (Patzold *et al.*, 1998; Sikora *et al.*, 2012a). These additional problems can be challenging for a child already coping with social communication difficulties; the whole impairment caused by multiple conditions often greater than the sum of effects of individual conditions (Gillberg, 2010).

6.2.1.2 Impact on siblings of children with ASD

Siblings of children with ASD are affected indirectly by reduced attention and care of parents towards them as well as reduced quality family time (Meyer *et al.*, 2011; Moyson and Roeyers, 2011). In addition, some siblings of children with ASD can also have sub-threshold symptoms of ASD (the Broader Autism Phenotype) as well as other mental health conditions, possibly reducing their resilience (Benson and Karlof, 2008; Meadan *et al.*, 2010; Pickles *et al.*, 2013). Parent involvement in providing information for siblings regarding ASD can be supportive in their own journey as well as to help them develop a more nurturing relationship with their own siblings with ASD (Sage and Jegatheesan, 2010).

6.2.1.3 Impact on parents of children with ASD

Co-existing conditions, be they feeding problems or sleep problems or anxiety, affect care of the child impacting parents and care-givers (Allik *et al.*, 2006a; Lecavalier *et al.*, 2006; Manning *et al.*, 2010). Taking an example of sleep

problems, a child who does not sleep well affects parents' in turn affecting parents' quality of life including health related quality of life (Delahaye *et al.*, 2014).

Parents of children with ASD are shown to have lower parenting efficacy and higher parent stress when compared to parents of children with other neurodevelopmental disorders (Orsmond and Seltzer, 2007a; Meadan *et al.*, 2010; Silva and Schalock, 2012). Co-existing conditions can have a strong influence on parent stress. They are shown to be the predominant factor (Peters-Scheffer *et al.*, 2012; Silva and Schalock, 2012; Estes *et al.*, 2013) as well as a compounding factor along with core symptoms of ASD (Davis and Carter, 2008; van Steensel *et al.*, 2012a) in affecting parents of children with ASD. Co-existing conditions are associated with more parenting stress (Estes *et al.*, 2013), mental health issues (Werner and Shulman, 2013), endocrine and immunity problems (De Andrés-García *et al.*, 2012), and fewer employment and recreation opportunities for parents (Montes and Halterman, 2007). Overall, there is a reduction in well-being and quality of life for parents of children with ASD compared with parents of typically developing children and of children with other developmental disorders (Cappe *et al.*, 2011; Silva and Schalock, 2012; Werner and Shulman, 2013).

6.2.1.4 Impact on the whole family unit

Compared to other developmental disorders, ASD has a higher impact on families as well (Meadan *et al.*, 2010). Multiple co-existing conditions can have additive impact along with core symptoms of ASD on families (Davis and Carter, 2008; Phetrasuwan and Shandor Miles, 2009; van Steensel *et al.*, 2012a; Foody *et al.*, 2014). Co-existing conditions such as challenging behaviour, sleep problems and hyperactivity are shown to affect the final family adaptation more than autism severity (Hall, 2012; Paynter *et al.*, 2013; Delahaye *et al.*, 2014; Foody *et al.*, 2014). ASD and its associated co-existing conditions can affect family dynamics ranging from parents' marriage to outings and holidays for the whole family (Montes and Halterman, 2007). These associated conditions can also increase the financial burden on families as they are associated with increased rates of hospitalisation, medication and therapy of children with ASD (Frazier *et al.*, 2001; Peacock *et al.*, 2012). In addition, the persistence of

autism symptoms as well as co-existing conditions in children with ASD can result in chronic stress on families resulting in multiple family crises over life course (White *et al.*, 2012).

To summarise, co-existing conditions are shown to impact families directly irrespective of the type of diagnosis of ASD in this thesis. This is in corroboration with the majority of evidence in the literature, where this study can add stronger evidence as it used ASD-specific measures to assess co-existing conditions. The measure used in this study, the IOF scale, had only factor and did not have subscales to analyse the impact on individuals including siblings and different aspects of impact. Future studies can explore these components using ASD-specific measures.

6.3 The role of services

As described in section 2.3 (page 18) formal services play a role as part of the resources to enable family and parent adaptation and can be analysed by the assessment of perceived unmet needs of parents and families. This section will analyse the quantity of unmet needs, the role of professionals and predictors and mediation effect of unmet needs of families of children with ASD before moving on to evaluate the current national recommendations, clinical service provision and recommendations from this study for autism related services.

6.3.1 Quantity of unmet parent needs

In this study, regarding all co-existing conditions, more than two-thirds of families reported at least one unmet need (page137). Similar high unmet need (74.3%) was reported for continuous service provision from other studies in ASD as well (Dillenburger *et al.*, 2010; Brown *et al.*, 2012). There are reports of high parent unmet needs in children with cerebral palsy and children with special health care needs (Graungaard *et al.*, 2011; Jackson *et al.*, 2011). However this is the first study to analyse unmet need for co-existing conditions of children with ASD.

This thesis has also shown that highest level of unmet need was reported for ‘other behavioural problems’ (behaviours other than hyperactivity, anxiety, sleep, feeding and sensory problems) (refer to page 137). Problem behaviours

are shown to contribute to increase in unmet needs of children with cerebral palsy, another neurodevelopmental disorder (Jackson *et al.*, 2011). In ASD, problem behaviours are shown to contribute to unmet parent needs in addition to communication needs of children in a community study of children with ASD (Papageorgiou and Kalyva, 2010). Problem behaviours in children with ASD, though very common, can be difficult to disentangle from the complexity of ASD itself (Matson *et al.*, 2010) and this might be contributing to high unmet needs reported for behaviour problems.

6.3.2 Role of professionals

Generally across all co-existing conditions and significantly for sleep problems, anxiety, hyperactivity, 'other behaviour problems' and sensory problems, parents who reported partial support met more professionals than those who reported enough support (page 138). Similarly more parents who reported partial support sought and received non-professional help than parents who reported enough support, (significantly for 'other behaviour problems' and sensory problems with non-significant differences seen for feeding and sleep problems, anxiety and hyperactivity, page 140). These findings point to a need-support mismatch, where support provided by services does not meet needs of the child and the family. Parents of children with ASD have reported that services are not efficient in meeting their needs and there is a mismatch between needs as perceived by parents and professionals (Dillenburger *et al.*, 2010; Keenan *et al.*, 2010).

There may be a subset of children with ASD who have complex needs and need to meet a number of professionals for help and support. For most of the children, there is also a possibility of a need-service mismatch where the professional might not be trained and experienced to meet the complex needs of the child. This might result in the family meeting a number of different professionals, each of whom suggests someone else might have the skills to intervene effectively, but finally parents end up with only partial support which is less than required for a particular co-existing condition; this could happen for many co-existing conditions simultaneously. By contrast, a very well trained and experienced professional might be able to provide intervention for a number of

co-existing conditions, leading to fewer consultations with other professionals. Ideally, that professional should also help the family to harness informal sources of support in addition.

6.3.3 Predictors for unmet parent needs

This study has revealed younger age, the number of moderate-severely impairing co-morbid types of psychopathology and externalising behaviour as predictors for total unmet parent needs regarding co-existing conditions (page 141). Though this thesis is the first study analysing unmet parents needs for co-existing conditions of children with ASD, findings of this study concur with those analysing unmet parent needs in general among parents of children with ASD (Ellis *et al.*, 2002; Papageorgiou and Kalyva, 2010).

As this study analysed unmet needs regarding co-existing conditions, it is not surprising that both co-morbid psychopathology and externalising behaviours are significant predictors. There is corroborative evidence from a large national survey in US that reported additional special health care needs including problem behaviours increased unmet needs of parents of children with ASD (Kogan *et al.*, 2008). Another study using the National Autism Society Database also reported similar findings for parents of children with ASD (Bromley *et al.*, 2004). The level of language ability was not a significant predictor in the current study unlike other studies (Bromley *et al.*, 2004; Kogan *et al.*, 2008) and might be related to the fact that this thesis analysed unmet needs regarding co-existing conditions only.

6.3.4 Mediation effect of total unmet parent needs on impact on the family

In the present study, total unmet parent needs mediated the impact on the family of co-morbid psychopathology and internalising behaviours (refer to page 143). This is in corroboration with the composite research model of stress and coping where services as part of resources mediate the relationship between stressors and the final adaptation. For families of children with ASD, the quantity of unmet needs is shown to be associated with parent distress (Foody *et al.*, 2014) as well as impact on family (Brown *et al.*, 2011). This thesis adds evidence to the already existing literature that the quantity of unmet parent needs regarding co-existing conditions not only is associated with impact on the

family, but also mediates the relationship between these conditions and the impact.

Summarising, this thesis highlights the high quantity of unmet parent needs regarding support for co-existing conditions and its association with the impact on the family. The results of the mediation analysis corroborate the composite research model of stress and coping, and strengthen the role of services as a mediator between stressors and the final family adaptation. Before moving on to recommendations, further sections analyse the current national recommendations and realities of clinical autism related services in the UK.

6.3.5 Current national recommendations in UK

The National Autism Plan for Children (NAP-C) published by National Autistic Society in collaboration with the Royal College of Psychiatrists and the Royal College of Paediatrics and Child Health made detailed recommendations about multi-agency, multi-disciplinary assessments (NAS, 2003). It stressed the need for local co-ordination between different agencies and the importance of individualised care plans to be relevant not just for each child but also for each family.

The National Institute for Health and Clinical Excellence (NICE) guideline, that sets the standards for clinical care of children and young people up to 19 years of age in UK, recommends early and appropriate assessments of all co-existing conditions (NICE, 2011; NICE, 2013; NICE, 2014). The NICE guidance for diagnosis released in 2011, a year before this project started, suggested establishment of a local autism multi-agency strategy group with representations from child health and mental health clinical services, education and social services, parents and non-governmental organisations (NICE, 2011). It suggested a multi-professional autism assessment and diagnosis team should ideally comprise a paediatrician, a child and adolescent psychiatrist, clinical and educational psychologists, occupational therapists and specialist teachers to evaluate children with ASD. It laid stress on proper co-ordination between different services catering to the needs of the child, and multi-agency staff to work with children with ASD.

