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Repetitive Behaviours, Anxiety and Sensory Problems in Children with Autism and Correlates of Anxiety in their Parents

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Thesis Summary

Repetitive and restricted behaviours have been considered as a core symptom of autism since the first descriptions provided by Leo Kanner (Kanner, 1943) and Hans Asperger (Asperger, 1944), and this view has been supported through all the incarnations of international diagnostic systems. However, when compared with other core features: problems in communication and problems in reciprocal social interaction, repetitive behaviours have been relatively neglected in terms of research. In particular, little is known about the triggers for repetitive behaviours and what functions they might serve for individuals with autism. Early theoretical accounts of autism proposed that sensory problems and anxiety are the key triggers for repetitive behaviours. Research in the field of developmental psychology has also demonstrated that in very young children, repetitive behaviours serve the function of constraining sensory unpredictability and warding off anxiety and this relationship has also been found in individuals with Obsessive Compulsive Disorder, a condition defined by anxiety and ritualism. However, the three way relationship between repetitive behaviours, sensory problems and anxiety has not been explored in the autism literature before. The main aim of the thesis (Part 1) was to explore this relationship in children with autism. In addition to this main focus, the work of the thesis also included a study of the mothers of the same children (Part 2), investigating mothers' own anxiety, sensory correlates of this anxiety and other factors.

The main effects of autism are on the individual who is on the autism spectrum, however, impact is also felt by families. Parents of children with autism have been found to have higher prevalence of affective disorders when compared to both parents of typically developing children and parents of children with other disabilities. The second main aim of the thesis was to explore correlates of anxiety in mothers of children with autism.

In Part 1, a series of studies are reported using questionnaire data to investigate repetitive behaviours (RRBs), anxiety and sensory problems in children with autism. Part 2, focused on anxiety in the mothers of these children and correlates of their anxiety.

The first empirical chapter (Chapter 3) examined RRBs in children with and without autism and validated the Repetitive Behaviour Questionnaire-2 (RBQ-2; Leekam et al., 2007) for the first time with children with autism. Results indicated that the RBQ-2 provides a highly reliable measure of repetitive behaviours for children with autism aged from 2 to 17 years. Factor analysis revealed that a two-factor structure with the repetitive motor and sensory behaviour (RSM) factor and the insistence on sameness (IS) factor best represented

the data. Internal consistency was high for both the total RBQ-2 scale and for each RSM and IS subscale separately. This chapter also explored moderators of repetitive behaviours in children and adolescents with autism and found that while RSM behaviours were negatively associated with lower chronological age, and lower expressive language levels, IS behaviours were not. Further analysis was conducted using a secondary dataset from a longitudinal study of RRBs in typically developing children at age 15, 24 and 72 months old. Results showed that IS and RSM behaviours represent relatively independent classes of behaviours in developmental terms.

The second empirical chapter (Chapter 4) examined anxiety in children with autism and provided validation for the school age and preschool version of the Spence Anxiety Scales. Using this scale it was found that as many as 49% of children with autism met the criteria for elevated overall anxiety and that separation anxiety and physical injury fears were the most prevalent anxiety subtypes. Good agreement on total anxiety scores between children's and parents' reports was found. Analysis on the mediators of anxiety suggested that anxiety was not associated with chronological age, impairments in communication and social interaction, and expressive language levels.

The third empirical chapter (Chapter 5) provided evidence that sensory problems in children and adolescents with autism are both multisensory and multimodal in nature. More precisely, only 2 children had problems in a single sensory modality while on the other hand, 40% of children had problems simultaneously across all five primary sensory modalities. Almost 91% of children and adolescents showed mixed types of sensory modulation problems, with 65.3% of children having problems in all four sensory quadrants (sensory hyper- and hypo-sensitivity, seeking, and avoidance). This chapter also explored the mutual relationship between the four sensory subscale (quadrants) of the Sensory Profile. Results indicated that children and adolescents with autism fluctuate between the states of hypo- and hyper-responsiveness; that avoidance behaviours are compensatory strategies related to being over-stimulated, and that seeking behaviours are more related to being over-stimulated rather than hypo-stimulated.

The final chapter in Part 1, Chapter 6, examined the interrelations between RRB, sensory problems and anxiety. Results showed that both insistence on sameness (IS) and repetitive motor (RM) behaviours were associated with sensory problems. However, only IS behaviours were associated with anxiety while RM behaviours were not. Mediation analyses models showed that sensory sensitivity and anxiety were reinforcing each other in the relationship with IS behaviours.

The two chapters in Part 2 examined the frequency and correlates of anxiety in mothers of children with autism. Findings from these two chapters (Chapters 8 and 9) suggested that anxiety is very prevalent among mothers of children with autism with 46% of mothers meeting the cut-off criteria for clinically significant anxiety. For the first time, sensory problems in mothers were examined, and were also found to be very prevalent in mothers, appearing in 60% per cent of mothers. The frequency of sensory features in mothers was correlated with the frequency of sensory features in their children. Further analysis suggested that higher levels of sensory over-sensitivity, higher levels of intolerance of uncertainty, and the dominant use of escape-avoidance coping style contributed to anxiety levels in mothers. Furthermore, it was found that sensory sensitivity in mothers was related to their higher use of escape-avoidant coping strategy and the mediation analysis suggested that sensory sensitivity, escape-avoidance coping and anxiety reinforced each other.

The importance of these findings for future theoretical and clinical work is considered in detail in the general discussion chapter (Chapter 10).

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Chapter 1: General Introduction

The dread of change and incompleteness seems to be a major factor in the explanation of the monotonous repetitiousness and the resulting limitations in the variety of spontaneous activity.

(Kanner, 1943, p246)

1.1. Repetitive Behaviours

Leo Kanner (Kanner, 1943) provided a detailed description of various behaviours that are now considered as repetitive behaviours. Behaviours that he observed in his original sample of 11 children included various motor mannerisms such as shaking head from side to side or jumping up and down repeatedly, use of objects in inflexible and highly repetitive manner (e.g., spinning round objects), unusual preoccupation and fascination with ordinary objects such as cardboard boxes and pencils, highly intense interests and very strong insistence that things need to be 'just so' (e.g. insistence that parts of the furniture and other objects need to be arranged in a certain way and becoming distressed if any change was made). Kanner considered these behaviours as an essential feature of then new disorder that has become known as autism and this view has not changed much to this day, exactly 70 years after his seminal paper was published.

Since Kanner's original description, various incarnations of international diagnostic criteria required repetitive behaviours to be present in order for ASD diagnosis to be established. The International Classification of Diseases (10th rev.; ICD-10; WHO, 1993) describe the following four subtypes of Restricted and Repetitive Behaviours (RRBs): (a) encompassing preoccupation with one or more stereotyped and restricted patterns of interest that is abnormal either in intensity or focus; (b) apparently inflexible adherence to specific, non-functional routines or rituals; (c) stereotyped and repetitive motor mannerisms (e.g., hand or finger flapping or twisting or complex whole-body movements); and (d) persistent preoccupation with parts of objects. The latest Diagnostic and Statistical Manual of Mental Disorders (5th ed.; DSM-V; APA, 2013) lists the following 4 types of repetitive behaviours: (a) extreme attachment to routines and patterns and resistance to changes in routines; (b) repetitive speech or movements; (c) intense and restrictive interests; and (d) difficulty integrating sensory information or strong seeking or avoiding behaviour of sensory stimuli.

Although there is a consensus that all these behaviours are linked by rigidity, repetitiveness, invariance and inappropriateness (Turner, 1999), it is still debated whether

they should be conceptualized as an uni-dimensional or alternatively as multi-dimensional construct that encompass several related but at the same time distinct behavioural categories. Based on clinical observations and a developmental approach, Prior and Macmillan (1973) and later Turner (1999) argued that RRBs can be classified into “lower level” behaviours such as dyskinesias, tics, stereotyped movements, repetitive manipulation of objects and self-injurious behaviours thought to be characteristic for younger and/or lower functioning children and higher level repetitive behaviours such as object attachments, repetitive language, insistence on sameness and circumscribed interests thought to be present in older and more able children.

Using a different approach to classification, Lewis & Bodfish (1998) argued that co-occurrence of different repetitive behaviours across various disorders as well as their co-occurrence within individuals suggests that there is likely a common pathophysiology behind various repetitive behaviours and that they can be considered as unidimensional construct (or “overall response class”) in this sense. Dysfunction in nigrostriatal dopaminergic pathway was considered as a key problem in the model proposed by Lewis and Bodfish. More recently, Langen and colleagues (Langen, et al., 2010a; b) proposed a more refined neurobiological model that conceptualizes repetitive behaviours as a multidimensional construct with motor stereotypies linked to motor and premotor cortex, rigidity and obsessiveness to cognitive/associative loop and compulsions to anterior cingulate-orbitofrontal cortex loop.

Over the last 15 years, considerable research effort in the form of factor analytic studies, has focused on the problem of how to describe and classify RRBs. This is not simply a semantic exercise but rather a crucial question and worthwhile enterprise as clear definitions can provide conceptual distinctions that differentiate subtypes of RRB which should facilitate more effective research on origins and outcomes of these behaviours and eventually lead to the development of effective intervention approaches. Therefore this thesis begins with a detailed summary of the classification of RRBs.

1. 1. 1. Summary of factor analytic studies that used ADI-R

Systematic search of the literature has identified 17 factor analytic studies that have investigated the structure of RRBs in autism. Detailed description of the studies is provided in the table 1. 1.

Out of the 17 studies, 11 studies used the Autism Diagnostic Interview-Revised (ADI-R; Lord, Rutter, & Le Couteur, 1994), four studies used the Repetitive Behaviour Scale-

Revised (RBS-R; Bodfish et al., 1999), Maudsley Item Sheet (Carcani-Rathwell, Rabe-Hasketh, & Santosh, 2006) and the Repetitive Behaviour Questionnaire (RBQ, Turner, 1995) were used in one study each. A detailed review of RRBs measures will be provided in the Chapter 3. However, before discussing factor analytic studies in more detail, it is appropriate to briefly summarize the above mentioned measures here.

The ADI-R is a semi-structured parental interview used in the diagnosis of Autism Spectrum Disorders (ASD). It contains 14 RRBs items with 12 items applicable to children of all age and ability levels and the following 2 items: circumscribed interests and repetitive language which require a certain cognitive ability level. The RBS-R is a 43 item parent questionnaire that asks parents to rate severity and report on the extent to which each item is a problem. RBS-R has the following 6 subscales: stereotyped behaviour, self-injurious behaviour, compulsive behaviour, ritualistic behaviour, sameness behaviour, and restricted behaviour. The RBQ is a 33-item parental questionnaire. RBQ assesses the following four types of RRBs: repetitive movements, sameness behaviour, repetitive use of language and circumscribed interests. The Maudsley Item Sheet is a psychiatric assessment which includes mental state examination and parent interview. It consists of 13 RRBs related items grouped into the following 3 categories: sensory behaviours, repetitive movements and cognitive rigidity symptoms.

Out of the 11 factor analytic studies that used the ADI-R, eight studies identified 2 factor solution, two studies identified 3 factor solution and one study identified only one factor. In studies that found a two factor solution, the two factors were generally labelled as repetitive sensory-motor (RSM) behaviours and insistence on sameness (IS) behaviours. The third factor was labelled as Restricted Interests/Preoccupations.

As can be seen from Table 1.1, the findings regarding RSMB factor were quite consistent across the studies. For example, in the examination of the RRBs of 292 individuals with autism (age range: 3-21 years), Cuccaro et al. (2003) used 12 ADI-R items and identified a RSMB factor consisting of the following items: hand and finger mannerisms, unusual sensory interests, repetitive use of objects or parts of objects, complex mannerisms or stereotyped body movements and rocking. With the exception of the rocking item, the RSMB factor identified by Cuccaro and colleagues contained identical items as the RSMB factor identified by Honey et al. (2006), Szatmari et al. (2006) and very similar items found by other 6 ADI-R factor analytic studies.

The situation was more complicated regarding the IS factor. Although the following items: difficulties with minor changes in routine or personal environment, resistance to trivial

changes in the environment and compulsions/rituals, consistently loaded onto an IS factor across all 9 studies, other items such as the unusual attachment to objects, unusual preoccupations and restricted interests showed inconsistent loading. More specifically, these items either loaded onto the IS factor (Mooney et al., 2009), did not load onto the IS factor and were not included in the final factor solution (Cuccaro et al., 2003) or, together with some other IS items constituted Restricted Interests/Preoccupations factor as in Lam, Bodfish & Piven (2008) and Honey et al. (2006) studies.

Two factors that resemble RSMB and IS factor identified in ADI-R factor analytic studies were also found by Carcani-Rathwell et al. (2006) who used Maudsley Item Sheet and by Honey et al. (2012) who used RBQ. Two studies that used the RBS-R to investigate the structure of repetitive behaviours identified more complex factor solutions than studies reviewed above. In a sample of 307 individuals with autism, Lam et al. (2007) found evidence for the existence of the following 5 factors: Rituals/Sameness, Self-injurious Behaviour, Stereotypic Behaviour, Compulsive Behaviour and Restricted Interests. A similar factor structure of RBS-R to that identified by Lam et al. (2007) was also reported by Bishop et al. (2013) in a large sample of 1825 individuals with autism aged 4-18 years. As can be seen from Table 1. 1., the age range of individuals with autism included in this study was very wide, ranging from 3-48 years. This limitation was addressed by Mirenda et al. (2010) who explored the structure of RRBs in a population 287 children aged 2-5 years. Factors identified by Mirenda et al. were very similar to the ones identified in the previous study by Lam and colleagues. The third factor analytic study that used the RBS-R study (Georgiades et al. 2010) found evidence for two factors in a sample of 205 individuals with autism aged 2-48 years. These findings are more in line with ADI-R and other studies reviewed above. Georgiades and colleagues found that all items from the following four original RBS-R subscales: Compulsive, Ritualistic, Sameness, and Restricted behaviours loaded onto a single factor resembling IS behaviours and that all items from the original RBS-R Stereotypy and Self-Injurious subscales loaded onto the factor that somewhat resembled RSM behaviours.

The studies reviewed above differed in the characteristics of participants and measures used. As can be seen from Table 1.1, some studies have examined RRBs structure in toddlers and very young children (Honey et al., 2008; Mooney et al., 2009), however, samples in the rest of the studies consisted of individuals with very wide chronological age range and also variable developmental level. Equally important is the question of the specific measure that was used. I will discuss this issue in depth in Chapter 3. However, it is important to highlight here that the type of measure that is used to assess RRBs sets the

boundaries for the phenomenon under focus and necessarily influences the conceptualization of RRBs. For example, although the ADI-R is a dominant measure of RRBs, it is limited in two ways. Firstly, it has been highlighted by several authors that because the ADI-R is used to diagnose children with autism, its use in research raises the issue of circularity (Lam et al., 2007; Leekam et al., 2011; Honey et al., 2012). Secondly, in addition to the issue of circularity the ADI-R has only 12 RRB items that are applicable to children of all ages and developmental levels and it has been reported that it undersamples RRBs (Lam et al., 2007; Honey et al., 2012). Further to this, as can be seen from Table 1. 1., studies were inconsistent in terms of the numbers of ADI-R items that are included in the analysis. Although the RBS-R is a measure that assesses a much wider range of behaviours when compared to ADI-R, most of the items are appropriate to individuals with lower developmental level only. In addition to this, the RBS-R includes self-injurious behaviours (SIBs) which are considered by a number of authors to be conceptually different than repetitive behaviours (a comprehensive review of this issue is provided in Matson, Dempsey, & Fodstad, 2009). In summary, as Honey et al. (2012) pointed out, it can be argued that it is difficult to achieve a consensus regarding the conceptualization of RRBs in the absence of a good measure.

The variability in the samples examined and in RRBs measures, necessarily limits conclusions that can be drawn from the reviewed literature. The reviewed studies seem to be relatively consistent regarding the existence and content of RSM factor that resembles behaviours that Turner (1999) termed as “low-level” behaviours. Findings regarding the group of behaviours that Turner considered as higher level behaviours are much less consistent. More precisely, these behaviours form either one or two categories of behaviours and specific behaviours that form these categories are highly variable between studies. However, in general, results seem to lend support to the categories proposed by Turner (1999) and listed in the current diagnostic criteria.

Table 1. 1. Factor Analytic Studies on Repetitive behaviours in autism

Author (year)	Autism Group (Number of participants, age, functioning/diagnostic group)	RRBs Measure		Factors		
			Sensory-Motor	Insistence on Sameness	Restricted Interests	Items not included in the final model
Cuccaro et al. (2003)	N= 292 individuals with autism, CA range: 3-21 years.	ADI-R (12 items)	hand and finger mannerisms, unusual sensory interests, repetitive use of objects or parts of objects, other complex mannerisms or stereotyped body movements and rocking	difficulties with minor changes in routine or personal environment, resistance to trivial changes in the environment and compulsions/rituals		unusual preoccupations, unusual attachment to objects, idiosyncratic negative responses, sensitivity to noise
Shao et al. (2003)	N= 221 individuals with autism, CA range: 3-21 yr.	ADI-R (13 items)	hand & finger mannerisms, repetitive use of objects, unusual sensory interests, complex mannerisms, rocking, unusual preoccupations, unusual attachment to objects	difficulties with minor changes in personal routine or environment, resistance to trivial changes in the environment, compulsions/rituals		circumscribed interests, sensitivity to noise, idiosyncratic negative responses
Tadevosyan-Leyfer et al. (2003)	N= 292 individuals with autism, age range: 2-47yr	ADI-R (98 items in total, number of RRB items was not specified)	Only one Factor (Compulsions): Stereotyped utterances, Unusual preoccupations, Compulsions/rituals, Resistance to trivial changes in the environment, Unusual attachment to objects. Repetitive motor behaviours didn't load to compulsions factor but loaded with items related to Social Interaction.			

Author (year)	Autism Group (Number of participants, age, functioning/diagnostic group)	RRBs Measure		Factors		
			Sensory-Motor	Insistence on Sameness	Restricted Interests	Items not included in the final model
Carcani-Rathwell et al., 2005	N= 319 individuals with PDD (183 with PDD+ MD and 119 with PDD without MD). Mean CA= 7.7 years (SD= 4.1)	Maudsley Item Sheet	thumb sucking, tongue sucking, rocking, masturbation, nail biting, scratching, head banging, whirling, flapping, twisting of hands, complex mannerisms	circumscribed interests and preoccupations, Resistance to change, Repetitive lining up		
Bishop, Richler & Lord (2006)	N=830 children with autism (N=560 autism, N=268 PDD-NOS, 2 AS). Mean CA 4.8 yr (range: 1.2 –11.9).	ADI-R (11 items)	repetitive use of objects, unusual sensory interests, complex mannerisms, hand & finger mannerisms, unusual preoccupations	difficulties with minor changes in personal routine or environment, resistance to trivial changes in the environment, compulsions/rituals, idiosyncratic negative responses, sensitivity to noise		unusual attachment to objects.
Szatmari et al. (2006)	N= 339 individuals with autism, mean CA: 8.4 yr (SD = 5.5) months. Mean IQ (Leiter) = 65.7 (SD = 28.7).	ADI-R (11 items)	difficulties with minor changes in personal routine or environment, resistance to trivial changes in the environment, compulsions/rituals	hand & finger mannerisms, repetitive use of objects, unusual sensory interests, complex mannerisms, rocking		circumscribed interests, unusual preoccupations, unusual attachment to objects didn't load on any of the factors.

Author (year)	Autism Group (Number of participants, age, functioning/diagnostic group)	RRBs Measure	Factors			
			Sensory-Motor	Insistence on Sameness	Restricted Interests	Items not included in the final model
Lam et al. (2007)	N= 307 individuals with autism. Mean CA= 15.34 years (age range: 3-48)	RBS-R	<p>Factor 1 (Rituals/Sameness): placement of objects, no new places, no interruption, walks certain way, sits certain place, appearance/behaviour of others, uses certain door, videotapes, difficult transitions, insists on routine, insists on time;</p> <p>Factor 2 (Self-injurious Behaviour): hits body, hits against surface, hits object, bites self, pulls hair/skin, rubs/scratches, inserts finger/object, picks skin;</p> <p>Factor 3 (Stereotypic Behaviour): body movements, head movements, finger movements, locomotion, object usage, sensory;</p> <p>Factor 4 (Compulsive Behaviour): ordering, completeness, washing, checking, counting, hoarding;</p> <p>Factor 5 (Restricted Interests): preoccupation with subject, attached to object, preoccupied with parts of object, preoccupation with movement.</p>			
Richler et al. (2007)	N= 165 AUTISM (CA < 3yr), N= 44 DD (1.1-2.9 yr) and N= 65 TD children (CA < 3yr). Children were assessed when they were 2, 3, 5 and 9yr old.	ADI-R (12 items)	repetitive use of objects, unusual sensory interests, hand/finger mannerisms, and other complex mannerisms	compulsions and rituals, difficulties with changes in routine, and resistance to trivial changes in the environment		unusual preoccupations, unusual attachment to objects, sensitivity to noise, and abnormal/idiosyncratic response to sensory stimuli
Honey et al. (2008)	N= 104 children with autism or language disorders, CA range: 2-4 yr.	ADI-R (12 items)	hand & finger mannerisms, repetitive use of objects, unusual sensory interests, complex mannerisms, unusual fears, self injury	difficulties with minor changes in personal routine or environment, resistance changes in environment, compulsions/rituals, unusual fears, idiosyncratic negative responses	unusual preoccupations, unusual attachment to objects	

Author (year)	Autism Group (Number of participants, age, functioning/diagnostic group)	RRBs Measure	Factors			
			Sensory-Motor	Insistence on Sameness	Restricted Interests	Items not included in the final model
Lam, Bodfish & Piven (2008)	N= 316 Individuals with autism. Mean CA=9.02 yr (range 1.7-29). Mean IQ=69.5 (range 20-133).	ADI-R (10 items)	repetitive use of objects, hand and finger mannerisms, and other complex mannerisms/stereotyped body movements items	difficulties with minor changes in personal routine and environment, resistance to trivial changes in the environment, and compulsions and rituals	circumscribed interests, unusual preoccupations and unusual attachment to objects	Rocking
Mooney et al. (2009)	N= 137 DD children with PDD and N= 61 DD children without PDD, all with CA 1.7-4.6 yr.	ADI-R (12 items)	hand & finger mannerisms, repetitive use of objects, complex mannerisms	difficulties with minor changes in personal routine or environment, resistance to trivial changes in the environment, compulsions/rituals, unusual attachment to objects		idiosyncratic negative responses, sensitivity to noise, unusual sensory interests, unusual preoccupations, self-injury
Georgiades et al. (2010)	N= 205 individuals with autism. Mean CA= 11.5 years (range: 2-48)	RBS-R	Factor I (Compulsive Ritualistic Sameness Restricted Behaviours; CRSRB): all items from the original RBS-R Compulsive, Ritualistic, Sameness, and Restricted subscales. Factor II (Stereotyped Self-Injurious Behaviours; SSIB): all items from the original RBS-R Stereotypy and Self-Injurious subscales.			
Mirenda et al. (2010)	N= 287 children with autism, mean CA3.4 yr, range: 2-5.3.	RBS-R	3 and 5 factor RRBs models were the preferable models. Model III comprised of Factor I: Compulsive Ritualistic Sameness Behaviours (CRSB); Factor II: Self Injurious Behaviours (SIB), Factor III: Restricted Stereotyped behaviours (RSB), items 1–6 and 40–43. Model V comprised of Factor I: Stereotyped Behaviours; Factor II: Self Injurious Behaviours (SIB); Factor III: Compulsive Behaviours (COMP); Factor IV: Ritualistic Sameness Behaviours (RITUAL/SAME) and Factor V: Restricted Behaviours (RESTR).			

Author (year)	Autism Group (Number of participants, age, functioning/diagnostic group)	RRBs Measure		Factors		
			Sensory-Motor	Insistence on Sameness	Restricted Interests	Items not included in the final model
Richler et al. (2010)	N= 192 children with autism who were referred for a diagnosis when they were under the age 3 and followed up at the age of 3, 5 and 9.	ADI-R	repetitive use of objects, unusual sensory interests, hand/finger mannerisms, and other complex mannerisms	compulsions and rituals, difficulties with changes in routine, and resistance to trivial changes in the environment		unusual preoccupations, unusual attachment to objects, sensitivity to noise, and abnormal/idiosyncratic response to sensory stimuli
Honey et al. (2012)	N= 180 individuals with autism. Mean CA= 45.53 months (age range: 37-192). N= 40 children had FSIQ< 70	RBQ	spin around, rock backward, forwards or side to side, pace or move around, hand and finger mannerisms, body movements	Insistence on Sameness/Restricted Interests Factor: insists things in the house stay the same, insists items put out or stored in the same way, plays the same music, game or video, insists on using same objects or items in specific situations, insists wearing the same clothes or refuse new clothes, certain items of clothing always worn, insists on eating same foods, insists on moving or travelling by same route, react to changes to surroundings at home, insists on same routines, rituals for everyday activities, rituals linked to particular occasions and places, negative reaction to changes in daily routine		says the same things or make same noises, talks about same topic, operate light switches, taps toilet flush, arrange toys or other items,

Author (year)	Autism Group (Number of participants, age, functioning/ diagnostic group)	RRBs Measure		Factors			
Bishop et al., 2013	N= 1825 Individuals with autism. Mean CA= 8.9 years (range: 4-18 years, SD: 3.5). Mean VIQ: 79.5 (range: 5-167, SD= 30.2), mean NVIQ: 86.5 (range: 9-161, SD= 25.1), mean FSIQ: 83 (range: 7-167, SD: 26.9)	ADI-R	Unusual preoccupations, repetitive use of objects, hand and finger mannerisms, complex mannerisms, unusual sensory interests	Compulsions/rituals, sensitivity to noise, abnormal/idiosyncratic response, difficulties with change, resistance to change, circumscribed interests		Self-injurious behaviours, Unusual attachment to objects	
Bishop et al., 2013	Same as above	RBS-R	Factor 1(Sensory motor): body movements, head movements, finger movements, locomotion, object usage, sensory, fascination with movements; Factor 2 (Self-injurious Behaviour): hits body, hits against surface, hits object, bites self, pulls hair/skin, rubs/scratches, inserts finger/object, picks skin; Factor 3 (Compulsive Behaviour): ordering, completeness, washing, checking, counting, hoarding, repeating; Factor 4 (Ritualistic/Sameness Behaviours): ordering, completeness, washing, checking, counting, hoarding; Factor 5 (Restricted Interests): preoccupation with subject, attached to object, preoccupied with parts of object, preoccupation with movement.				

1. 1. 2. How do repetitive behaviours in autism change over time?

Another way to investigate whether proposed distinctive and yet related categories of RRBs are valid is to look at whether there is a difference in the way that these behaviours are related to other characteristics of individuals with autism, in particular their chronological age and developmental level. This literature will be reviewed below (studies are also summarized in Table 1, Appendix 1).