The NICE guidelines for management and support of children and young person with ASD published in 2013, during the course of this thesis, suggested in addition to the existing recommendations, interventions specifically for co-existing mental health and medical problems, challenging behaviours and sleep problems (NICE, 2013). This new clinical guideline laid stress on appropriate training for health and social care workers to meet the individual needs of children and young people. It also added provisions for medical treatment of co-existing conditions including anti-depressants. The NICE quality standards on autism published this year recognise the impact of co-existing conditions on the quality of life of individuals and their families (NICE, 2014). The NICE standards stress a 'person-centred, integrated and co-ordinated approach to providing services' and recommend that family members of children and young people with ASD be involved in 'decision making process about investigations, treatment and care'.

The NICE guidelines recognise the importance of empowering parents and carers in addition to the capacity building of services. The parents need to be empowered to recognise and support their child's co-existing conditions. The role of social support including that of extended family to empower parents in this process also needs to be strengthened. However, the pressing need is to reinforce services to provide support for co-existing conditions of children with ASD and their families. NICE suggestions and recommendations are in the right direction and can make a huge difference to families of children with ASD.

6.3.6 Clinical services for children with autism in the UK

In the background of these recommendations since 2003, how are the services in the UK performing regarding these provisions? Though improvements were noted in diagnostic services for children with ASD since the publication of NAP-C guidelines, there were inequalities in standards across different services in completing diagnostic assessments within the targeted timescale (Palmer *et al.*, 2011). Families had a greater access to members of the multidisciplinary child development team (CDT) in 2007 than in 2001, after the publications of these guidelines.

However, a recent study covering CDTs in the UK reported a decline in the number of professionals especially psychologists working within the multi-disciplinary team (Parr *et al.*, 2013). There was reduced funding reported over the years. The recommended national initiatives could be only partially adopted. The inadequate resources at the CDT could be one of the reasons for the high frequency of unmet parent needs reported in this present study.

This current thesis also shows discrepancies in service provision where parents who reported partial support actually met a greater number of professionals than the parents who reported enough support. It can be hoped that adequate training of professionals regarding co-existing conditions can improve service provisions for children with ASD. Professionals need to be trained to recognise, monitor and intervene and to work closely with families to support co-existing conditions of children with ASD. New evidence based intervention and support options need to be made available to families and professionals to support families and children in coping with these conditions.

Co-existing conditions including problem behaviours are one of the main reasons families of children with ASD seek support from services (Matson *et al.*, 2010). Support for co-existing conditions remain an important unmet need for these families (Papageorgiou and Kalyva, 2010; Brown *et al.*, 2012) and contribute substantially to health care cost of children with ASD (Peacock *et al.*, 2012). A recent article evaluating costs of ASD in the UK and the US reported ASD as the most costly condition in the UK, more than any other chronic condition (Buescher *et al.*, 2014). Medical costs were found to be higher for adults with ASD than for children in this review. Reviews have recognised the relevance of co-existing conditions in contributing to the costs and resources needed to support individuals with ASD (Matson and Nebel-Schwalm, 2007b; Peacock *et al.*, 2012). Further prospective research need to be done to evaluate if advice and support for co-existing conditions of children with ASD can reduce the financial costs needed to support these individuals as they reach adulthood.

All different service providers including education services, social support services and child health and mental health services need to integrate efficiently and effectively to support families of children with ASD. Families should be

empowered to support co-existing conditions keeping in mind the parental factors of personality, resilience, self-efficacy and well-being. Improvements in diagnostic services have been reported for children with ASD following the publication of the NAP-C guidelines, but there were inequalities persisting in the service provision (Palmer *et al.*, 2011). It can be hoped that similarly, NICE guidelines also will improve services for support of co-existing conditions of children with ASD and their families.

6.4 Strengths of this thesis

This thesis proposed to evaluate the impact of co-existing conditions of children with ASD on their families in a broad scale using two large ASD-specific databases and has been fairly successful to an extent. This is the first study to analyse the impact of all co-existing emotional and behavioural conditions as holistically as possible. It used autism specific measures to analyse co-existing conditions and co-morbid psychopathology of children with ASD. To analyse individual conditions such as feeding problems and anxiety, autism specific measures or the next best measures available were used. This is the first study to show that there are differing influences for co-morbid psychopathology and problem behaviours of children with ASD.

The present thesis is the largest study so far to analyse the impact of these co-existing conditions on families of children with ASD and has brought out to fore the high impact of these conditions. This study is the first one so far to analyse unmet parents needs for support of these co-existing conditions, develop a unique tool to measure them and also demonstrate high rates of unmet parent needs for these conditions. The mediation analysis of unmet parent needs on the relationship between co-existing conditions and the impact on the family also demonstrated the role of services to alleviate some of these conditions, for the first time in literature.

This thesis has made an attempt to compile the evidence from this study, the available literature in paediatric neuro-disability and the clinical experiences of the primary researcher to suggest a more holistic view of a child with disability and his/her family for both clinical and research purposes.

6.5 Limitations

The present study used two large ASD databases in UK for recruiting participants. Both these databases enlist parents by voluntary enrolment and use data provided by them. There is a possibility of leaving out a segment of parents who are less enthusiastic about enrolment. Though parent reports are shown to be fairly reliable and valid, there can be limitations on the quality of the information they provide through databases (Lampi *et al.*, 2010). The response rate to the present study has been around 40%, indicating that the majority of eligible parents did not participate.

This thesis used standardised parent report questionnaires to analyse co-existing conditions. Though parent report questionnaires are used to screen emotional and behavioural conditions of children, they can be of limited value compared to a detailed clinical interview, especially in cases of overlap of symptoms and multiple co-existing conditions (Nauta *et al.*, 2004; Bauer and Blunden, 2008; Meltzer, 2008). For sleep problems, subjective assessment including sleep questionnaires has been shown not to match the objective assessment of sleep, limiting the utility of parent report questionnaires (Bauer and Blunden, 2008).

The ASD-specific measures used in this study, the Autism Spectrum Disorders – Comorbidity for Children (ASD-CC) and the Autism Spectrum Disorders – Problem Behaviours in Children (ASD-PBC) are recently developed tools. Other measures used in this study such as the Children's Sleep Habits Questionnaire (CSHQ), the Impact on Family scale, the Conner's Rating Scales and the Spence Children's Anxiety Scale – Parent version (SCAS-P) were developed for the general paediatric population though used extensively in the ASD population; they await specific validation and adaptation for the ASD population (Grondhuis and Aman, 2012; Wolraich *et al.*, 2013; Lecavalier *et al.*, 2014). The sensory profile sets of questionnaires developed for the general population have acceptable discriminant validity for children with ASD (Ben-Sasson *et al.*, 2009). The Brief Autism Mealtime Behaviour Inventory (BAMBI) is a measure developed specifically for children with ASD (Lukens and Linscheid, 2008). The measure used in this study to analyse the impact on the family, the IOF scale had only one factor. It did not have sub-domains to analyse the impact on

individuals like siblings and parents and the type of impact. Despite limitations regarding appropriate measurement tools, this is the first study to use ASD-specific measures to analyse co-existing conditions to understand their impact on the family.

As there was no standardised measure to assess unmet needs for support of co-existing conditions in children with ASD, this project developed a schedule to analyse unmet parent needs, the Needs and services questionnaire. There were invalid returns for this questionnaire, which might have affected the results, though there was no significant difference between those included in the analysis and those excluded in rates of frequent co-existing conditions in the screening questionnaire. Though this measure was developed with the help of parents and its results across different co-existing conditions were consistent, it requires further validation in research and clinical contexts. Despite these limitations, the present study has notable findings that co-existing conditions impact families and appropriate services can ameliorate some of that impact.

6.6 Implications and recommendations for research

The findings of this study must encourage researchers to 'look beyond the diagnosis of ASD' at various factors impacting families of children with ASD. In future, a further exploration of different factors including parent personalities, personal resilience and family hardiness that impact or buffer families, can be undertaken by a qualitative analysis. Such a detailed analysis can shed light on the complex subjective nature of the impact of co-existing conditions as well as facilitators and barriers for families to access services. A direct objective analysis of services at the service provider level will add additional information about what needs to be done for the capacity building of services.

There is a need to develop ASD-specific measures to assess co-existing conditions of children with ASD. There is only limited information about the psychometric properties of many measures currently used in the ASD population including those for anxiety (Grondhuis and Aman, 2012; Lecavalier *et al.*, 2014), hyperactivity (Bard *et al.*, 2013), repetitive behaviours (Scahill *et al.*, 2014) and problem behaviours (Matson and Nebel-Schwalm, 2007a). Some of the measures developed for the general population, but now extensively used

for children with ASD, need to be adapted for the ASD population. It is hoped that the findings of this study will accelerate efforts of researchers in developing and adapting appropriate tools for the ASD population.

It needs to be explored whether an understanding of co-existing conditions of children with ASD will empower parents. A prospective study can evaluate whether regular screening of co-morbid psychopathology at the time of diagnosis itself, and further appropriate psycho-educational courses for care givers, will empower parents to reduce the future family impact. It will be interesting to evaluate the relationship between co-morbid psychopathology and problem behaviours in such circumstances.

The appropriate interventions for these co-existing conditions of children with ASD also must be evolved. Though there are interventions for most of the co-existing conditions in the general population, the efficacy of these interventions in the ASD population must be explored. Wherever relevant, appropriate adaptations need to be undertaken for the ASD population. At the same time, new specific methods of interventions should be developed for children with ASD. There are encouraging signs in the right direction as specific interventions are being developed to target anxiety of children with ASD (White *et al.*, 2009; Maskey *et al.*, 2014).

The relevance of findings of this study must be explored in different settings of population. The results of this study are relevant to all countries where services support children with ASD. It will be interesting to evaluate the frequency of co-existing emotional and behavioural conditions and their impact in other developed as well as low and middle income countries (LMIC) settings and explore whether present findings hold true in a completely different setting.