The current empirical findings are mixed regarding the association between age and the number and severity of RRBs in autism. In a retrospective study of changes in the diagnostic triad, Piven, Harper, Palmer & Arndt (1996) used the ADI-R to examine the current behaviours of 38 high functioning adults and adolescents with autism (mean age= 17.6 years). These behaviours were then compared with the behaviours that these individuals showed at 5 years of age (according to their parents' retrospective reports). It was found that repetitive behaviours improved in only 50% of the sample. Similar methodology was also employed by Fecteau, Mottron, Berthiaume and Burack (2003) who examined changes in the triad features of 28 children and adults with autism. Fecteau and colleagues identified more positive changes in repetitive behaviours over time, although not as great as those seen for social and communication impairments. They found that the greatest improvements were in the repetitive use of objects. More precisely, of 17 children who were reported to engage in the repetitive use of objects at 4 to 5 years of age, 76.5% appeared to show a decrease in these behaviours with age. Also, with the exception of verbal rituals, no behaviours got worse over time.

The findings above were based on retrospective report of changes. Several longitudinal studies have also been carried out. For example, longitudinal study by Moore & Goodson (2003) examined twenty children at 2 years and again at 4 to 5 years using the ADI-R. While there was little change in social interaction and communication skills over time, repetitive behaviours became more apparent. Changes in the ADI-R scores of 29 children with autism were examined by Charman, Taylor, Drew, Cockerill, Brown and Baird (2005). This study showed that repetitive behaviours increased between 3 years and 4 to 5 years before decreasing from 4-5 years to 7 years. In a study that examined the way in which repetitive behaviours changed from 2 to 9 years of age in a sample of 172 children with autism and PDD-NOS, Lord, Risi, DiLavore, Thurm & Pickles (2006) found that repetitive behaviours increased with age. Finally, using the Autism Diagnostic Observation Schedule

(ADOS; Lord et al., 2002), Kim and Lord (2010), did not find that chronological age was associated with the severity of repetitive behaviours in a sample of 121 children with autism and 71 children with PDD-NOS divided into the following 6 age cohorts (younger than 18 months, 19–24; 25–30; 31–36; 37–42; and 43–56 months).

The reasons for reported inconsistencies in terms of the relationship between chronological age and repetitive behaviours are two-fold. Firstly, as studies by Evans, Gray and Leckman (1999) and Berkson and Tupa (2000) show, developmental level is negatively associated with repetitive behaviours in severely delayed populations. These findings, together with the fact that although chronological and developmental age are parallel in typical development, developmental delay is common in children with autism, suggest that it is necessary to consider the developmental level of children with autism when examining presence and changes in their behaviours. Secondly, as highlighted in the section on the structure of repetitive behaviours, it has been suggested that different types or classes of repetitive behaviours are differently associated with chronological and developmental age. The evidence for a relation between repetitive behaviour and developmental ability will be reviewed below.

In addition to the question of how repetitive behaviours change with chronological age, Piven et al. (1996) also examined differences based on cognitive level. When the autism group was split according to IQ, it was found that the greatest decrease in repetitive behaviours with age was for a group of Individuals with autism with an IQ over 70 suggesting that the greater the cognitive capacity of an individual, the greater their decrease in repetitive behaviours with age. Gabriels, Cuccaro, Hill, Ivers & Goldson (2005), like Piven et al., divided individuals with autism into high nonverbal mental age group (NVIQ > 97) and low nonverbal mental age group (NVIQ < 56). Using RBS-R to examine differences in repetitive behaviours between groups, they found that total RRBs scores were significantly higher in lower NVIQ group. After adjusted for multiple comparisons it was revealed that the groups differed significantly on only the Sameness scale (i.e., difficulties with change) which was more severe in the Low NVIQ group. A study by Bishop, Richler & Lord (2006) used the ADI-R in order to examine the effects of non-verbal IQ and chronological age on repetitive behaviours in 830 children with autism (age range: 6 to 12 years). This study examined the prevalence and severity of thirteen repetitive behaviours, that is, whether behaviours were present or not and to what extent they interfere with daily life. With respect to age, analysis revealed significant improvements in the prevalence of some ADI-R

repetitive behaviour items including the following: sensitivity to noise, circumscribed interests, difficulties with changes in routine and resistance to trivial changes in the environment. On the other hand, the repetitive use of objects and unusual sensory interests were found to increase significantly. With respect to non-verbal IQ, lower IQ indicated a larger number of behaviours including the repetitive use of objects while higher IQ levels indicated the higher presence of circumscribed interests. Esbensen et al. (2009) used RBS-R to examine age related differences in 700 individuals with autism aged 2 to 62 years. They found that older individuals had significantly lower RBS-R total scores as well as lower scores for all subscales. Furthermore, individuals with autism and comorbid ID had significantly more stereotyped movements and SIB than autistic individuals without the comorbid ID. ID was not significantly correlated with expression of ritualistic/sameness behaviours, compulsions and restricted interests. More recently, Richler, Huerta, Bishop & Lord (2010) used the ADI-R to track longitudinal change in RRBs in children with ASD over a 7 year period (RRBs were assessed when children were 2, 3, 5, and 9 years of age). Results showed that RSM behaviours remained relatively stable over a 7 year period but RSM behaviours did decrease in children with higher NVIQs. Unlike RSM behaviours, IS behaviours, gradually (moderately) increased over time. In addition to older chronological age, higher IS scores were also associated with milder social and communication impairments. Possible reason for this finding was the fact that age/IQ ratio was too low.

The findings reviewed above and summarized in Table 1 (Appendix 1) highlight the importance of considering the role of developmental level in addition to the role of chronological age and suggest that different types of RRBs are differently associated with chronological and developmental age. The majority of studies seem to support Turner's (1999) conceptualization of RSM behaviours as "lower level" behaviours as they are generally found in younger children and individuals with lower developmental level. Conceptualization of IS behaviours as higher order is less straightforward. Several studies failed to find association between this factor and developmental level. However, it is important to highlight that these findings need to be considered against what we know about the structure and change of repetitive behaviours during typical development. A brief summary of repetitive behaviours in typical development will be reviewed below.

1. 1. 3. Repetitive Behaviours in Typical Development

Repetitive behaviours are seen throughout childhood in the normative population and are considered to be an important feature of development. A variety of rhythmic and stereotyped motor and sensory behaviours are very prevalent during the first year of life (Thelen, 1979; 1981). It has been suggested that these behaviours serve adaptive purposes such as neuromuscular and motor skills development (Wolff, 1968; Thelen, 1979) and general central nervous system maturation (Sprague & Newell, 1996) and begin to reduce after the child's first birthday. For example, Leekam et al. (2007) used the Repetitive Behaviour Questionnaire-2 (RBQ-2) to explore repetitive sensory and motor behaviours in 675 two-year old TD children and found that, at that age, these behaviours were common, with each of RBQ-2 items being endorsed by between 18 and 30% of children. Arnott et al. (2010) explored the frequency of RRBs in the same sample of children as Leekam and colleagues (children were 15 month old) and found that motor behaviours were more frequent than when children were 24 months old, with some items such as repetitively fiddling with toys, being endorsed by 60% of children.

At the same time, when sensory and motor repetitive behaviours such as kicking and banging start to decrease, rigid types of behaviours such as a need for sameness start to increase. Gessell and colleagues (1928; 1974) observed that between 2 to 3 years of age, typically developing children show compulsive like patterns of behaviours such as insistence on sameness, repetitive and ritualised behaviours, rigidity in likes and dislikes and acute sensory perception for minute details. In a parental-report study of 8 to 72 month-old typically developing children, Evans, Leckman, Carter, Reznick, Henshaw, King & Pauls (1997) used the Childhood Routines Inventory (CRI, Evans et al., 1997) to examine the developmental trajectory of two components of 'compulsive-like' behaviours. These were 'just right' behaviours which included higher level behaviours (such as a preference for the same daily routine, repetition of certain actions over and over, acting out the same sequence over and over in pretend play), and 'repetitive behaviours & insistence on sameness' (including items such as: preference for the same household schedule or routine every day, preference for certain foods). With respect to 'just right' behaviours, they occurred significantly more in children aged 24 to 48 months than in children aged 12 months and 72 months who displayed similar lower levels of these behaviours. Children younger than 12 months demonstrated the lowest levels of 'just right' behaviours. With respect to 'repetitive behaviours and insistence on sameness', these behaviours were present to a similar degree as

'just right' behaviour, although these behaviours emerged earlier and had a steeper increase over time.

In a longitudinal study using the RBQ-2 described above, Leekam et al. (2007) found that at the age of two, items: 'fascination with specific objects' and 'carry special objects around' were reported as 'marked' behaviours in more than 30% of the sample and as 'mild' and 'occasional' in 40% and 28% of their sample of 675 TD children respectively.

The findings reviewed above suggest that RSM and IS behaviours have somewhat distinctive developmental trajectories. RSM behaviours seem to be apparent very early in development and start to quickly reduce in the frequency and intensity from around 15-18 months while IS behaviours start to increase, about the same time (15-18 months) and are prominent up to around 4 years when they start to decrease. It is not clear whether RSM behaviours are precursors or earlier developmental forms that are then replaced by the IS behaviours or whether, these two behaviours represent independent classes of behaviours with separate genetic and neurophysiological make-up. Exploring this question is of crucial importance for better understanding of RRBs in autism as well as in other disorders.

Repetitive behaviours are not only present during early typical childhood and are not only found in individuals with autism but also occur in a wide variety of neurodevelopmental conditions and individuals with developmental disabilities, psychiatric disorders (for example schizophrenia, obsessive-compulsive disorder), and neurological conditions such as Tourette's Syndrome for example. It is important to determine whether any of repetitive behaviours are specific to autism. The literature on repetitive behaviours in other conditions will be very briefly summarized below and the question of distinctiveness of repetitive behaviours between autism and other clinical groups will be discussed.

1. 1. 4. Repetitive Behaviours in other neurodevelopmental and psychiatric conditions

Stereotyped hand and body mannerisms are common in individuals with mental retardation, individuals diagnosed with schizophrenia, Tourette's syndrome, attention deficit hyperactivity disorder (ADHD), Alzheimer's Disease and other forms of dementias, in individuals with sensory impairments such as loss of hearing and vision (Bodfish, Symons, Parker, & Lewis, 2000; Mahone, Bridges, Prahme, & Singer, 2004). Rigid behaviours in the form of obsessions and compulsions and behaviours with insistence on sameness component are a defining feature of OCD as well as in anxiety spectrum conditions (Robinson, 1998). It has been found that in some instances certain repetitive behaviours can be considered

characteristic of certain syndrome groups. For example, lick and flip and self-hug behaviours are frequently described in Smith Magenis syndrome (Finucane, Dirrigl, & Simon, 2001). Attachment to objects is very prevalent in Cri du Chat syndrome (Cornish & Pigram 1996) and skin picking is present in majority of individuals with Prader-Willi Syndrome (PWS; Dykens, Leckman, & Cassidy, 1996; Dykens, Cassidy, & King, 1999; Butler et al. 2004; Torrado et al. 2006).

The question naturally arises, what distinguishes repetitive behaviours seen in individuals with autism from repetitive behaviours observed in conditions reviewed above?

1. 1. 5. Distinctiveness of repetitive behaviours in autism when compared with other clinical groups

Turner (1999) suggested that in addition to being associated to younger chronological age and lower developmental age, repetitive sensory and motor behaviours do not distinguish the population with autism from other clinical groups where cognitive impairments are present. Findings from several comparative studies seem to confirm this hypothesis (Greaves, Prince, Evans, & Charman, 2006; Moss & Oliver, 2009; Flores et al., 2011). Evidence that insistence on sameness type of behaviours is more specific to autism are mixed. These types of behaviours are, as mentioned above, certainly present in other conditions and are even considered as defining characteristics of some disorder such as OCD for example. A study by Greaves et al. (2006) used the CRI to examine RRBs in 89 children with autism and 80 children with PWS and didn't find differences between children with PWS and children with autism on either total CRI scores or 'just right' and 'repetitive' behaviour subscale scores. However, more recent research by Flores et al. (2011) found that RRBs were significantly less frequent and less severe in the PWS sample when compared with the autistic sample (RBS-R was used). The use of different measures could have been the contributing factor to the inconsistent findings. Two studies compared the repetitive behaviours of individuals with autism and OCD. Zandt, Prior, & Kyrios (2007) used the RBQ and Children's Yale Brown Obsessive-Compulsive Scale (CY-BOCS; Scahill et al., 1997) to examine repetitive behaviours in 19 children with autism and CA and IQ matched children with OCD (N=17). They found that although both groups showed similar levels of IS behaviours, children with OCD had more compulsions and obsessions than children with autism. There were also differences in terms of how age influenced the expression of repetitive behaviours, with younger children with OCD showing more IS behaviours while for children with autism, age

was not significantly associated with the expression of IS behaviours, repetitive movements, compulsions or obsessions. Similar findings to the ones reported by Zandt et al. (2007) in children were also found when comparing adult ASD and OCD populations (McDougle et al., 1995).

The picture that seems to be emerging from reviewed studies is that it is the frequency of RRBs rather than their form or pattern that distinguishes autism from other clinical groups. In other words, while more specific behaviours can be seen in other clinical group, autistic group tends to express a wide range of RRBs at the same time.

1. 1. 6. Distinctiveness of RRBs as a core impairment or the relationship between repetitive behaviours and other core autism symptoms

Traditionally, it is assumed that repetitive behaviours are related to the impairments in reciprocal social interaction and deficits in communication. However, this assumption has been questioned by several researchers (Constantino, Gruber, Davis, Hayes, Passanante, & Przybeck, 2004; Ronald, Happe & Plomin, 2005; Happe, Ronald, & Plomin, 2006; Happe & Ronald, 2008). For example, Constantino et al. (2004), using both ADI-R and Social Responsiveness scale across different psychiatric conditions found that a single, unitary factor solution, rather than three factor, provided the best description of their data. On the other hand, Happe et al. (2006) and Happe and Ronald (2008) argued for the “fractionation” of social and non-social (RRB) symptoms of autism. They suggested that social and non-social impairments have distinctive genetic etiologies and that their association in children with autism is coincidental. This suggestion was supported to an extent by findings from Ronald et al. (2005) who found only modest correlation between RRBs, social and communication problems. Furthermore, evidence that pointed out to the independent, at least to certain extent, genetic influence on social and non-social symptoms were found.