6.7 Implications and recommendations for clinical practice

It is recommended to have regular monitoring and follow-up of co-existing conditions of children with ASD in community clinics. A key worker must work closely with the family to support families in managing these co-existing conditions in community settings in the UK. Irrespective of the type of clinical and education services available in different parts of the world, it will be useful

for families to have these support services along with diagnostic services for ASD. The stable nature of co-morbid psychopathology necessitates an evaluation of a child or young person with ASD irrespective of age, gender, ability levels or type of diagnosis. It may be also prudent to evaluate and manage both co-morbid psychopathology and problem behaviours independently.

Community clinicians and other community workers must be encouraged to open discussion about these conditions with parents of children with ASD. Non-governmental support organisations also must be enlisted to empower parents with relevant information and behavioural management techniques about co-existing conditions. Parents can also be actively encouraged to partake in the planning and implementation of measures to manage co-existing conditions of their children.

Regular capacity building exercises must be undertaken to empower professionals to identify, support and manage co-existing conditions of children with ASD. Child mental health service providers also have to be trained to understand how to work with children with ASD. These measures and training should not be limited to clinical professionals, but also to teachers, community workers and other professionals involved in supporting the child or young person with ASD.

The service planners must need to plan how best the available resources can be used to support families of children with ASD. Depending upon the level and variety of services offered, this can include the effective co-ordination of the available services, ensuring adequate number of professionals in existing child development teams and the development of a few multi-disciplinary specialist teams with added expertise.

6.8 Summary and conclusions

There are discussions and disagreements about what constitutes ASD, and these have been amplified recently due to the new DSM-5 criteria (APA, 2013). Moreover, there is no consensus about co-existing conditions in children with ASD regarding what to be included, how to categorise them, etc. (refer to 1.7;

page 5). The focus should not be lost from what matters to families amidst the academic/clinical conceptual discussions of what constitutes or does not constitute ASD or co-existing conditions. Robust discussions and disagreements about these concepts add richness to our understanding of ASD; however, waiting for these concepts to be tangible should not limit the extent of our support to children with ASD and their families. It is time to 'look beyond the diagnosis of ASD' to increase awareness about co-existing conditions of children with ASD among parents, caregivers, professionals, support organisations and service planners.

This thesis has shown a high frequency of co-existing conditions in children with ASD. Around 90% of children were reported to have at least one co-existing condition in the screening questionnaire and 80% reported to have at least one moderate-severely impairing co-morbid psychopathology in the autism specific questionnaire. This cross-sectional study also highlighted the stable nature of co-morbid psychopathology across different groups recommending its evaluation irrespective of age, gender, ability levels or type of diagnosis. This study showed clear distinction between co-morbid psychopathology and problem behaviours. The problem behaviours were shown to be more common in younger children, children diagnosed to have autism and children with lower language ability. The thesis also demonstrated that both co-morbid psychopathology and problem behaviours impacted on the family irrespective of the type of diagnosis of ASD.

This study has highlighted the high prevalence of unmet parent needs for support in managing these co-existing conditions. It has also shown the mediating role of services, as measured by unmet parent needs, in alleviating the impact of co-morbid psychopathology and externalising behaviours on families. It is sincerely hoped that findings of this thesis will empower and encourage both researchers and clinicians to look 'beyond the diagnosis of ASD' at co-existing conditions of children and to explore whether additional support and services might ameliorate the impact of these conditions on families of children with ASD.

7 Appendices

7.1 Appendix 1 – Table of measures analysed in the study

Behaviour Questionnaire

Name of Tool	Domains measured	Age	Respondents and format	Psychometric reliability Internal consistency (α); stability (trt – κ1); equality (interrater) κ2	Psychometric validity	strengths	weakness	Cost
CBCL (Child Behaviour CheckList) (Achenbach, 1991)	internalizing (i.e., anxious, depressive, and overcontrolled) and externalizing (i.e., aggressive, hyperactive, noncompliant, and undercontrolled) behaviours, score of 0-2, 0 – not true, 1 – somewhat or sometimes true, 2- always true.	1.5 – 18 yrs (6-18 yrs)	parent questionnaire;120 q	κ2overall icc 0.93 for 20 competence items & 0.96 for 118 specific probs κ1 overall icc 1 for 20 comp itms & 0.95 for 118 specific probs α ind – ranges 0.63 – 0.79; overall 0.9	Checked with teacher's report form and youth self report forms in general population	dsm orient; extensively used in ASD; almost complete (Karabekiroglu and Aman, 2009; Pandolfi *et al.*, 2012)	Long; psychometric properties in ASD not clear	Hand scoring forms = $7 each or x50=$25 Parent forms x50 = $25 Computer upgrade = $195 Compute

								r scoring kit = $395
SDQ (Strength & Difficulties Questionnaire) (Goodman, 1997; Goodman, 2001)	five-factor structure (emotional, conduct, hyperactivity-inattention, peer, prosocial) missing, not true, somewhat true, certainly true	4-17 year old	parent questionnaire25 items	α-0.73 κ1-0.62	Correlated with dsm diagnoses	Short, Has an impact supplement; used in ASD & HFA (Simonoff *et al.*, 2008; Iizuka *et al.*, 2010)	psychometric properties in ASD not clear; needs teacher report also	free
Conner's parent long form	Oppositional, Cognitive Problems/Inattention, hyperactivity, Anxious-Shy, Perfectionism, Social Problems,Psychosomatic; uses 0-2 likert scale	3-17 yrs	Parent q, 80 items	α-0.75-0.92 κ1 -0.47	Correlated with teacher and self-forms	Definite numbers for future follow-up	needs teacher forms; ?cost ?ASD - psychometric properties in ASD not clear	Long versions: $43.25 Short versions: $40.50 (packs of 25) $425 per complete kit including manual

Child symptom inventory-4 (Gadow *et al.*, 2005; Gadow *et al.*, 2009)	15 emotional and behavioural disorders ODD, OCD, CD, Dep, GAD, SAD, Social phobia, motor and vocal tics, asperger, pdd, dysthymia, schizophrenia 4 point scale – never, sometimes, often, very often	5-12 yrs; above 12 yr – ASI, YI	Parent q, 97 item; has teacher checklist	κ1-0.46-0.87 κ2-0.74-0.94	Good corr with cbcl	Used in asd, dsm based diagnosis; gives both symptom count scores (diagnostic model) or Symptom Severity scores (normative data model) (Gadow *et al.*, 2005; Gadow *et al.*, 2009)	Advantage over cbcl; will they score high on pdd, Asperger - psychometric properties in ASD not clear	$42.00 per version (pack 50)
BASC 2 (Behavioural Assessme	ADL, Adaptability, Aggression, Anxiety, Attention Problems, Atypicality, Conduct	2-5;6-11; 12-	Parent q, 134-160 q Has teacher and self	α -0.77-0.94 – preschool 0.73 to 0.95 - school	Validated in asd	Both clinical (ext+int) &	Long psychometric propertie	Manual = $89. Hand-scored

nt System for Children) (Reynolds, 2004)	Problems, Depression, Functional communication, Hyperactivity, Leadership, Learning Problems, Social Skills, Somatization, Study Skills, Withdrawal 4 point response- never, sometimes, often, always	21	report forms	0.76 to 0.95 adolescent κ1 -0.77, 0.84 and 0.81 κ2- 0.74, 0.69 and 0.77		adaptive scales, dsm based Gets a chart based data; Used in asd	s in ASD not clear	forms x25 = $33.50. Computer entry forms = $28
Autism Spectrum Disorders – Comorbid for Children (ASD- CC) (Matson and Wilkins, 2008; Matson *et al.*, 2009b; Thorson and Matson, 2012)	7 subscales ; Tantrum Behaviour, Repetitive Behaviour, Worry/Depressed, Avoidant Behaviour, Under-Eating, Over-Eating, and Conduct Response; 0- not a prob, 1- mild prob, 2-severe prob, x-don't know	2-16 yrs	30 items; checklist	High α (.91), moderate k1 (.46),k2 (.51)	With BASC-2; all factors other than avoidant behaviour had acceptable validity	Validated in asd dsm-iv and icd-10 based; developed for asd	No hyperactivity, anxiety, aggression	Whole battery 325$ - manual +50 forms
ASD-	Problem behaviour	2-16			With			Whole

Problem Behaviours for Childern (ASD-PBC) (Matson *et al.*, 2008a; Mahan and Matson, 2011)	checklist has internalizing and externalizing factors Response 0,1,2 as above for ASD-CC	yrs			BASC-2			battery 325$ - manual +50 forms
Aberant Behaviour Checklist (Aman *et al.*, 1987)	1. Irritability, Agitation, Crying (15 items); 2. Lethargy, Social Withdrawal (16 items); 3. Stereotypic Behaviour (7 items); 4 Hyperactivity, Noncompliance (16 items); and 5. Inappropriate Speech (4 items).	6-54 yrs	58 items four-point scale ranging from 0 (‘‘not at all a problem’’) to 3 (‘‘the problem is severe in degree’’).	α-0.86-0.94 κ1 -0.63	With cbcl	Used in asd	Not developed for asd specifically; psychometric properties in ASD not clear	
Development Behaviour Checklist short (Taffe *et*	Long one structure similar to CBCL	4-18 yrs	24 items (long-96 items) Three point scale	α-0.85		Used in asd children	Originally developed for ID, no subscale scores;	

al., 2007)							psychometric properties in ASD not clear	
Child's Challenging Behaviour Scale (Bourke-Taylor *et al.*, 2010)	Problem behaviours in children	5-18 yrs	11 item	α-0.89	With PedsQL Psychosocial Health Summary Score	Has mother's coping abilities	Developed for disability, had asd children; psychometric properties in ASD not clear	
The Eyberg Child Behaviour Inventory (Eyberg & Pincus 1999)	Problem behaviours at home and school; externalising symptoms, general symptomatology	2-16 yrs	36-item scale; both problem (Y/N) scale and intensity(1-Never to 7-always) scale	α-0.93-0.94 κ1 -0.75 k2-0.61-0.79	Good construct validity		?in ASD - psychometric properties in ASD not clear	
Rutter Child Behaviour Questionn	Normal and abnormal behaviour	7-13 yrs	3 sections 1-8 items, 2-5 items, 3-18 items	α-0.93-0.94 κ1 -0.74 k2-0.64	good		? in asd - psychometric propertie	

aire (Rutter, 1967)							s in ASD not clear	
Behaviour Problem Inventory (Rojahn *et al.*, 2012)	self-injurious, stereotypic, and aggressive/destructive behaviour	14-91yrs	50 items; 5 point frequency scale n 4-point severity scale	α-0.61-0.82 κ1 -0.65-0.76 k2-0.69-0.96	Compared with APA stds		Developed for ID ?in ASD - psychometric properties in ASD not clear	
Nisonger Child Behaviour Rating Form (Aman *et al.*, 1996)	problem behaviour and social competencies	3-16yrs	2 sections – problem behaviour + adaptive behr	α-0.77-0.91	Derived from CBCL		Developed for ID ?in ASD - psychometric properties in ASD not clear	
PDD Behaviour Inventory (Cohen *et al.*, 2003)	Adaptive and maladaptive behaviour; 10 subscales	1-12 yrs	180 items, 0 to 3 Likert scale (0 5 never; 1 5 Rarely; 2 5 Sometimes/	α-0.79-0.97	Good criterion, construct validity; with ADI-R	For ASD	Developed for follow-up for core autism features	

			Partially, and 3 5 Often / Typically).					

Anxiety questionnaires

Name of Tool	Domains measured	Age	Respondents and format	Psychometric reliability Internal consistency (α); stability (trt – κ1); equality (interrater) κ2	Psychometric validity	strengths	weaknes s	Cost
Spence children's anxiety scale-parent version (Spence, 1998)	social phobia, separation anxiety, panic attack/agoraphobia, obsessive-compulsive disorder, generalized anxiety and physical injury fears Likert of never, st, often, always	2.5-6.5yrs & 8-18 yrs	35-45 items;	Good α, κ	Good correlation	Parent report, better age range	? in ASD; psychometric properties in ASD not clear	free
Screen for Child anxiety related	GAD,SAD, social phobia, panic disorder, school phobia. 0-not true, 1-	8 - 18 yrs	41 tems	α-0.76-0.91	Good correlation with other measures	Parent report,	?n ASD; psychometric propertie	Free for downloa d

disorders (SCARED) parent (Birmaher *et al.*, 1997)	somewhat/sometime true, 2- very /often true						s in ASD not clear	
Autism Spectrum Disorders – Comorbid for Children (ASD- CC) (Matson *et al.*, 2009b)	Subscale of worry/depressed	2-16 yrs	5 items	High α, moderate k1,k2	With basc-2	Validated in asd dsm-iv and ocd-10 based	Does not evaluate the type of anxiety	Whole battery 325$ - manual +50 forms
Child symptom inventory-4 (Gadow, 2002)	Has GAD, SAD, social phobia 4 point scale – never, sometimes, often, very often	5-12 yrs; above 12 yr – ASI, YI	Total parent q, 97 item; has teacher checklist	κ1-0.46-0.87 κ2-0.74-0.94	Good corr with cbcl	Used in asd, dsm based diagnosis; gives both symptom count scores and Symptom	Advantage over cbcl; will they score high on pdd, Asperger - psychometric propertie	$42.00 per version (pack 50)

						Severity scores	s in ASD not clear	
BASC 2 (Behavioural Assessment System for Children) (Reynolds, 2004)	One scale covers attention 4 point response- never, sometimes, often, always	2-5;6-11; 12-21	Total Parent q, 134-160 items Has teacher and self report forms	α -0.77-0.94 – preschool 0.73 to 0.95 - school 0.76 to 0.95 adolescent κ1 -0.77, 0.84 and 0.81 κ2- 0.74, 0.69 and 0.77	Validated in asd	Both clinical (ext+int) & adaptive scales, dsm based Gets a chart based data; Used in asd	Long - psychometric properties in ASD not clear	Manual = $89. Hand-scored forms x25 = $33.50. Computer entry forms = $28
CBCL (Achenbach, 1991)	Anxiety part of internalising behaviour, score of 0-2, 0 – not true, 1 – somewhat or sometimes true, 2-always true.	1.5 – 18 yrs (6-18 yrs)	Total parent questionnaire;120 q	κ2overall icc 0.93 for 20 competence items & 0.96 for 118 specific probs κ1 overall icc 1 for 20 comp itms & 0.95 for 118 specific probs α ind – ranges 0.63 – 0.79;	Checked with teacher's report form and youth self report forms	dsm orient; extensively used in ASD; almost complete q	psychometric properties in ASD not clear	Hand scoring forms = $7 each or x50=$25 Parent forms x50 = $25 Computer upgrade

				overall 0.9				= $195 Computer scoring kit = $395

Hyperactivity questionnaires

Name of Tool	Domains measured	Age	Respondents and format	Psychometric reliability Internal consistency (α); stability (trt – κ1); equality (interrater) κ2	Psychometric validity	strengths	weakness	Cost
Child symptom inventory-4 (Gadow, 2002)	Attention and oppositional behaviour analysed 4 point scale – never, sometimes, often, very often	5-12 yrs; above 12 yr – ASI, YI	Total parent q, 97 item; has teacher checklist	κ1-0.46-0.87 κ2-0.74-0.94	Good corr with cbcl	Used in asd, dsm based diagnosis; gives both symptom count scores (diagnost	psychometric properties in ASD not clear	$42.00 per version (pack 50)

						ic model) or Symptom Severity scores (normative data model)		
BASC 2 (Behavioural Assessment System for Children) (Reynolds, 2004)	Attention Problems, Hyperactivity subscales 4 point response- never, sometimes, often, always	2-5;6-11; 12-21	Total parent q, 134-160 q Has teacher and self report forms	α -0.77-0.94 – preschool 0.73 to 0.95 - school 0.76 to 0.95 adolescent κ1 -0.77, 0.84 and 0.81 κ2- 0.74, 0.69 and 0.77	Validated in asd	Both clinical (ext+int) & adaptive scales, dsm based Gets a chart based data; Used in asd	psychometric properties in ASD not clear	Manual = $89. Hand-scored forms x25 = $33.50. Computer entry forms = $28
Aberant Behaviour Checklist (Aman *et al.*, 1985)	Has subscale of hyperactivity,	6-54 yrs	Total 58 items four-point scale ranging from 0 (“not	α-0.86-0.94 κ1 -0.63	With cbcl		Not for asd specifically	

			at all a problem'') to 3 (''the problem is severe in degree'').					
SDQ (Goodman, 2001)	five-factor structure (emotional, conduct, hyperactivity-inattention, peer, prosocial) missing, not true, somewhat true, certainly true	4-17 year old	Total parent questionnaire25 items	α-0.73 κ1-0.62	Correlated with dsm diagnoses	Short, Has an impact supplement; used in HFA	psychometric properties in ASD not clear	free
Conner's parent long form	Has subscales of oppositional, cognitive Problems/Inattention, and hyperactivity; uses 0-2 likert scale	3-17 yrs	Total parent q, 80 items	α-0.75-0.92 κ1 -0.47	Correlated with teacher and self-forms; validated with CBCL	Definite numbers for future follow-up	needs teacher forms; high cost; not validated in ASD	Long versions: $43.25 Short versions: $40.50 (packs of 25) $425 per complete kit including manual
CBCL (Achenbach, 1991)	Hyperactivity part of externalising behaviours score of 0-2, 0 – not true,	1.5 – 18 yrs	Total parent questionnaire;120 q	κ2overall ICC 0.93 for 20 competence	Checked with teacher's	dsm orient; extensiv	psychometric propertie	Hand scoring forms =

	1 – somewhat or sometimes true, 2- always true.	(6-18 yrs)		items & 0.96 for 118 specific probs κ1 overall icc 1 for 20 comp itms & 0.95 for 118 specific probs α ind – ranges 0.63 – 0.79; overall 0.9	report form and youth self-report forms	ely used in ASD; almost complete q	s in ASD not clear	$7 each or x50=$25 Parent forms x50 = $25 Computer upgrade = $195 Computer scoring kit = $395
Conner's ADHD index (Erhart *et al.*, 2008)	Hyperactivity and attention	8-18 yrs	10 items	Good internal consistency and reliability (on main forms)	Validated with cbcl	Short; separate male and female, age norms	psychometric properties in ASD not clear	
Multidimensional scale for pervasive developmental disorders and	14 domains: 5 domains for ASD,2 domains for developmental co-ordination disorder, 3 domains for ADHD, 4 other general disability scale for sensory, sleep, language and	3-49 years	9 point scale; eg: 1 – no sign; 2 – somewhat but no need to support to 5 – still very difficult even	Satisfactory inter-rater reliability		Covers both PDD and ADHD		

attention deficit/hyperactivity disorder (MSPA) (Funabiki *et al.*, 2011)	development		with full support in groups					