Several studies have explored the relationship between RRBs and social/communication problems (Charman et al., 2005; Gabriels et al., 2005; Szatmari et al., 2006; Lam, Bodfish & Piven, 2008; Paul, Chawarska, Cicchetti, & Volkmar, 2008; Watt Wetherby, Barber, & Morgan, 2008; Richler et al., 2010; Ray-Subramanian & Weismer, 2012) and in general, found that higher RRBs scores were associated with more social/communication problems. For example, Ray-Subramanian & Weismer (2012) examined the association between repetitive behaviours, and language and nonverbal

cognitive skills in a sample of 115 children with autism. All the variables were assessed at two age points: at the ages of 2 and 3. They found that, by the age of 3, higher RRBs scores were correlated with lower levels of receptive and expressive language, and NVIQ. It was also reported that the increase in language levels between ages 2 and 3 predicted decrease in RRBs. A study by Paul et al. (2008) looked at the language outcomes in 37 children with autism. Children were assessed at two time points-when they were 15-25 months old and 12 months later using ADI-R and VABS. It was found that lower levels of repetitive behaviours during the initial assessment (together with higher receptive language VABS scores) were significant predictors of VABS expressive language scores 12 months later. Studies that looked at subtypes of repetitive behaviours found that they were differently associated with communication and social abilities. For example, Lam, Bodfish & Piven (2008) used ADI-R in a sample of 316 children with autism (mean age= 9 years) to look at the relationship between different RRBs subtypes and social/communication deficits. They found that both higher repetitive motor behaviours and insistence on sameness were significantly associated with more social/communication problems. No association between circumscribed interests and social/communication deficits was found. Also, Szatmari et al. (2006) found that while repetitive sensory-motor behaviours (ADI-R) were negatively associated with all VABS subscales and ADI-R communication scores, insistence on sameness behaviours showed positive association with VABS and ADI-R communication scores.

1. 1. 7. Neurobiology of Repetitive Behaviours

Basal ganglia are the structures most often connected with repetitive behaviours in autism although the structural and functional findings have been somewhat inconsistent (Langen et al., 2010). Hollander et al. (2005) found a 10% larger right caudate volume in a group of 17 individuals with autism (mean age= 29.4 years, SD= 9.08) and a lack of asymmetry between left and right caudate when compared with typically developed controls. The insistence on sameness repetitive behaviour scores (measured with ADI-R) showed a positive correlation with the right caudate and putamen volumes. However, Sears et al. (1999) found no differences in the volume of caudate (and no evidence for symmetry reversal), putamen and globus pallidus between 35 individuals with autism (age range: 12-29 years) and typically developed controls. Caudate volume was negatively associated with the following ADI-R items: complex mannerisms, compulsions/rituals, and difficulties with

minor changes in routine were significantly negatively correlated with caudate volume (ADI was used).

Other structures, such as anterior cingulate cortex (ACC) and amygdala, have also been implicated in the neurobiology of repetitive behaviours. For example, Thakkar et al. (2008) examined the association between ACC structure and RRBs. They found reduced fractional anisotropy in white matter underlying dorsal and rostral ACC in 12 adults with autism. Lower fractional anisotropy was significantly associated with higher repetitive behaviour scores (ADI-R). Furthermore, individuals with autism made more antisaccade errors, showed increased rostral ACC activation to both correct and error responses, and increased activation in rostral ACC to correct responses was positively associated with ADI-R repetitive behaviour scores. Although amygdala abnormalities have been traditionally associated with the social impairments (Baron-Cohen et al. 2000; Ashwin et al., 2006), Amaral and Corbett (2003) suggested that it might contribute to anxiety and repetitive behaviours. Indeed, Dziobek et al. (2006) found that higher imposition of routines and rituals (ADI-R) was associated with smaller amygdala volume in 17 individuals with Asperger syndrome (mean age= 41.4). However, although interesting, this finding needs to be replicated.

It is important to emphasize that both methodological differences such as manual anatomic tracing versus automated MRI morphometry, as well as differences between participants, may have contributed to the above mentioned inconsistencies in findings between the studies. For example, one particularly interesting issue might be the age difference between samples. Langen et al. (2009) investigated the structural brain development of caudate nucleus in 99 individuals with high functioning autism (mean age= 12.89 years, SD= 4.45) and found that while caudate volume in the autism group increased with age it decreased in the control group.

In summary, it can be argued that in order to gain better understanding of the neurobiology of repetitive behaviours, it is necessary to explore these behaviours across different conditions and also to go beyond separate brain structure and investigate neural circuits and the role of their dysfunction in repetitive behaviours. This approach was advocated by Langen and colleagues in two recent review articles (Langen et al., 2010a, b). They proposed that motor stereotypies can be linked to the dysfunction in motor and premotor cortex, rigidity and obsessiveness to cognitive/associative loop and compulsions to

anterior cingulate-orbitofrontal cortex loop. Future experimental work will need to test and further refine this proposal.

1. 1. 8. Summary

Despite being recognized as a core autism feature from the original description by Kanner and Asperger to the latest instalments of the current diagnostic criteria, there is still much to be learned about the definition and structure of RRBs in autism, their cause and potential to change, their relationship with other core autism characteristics. Although several new measurements of RRBs have been developed, as could be seen from the brief overview of the literature presented in this section, the majority of research is still dominated by the ADI-R whose use in the research on RRBs raises the issue of circularity and undersampling of RRBs. As has been emphasized in this section, characteristics of the measure that is used to assess RRBs (or any other behaviour or phenomenon for that matter) sets the boundaries and necessarily influences the conceptualization of RRBs. Having this in mind, one of the main aims of the first empirical chapter on RRBs (Chapter 3) will be to provide a more detailed review of the RRBs measures and how the RRBs measure used in this thesis was selected. This chapter will then present results of a factor analysis and exploration of the psychometric properties of the selected RRBs measure. The second focus of Chapter 3 will be to look at the developmental trajectory of two hypothesised classes of RRBs: RSM and IS in typical development. As has been discussed above, a better understanding of the change in RRBs in typical development is of crucial importance for the better understanding of these behaviours in autism. Finally, Chapter 3 will also address the issue of how different types of RRBs are related to the core autism features in children and adolescents with autism. In subsequent chapters the focus moves to the issue of potential factors for the maintenance of RRBs in children with autism. More precisely, I focus on the possibility that anxiety and sensory problems serve as important triggers for RRBs.

1.2. Anxiety and Autism

The presence of anxiety in individuals with autism was noted in the original descriptions by Leo Kanner (Kanner, 1943) and Hans Asperger (Asperger, 1944). Fear of unusual objects and situations such as sound of egg beater, vacuum cleaner, playgrounds and various toys were frequent in children that both Kanner and Asperger described. Kanner noted that children with autism displayed an ‘anxiously obsessive desire for the maintenance of sameness’ and further speculated that some of the core autism features might be, at least in part, driven by high levels of anxiety. Since these original observations, numerous clinical and autobiographical accounts demonstrated the negative impact of anxiety in autism. Vivid descriptions of the negative impact of these difficulties were provided by Joliffe, Landsdown and Robinson (1992), with one description stating:

‘reality to an autistic person is a confusing, interacting mass of events, people, places, sounds and sights. There seem to be no clear boundaries, order or meaning to anything. A large part of my life is spent trying to work out the pattern behind everything. Set routines, times, particular routes and rituals all help to get order into an unbearably chaotic life. Trying to keep everything the same reduces some of the terrible fear.’

The problem of anxiety in autism has been largely neglected until relatively recently despite the clear indications of its pervasiveness and debilitating influence on the lives of Individuals with autism. This is illustrated by the fact that the first systematic review of anxiety in autism was published in 2009 (White, Oswald, Ollendick, & Scahill, 2009). Discussion in this section will focus on the prevalence, moderators and current theories of anxiety in autism, briefly touching on the issue of assessment. Assessment of anxiety in populations with autism will be discussed in more depth in Chapter 4.

1. 2. 1. Prevalence of anxiety in autism

The existing research studies show that individuals with autism exhibit higher levels of anxiety than typically developed individuals (Kim, Szatmari, Bryson, Streiner, & Wilson, 2000; Bellini, 2004; Bradley, Summers, Wood, & Bryson, 2004; Gadow, DeVincent, Pomeroy, & Azizian, 2004; Weisbrot, Gadow, DeVincent, & Pomeroy, 2004; Thede & Coolidge, 2007; Lopata et al., 2010), individuals from other clinical groups, such as Down

Syndrome (Evans, Canavera, Klinepeter, Taga, & Maccubbin, 2005), Conduct Disorder (Green, Gilchrist, & Cox, 2000), Specific Language Impairment (Gillott, Furniss, & Walter, 2001), Williams Syndrome (Rodgers, Riby, Janes, Connolly, & McConachie, 2012a) and non-autism individuals with learning disabilities (Gadow et al., 2004; Gillot & Standen, 2007). Two studies have compared anxiety levels between autism and clinically anxious individuals (Russell & Sofronoff, 2005; Farugia & Hudson, 2006) and in general, found that the two groups showed comparable levels of anxiety. However, Russell and Sofronoff (2005), using the Spence Anxiety Scales (both parent and children version), found that while children with AS (aged 10-13 years) and clinically anxious comparison group had similar anxiety scores on childrens' report, parents of these children reported that ASD children had higher total anxiety scores as well as higher obsessive-compulsive and physical injury fears scores. This study points to the issue of discrepancy between parents' and children's reports of anxiety that will be further discussed in the measurement section.

In addition to comparing the levels of anxiety between autism group and TD controls and/or other clinical groups, several studies have looked at differences between levels of anxiety across different subtypes of autism (Muris, Steerneman, Merckelbach, Holdrinet, & Meesters, 1998; Tonge, Brereton, Gray, & Einfeld, 1999; Kanai, Koyama, Kato, Miyamoto, Osada, & Kurita, 2004; Weisbrot et al., 2005; Pearson et al., 2006; Thede & Coolidge, 2007; Kuusikko et al., 2008; Davis et al., 2011; Matson, Hess, & Boitsjoli, 2010) with mixed findings. In a large study that looked at anxiety in 182 preschool and 301 school children with AS, AD and PDD-NOS, Weisbrot et al. (2005) found no significant differences between 3 autism subgroups, either on parent or child version of Spence anxiety scale. However, in an older sample (6-12 years), children with AS showed significantly higher levels of anxiety than both PDD-NOS and AD children and PDD-NOS children had higher levels when compared with AD children. Three other studies compared levels of anxiety between AS and HFA individuals (Tonge et al. 1999; Thede & Coolidge, 2007; Kuusikko et al., 2008). A study by Tonge et al. (2008) found that AS group had higher levels of anxiety than HFA group. These results were partially supported by the findings of Thede and Coolidge (2007) whose results showed that while AS and HFA individuals had similar levels of overall anxiety the AS group had significantly higher scores for generalized anxiety. However, Kuusikko et al. (2008) found no differences in levels of anxiety between AS and HFA individuals. Four other studies compared PDD-NOS and AD group, with three studies finding higher levels of anxiety in PDD-NOS group (Muris et al., 1998; Davis et al., 2010; Matson et

al., 2010) and one study finding no differences (Pearson et al., 2006). Finally, Kanai et al. (2004) compared 53 HFA children with HFA (mean age= 6 years) and 21 children with AD (mean age: 8.2 years) using CARS-TV questionnaire and reported that individuals with HFA had significantly higher levels of anxiety.

From the brief overview of the literature provided above it is clear that there is an agreement between the studies regarding the fact that anxiety is more prevalent in individuals with autism than in both general population and various clinical groups (apart from the clinically anxious group). However, results of anxiety prevalence studies have been much less consistent and this has been complicated by trying to divide individuals with autism into diagnostic subgroups, using different measures and including different age groups. In one of the first studies that examined the presence of co-occurring anxiety symptoms in autism, Muris et al. (1998) reported that 84.1% of children met criteria for at least one DSM anxiety disorder. Muris and colleagues used the Anxiety Disorders section of the Diagnostic Interview Schedule for Children (DISC; Version 2.3; National Institute of Mental Health, 1992) and their sample included 44 children with autism aged between 2 and 18 years. In contrast, Kim et al. (2000), using the Ontario Child Health Study Questionnaire Revised, found that only 13.6% of 59 children with autism (aged 9-14 years) met the criteria for elevated anxiety. However, most of the studies found the prevalence of anxiety to be between 35 and 55% (Bellini, 2004; Bradely et al., 2004; Gadow et al., 2005; Leyfer et al., 2006; De Bruin, Ferdinand, Meester, De Nijs, & Verheji, 2007; Sukhodolsky et al., 2007; Kuusikko et al., 2008; Simonoff, Pickles, Charman, Chandler, Loucas, & Baird, 2008; Mattila et al., 2010; Rodgers, Glod, Connolly, & McConachie, 2012; Strang et al., 2012). To briefly summarize, studies looking at the prevalence of anxiety subtypes in ASD have been inconsistent but overall, it seems that the most frequent types of anxiety disorders are specific phobias, generalized anxiety disorder, separation anxiety disorder, obsessive-compulsive disorder, and social phobia (Muris et al., 1998; Evans et al., 2005; Gadow et al., 2005; Weisbrot et al., 2005; De Bruin et al., 2006; Gillot et al., 2007; Sukhodolsky et al., 2007; Mazefsk, Kao, & Oswald, 2011).

1. 2. 2. Moderators of anxiety

It has been suggested that in typical population, types of anxiety symptoms change during different stages of development (Warren & Sroufe, 2004). In general, separation anxiety and phobias of animals are prevalent in children between 6 and 9 years of age,

generalized anxiety symptoms and death phobias in children aged 10-13 years, and social anxiety in adolescents aged 14 to 17 years. Several studies have supported this model. For example, Kashani and Orvaschel (1990) found that symptoms related to social anxiety steadily increased between the ages of 8 and 17.