Sleep questionnaires

Name of Tool	Domains measured	Age	Respondents and format	Psychometric reliability Internal consistency (α); stability (trt – κ1); equality (interrater) κ2	Psychometric validity	strengths	Weakness	cost
Children's Sleep Habits questionnaire (Owens *et al.*, 2000)	3 point scale rarely to usually; Sleep initiation, maintenance, nightwakings, parasomnias, sleep disordered breathing	4-10 yrs (2.5-5 yrs)	Parent report 35 item; 8 subscales; 1 week retrospective report; has abbreviated form – 22	α – 0.36-0.70 κ1 – 0.62-0.79	With polysomnography (PSG) and parent reports; best available multidimensional;lewandowski (6)	Best available Multidimensional, well established,used in asd	Not validate for ASD	free for our study

			questions, 5 point scale –never to always					
Family Inventory of sleep habits (Malow *et al.*, 2009)	never-1 to always-5 daytime and prebedtime habits, bedtime routine, sleep environment	3-10 yrs	Parent report I month retrospective, 12 q	α053-0.61 κ10.82-asd, 0.56 –n		Used in asd	Only habits	
Paediatric sleep questionnaire (PSQ) (Spruyt and Gozal, 2011)	Sleep related breathing problems, snoring, daytime sleepiness, sleep initation n maint; answers – yes/no/don't know	2-18 yrs	Parent report 69 items; 8 subscales	α -0.77-0.89 κ1 - -.66-0.92	Validated with PSG	Well established	Validity in asd not established	Cost depends on use
Modified Simonds & Parraga Sleep Questionnaire (MSPSQ	bedtime resistance/ struggles, sleep onset delay, parasomnias, sleep disordered breathing, sleep anxiety,	5-18 yrs	36/51 items never = 1; about once a month = 2; a few times a month = 3; once or	α -0.8 k 1 – 0.83-1.0	Validated with sleep disturbance scale for children r=0.79 With CSHQ r= 0.7		Validity in asd not established	

) (Wiggs and Stores, 2004)	and daytime sleepiness.		twice a week = 4; and many times a week or daily = 5. Likert scale or yes/no					

Feeding questionnaires

Name of Tool	Domains measured	Age	Respondents and format	Psychometric reliability Internal consistency (α); stability (trt – κ1); equality (interrater) κ2	Psychometric validity	strengths	weakness	cost
Children Eating Behaviour Inventory (Archer *et al.*, 1991)	Eating and mealtime problems; 5 point likert scale never, seldom, sometimes, often, always	2-13 yrs	Parent report 40q	α>0. 70 (except single parent/more than 1 child group -0.58) κ1 – 0.87	Asd children in study	2scores 1. Total eating problem score 2. Total no of items perceived a problem by respondent	Validity in asd not established	

Brief Autism Mealtime Behaviour Inventory (Lukens and Linscheid, 2008)	Likert scale, 5 point likert scale as above	3-11 yrs	Parent report 18 items	α -0.87 κ1 – 0.88	Compared with Behavioural Pediatric Feeding Assessment Scale	Looks at ltd variety, food refusal. 2 scores as above. Short and specific for ASD		free
Behavioural Paediatric Feeding Assessment Scale. (Crist and Napier-Phillips, 2001)	35 item measure looks at child behaviour and parents strategies	9 mt – 7 years	In clinic, 35 item	α > 0.70	Good construct validity		Validity in asd not established	
STEP – Screening Tool of Feeding Problems – Child (Seiverling *et al.*, 2011)	Looks at aspiration risk, selectivity, food refusal, feeding skills, nutrition related behavioural problems	10-87 years	23 item	α =0.68 k1 = 0.72 k2 =0.71		Developed for MR	Validated in asd	
Mealtime Behaviour Questionnaire	Looks at Structure of family meals, Problematic Child Mealtime Behaviour,	3-11.9 yrs	Past 1 week parent report,	α =0.72 k1 = 0.8-0.95	Good construct validity	For typically developing children		

(Anderson *et al.*, 2012)	use of food as a reward, parental concern about child's diet, spousal stress, influences of child's preferences on family's choices		different likert scales					
Parent Mealtime Action Scale (PMAS) (Hendy *et al.*, 2013)	Both child and parent mealtime behaviour snack limits, positive persuasion, daily fruits and vegetables availability, use of rewards, insistence on eating, snack modeling, special meals, fat reduction, and many food choices	2-12yrs	31 item q	α =0.42-0.81 k1 = 0.51-0.78	Compared with CEBI	Parent behaviour contribution	Not spec addressed to asd	

SI problems

Name of Tool	Domains measured	Age	Respondents and format	Psychometric reliability Internal	Psychometric validity	strengths	weaknes s	cost

				consistency (α); stability (trt – κ1); equality (interrater) κ2				
Short sensory profile (Dunn and Brown, 1997)	tactile sensitivity, taste/smell sensitivity, movement sensitivity, underresponsive/seeks sensation, auditory filtering, low energy/weak and visual/auditory sensitivity 5 point scale – always, freq, occ, seld, never	3 (5)-10 yrs	38 item parent q	α -0.7-0.91	As of long version	Shorter, sensory modulation, preferred tool, better discrimination		Sensory profile kit with manual, 25 short sensory profile -186 $; 25 short sensory profile – 28.50$
Sensory profile. (Dunn and Brown, 1997)	Sensory processing (A,V,T,Vest,multi, oral), sensory modulation and	3 (5)-10 yrs	125 item parent q; typical,probable diff and	α -0.47-0.91	Content validity ok, high correlation	Covers all domains	Long, no total score	

	behavioural responses		definite diff		with school function assessment performanc e			
Sensory experiences questionnaire,(Baranek *et al.*, 2006)	Sensory hypo/hyper and social set up	5-80mts	21 item	α -0.8		Has sensory orienting threshold and a sensory aversion threshold	Not validate in ASD	
Evaluation of Sensory Processing, (Johnson-Ecker and Parham, 2000)	Sensory modulation, processing	preschoolers	84 item	Skewed results			long	

Impact questionnaires

Name of Tool	Domains measured	Age	Respondents and format	Psychometric reliability Internal consistency (α); stability (trt – κ1); equality (interrater) κ2	Psychometric validity	strengths	weakness	cost

Impact on family Scale (Stein and Jessop, 2003)	Financial Impact; Familial-Social Impact, Personal Strain, Mastery 4 point Likert-type scale (strongly agree – strongly disagree).	2-17 years	27 item scale/ 15 item scale	α –total impact – in the high 0.80s, financial-.0.68-0.79, coping 0.46-0.52.	high	Looks at financial implications	Not validated in ASD	Author contacted
Autism Parent Stress Index (Silva and Schalock, 2012)	Core and co-morbid symptoms in autism	2-6 years	125 item parent q; typical,probable diff and definite diff	α -0.732-0.834 k1-0.882	Discrimination good	Autism specific	Validity, narrow age range not relevant to our study	
Family Impact Questionnaire (Baker *et al.*, 2002)	Impact on social relationships, Negative feelings about parenting, Positive feelings about parenting, Financial impact, Impact on marriage, Impact on siblings; 4 point likert scale	3-19	50 item parent questionnaire	Fair test retest	Compared with PSI	Used in ASD,		

SF-12	Physical component summary and mental component summary		12 item		Compared with sf-36, correlations 0.92-0.96	Used in ASD	Not validated in ASD	

7.2 Appendix 2 – Results of literature search

Results of literature search on Scopus (all years – October 2012)

Search term number	Search terms used	Search results
1.	Autis* in 'title-abstract-keyword'	33,050
2.	Anxiety in 'title-abstract-keyword'	208,261
3.	Search term number 1 and 2 combined	2,124
4.	Search term number 3 and 'questionnaire or tool or measure' combined	857
5.	Hyperactivity in 'title-abstract-keyword'	94,593
6.	Search term number 1 and 5 combined	4,201
7.	Search term number 6 and 'questionnaire or tool or measure' combined	1,884
8.	Sleep in 'title-abstract-keyword'	162,435
9.	Search term number 1 and 8 combined	941
10.	Search term number 9 and 'questionnaire or tool or measure' combined	359
11.	Feeding in 'title-abstract-keyword'	347,202
12.	Search term number 1 and 11 combined	335
13.	Search term number 12 and 'questionnaire or tool or measure' combined	85
14.	Sensory in 'title-abstract-keyword'	229,044
15.	Search term number 1 and 14 combined	1,190
16.	Search term number 15 and 'questionnaire or tool or measure' combined	471
17.	'Impact on family' in 'title-abstract-keyword'	53,365
18.	'Quality of life' in 'title-abstract-keyword'	336,866
19.	Search term number 17 or 18 combined	384,953
20.	Search term number 1 and 19 combined	847
21.	'Needs' in 'title-abstract-keyword'	69,540

22.	Search term number 1 and 21 combined	233

Results of literature search on Ovid-medline (1996 – October 2012)

Search term number	Search terms used	Search results
1.	Autistic disorder.mp or autistic disorder in title	10,338
2.	Anxiety.mp in title	27,315
3.	Search term number 1 and 2 and 'questionnaire.mp or tool.mp or measure.mp' combined	13
4.	Hyperactivity.mp or hyperkinesis or attention deficit hyperactivity disorder in title	23,299
5.	Search term number 1 and 4 and 'questionnaire.mp or tool.mp or measure.mp' combined	66
6.	Sleep.mp in title	16,007
7.	Search term number 1 and 6 and 'questionnaire.mp or tool.mp or measure.mp' combined	6
8.	Feeding.mp or feeding behaviour or feeding methods or feeding and eating disorders of childhood in title	91,013
9.	Search term number 1 and 8 and 'questionnaire.mp or tool.mp or measure.mp' combined	9
10.	Sensory.mp	81,025
11.	Search term number 1 and 10 and 'questionnaire.mp or tool.mp or measure.mp' combined	41
12.	Exp quality of life/psychology in title	11,433
13.	Search term number 1 and 2 combined	38
14.	Exp Health services, needs and demands	31,182
15.	Search term number 1 and 14 combined	68

7.3 Appendix -3: Screening questionnaire (Ten item parent report database questionnaire)

Problem behaviours

So that we can estimate children's needs for extra support, please indicate whether your child has or has had the following behaviour problems:

Ratings: F = frequent, ie. Behaviour is apparent several times a week (3 or more)

S = sometimes, ie. Behaviour occurs once or twice a week

N = never or rare P = in past only

	F	S	N	P
sleep problems (including settling and night waking)				
toileting problems (including constipation, retaining faeces, smearing faeces, diarrhoea, wetting self after it is usual for his/her age group)				
'hyper' periods, very restless and irritable				
temper tantrums when not able to do what s/he wants				
aggression to other people				
injury to self (such as head-banging, biting hand)				
very reluctant to separate from one parent				
anxiety, fears or phobias				
selective about eating				
sensory reactions (such as great distress at noises, hair cutting, dentist; or unusual sensory interests)				
other: (*please give description)*				

7.4 Appendix 4 – Needs and services questionnaire

In which of these areas, have you wanted support and advice for your child in the last TWO years (please tick all that apply)					
Feeding / eating issues	Sleep	Anxiety	Hyperactivity	Other Behavioural issues	Sensory issues
○	○	○	○	○	○

For each of these areas, who did you meet for discussions and for support? Please tick ALL professionals who supported you in that area						
	Feeding / eating issues	Sleep	Anxiety	Hyperactivity	Other Behavioural issues	Sensory issues
Family doctor/ GP	○	○	○	○	○	○
Social Worker	○	○	○	○	○	○
Speech and language therapist	○	○	○	○	○	○
Family Health visitor	○	○	○	○	○	○
Community child doctor(paediatrician)	○	○	○	○	○	○
Occupational therapist	○	○	○	○	○	○
Physiotherapist	○	○	○	○	○	○
Educational psychologist	○	○	○	○	○	○
Clinical psychologist	○	○	○	○	○	○
Psychiatrist	○	○	○	○	○	○
Community Child Health mental nurse	○	○	○	○	○	○
Child's teacher	○	○	○	○	○	○
School special education needs co-ordinator	○	○	○	○	○	○
Special support assistant in school (SSA/LSA/TA)	○	○	○	○	○	○
School nurse	○	○	○	○	○	○
Community learning disability team	○	○	○	○	○	○
Voluntary organisations/charity	○	○	○	○	○	○
Other parents	○	○	○	○	○	○
We tried to solve this on our own	○	○	○	○	○	○

Appendix 4 - Needs and services questionnaire

How much support have you got from professionals in the last TWO years? Please complete this for all areas that apply to you.						
	Feeding/ eating issues	Sleep	Anxiety	Hyperactivity	Other Behavioural issues	Sensory issues
Got enough support	○	○	○	○	○	○
Got support, but wanted more	○	○	○	○	○	○
Have not got support, but did not want it	○	○	○	○	○	○
Have not got support, but wanted it	○	○	○	○	○	○
No support was needed	○	○	○	○	○	○

In each of the areas that apply to you, was the support provided by professionals useful?						Comments
Feeding/ eating issues	Sleep	Anxiety	Hyperactivity	Other Behavioural issues	Sensory issues	
○ Yes ○ No	○ Yes ○ No	○ Yes ○ No	○ Yes ○ No	○ Yes ○ No	○ Yes ○ No	

In each of these areas, do you still want support regarding any of these issues NOW from professionals?						Comments
Feeding/ eating issues	Sleep	Anxiety	Hyperactivity	Other Behavioural issues	Sensory issues	
○ Yes ○ No	○ Yes ○ No	○ Yes ○ No	○ Yes ○ No	○ Yes ○ No	○ Yes ○ No	

7.5 Appendix 5 – Approvals

7.5.1 NRES committee Approvals

National Research Ethics Service

NRES Committee West Midlands - The Black Country

HRA NRES Centre Manchester

3rd Floor, Barlow House

4 Minshull Street

Manchester

M1 3DZ

Telephone: 0161 625 7832

Facsimile: 0161 625 7299

11 March 2013

Dr Beena Koshy

Research Fellow

Institute of Neurosciences

James Spence Building – Third Floor

Royal Victoria Infirmary

Newcastle upon Tyne

NE1 4LP

Dear Dr Koshy

Study title:	ASD+ Study - Co-existing emotional and behavioural conditions in children with Autism Spectrum Disorder - frequency, severity and correlates

	from two large UK databases
REC reference:	13/WM/0098
IRAS project ID:	117526

The Proportionate Review Sub-committee of the NRES Committee West Midlands - The Black Country reviewed the above application on 27 February 2013.

We plan to publish your research summary wording for the above study on the NRES website, together with your contact details, unless you expressly withhold permission to do so. Publication will be no earlier than three months from the date of this favourable opinion letter. Should you wish to provide a substitute contact point, require further information, or wish to withhold permission to publish, please contact the Co-ordinator Miss Helen Penistone, nrescommittee.westmidlands-blackcountry@nhs.net.

Ethical opinion

On behalf of the Committee, the sub-committee gave a favourable ethical opinion of the above research on the basis described in the application form, protocol and supporting documentation, subject to the conditions specified below.

Ethical review of research sites

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

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

Yours sincerely

On behalf of

Dr Jeff Neilson

Chair

7.5.2 Approval by NTW trust

Research & Clinical Effectiveness Department
St Nicholas Hospital
Jubilee Road
Gosforth
Newcastle upon Tyne NE3 3XT
Tel: (External) 0191 223 2338
(Internal) 32338
Fax: 0191 223 2341
18 April 2013
RES-13-004
Dr Jeremy Parr
Clinical Senior Lecturer
Consultant Neurodisability Paediatrician
Institute of Neuroscience
Newcastle University
Sir James Spence Building
RVI
NE1 4LP

Dear Dr Parr
Re: ASD + Study – co-existing conditions in children with ASD
CSP Ref: 117526
I write to confirm that Northumberland, Tyne and Wear NHS Foundation Trust are happy to support and approve the above study. Please accept this letter as verification of Trust approval.
Approval is granted with the condition that the R&D Department are notified of:
Commencement and completion of the study
Any significant changes to the study design
Suspension or abandonment of the study
All publications and/or conference presentation of the study findings

The Department of Health's minimum standards for research governance state that at least 10% of projects should be routinely audited. It is a condition of our approval that the researchers accept the Trust's right to include this project in the auditing and monitoring process.

Best wishes

Yours sincerely

Simon Douglas

Senior Manager for Research, Innovation and Clinical Effectiveness

7.5.3 NIHR clinical research portfolio approval

Dear Dr Parr

Re: 117526 - ASD+ Study - Co-existing conditions in children with ASD

Thank you for your R&D submission.

We are pleased to confirm that your study is proceeding through NIHR CSP.

Please contact the Lead CLRN, Northumberland, Tyne and Wear, for this study if you require any further information.

If new research sites or PICs are added to the study, you should amend the list of sites in Part C of the R&D Form and resubmit via IRAS. Please inform the Lead CLRN at this time.

Further information on NIHR CRN Portfolio and NIHR CSP can be found at http://www.crncc.nihr.ac.uk.

Should you decide not to proceed with this study, please inform the Lead CLRN as soon as possible.

Regards

Jane Darnbrough

Research Approvals Facilitator, CSP Unit

NIHR Clinical Research Network Coordinating Centre

Email crncc.cspunit@nihr.ac.uk

7.6 Appendix 6: Coding and cleaning of data and missing data corrections for each questionnaire

The detailed description below provides information about how each questionnaire was handled regarding data entry and analysis.

7.6.1 The ASD-CC (Autism Spectrum Disorder Comorbidity – Child Version)

The ASD-CC was entered as parents have marked for each item:

0 – 'Not a problem or impairment'

1 – 'Mild problem or impairment'

2 – 'Severe problem or impairment'

The item, parent has marked as X indicating 'does not apply', was entered as 3. Missing data was entered as 9. In any instance, when a parent had marked a number in a column, the column was taken as relevant – for example if a parent marked 1 in the column of 'not a problem or impairment'; entry was made as 0, as 'not a problem or impairment' stood for 0. In instances where parent has marked two columns or marked on the border of two columns indicating a score between two values, the higher value was taken.

The data entered was analysed for any wrong entry and internal consistency was done for the whole scale. We recoded 3 as 0. Missing value analysis done as described below in 1.3.

If any questionnaire had items missing more than 20% or any of the seven subscales had items missing more than 20%, the whole questionnaire was excluded from analysis. If there were less than 20% of missing items, the mean of the subscale for that item replaced the missing value.

The total ASD-CC score was calculated by adding all the items and the subscale scores were calculated by adding the subscale items.

7.6.2 The ASD-PBC (Autism Spectrum Disorder Problem Behaviour – Child Version)

The ASD-PBC was entered as parents have marked for each item:

0 – 'Not a problem or impairment'

1 – 'Mild problem or impairment'

2 – 'Severe problem or impairment'

Though this questionnaire does not have an option of 'does not apply', some parents continued to mark X outside the columns. This was entered as 3. Missing data was entered as 9. In any instance, when a parent had marked a number in a column, the column was taken as relevant – for example if a parent marked 1 in the column of 'not a problem or impairment'; entry was made as 0, as not a problem or impairment stood for 0. In instances where parent has marked two columns or marked on the border of two columns indicating a score between two values, the higher value was taken.

The data entered was analysed for any wrong entry and internal consistency was done for the whole scale. We recoded 3 as 0. Missing value analysis done as described in 1.3.

If any questionnaire had items missing more than 20% or any of the two subscales had items missing more than 20%, the whole questionnaire was excluded from analysis. If there were less than 20% of missing items, the mean of the subscale for that item replaced the missing value.

The total ASD-PBC score was calculated by adding all the items and the subscale scores were calculated by adding the subscale items.

7.6.3 The IOF (Impact on Family)

The scale was entered as marked by parents to each statement:

1 - Strongly Agree

2 - Agree

3 – Disagree

4 – Strongly Disagree

Missing data was entered as 9. The data entered was analysed for any wrong entry and internal consistency was done for the whole scale. We reverse scored all items as per the manual: 1 as 4, 2 as 3, 3 as 2 and 4 as 1. Missing value analysis done as described in 1.3.

If any questionnaire had items missing more than 20% the whole questionnaire was excluded from analysis. If there were less than 20% of missing items, the mean replaced the missing value (Revised IOF has only one factor structure).

The total Revised Impact on Family score was calculated by adding all the coded items.

7.6.4 The Needs and services Questionnaire

This was a questionnaire developed by the researcher.

For Question 1 (wanted support and advice in the last two years), parents' positive response (included answers of tick/cross/dot/blackened circle) was entered as 1 and negative response as 0.