Following these findings from the typically developing population several studies have examined the role of age, developmental level in the expression of anxiety in autism, with mixed findings. Several studies found evidence that higher cognitive ability was associated with higher levels of anxiety (Weisbrot et al., 2005; Lecavalier, 2006; Mazurek & Kanne, 2010). For example, Weisbrot et al. (2005) found that anxious children with autism (n= 44) had higher IQ scores than non-anxious children with autism (n= 62). However, other studies failed to find association between IQ levels and anxiety (Sukhodolsky et al., 2007; White et al., 2009; Mazefsky et al., 2011; Strang et al., 2012).

The findings regarding the relationship between chronological age and anxiety in autism have also been inconsistent, with some studies finding no association between chronological age anxiety (Sukhodolsky et al., 2007; Strang et al., 2012) and other studies suggesting that anxiety severity increases with age (Kuusikko et al., 2008; Davis et al., 2010, Green et al., 2012). It is important to point out that these findings are limited by the fact that very few studies have investigated anxiety in very young children and older adults with Autism. Three recently published studies (Davis et al., 2011; Matson et al., 2010; Green, Ben-Sasson, Soto, & Cater, 2012) have demonstrated that infants with autism and toddlers show more severe anxiety related symptoms than TD controls. Furthermore, Green et al. (2012) assessed anxiety in toddlers with autism at two time points, when they were aged 25-31 months and 12 months later and found significant increase in anxiety between two time points. A recent cross-sectional study by Davis et al. (2011) compared the levels of anxiety in 131 toddlers (aged 17–36 months), children (aged 3–16 years), and adults (aged 20–65 years) with autism and found that anxiety levels increased from toddlerhood to childhood, decreased from childhood to young adulthood, and again increased from young adulthood into older adulthood, was reported. Although the study by Davis and colleagues is necessarily limited by its cross-sectional design and the use of different anxiety measures for different age groups, it does provide an initial insight into how anxiety might change over the life-span in individuals with autism.

1. 2. 3. Relationship between core autism symptomatology and anxiety

Very few studies have examined the relationship between the social and communication difficulties, and anxiety difficulties. This is surprising if we have in mind that research on anxiety in the general population has consistently shown positive association between the impairments in language and communication and levels of anxiety (Beitchman et al., 2001; Pinborough-Zimmerman, Satterfield, Miller, Bilder, Hossain, & McMahon, 2007; Bornstein, Hahn, & Suwalsky, 2013), and considering the fact that the problems in communication and reciprocal social interaction are core features of autism. A study by Sukhodolosky et al. (2007) showed that anxiety levels were correlated with all three core autism symptoms. More precisely, increased levels of anxiety were significantly associated with higher language levels and also significantly, but negatively associated with non-verbal communication. The relationship between repetitive behaviours and anxiety found in this study will be discussed in depth in section 1. 4. Sukhodolsky and colleagues, similarly like Tsai (1996) and Strang et al. (2012) suggested that children with very basic communicative abilities may be less able to communicate their fears and anxieties and that language and communication problems affect expressed, rather than experienced levels of anxiety. In other words, lower levels of anxiety found in children without or with very limited language/communication abilities might be a consequence of children not being able to verbalize their problems rather than a true reflection of lower levels of anxiety in this population.

Studies examining the relationship between overall autism severity (defined as total ADI-R scores) and anxiety have yielded mixed results with Mazurek and Keane (2010) finding a significant negative association between autism severity and anxiety levels and Lopata et al. (2010) reporting no significant association. In summary, mixed findings regarding the relationship between anxiety and core autism symptoms do not yet lend strong support to those theories of anxiety that propose that problems in social functioning are the main contributing factor for the development of anxiety in this population. A brief overview of the current theories of anxiety in autism will be summarized in the section below.

1. 2. 4. Theories of Anxiety in autism

Numerous authors have suggested that the problems in social functioning, a core feature of autism, contribute to the development of anxiety in this population (Wing, 1992; Attwood, 1998; Tantam, 2000; Bellini, 2004; 2006). It has been hypothesized that early

deficits in social functioning can limit the individual's opportunity to interact with and learn from others, interfering with the ability to establish friendships and romantic relationships. Repeated social failures would cause increased emotional pain, leading to increased isolation which would then contribute to the development of anxiety (Wing, 1992). This model does provide plausible explanation for the presence of anxiety in individuals with autism who are cognitively able enough to realize their own problems and failures in the area of social interaction. However, as noted above, anxiety has also been found in younger and less cognitively able individuals with autism who are not likely to be aware of their own social deficits or be bothered by them. Anxiety in this population has been suggested to be a consequence of the constant overreactions to insignificant environmental sensory stimuli (Green & Ben-Sasson, 2010; Pfreiffer, Kinnealey, Reed, & Herzberg, 2005).

It is likely the combination of both social deficits and sensory problems together with other cognitive deficits that puts individuals with autism at the risk for the development of anxiety. Because of problems in understanding of emotional states and intentions of others, together with over-reaction and inability to block-out insignificant environmental sensory stimuli, individuals with autism might experience their world as highly unpredictable, making even the most simple situations and events uncertain and difficult. Support for this hypothesis comes from the recent exploratory study by Ozsivadjian, Knott & Magiatti (2012). Ozsivadjian et al. (2012) found that all five groups of parents of children and adolescents with autism provided fairly similar descriptions of the triggers for anxiety. Ozsivadjian and colleagues classified these triggers into the following categories: change or routines; social or language-related triggers; specific fears or phobias; sensory triggers; triggers relating to obsessions; and triggers relating to high expectations in performance or organization. In reflecting on findings from the literature discussed so far, my proposal would be that what most of these triggers have in common is the element of change and unpredictability. This indeed makes sense in the light of the general anxiety literature that identifies negative reactions when dealing with uncertain and unpredictable situations as one of the key factors in the development of anxiety (McEvoy & Mahoney, 2011).

However, it is important to note that theories briefly summarized here have not been extensively tested and when testing has been attempted, results have been inconsistent. It can be argued that, similarly like with repetitive behaviours, our understanding of anxiety in autism is hindered by the lack of appropriate measures. The section below will very briefly

identify the key issues in the way that anxiety was assessed in the autism studies conducted thus far. These issues will be discussed in more depth in Chapter 4.

1. 2. 5. Main issues in the measurement of anxiety in population with autism

Several authors have suggested that one of the key reasons for the conflicting findings in the literature on anxiety in autism, is the fact that at present, there is a lack of appropriate assessment of anxiety in this population (White et al., 2009; MacNeill, Lopes, & Minnes, 2009; Reaven, 2010; van Steensel et al., 2011). At present, the following six questionnaires and interviews have been designed to assess a broad range of pathologies (including, but not limited to anxiety) in individuals with developmental disabilities: Diagnostic Assessment for the Severely Handicapped-II (DASH-II; Matson, 1995), Autism Comorbidity Interview–Present and Lifetime Version (ACI-PL; Leyfer et al., 2006), Autism Spectrum Disorders–Comorbid for Children (Autism-CC; Matson & Gonzalez, 2007), Baby and Infant Scale for Children with Autistic Traits (BISCUIT; Matson, Boisjoli, & Wilkins, 2007), Behavioural Assessment System for Children-2 (BASC-2; Reynolds & Kamphaus, 2004). These assessment will be reviewed in more detail in Chapter 4 but in general, although some of these measures provide a fairly comprehensive assessment of anxiety related symptoms, their use is limited to either very young children or individuals with a low cognitive level and therefore not appropriate for older and more high functioning individuals with autism.

Currently, there are no measures designed specifically for the assessment of anxiety in autism population. However, it is worth emphasizing the point made by Wood and Gadow (2010) that anxiety in autism should not be considered in isolation, in other words, it is necessary to compare phenomenology of anxiety in autism and general population. In order to do this, it is debatable whether, at this point, we necessarily need a measure developed specifically for autism or is it a more fruitful approach to carefully select a measure that is currently considered as a gold standard in the anxiety assessment in other populations and validate it in population with autism. The use of the same (or slightly modified) assessment across different populations will enable us to first identify common features of anxiety in autism and other populations and then to apply existing knowledge on anxiety disorders to this problem in autism. It is surprising that the majority of studies that examined various aspects of anxiety in autism did not employ measures that are currently considered to be a gold standard in the anxiety assessment in the general population. In fact, most of the studies have used assessments whose psychometric properties remain unexplored in general

population, let alone in autism population. Also, studies that used measures standardized in the general population failed to examine or at least to report their psychometric properties when used with population with autism. The second issue related to the assessment of anxiety in autism is on whose report (child's or caregiver's) to rely on. Individuals with autism are presented with a unique set of difficulties such as impairments in communication, problems with emotional self-awareness (Baron-Cohen et al., 2002; Losh et al., 2006) that might limit their ability to first identify and then articulate anxiety-related issues that they may face. However, this issue remains largely unaddressed in autism anxiety research.

1. 2. 6. Summary

Research conducted so far confirmed the initial observations by Leo Kanner and Hans Asperger about the association between autism and anxiety by demonstrating that anxiety is a very frequent problem in this population. Results of studies looking at the relationship between chronological and developmental age and anxiety levels in population with autism have not been consistent so far. We still do not know whether communication and social problems in autism play a crucial role in the development of anxiety in this population or, as suggested by Tsai (1996), have no or little influence on the actual levels of anxiety but only on the ability of individuals with autism to report anxiety. It is clear that some of the inconsistencies in the research on anxiety in autism are due to the problems of how to best assess anxiety in this population. Two major issues related to the assessment of anxiety are: the choice of appropriate measure and the informant issue. Although the need to design autism specific anxiety measures has been stressed by several authors (White et al., 2009; van Steensel et al., 2012), as discussed above, this thesis will argue that before designing such measure, it is first necessary to use anxiety measures that are considered as a gold standard for the anxiety assessment in general population and examine their psychometric properties in population with autism before opting for an autism specific measure. The issues of anxiety measurement, anxiety prevalence and the relationship between anxiety and chronological age, social and communication difficulties will be explored in Chapter 4. The relationship between anxiety and sensory problems will be addressed in Chapter 6.

1. 3. Sensory Problems in Autism

Initial description of what would currently be considered sensory symptoms date back to Leo Kanner (1943). Fascination with various sensory stimuli such as light reflecting from various surfaces as well as increased sensitivity and extreme

distress due to various sounds, moving objects or covering one's face are just some of the various atypical reactions to sensory stimuli that Kanner observed in 7 of his 11 original cases. Although Kanner himself (Kanner, 1943) rejected sensory hypothesis of autism (Rogers & Ozonoff, 2005), several pioneers in autism research did consider the possibility that sensory problems are behind some of the core autism features (Bergman & Escalona, 1949; Hut, Hutt, Lee & Ounsted, 1964; Ornitz, 1974; Ornitz, Guthrie, & Farley, 1977; Zentall & Zentall, 1983; Lovaas, Newsom, & Hickman, 1987). Kanner's early descriptions were corroborated by the subsequent clinical observations, autobiographical accounts and research studies. For example, in one of the earliest studies that systematically looked at the sensory processing in children with autism, Lorna Wing (1969) found responses of children with autism to sound, visual, tactile and olfactory stimuli to be markedly different from reactions of typically developing children, children with Down syndrome, receptive and expressive aphasia. Wing noted that responses of children with autism to different sensory stimuli resembled those of deaf-blind children.

Strong, unpleasant reactions to sounds, visual and tactile stimulation and other sensory abnormalities are a prominent and recurring theme running throughout autobiographical accounts of individuals with autism as described here (Williams, 1994; O'Neil, 1999; Grandin, 1992; Jones, Quigney, & Huws, 2003).

“The sharp sounds and bright lights were more than enough to overload my senses. My head would feel tight, my stomach would chum and my pulse would run my heart ragged until I found a safety zone.” (Willey)

“I was also very touch sensitive; scratchy petticoats felt like sandpaper ripping off my skin. There is no way a child is going to function in a classroom if his or her underwear feels like it is full of sandpaper.” (Temple Grandin)

“The sensory overload caused by bright lights, fluorescent lights, colours and patterns makes the body feel like being attacked or bombarded.” (Williams)

It is obvious that these problems present significant obstacles in the lives of people with autism. Despite this, sensory issues are currently not part of the diagnostic criteria for ASD (APA, 2000; WHO, 1944) but they are included in the latest version of the DSM-V criteria

(5th ed.; DSM-V; APA, 2013). Inclusion of sensory problems in the latest diagnostic criteria are not at all surprising. As pointed out by Wing, Gould and Gillberg (2011) enough evidence accumulated over the years to support such change. Firstly, the majority of studies have demonstrated that more than 90% of autistic individuals exhibit various sensory problems (Rogers & Ozonoff, 2005; Leekam, Nieto, Libby, Wing, & Gould, 2007; Ben-Sasson, Hen, Fluss, Cermak, Engel-Yeger, & Gal, 2009). Secondly, several studies have found that atypical reactions to sensory stimulation in children as young as 12 months predicted later diagnosis of autism (Osterling & Dawson, 1994; Baranek et al., 1999). For example, Baranek and colleagues (Baranek et al., 1999) found that decreased orienting to visual stimuli, increased mouthing of objects and aversion to touch distinguished 9-12 months infants who were later diagnosed with autism from the ones who did not receive diagnosis. Finally, sensory problems have been found to be stable across the life-span of individuals with autism. In a follow-up study with 105 individuals with autism and adults who were diagnosed as children, Billstedt, Gillberg, & Gillberg (2007) found that all individuals showed sensory problems during original diagnostic assessment and more importantly, that 93% of those individuals continued to have impairing sensory abnormalities as adults. Billstedt and colleagues used the Diagnostic Interview for Social and Communication disorders (DISCO) that provides detailed information on sensory problems during both assessments.