For Question 2 (tick all professionals who parent met for support), parents' positive response was entered as 1 and negative response as 0.

For Question 3 (how much support), parents' positive response was entered as below:

1 – Got enough support

2 – Got support, but wanted more

3 – Have not got support, but did not want it

4 – Have not got support, but wanted it

5 – No support was needed

The response was noted as missing (9) if there was no entry. In instances where parent has marked two rows or marked on the border of two rows indicating a score between two values, the value indicating more need was taken.

For Question 4 (useful support), if parents marked yes; this was entered as 1 and no as 0. Missing value was entered as 9.

For Question 5 (still want support), if parents marked yes; this was entered as 1 and no as 0. Missing value was entered as 9.

Incomplete questionnaires were excluded from analyses (2nd page of the questionnaire left blank).

If Question 3 had a missing value and the problem (Question 1) was scored 0, Question 3 was recoded as 5 for that problem. If Question 3 had a missing value and Question 4 (useful support) was 0 and/or Question 5 (still need support) was 1, Question 3 was recoded as 4 for that problem. If Question 3 had a missing value and Question 4 was 1, Question 3 was recoded as 2 for that problem.

If the coded entry for Question 3 for a problem was 2 (got support, but wanted more) or 4 (have not got support, but wanted it), this was recoded as unmet need for that problem. Missing value analysis done as described in 1.3. Similarly, unmet need was calculated for each problem. Total unmet need was calculated by adding the individual unmet need and can have a maximum of 6 (feeding, sleep, anxiety, hyperactivity, other behavioural issues, sensory issues).

7.6.5 The Short Sensory Profile (SSP)

As per the manual, the individual responses were entered as below:

1 – Always

2 – Frequently

3 – Occasionally

4 – Seldom

5 – Never

In instances where parent has marked two columns or marked on the border of two columns indicating a score between two values, the higher frequency was taken. Eleven parents recorded limited food choices for Questions 9 (will only eat certain tastes: list) and/or 10 (limits self to particular food textures/temperatures: list), but did not mark the frequency. These were entered as 1. Missing data was entered as 9. The data entered was analysed for any wrong entry and internal consistency was done for the whole scale. Missing value analysis done as in 1.3.

If any questionnaire had items missing more than 20% or any of the seven subscales had items missing more than 20%, the whole questionnaire was excluded from analysis. If there were less than 20% of missing items, the mean of the subscale for that item replaced the missing value.

The total SSP score was calculated by adding all the items and the subscale scores were calculated by adding the subscale items.

7.6.6 The Conners 3 AI – Parent Questionnaire

The items were entered as marked by each parent on the questionnaire. Missing data was entered as 9. The data entered was analysed for any wrong entry and internal consistency was done for the whole scale. Missing value analysis done as in 1.3.

If any questionnaire had items missing more than 20% the whole questionnaire was excluded from analysis. If there were less than 20% of missing items, the mean replaced the missing value (Conners 3 AI has only one factor structure).

The total score was calculated by adding all the items.

7.6.7 The Spence Children's Anxiety Scale – Parent version

The parent responses were scored as per the manual:

0 - Never

1- Sometimes

2 - Often

3 - Always

Missing data was entered as 9. In instances where parent has marked two columns or marked on the border of two columns indicating a score between two values, the higher frequency was taken.

The data entered was analysed for any wrong entry and internal consistency was done for the whole scale. Missing value analysis done as in 1.3.

If any questionnaire had items missing more than 20% or any of the six subscales had items missing more than 20%, the whole questionnaire was excluded from analysis. If there were less than 20% of missing items, the mean of the subscale for that item replaced the missing value.

The sub-scale scores are computed by adding the individual item scores. The total score is the sum of all these six sub-scale scores.

7.6.8 The Brief Autism Mealtime Behaviour Inventory (BAMBI)

The parent responses were entered as per the questionnaire for frequency of feeding habits

1 – Never/Rarely

2 – Seldom

3 – Occasionally

4 – Often

5 – At almost every meal

For each item, if parent marked ‘Yes’ indicating a problem, this was scored as 1 and ‘No’ as 0. Missing data was entered as 9. The data entered was analysed for any

wrong entry and internal consistency was done for the whole scale including the frequency of feeding habits. Missing value analysis done as in 1.3. Items 3, 9, 10 and 15 were reverse scored as per the manual.

If any questionnaire had items missing more than 20% or any of the three subscales had items missing more than 20%, the whole questionnaire was excluded from analysis. If there were less than 20% of missing items, the mean of the subscale for that item replaced the missing value.

The total BAMBI score was calculated by adding all the items and the subscale scores were calculated by adding the subscale items.

7.6.9 The Children's Sleep Habits Questionnaire (CSHQ)

The parent responses were entered as per the questionnaire for frequency of sleep habits

3 – Usually

2 – Sometimes

1 – Rarely

For each item, if parent marked 'Yes' indicating a problem, this was scored as 1 and 'No' as 0. Missing data was entered as 9. The data entered was analysed for any wrong entry and internal consistency was done for the whole scale including the frequency of sleep habits. Missing value analysis done as in 1.3. Items 1, 2, 3, 10, 11 and 26 were reverse scored as per the manual.

If any questionnaire had items missing more than 20%, the whole questionnaire was excluded from analysis. If there were less than 20% of missing items, the mean replaced the missing value.

The total CSHQ score was calculated by adding all the items.

7.7 Appendix 7 - Variables for ASD+ study

- Type of diagnosis recoded as below

1- Autism

3- Asperger syndrome

2- All other ASD

- Diagnosis coded for regression analysis

1 – autism

0 – all others

- School recoded as below

1 - Special school

0 – all others

- Language level

1- Sentences

0 – All other language levels

- Gender

1 – male

0– female

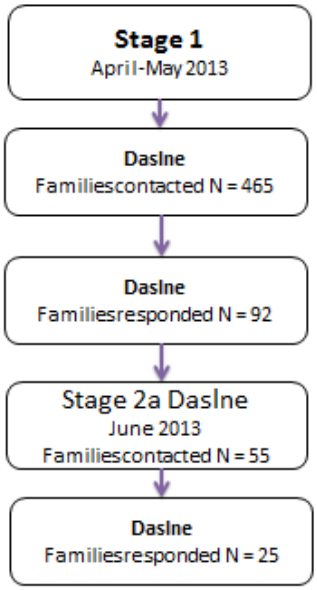
- Age as continuous variable calculated from date of birth and 1st May 2013. Age 2 coded 8 and less – 1 and above 8 - 2
- Siblings with ASD recoded as 1 if yes and 0 if no

7.8 Appendix 8 - Methodology of contacting families

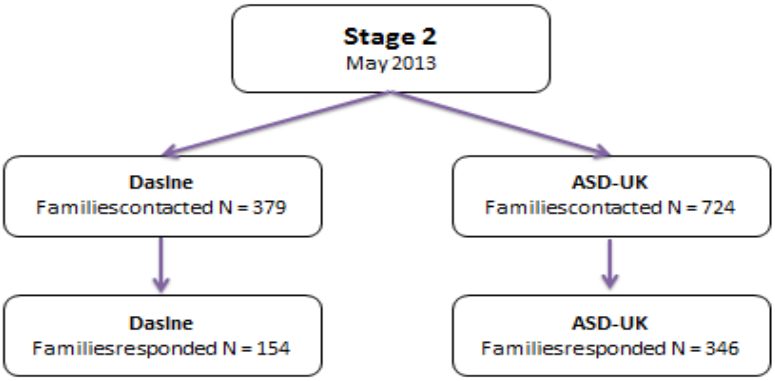
We contacted families in four stages

1. Stage 1: We contacted Daslne families who did not respond to the prior concluded parent update. All these families received Pack 1 along with the ten item parent questionnaire. This stage was sent out in April and May 2013.
2. Stage 2: We contacted both Daslne and ASD-UK families in this stage. Parents received Pack 1 and customised Pack 2 based on their report on ten item parent questionnaire. This stage was sent out in May 2013
3. Stage 2a: Parents who responded to Stage 1 received appropriate Pack 2 questionnaires based on their report on the ten item parent questionnaire, if they agreed to be contacted again on their consent form. This stage was sent out in June 2013.
4. Stage 3: Parents who joined the databases after April 2013 were not included in Stage 1. These families were included in this stage and the mails were sent out in July 2013.

   <u>Numbers contacted and responded in different stages</u>



Appendix 8 – Methodology of contacting families



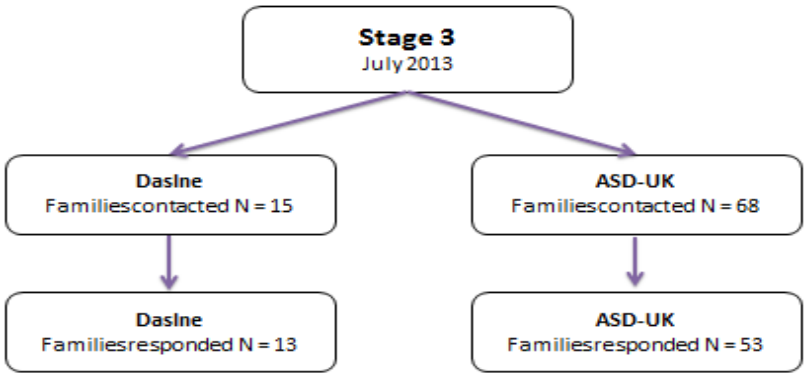


Table showing responses from different databases

	Stage 1	Stage 2	Stage 3	Total
Daslne (%)	92/465 (20%)	154/379 (40%)	13/15 (87%)	259/859 (30%)
ASD-UK (%)	-	346/724 (48%)	53/68 (78%)	399/792 (50%)

Numerator shows the number of responses and denominator the number contacted

7.9 Appendix 9 - Raw data for mediation models

Table 46: The details of mediation model of total unmet needs using Hayes' model 4 with number of moderate-severely impairing co-morbid psychopathology as X variable

```
Run MATRIX procedure:

************* PROCESS Procedure for SPSS Beta Release 140712 *************

        Written by Andrew F. Hayes, Ph.D.   http://www.afhayes.com

**************************************************************************
Model = 4
    Y = totalIOF
    X = numbermo
    M = TOTALUNM

Statistical Controls:
CONTROL= genderco sibscode diagnosi language schoolco age      EXTERNAL INTERNAL

Sample size
        517

**************************************************************************
Outcome: TOTALUNM

Model Summary
          R       R-sq          F        df1        df2          p
      .4487      .2013    14.2006     9.0000   507.0000      .0000

Model
              coeff         se          t          p       LLCI       ULCI
constant     1.3209      .4761     2.7742      .0057      .3855     2.2564
numbermo      .2221      .0550     4.0413      .0001      .1141      .3300
genderco     -.1270      .2050     -.6195      .5359     -.5298      .2758
sibscode      .2307      .2643      .8728      .3832     -.2886      .7500
diagnosi      .0545      .1353      .4030      .6871     -.2113      .3203
language      .1248      .1798      .6942      .4879     -.2284      .4780
schoolco      .0830      .1369      .6064      .5445     -.1859      .3519
age          -.0632      .0233    -2.7104      .0069     -.1090     -.0174
```

```
EXTERNAL      .1041      .0269     3.8658      .0001      .0512      .1570
INTERNAL      .0405      .0275     1.4722      .1416     -.0135      .0945

************************************************************************
Outcome: totalIOF

Model Summary
          R       R-sq          F        df1        df2          p
      .6189      .3830    31.4161    10.0000   506.0000      .0000

Model
              coeff         se          t          p       LLCI       ULCI
constant    31.4938     2.1061    14.9537      .0000    27.3560    35.6316
TOTALUNM      .8613      .1950     4.4178      .0000      .4783     1.2444
numbermo      .9209      .2451     3.7571      .0002      .4393     1.4024
genderco    -1.4119      .9004    -1.5681      .1175    -3.1809      .3571
sibscode     2.9261     1.1612     2.5198      .0120      .6447     5.2076
diagnosi    -1.0150      .5940    -1.7088      .0881    -2.1820      .1520
language     -.8197      .7895    -1.0382      .2997    -2.3709      .7314
schoolco      .3038      .6010      .5054      .6135     -.8770     1.4846
age           .2409      .1031     2.3361      .0199      .0383      .4435
EXTERNAL      .5812      .1200     4.8447      .0000      .3455      .8169
INTERNAL      .4404      .1210     3.6407      .0003      .2028      .6781

****************** DIRECT AND INDIRECT EFFECTS *************************

Direct effect of X on Y
     Effect         SE          t          p       LLCI       ULCI
      .9209      .2451     3.7571      .0002      .4393     1.4024

Indirect effect of X on Y
            Effect    Boot SE   BootLLCI   BootULCI
TOTALUNM      .1913      .0612      .0950      .3510
```

Table 47: The details of mediation model of total unmet needs using Hayes' model 4 with externalising behaviour as X variable

```
Run MATRIX procedure:


************* PROCESS Procedure for SPSS Beta Release 140712 *************

        Written by Andrew F. Hayes, Ph.D.   http://www.afhayes.com

**************************************************************************
Model = 4
    Y = totalIOF
    X = EXTERNAL
    M = TOTALUNM

Statistical Controls:
CONTROL= numbermo genderco sibscode diagnosi language schoolco age      INTERNAL

Sample size
        517

**************************************************************************
Outcome: TOTALUNM

Model Summary
          R       R-sq          F        df1        df2          p
      .4487      .2013    14.2006     9.0000   507.0000      .0000

Model
              coeff         se          t          p       LLCI       ULCI
constant     1.3209      .4761     2.7742      .0057      .3855     2.2564
EXTERNAL      .1041      .0269     3.8658      .0001      .0512      .1570
numbermo      .2221      .0550     4.0413      .0001      .1141      .3300
genderco     -.1270      .2050     -.6195      .5359     -.5298      .2758
sibscode      .2307      .2643      .8728      .3832     -.2886      .7500
diagnosi      .0545      .1353      .4030      .6871     -.2113      .3203
```

```
language      .1248      .1798      .6942      .4879     -.2284      .4780
schoolco      .0830      .1369      .6064      .5445     -.1859      .3519
age          -.0632      .0233    -2.7104      .0069     -.1090     -.0174
INTERNAL      .0405      .0275     1.4722      .1416     -.0135      .0945

**************************************************************************
Outcome: totalIOF

Model Summary
          R       R-sq          F        df1        df2          p
      .6189      .3830    31.4161    10.0000   506.0000      .0000

Model
             coeff         se          t          p       LLCI       ULCI
constant   31.4938     2.1061    14.9537      .0000    27.3560    35.6316
TOTALUNM     .8613      .1950     4.4178      .0000      .4783     1.2444
EXTERNAL     .5812      .1200     4.8447      .0000      .3455      .8169
numbermo     .9209      .2451     3.7571      .0002      .4393     1.4024
genderco    -1.4119      .9004    -1.5681      .1175    -3.1809      .3571
sibscode     2.9261     1.1612     2.5198      .0120      .6447     5.2076
diagnosi    -1.0150      .5940    -1.7088      .0881    -2.1820      .1520
language     -.8197      .7895    -1.0382      .2997    -2.3709      .7314
schoolco      .3038      .6010      .5054      .6135     -.8770     1.4846
age           .2409      .1031     2.3361      .0199      .0383      .4435
INTERNAL      .4404      .1210     3.6407      .0003      .2028      .6781

******************** DIRECT AND INDIRECT EFFECTS *************************

Direct effect of X on Y
     Effect         SE          t          p       LLCI       ULCI
      .5812      .1200     4.8447      .0000      .3455      .8169

Indirect effect of X on Y
            Effect    Boot SE   BootLLCI   BootULCI
TOTALUNM      .0897      .0325      .0410      .1759
```

Table 48: The details of mediation model of total unmet needs using Hayes' model 4 with internalising behaviour as X variable

```
Run MATRIX procedure:

************* PROCESS Procedure for SPSS Beta Release 140712 *************

        Written by Andrew F. Hayes, Ph.D.   http://www.afhayes.com

**************************************************************************
Model = 4
    Y = totalIOF
    X = INTERNAL
    M = TOTALUNM

Statistical Controls:
CONTROL= numbermo genderco sibscode diagnosi language schoolco age      EXTERNAL

Sample size
        517

**************************************************************************
Outcome: TOTALUNM

Model Summary
          R       R-sq          F        df1        df2          p
      .4487      .2013    14.2006     9.0000   507.0000      .0000

Model
             coeff         se          t          p       LLCI       ULCI
constant    1.3209      .4761     2.7742      .0057      .3855     2.2564
INTERNAL     .0405      .0275     1.4722      .1416     -.0135      .0945
numbermo     .2221      .0550     4.0413      .0001      .1141      .3300
genderco    -.1270      .2050     -.6195      .5359     -.5298      .2758
sibscode     .2307      .2643      .8728      .3832     -.2886      .7500
diagnosi     .0545      .1353      .4030      .6871     -.2113      .3203
```

```
language      .1248      .1798      .6942      .4879     -.2284      .4780
schoolco      .0830      .1369      .6064      .5445     -.1859      .3519
age          -.0632      .0233    -2.7104      .0069     -.1090     -.0174
EXTERNAL      .1041      .0269     3.8658      .0001      .0512      .1570

************************************************************************
Outcome: totalIOF

Model Summary
          R       R-sq          F        df1        df2          p
      .6189      .3830    31.4161    10.0000   506.0000      .0000

Model
             coeff         se          t          p       LLCI       ULCI
constant   31.4938     2.1061    14.9537      .0000    27.3560    35.6316
TOTALUNM     .8613      .1950     4.4178      .0000      .4783     1.2444
INTERNAL     .4404      .1210     3.6407      .0003      .2028      .6781
numbermo     .9209      .2451     3.7571      .0002      .4393     1.4024
genderco    -1.4119      .9004    -1.5681      .1175    -3.1809      .3571
sibscode     2.9261     1.1612     2.5198      .0120      .6447     5.2076
diagnosi    -1.0150      .5940    -1.7088      .0881    -2.1820      .1520
language     -.8197      .7895    -1.0382      .2997    -2.3709      .7314
schoolco      .3038      .6010      .5054      .6135     -.8770     1.4846
age           .2409      .1031     2.3361      .0199      .0383      .4435
EXTERNAL      .5812      .1200     4.8447      .0000      .3455      .8169

****************** DIRECT AND INDIRECT EFFECTS *************************

Direct effect of X on Y
     Effect         SE          t          p       LLCI       ULCI
      .4404      .1210     3.6407      .0003      .2028      .6781

Indirect effect of X on Y
            Effect    Boot SE   BootLLCI   BootULCI
TOTALUNM      .0349      .0295     -.0179      .1049
```

8 Publications and presentations from this thesis

1. International Meeting for Autism Research (IMFAR) 2013. Poster presentation. *Beena Koshy, Morag Maskey, Frances Warnell, Mary Johnson, Helen McConachie, Ann Le-Couteur & Jeremy Parr.* ASD+ Study - Co-existing conditions in children with ASD: evidence from two large UK databases.
2. European Academy of Childhood Disability (EACD) meeting 2013. Poster presentation. *Beena Koshy, Morag Maskey, Frances Warnell, Mary Johnson, Helen McConachie, Ann Le-Couteur & Jeremy Parr.* ASD+ Study - Co-existing conditions in children with ASD: evidence from two large UK databases.
3. EACD meeting 2013. Poster presentation. *Abigail Soul, Beena Koshy, Helen McConachie and Jeremy Parr*. Impact on the Family of Co-existing Conditions in Autism Spectrum Disorders.
4. IMFAR 2014. Poster presentation. *Beena Koshy, Jacqui Rodgers, Ann LeCouteur, Helen McConachie & Jeremy Parr*. The UK ASD+ Study – Co-existing conditions of children with ASD, unmet needs for services and impact on the family.

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