Despite the significant progress in the understanding of sensory problems in autism it is still not clear which sensory problems are specific to autism, how sensory problems are associated with the core autism symptomatology and how sensory problems change with age. This section will review the literature relevant to these topics. Before this, it is important to highlight that the term atypical sensory processing has been used in inconsistent ways throughout the literature. Some authors referred to what is defined as Sensory Discrimination Disorder by the Interdisciplinary Council on Developmental and Learning Disorders (ICDL; 2005; see also Miller, Anzalone, Lane, Cermak, & Osten, 2007), in other words, to the difficulties in “interpreting qualities of sensory stimuli and/or inability to perceive similarities and differences among stimuli” (ICDL, 2005). Other authors have used the term atypical sensory processing to denote the difficulties in responding to sensory input with behaviour that is appropriate to the nature and/or intensity of the sensory stimuli. These difficulties are termed as Sensory Modulation Disorders (ICDL, 2005) and throughout this thesis, the term atypical sensory processing will refer to the Sensory Modulation Disorder. Although Sensory Discrimination Disorder will not be the topic of this thesis, for the sake of

comprehensiveness, a short overview of these difficulties will be provided before moving to the issues of prevalence, specificity and phenomenology of Sensory Modulation Disorders in autism.

1. 3. 1. Sensory Discrimination Disorder

Atypical sensory discrimination is well documented across all modalities in autism. Behavioural and electrophysiological studies showed both enhanced and impaired performance in different areas of visual processing. Higher levels of visual acuity in autism group when compared to TD participants were found by Ashwin, Ashwin, Rhydderch, Howels, & Baron-Cohen (2009). However, several other studies failed to replicate these findings (Bolte et al., 2012; Tavassoli, Latham, Bach, Dakin, & Baron-Cohen, 2011). Results regarding colour discrimination, biological motion perception and visual search seem to be more consistent with several studies reporting impaired colour discrimination in individuals with autism (Franklin, Sowden, Burley, Notman, & Adler, 2008; Heaton, Ludlow, & Roberson, 2008), intact biological motion perception apart from processing emotional expressions (Atkinson, 2009; Murphy, Brady, Fitzgerald, & Troje, 2009; Kaiser, Demolino, Tanaka, & Shiffrar, 2010; Koldewyn, Whitney, & Rivera, 2010) and superior visual search performance (O’Riordan, Plaisted, Driver, & Baron-Cohen, 2001; Jarrold, Gilchrist, & Bender, 2005).

Atypical responses to auditory stimuli are frequently reported in questionnaire studies (Kientz & Dunn, 1997; Talay-Ongan & Wood, 1999; Dunn, Myles, & Orr, 2002; Rogers, Hepburn, & Wehner, 2003; Tomchek & Dunn, 2007, Baker, Lane, Angley, & Young, 2008; Wiggins, Robins, Bakeman, & Adamson, 2009; Hilton, Harper, Kueker, Lang, Abbacchi, Todorov, & LaVesser, 2010, Klintwall et al., 2010). Behavioural studies have consistently found increased pitch perception for both simple and complex tones as well as speech sounds (Bonnel, Mottron, Peretz, Trudel, & Gallun, 2003; Gomot, Belmonte, Bullmore, Bernard, & Baron-Cohen, 2008; Jones et al., 2009). Results regarding intensity discrimination seem less consistent with some studies finding evidence for hyper-acusis (Khalifa et al., 2004) and others finding no differences in intensity discrimination between the autism and TD groups (Jones et al., 2009). Lastly, evidence suggest that Individuals with autism are better at local processing of sounds (Mottron, Peretz, & Menard, 2000; Jarvinen-Pasley & Heaton, 2007).

Several experimental studies have shown that Individuals with autism have higher thresholds for higher (but not lower) frequency and difference placement of tactile stimuli

(Blakemore, Tavassoli, Calo, Thomas, Catmur, Frith, & Haggard, 2006; Cascio et al., 2008) as well as lower pain thresholds for thermal tactile stimuli (Cascio McGlone, Folger, Tannan, Baranek, Pelphrey, & Essick, 2008).

Systematic research on the olfactory and taste processing in autism has started only recently. Conflicting results have been found regarding the olfactory detection thresholds with Tavassoli and Baron-Cohen (2012a) finding no differences between autism and TD controls and Dudova, Vodicka, Havlovicova, Sedlacek, Urbanek, and Hrdlicka (2011), reporting impaired olfactory detection thresholds in group with autism. Both Tavassoli and Baron-Cohen (2012a) and Dudova et al. (2011) used the standardized Sniffin' Sticks olfaction task. Mixed results have also been found regarding the odour identification (Suzuki, Critchely, Rowe, Howlin, Murphy, 2003; Dudova et al., 2011; May, Brewer, Rinehart, Enticott, Brereton, & Tonge, 2011). Only two studies have examined taste identification in autism finding that Individuals with autism were impaired at taste identification (Bennetto, Kushner, & Hyman, 2007; Tavassoli & Baron-Cohen 2012b). Sensitivity to and fascination with smells have been constantly reported in questionnaire studies (Kientz & Dunn, 1997; Dunn et al., 2002; Rogers et al., 2003; Leekam et al., 2007; Tomchek & Dunn, 2007; Schoen, Miller, Brett-Green, & Nielsen, 2009; Wiggins et al., 2009; Hilton et al., 2010; Kintwall et al., 2010).

1. 3. 2. Sensory Modulation Problems in autism

Sensory modulation disorder (SMD) is defined as the failure to behaviourally respond in a way that is appropriate to the intensity and nature of sensory stimulation. Dunn (1997) proposed four SMD subtypes patterns:

- a. sensory hyper-sensitivity-characterized by distress to innocuous stimuli and passive withdrawal from unpleasant situations;
- b. sensation avoiding-also characterized by distress but also active avoidance;
- c. low registration (or sensory hypo-responsiveness)-characterized by either complete lack of or delayed response to a stimulus or a higher response threshold (e.g., only responding to a more intense stimuli);
- d. sensation seeking-behaviours that either perpetuate or intensify a sensory experience.

A very similar classification of SMD has been proposed by the Interdisciplinary Council on Developmental and Learning Disorders (ICDL Work Groups, 2005). ICDL

proposed that SMD can be classified into: (a) Sensory Overresponsivity; (b) Sensory Underresponsivity; and (c) Sensory Seeking. As can be seen, the only difference between the two classification systems is that whereas Dunn's model distinguishes between sensory sensitivity and avoidance as two separate (though related) expressions of sensory over-responsiveness (SOR), ICDL classification considers SOR unitary construct.

1. 3. 3. Prevalence of Sensory Modulation Disorders in Autism

Over 20 questionnaire studies have compared sensory responses of individuals with autism to both typically developed individuals and individuals with various disorders. In general, studies have consistently shown that Individuals with autism have more SMD problems when compared to TD individuals. Most of the studies used the Sensory Profile questionnaire. The Sensory Profile (SP; Dunn, 1999) is a 125 item, norm-referenced, parent-report questionnaire which measures a child's/young person's responses to everyday sensory experiences. Parent rate frequency of each items on a 5-point Likert scale ranging from always to never. SP assesses sensory processing across different modalities and can also be used to obtain four SMD patterns (or sensory quadrants in Dunn's terminology). In one of the earliest questionnaire studies, Kientz and Dunn (1997) used the original Sensory Profile to examine sensory processing in 38 children with autism (agerange: 3-13 years) and 64 TD children aged 3-10 years. It was found that 85% of the Sensory Profile items differentiated children with autism from TD children. Children with autism had significantly lower scores (indicating poorer performance) across all six modalities and on both sensory hyper and hypo-sensitivity. Consequent studies that used the Sensory Profile largely replicated results found by Kientz & Dunn in individuals with autism across all ages (Watling, Deitz, & White, 2001; Dunn et al., 2002; Smith Myles et al., 2004; Kern et al., 2007; Ben-Sasson, Cermak, Orsmond, Carter, Kadlec, & Dunn, 2007; Tomchek & Dunn, 2007; Crane, Goddard, & Pring, 2009; Hilton et al., 2010). For example, Crane et al. (2009) found that a group of 18 adults with autism (age range: 18-65 years) had poorer performance in terms of overall Sensory Profile scores as well as on the hypo-sensitivity, sensory sensitivity and sensation avoidance quadrants, but not on the sensation seeking quadrant when compared to 18 age and IQ matched TD controls. In one of the rare studies that compared sensory responses of autism and TD toddlers, Ben-Sasson et al. (2007) found that autism toddlers (age range: 13-33 months) had higher frequency of sensory under-responsiveness and avoidance, and lower frequency of sensory seeking behaviours than CA and MA matched TD controls. Differences

between autism and TD individuals in sensory processing found using Sensory Profile were largely replicated in studies that used other questionnaires/interviews, most notably Diagnostic Interview for Social and Communication Disorders (DISCO; Wing et al., 2002), Sensory Sensitivity Questionnaire (Talay-Ongan & Wood, 2000) and Sensory Experiences Questionnaire (Baranek, David, Poe, Stone, & Watson, 2006). For example, Leekam et al. (2007), using the sensory items from DISCO, found that more than 90% of children with autism had sensory abnormalities in at least one sensory domain.

Relatively few studies have compared the sensory processing of individuals with autism with other non-typical populations. In a study that compared children with autism and ADHD, Ermer and Dunn (1998) found that ADHD children had higher incidence of sensory seeking and inattention/distractibility than children with autism who in turn had more hyper-sensitive reactions across different modalities and also scored higher on the fine motor/perceptual difficulties factor that is indicative of hypo-sensitivity. Rogers et al. (2003) using the Short Sensory Profile found that while children with autism had more sensory symptoms (especially increased sensitivity to tactile and taste/smell stimuli) than both DD and TD groups, they were not significantly different when compared to the children with Fragile X syndrome. Several other studies compared sensory processing of Individuals with autism and DD individuals and in general, individuals with autism were found to have more sensory problems than DD individuals (Baranek, David, Poe, Stone, & Watson, 2006; Leekam et al., 2007; Wiggins et al., 2009; Watson, Patten, Baranek, Poe, Boyd, Freuler, & Lorenzi, 2011). A very important question when comparing sensory problems in autism with both TD and various clinical groups is whether there is a particular type of sensory problems that is specific to the group with autism. Discussion below will consider findings regarding the specificity of sensory hyper-, hypo-sensitivity and sensory seeking separately.

1. 3. 3. 1. Sensory hyper-sensitivity

Evidence regarding the specificity of sensory hyper-sensitivity in individuals with autism have been somewhat inconsistent but in general, results seem to suggest that hyper-sensitivity is not specific to the population with autism. Several studies reported a higher frequency of sensory hyper-sensitivity in children with autism when compared to TD children (Baranek et al., 2006; Dunn, Smith-Myles, & Orr, 2002), however, other studies failed to find evidence that sensory hyper-sensitivity distinguished children with autism from either TD children (Ermer & Dunn, 1998; Watling et al., 2001), from children with various

developmental delays (Baranek et al., 2006) or individuals with ADHD (Ermer & Dunn, 1998). For example, using the Sensory Experiences Questionnaire, Baranek et al. found that although 56% of children with autism aged 5-80 months showed extreme sensory hyper-sensitive behaviours, these behaviours did not distinguish them from CA matched DD children. It has been suggested by several authors (e. g. Rogers & Ozonoff, 2005) that sensory hyper-sensitivity is a consequence of lower developmental level rather than specific to autism. However, it is important to point out that hyper-sensitivity is found widely outside of learning disability populations, for example in individuals with anxiety (Hofmann & Bitran, 2007; Farrow & Coulthard, 2012), Schizophrenia (Brown, Cromwell, Filion, Dunn, & Tollefson, 2002) and as pointed out above, ADHD (Ermer & Dunn, 1998) .

1. 3. 3. 2. Sensory hypo-sensitivity

Sensory hypo-sensitivity has been suggested to be distinctive of children with autism (Baranek et al., 2006; Rogers & Ozonoff, 2005). Findings from several studies that showed that individuals with autism exhibit a significantly higher frequency of hypo-sensitivity behaviours when compared to both CA matched TD individuals (Baranek et al., 2006; Dunn et al., 2002; Watling et al., 2001) and either TD or other clinical groups matched on mental age (MA) (Baranek et al., 2006) seem to lend support to the claim that sensory hypo-sensitivity is specific to autism. For example, Baranek, et al., (2006) found that 63% of children with autism showed sensory hypo-sensitivity and that a pattern that differentiated autism from both TD and DD groups was hypo-sensitivity to both social and non-social stimuli. However, several studies failed to find a support for hypo-sensitivity as distinguishing factor between individuals with autism and CA matched TD children, children with ADHD or children with Fragile X syndrome (Ermer & Dunn, 1998; Rogers et al., 2003). Furthermore, sensory hypo-sensitivity has also been found in clinical disorders without the co-morbid learning disability, for example in Schizophrenia (Brown et al., 2002)

1. 3. 3. 3. Sensory Seeking

Results regarding sensory seeking behaviours seem to be the least consistent. While two studies failed to find the difference in sensory seeking behaviours between autism and TD groups, in both toddlers (Dunn, 2002) and adults (Crane et al., 2009), other studies found increased frequency of sensory seeking in children with autism aged 3-6 years (Watling et al., 2001) and in individuals with autism aged 3-43 years (Kern et al., 2006). All these studies used Sensory Profile. Studies that compared the group with autism with other clinical groups

also reported inconsistent results. For example, Ermer & Dunn (1998) found that the group with autism showed significantly lower frequency of sensory seeking behaviours than the children with ADHD. On the other hand, a study by Watson et al. (2011) found that toddlers with autism showed significantly more sensation seeking behaviours than CA matched DD children.

As can be seen from the brief review of the literature, it is difficult to draw conclusions regarding the question of which, if any, type of sensory problems differentiates individuals with autism from both general population and other clinical groups. Hyper-responsiveness does not seem to be specific to autism as it can be seen in various other disorders and as suggested by Rogers and Ozonoff (2005) it may well be a consequence of lower developmental level rather than any specific pathology. However, as pointed out, sensory hyper-sensitivity can be found in several disorders that are not characterized by low developmental level, for example anxiety. It also seems that hypo-responsiveness might be more prevalent in the group with autism although, as reviewed above, the results are inconsistent. It is not possible to draw any conclusion regarding the specificity of sensory seeking behaviours in autism.

Previous discussion regarding the specificity of particular type of sensory problems in the population with autism usually does not take into the consideration the fact that several studies have found that individuals with autism tended to show a mixed type of sensory responsiveness, in other words an individual would show atypical scores across all Sensory Profile quadrants i. e. sensory hyper-, hypo-sensitivity, seeking and avoidance (Baranek et al., 2006; Ben-Sasson et al., 2007; Tomchek & Dunn, 2007). It is possible that, like repetitive behaviours, it is the frequency of sensory modulation disorders rather than their systematic form that distinguishes individuals with autism from both TD individuals and other clinical groups.

The simultaneous presence of both sensory hyper- and hypo-responsiveness in autism has not been properly looked into so far. One line of research seems to suggest that a subgroup of individuals with autism shows hyper-responsiveness to environmental stimuli from infancy and toddlerhood. This state of permanent heightened responsiveness would lead to chronic stress with alteration in functioning of hypothalamic-pituitary-adrenal axis and its hormones, change in cytokines metabolism and change in the autonomic nervous system functioning. These changes, although generally adaptive in the short term, if persist over a longer period of time lead to neurotoxic effects to limbic system and permanent alterations in

endocrine system functioning. This model is supported by findings from the research on the relationship between anxiety and depression where it has been proposed that anxiety and a state of sensory hypersensitivity over time may precede a state of depression and hyposensitive responses to stimuli in the environment (Markram & Markram, 2010; McEwen, 1998; McEwen & Wingfield, 2010; Ganzel, & Morris, 2011).

Other researchers have suggested more circular relationship between sensory hyper- and hypo-sensitivity (Lane, Young, Baker, & Angley, 2010; Lane, Dennis, & Geraghty, 2011). In other words, it has been hypothesized that individuals with autism have problems in finding and maintaining levels of arousal in “the middle ground between hyper- and hyposensitivity“ (Pfeiffer et al., 2005). Furthermore, it has also been suggested that sensory seeking and sensory avoidance behaviours might represent an attempt to modulate the current state of arousal, in other words, sensory seeking might represent a soothing mechanism when an individual is in the state of over-arousal, and for raising stimulation in cases of under-arousal; and sensory avoidance might represent a mechanism for coping with the state of over-arousal and consequent over-stimulation (Lane et al., 2011). The two accounts described above will be explored in Chapter 5.

At this point it is important to highlight the fact that although studies were very consistent in terms of the measure used-most studies using either the full or abbreviated Sensory Profile, there was a wide variability between the studies in terms of the characteristics of the group with autism with some studies being limited to older, higher functioning individuals and other studies including individuals with a wide age range and abilities. This, together with the fact that in some cases the group with autisms were matched with control groups on chronological age, and in other on developmental age necessarily limits generalizability of the findings. Indeed, it has been suggested that sensory problems can change over time (Rogers & Ozonoff, 2005; Ben-Sasson et al., 2009) and when considering sensory problems, it is necessary to take into account chronological age and developmental level. Before defining the questions that this thesis will try to answer, in the following section I will provide a brief review of the literature examining the association between sensory problems and chronological and developmental age as well as a small body of literature that looked at the relationship between core autism features and sensory problems.

1. 3. 4. The relationship between sensory modulation problems, chronological age and developmental level

The results of the studies that examined occurrence of sensory problems across development have been very inconsistent. A cross-sectional study by Kern et al. (2006) reported that sensory problems were less frequent at later ages in their sample of individuals with autism (age range 3-56 years), that older individuals with autism became more similar to TD controls in the way that they respond to sensory stimuli. The only exception was tactile hyper-sensitivity that did not improve over time. However, other studies did not find evidence for the improvement in sensory problems over time (Talay-Ongan & Wood, 2000; Leekam et al., 2007). Using the Sensory Sensitivity Questionnaire with a group of 30 children with autism age between 4 and 14 years, Talay-Ongan and Wood (2000) found that sensory problems were more common in a subgroup of 10 to 14 year olds than in a subgroup of 6 to 9 year olds who in turn had more problems than 4 to 5 year olds. Leekam et al. (2007) found the evidence that, with the exception of visual symptoms, other sensory problems persisted over time supporting numerous autobiographical accounts which pointed out that sensory problems were an on-going problem regardless of the age of individual (Jones et al., 2003). Results from a relatively recent meta-analysis by Ben-Sasson et al. (2009) which summarized results from 14 studies suggest a non-linear course of sensory problems in autism with an increase in total sensory problems, sensory hyper-sensitivity and sensory seeking up to ages 6-9 and a decrease after the age of 9. Meta-analysis failed to find evidence for a consistent course of sensory hypo-sensitive behaviours.

As has been pointed out in the section on the development of repetitive behaviours, although in typical population developmental age parallels chronological age, this is not necessarily the case in autism, and differences in the developmental level might have contributed to the mixed findings regarding the relationship between chronological age and sensory problems. However, results regarding the influence of cognitive level on the expression of sensory problems have also been very inconsistent. Rogers et al. (2003) failed to find an association between developmental level and any of the Sensory Profile scores. Similar findings were reported by several other studies (Kientz & Dunn, 1997; Baranek et al., 2006; Leekam et al., 2007; Baker et al., 2008; Lane et al., 2010). On the other hand, Crane et al. (2009) found that IQ level was negatively associated with three of four sensory profile quadrants-sensory hypo- and hyper-sensitivity and avoidance but not with sensory seeking behaviours.

1. 3. 5. Sensory Problems and Core Autism Symptoms

The majority of research on sensory problems in individuals with autism has focused on comparing the frequency/severity of sensory problems between individuals with autism with both TD individuals and DD individuals and relatively few studies have examined the relationship between sensory problems and core autism symptoms. Several studies have reported associations between overall autism severity and the presence of more sensory problems (Adamson, O'Hare, & Graham, 2006; Kern et al., 2006; Ben-Sasson, Cermak, Orsmond, Tager-Flusberg, Carter, & Kadlec, 2008). For example, using the CARS (Schopler et al. 1994) as a measure of autism severity, Kern et al. (2006) found that total Sensory Profile scores correlated with autism severity in children with autism. It is interesting to note that Kern and colleagues did not find correlations between sensory problems and autism severity in adolescents and adults.

However, from looking at the overall autism severity, it is difficult to interpret the relative contribution of individual core symptoms to the significant correlations found in the previously mentioned studies. A detailed review on the relationship between repetitive behaviours and sensory problems will be provided in Section 1. 4. of this chapter. In this section I will focus on the studies that have examined the relationship between sensory problems and other two core autism features. Only four studies have examined the association between social/communicative deficits and sensory problems (Liss et al., 2006; Hilton, Graver, & LaVesser, 2007; Hilton et al., 2010; Watson et al., 2011). Hilton et al. (2007) compared the levels of sensory problems assessed by the Sensory profile between a group of individuals with autism with Social Responsiveness Scale (SRS; Constantino & Gruber, 2005) score in the severe range of functioning and a group of individuals with SRS score that suggested milder impairments. It was found that the individuals with autism with higher SRS scores had significantly more problems on all 4 Sensory Profile quadrants and that sensory hyper-sensitivity and avoidance had strong negative correlations with SRS total scores while sensation seeking and hypo-responsiveness were moderately negatively correlated. Similar findings were reported by Watson et al. (2011), apart from the fact that they did not find significant correlations between social/communication problems and sensory hyper-sensitivity. Finally, Liss, Saulnier, Fein, & Kinsbourne (2006) found positive correlations between both hypo-responsiveness and sensory seeking, and social and communication symptom severity in children and adults with autism spectrum disorders. Hyper-responsiveness did not correlate with either social or communication symptoms.

1. 3. 6. Neurobiology of Sensory Problems in Autism

Despite the prevalence and clinical importance of sensory problems in autism, there is a relative paucity of studies exploring the neurobiological basis of these problems. In one of the few functional magnetic resonance imaging (fMRI) studies on the processing of non-social sensory stimuli in autism, Gomot, Belmonte, Bullmore, Bernard and Baron-Cohen (2008) found that children with autism (N= 12, mean age= 13.5 years, SD= 1.6) had higher activity in the right prefrontal-premotor and the left inferior parietal regions when presented with novel auditory stimuli than typically developing children (N= 12, age and IQ matched to ASD children). Interestingly, there were no group differences in the primary auditory cortex activity. On a behavioural level, children with autism were faster than TD children in their ability to detect novel auditory stimuli. Gomot and colleagues suggested that although there were no differences in terms of initial registration of novel auditory stimuli (at the level of primary auditory cortex), these stimuli were differently processed (attended) by higher prefrontal regions. fMRI evidence of hyperactivity to auditory stimuli are also supported by electrophysiological findings (Gomot, Giard, Adrien, Barthelemy, & Bruneau, 2002; Gomot, Blanc, Clery, Roux, Barthelemy, & Bruneau, 2011; but see Ceponiene et al., 2003; Lepisto et al., 2006). For example, Gomot et al. (2011) used the mismatched negativity (MMN) and P3a event related potentials (ERP) to explore the neural basis of novelty auditory detection in 27 children with autism (age range: 5-11 years) and 27 gender, age and IQ matched TD children. It was found that ASD children showed significantly shorter MMN latency and greater P3a amplitude when compared to TD children. In a very recent study Green and colleagues (Green et al., 2013) used fMRI to explore brain responses of 25 individuals with ASD (mean age= 13.13 years, SD= 2.29) and 25 matched TD controls to mildly aversive sensory stimuli (presented in visual and auditory modality). It was found that ASD group showed increased responses in the primary visual and auditory cortex, thalamus, hippocampus, amygdala and orbitofrontal cortex suggesting that sensory problems might be the results of impairments in both bottom-up as well as in top-down processing.

In addition to the neuroimaging methods reviewed above, another approach to exploring the biological basis of sensory problems is the use of non-invasive measures of activity of sympathetic and parasympathetic branches of autonomic nervous system. An influential paper by Rogers and Ozonoff (2005) provided a systematic assessment of the literature on physiological responses to sensory stimuli in individuals with ASD and despite inconsistencies in the literature (for example Bernal and Miller (1970) and van Engeland

(1984) reported reduced activity and Stevens and Gruzelier (1984) increased activity of sympathetic nervous system) concluded that ASD individuals are characterized by hypo-activity to sensory stimuli. It is important to emphasise that physiological studies reviewed by Rogers and Ozonoff were published before 2000 (and majority in 1970s and 80s) and were limited in terms of both the equipment used and by the fact that some of the studies were published prior to the DSM-III-R classification which raises some concerns about the reliability of the diagnosis. However, in recent years several studies have explored the profile of autonomic nervous system responses to sensory stimuli in ASD individuals and these studies will be summarized below.

In a study that explored the activity of sympathetic and parasympathetic systems in 59 children with ASD (mean age= 92 months, SD= 13 months) and 29 typically developing children (mean age= 98 months; SD= 15 months), Schaaf, Benevides, Leiby and Sendeck (2013) used the Sensory Challenging Protocol to present sensory stimuli. The Sensory Challenging Protocol (McIntosh, Miller, & Shuy, 1999) consists of 3 minute baseline condition followed by the presentation of 48 sensory stimuli (8 stimuli in each of the primary sensory domains) with each stimulus lasting 3 seconds. The stimuli are divided with variable 12-17 seconds inter-stimulus interval. Respiratory sinus arrhythmia (RSA), naturally occurring heart rate variability during respiration, was used as an index of parasympathetic system activity and pre-ejection period (PEP), the interval from the beginning of electrical stimulation of the ventricles to the opening of the aortic valve, was used as an index of sympathetic system activity. While TD participants exhibited a decrease in RSA in response to challenging stimuli, suggesting intact functioning of the “vagal brake” (Porges, 2007), this pattern was not evident in children with ASD. This suggests that children with ASD had problems in utilizing the activity of parasympathetic system in regulating their responses to sensory stimulation. Although there children with ASD did not differ from TD children in terms of their sympathetic activity, PEPs were lower in ASD children (with high effect sizes for auditory, olfactory and movement domains of the Sensory Challenging protocol) suggesting higher activity of the sympathetic system in ASD children. Woodard et al. (2012) compared the heart rate reactivity to sensory stimulation between 8 children with ASD and 8 TD children (both groups had the age range between 2 and 3.15 years). The sensory protocol consisted of the presentation of visual, auditory, olfactory, tactile and taste stimuli, 22 in total. Stimulus presentation was counterbalanced (with the exception of the taste stimuli which was always presented at the end of the protocol) with 30s rest interval between each

stimulus presentation. Heart rate responses were significantly higher in children with ASD during both baseline and across sensory stimuli presentations suggesting sympathetic stimuli hyper-activity.

Unlike Schaaf et al. (2013) and Woodard et al. (2012) who found evidence of increased autonomic nervous system reactivity to sensory stimuli, Schoen, Miller, Brett-Green and Nielsen (2009) reported evidence for under-reactivity of ANS system. They found that skin conductance responses during the Sensory Challenging Protocol were lower in ASD children (N= 40; mean age= 9.3 years, SD= 2.74) when compared to both typically developing children (N= 33, mean age= 8.1 years, SD= 2.44) and children with Sensory Modulation Disorder (N= 31, mean age= 8 years, SD= 1.93).

Inconsistent findings highlighted above can be explained by the results from the study by Schoen, Miller, Brett-Green and Hepburn (2008) that suggested the existence of two subgroups of ASD children (total N= 40, age range: 5-15 years) based on their pattern of skin conductance responses during the sensory stimulation. One group of children was characterized by low skin conductance levels amplitude and variability (slower latency and faster habituation) and the other with high skin conductance levels amplitude and more SCR variability (faster latencies and slower habituation). These findings were in line with previous findings from Hirstein, Iversen and Rachandran (2001) who identified that around 30% of their ASD participants showed low baseline skin conductance and low reactivity to stimulation while the rest of their sample had high baseline arousal levels and were hyperactive to sensory stimuli.

In summary, despite the strong evidence (reviewed in previous subsections) suggesting that majority of ASD individuals exhibit some form of sensory problems, neurophysiological basis of these problems are currently not clear. What is clear from the brief overview of the literature is that future work will need to develop more carefully designed, ecologically valid experimental sensory protocols. Furthermore, more dimensional approach to ASD combined with exploration of neurophysiological basis of sensory problems across other conditions will be necessary in order to gain better understanding of biological basis of sensory problems. Some of the future directions will be discussed in chapter 10 of this thesis.

1. 3. 7. Summary

As can be seen from the short review provided above, one consistent finding from the literature on sensory problems in autism is that sensory atypicalities are a very frequent problem in this population. However, it is still not clear which of these problems is the most pervasive problem in autism, what is the relationship between sensory modulation problems and chronological age, developmental level, and core autism features. Also, previous research has not addressed the question of what is the mutual relationship between different sensory modulation problems in autism. Having all this in mind, Chapter 5 of this thesis will aim to provide a detailed description of sensory processing in a sample of children and adolescents with autism. It will also examine the mutual relationship between sensory hyper-, hypo-sensitivity, sensory avoidance and sensory seeking types of sensory responsiveness. The role of chronological age, language level and core autism symptomatology will be examined. Also, the influence of different sensory modulation difficulties on adaptive abilities will be examined.

1. 4. Repetitive behaviours, sensory problems and anxiety in autism

'I have suggested that the bizarre and repetitive motor output may actually be a compensatory activity which helps the autistic child to make sense out of sensation.'

(Ornitz, 1974, p203).

The potential role of repetitive behaviours as a way of controlling sensory unpredictability and warding off anxiety was hinted in the original descriptions of Leo Kanner (Kanner, 1943). He noted that “the child’s behaviour is governed by an anxiously obsessive desire for the maintenance of sameness...changes of a pattern, of the order in which every day acts are carried out, can drive him to despair...The dread of change and incompleteness seems to be a major factor in the explanation of the monotonous repetitiousness and the resulting limitations in the variety of spontaneous activity.” More detailed account of the role of repetitive behaviours as a means for providing internally-controlled mechanisms for maintaining a homeostatic state of arousal was developed by Hutt, Hutt, Lee, and Ounsted (1964). Hutt and colleagues suggested that over-arousal and hyper-sensitivity to sensory stimulation led to the development of stereotypical motor behaviours. This hypothesis was based on several lines of evidence, in particular a series of studies using

EEG which suggested stereotypical pattern of behaviours provided a means of avoiding sensory stimulation.

A more comprehensive account of the relationship between sensory problems, arousal and repetitive behaviours was provided by Ornitz and Ritvo (1968) and later by Kinsbourne (1980). Ornitz and Ritvo suggested that the problems in sensory integration and maintenance of optimal levels of arousal were the primary symptoms of autism which led to the development of repetitive behaviours and problems in language, social and other abnormalities which were considered as secondary symptoms. Both Ornitz and Ritvo as well as Kinsbourne suggested that individuals with ASD, due to brainstem abnormalities, were constantly fluctuating between the states of over-excitation and over-inhibition and suggested that repetitive movements could be compensatory strategies for both of these states rather than compensatory strategies for just one or the other. These authors hypothesized that the individual with autism is likely to experience sensory problems as distressing and that due to this the individual may subconsciously search for ways to calm down and reduce the unpredictability by insisting on rigid patterns of routines.

Such hypotheses about the role of repetitive behaviours as a coping mechanism for dealing with problems in regulating and maintaining optimal levels of arousal and consequent hyper/under-responsivity and anxiety, have been supported by findings from the studies in typical population. For example, motor stereotypies such as leg swinging, rocking, repetitive finger movements and nail biting are often seen in typically developed individuals in the situations of boredom/understimulating environment with a function of increasing stimulation (Mason, 1991) and in demanding situations where they have been hypothesized to serve to block excessive stimulation (Wehmeyer, 1989). A potential link between stereotyped movements and sensory processing is further supported by the fact that these behaviours are observed in individuals with hearing and visual impairments (Troster, Brambring, & Beelmann, 1991, Gal, Dyck & Passmore, 2002; 2010). Troster et al. (1991) used the Bielefeld Parents' Questionnaire for Blind and Sighted Infants and Preschoolers to examine the frequency and duration of stereotypic behaviours in 85 blind children aged 10-72 months and found that all children exhibited at least one type of motor stereotypy with body rocking and eye poking being the most prevalent. They also found that heightened arousal and monotony were the typical situations when stereotypies occurred.

It has been hypothesized that ritualistic and compulsive behaviours in early childhood have emotion regulation role and serve to ward off anxiety (Gesell et al., 1974; Evans et al.,

2005). The link between childhood rituals and anxiety are very interesting in the light of the fact that some of these rituals are very similar to OCD symptoms which have also been hypothesized to be associated with anxiety (Evans et al., 1997; Mataix-Cols, & Rosario-Campos, & Leckman, 2005).

However, despite early theories suggesting the relationship between repetitive behaviours, sensory problems and anxiety in individuals with autism, and above mentioned findings from the typical development and other conditions where repetitive behaviours are a prominent or even a diagnostic feature, this relationship has not been thoroughly examined in the autism literature. Systematic review of the literature conducted for this thesis (with January 2013 as the last date of search) has identified 17 studies that examined the relationship between repetitive behaviours, sensory problems and anxiety. The relationship between repetitive behaviours and sensory problems was explored in 8 studies, between repetitive behaviours and anxiety in 5 studies and between anxiety and sensory problems also in 4 studies. At the time of writing this thesis, no published studies have examined the three way relationship between repetitive behaviours, sensory problems and anxiety. Studies identified in the literature search will be briefly reviewed below and described in more detail in Chapter 6.

1. 4. 1. Repetitive behaviours and sensory problems

As stated above, 8 published studies have addressed the question of the potential relationship between repetitive behaviours and sensory problems. Three studies (Gabriels et al., 2008; Chen, Rodgers, & McConachie, 2008; Boyd, McBee, Holtzclaw, Baranek, & Bodfish, 2009) have explored the relationship between sensory problems and repetitive behaviours (RRBs were treated as a unitary category) and in general, found that higher levels of repetitive behaviours were associated with more sensory problems. For example, Gabriels et al. (2008) found a positive relationship between repetitive behaviours and sensory processing problems in a sample of 70 children with autism, aged 3-19.7 years. This relationship stayed significant after controlling for IQ. Gabriels and colleagues also identified a subgroup of participants with autism that had high rates of both sensory processing problems, repetitive behaviours and high rates of co-morbid psychiatric problems, and did not differ from other participants in terms of chronological age and IQ. Despite providing support for the hypothesis that higher levels of total repetitive behaviours are associated with more sensory problems, the study by Gabriels et al. (together with studies by Boyd et al. 2009 and

Chen et al. 2008) is limited in the sense that it did not examine whether different subtypes of repetitive behaviours are differently associated with different sensory behavioural patterns. This limitation was addressed by Baranek, Foster and Berkson (1997), Boyd et al. (2010), Gal et al. (2002) and Gal et al. (2010). However, results of these studies have been somewhat conflicting with some studies suggesting that all subtypes of repetitive behaviours were associated with sensory hyper-sensitivity but not sensory hypo-sensitivity or sensory seeking (Boyd et al., 2010), other studies finding that hyper-responsiveness was associated only with the insistence on sameness behaviours but not motor stereotypies (Baranek et al., 1997) and that hypo- rather than hyper-sensitivity was associated with motor behaviours (Gal et al., 2010). It is important to stress the fact that these studies used different measures for assessing both repetitive behaviours and sensory problems and differed in terms of sample characteristics which further complicates interpretation of findings.

1. 4. 2. Repetitive Behaviours and Anxiety

Studies that examined the relationship between repetitive behaviours and anxiety have consistently found that higher levels of anxiety were associated with elevated levels of repetitive behaviours (Sukhodolsky et al., 2008; Rodgers, Riby, Janes, Connolly, & McConachie, 2012; Rodgers et al., 2012b). For example, Rodgers and colleagues (2012) used the RBQ to compare the levels of repetitive behaviours between children with autism who met cut-off criteria for elevated anxiety on SCAS-P and children with autism who did not meet the criteria. Children in high anxiety group had higher total repetitive behaviours, sameness behaviours/circumscribed interests, and sensory-motor behaviours than children in the low anxiety group.

1. 4. 3. Sensory Problems and Anxiety

The relationship between sensory problems, more specifically sensory hyper-sensitivity and anxiety have been found in general population (Aron & Aron, 1997) and it is not surprising that this relationship has been consistently replicated in population with autism (Green et al., 2012; Ben-Sasson et al., 2008; Mazurek et al., 2013; Pfeiffer et al., 2005). For example, Pfeiffer et al. (2005) examined the relationships between sensory problems and the affective disorders in a group of 50 children and adolescents aged 6-17 years diagnosed with Asperger's syndrome by using the Sensory Profile and the Revised Children's Manifest Anxiety Scale Adapted-Parent's Version (RCMAS; Reynolds & Richmond, 2000). They

found a relationship between sensory hypersensitivity, avoidance and anxiety for the entire group as well as for younger (N= 30; age range: 6-10 years) and older (N= 20; age range: 11-17 years) children. There was also a significant relationship between the sensory hyposensitivity and symptoms of depression for the whole group and in the older age group but not in the younger group.

1. 4. 4. Summary

Results of the studies reviewed above seem to provide some support for the arousal theory of repetitive behaviours. More specifically, studies have found that repetitive behaviours were associated with sensory modulation problems, e. g. either hyper- or hyposensitivity. Furthermore, several studies have found an association between repetitive behaviours and anxiety. Finally, anxiety was found to be associated with sensory hypersensitivity in all studies that explored the relationship between anxiety and sensory processing. However, none of the studies have looked at the three-way relationship between repetitive behaviours, sensory problems and anxiety. Chapter 6 of this thesis will examine the inter-relationship between these behaviours. Furthermore, this chapter will examine our proposal (Leekam Prior, & Uljarevic, 2011) that different subtypes of repetitive behaviours are differently associated with anxiety and sensory problems

Chapter 2: General method Section

2.1 Background

This chapter provides details of the participants and measures for this thesis. Both primary and secondary datasets were used. The primary data set was based on the data collected by the author for the *Children's and Parents' Behaviour Study*, a research project carried out in the local region (South Wales). For this project, parents completed a number of questionnaires about their children. The majority of questionnaires related to children's behaviour. These included questionnaires selected to study repetitive behaviours, sensory processing and anxiety. As discussed in Chapters 7-9, parents also completed questionnaires about themselves. Measures relevant to parents included questionnaires on anxiety, coping and other parents' variables. In order to carry out more extensive analysis of data specifically on repetitive behaviours, it was possible to access two additional secondary datasets. These are detailed in the sections 2.4 and 2.5. These secondary datasets gave the opportunity for new analyses to be conducted for the first time to answer specific questions raised by the thesis.

The first section of this chapter will provide details on the measurement selection process and the detailed description of the measures used in this thesis. The second section will provide a description of the recruitment process and primary as well as secondary datasets used in this thesis.

2.2. Measures

As described in the thesis statement and in Chapter 1, this thesis had two main aims: (1) to explore the relationship between repetitive behaviours, sensory problems and anxiety in children and adolescents with autism; and (2) to explore the prevalence of anxiety in parents of children with autism and the key factors associated with their anxiety problems. Such a broad scope dictated the choice of measures. The measures had to provide detailed assessment of behaviours as heterogeneous as repetitive behaviours and sensory modulation problems in children and concepts as complex as coping strategies and intolerance of uncertainty in parents. Also, it was necessary to collect data on levels of anxiety, autism severity, language levels in children and data on anxiety levels, sensory processing, autism-related traits and levels of support for parents. It is clear that, as mentioned, measures had to provide detailed and valid information on the above mentioned behaviours and traits but at

the same time, due to the scope of behaviours and traits examined, every measure had to be economical in terms of time and effort on the part of parents and children because, in addition to being concerned with obtaining quality data that would allow the thesis to answer the questions that were set-up at the beginning, equal concern was applied to not overburdening participants. Having this in mind, the decision was to opt for the use of questionnaires as the source of information for this thesis. Questionnaires are the primary source of information in the literature on anxiety, coping, autistic traits, and intolerance of uncertainty in parents. However, in terms of repetitive behaviours and sensory modulation problems, there are plenty of experimental protocols and observational approaches as well as autobiographical accounts that allow researchers to collect data on the phenomenon in question. Although for the pragmatic reasons, questionnaires were the most feasible method for gathering relevant information on the behaviours of interest, the use of questionnaires was also justifiable at a theoretical level.

In terms of repetitive behaviours, the goal of this thesis was to examine the structure of this heterogeneous category of behaviours which is not possible to do by using observational measurements. More precisely, laboratory-based observations allow for a detailed assessment and analysis of repetitive motor behaviours, either with or without objects, however, insistence on sameness types of behaviours are difficult to observe in laboratory settings and it has been suggested that these behaviours are better captured by parents' reports (Leekam et al., 2011; Honey et al., 2012). Also, the use of questionnaire measures of repetitive behaviours provides information on behaviours across time and across a range of situations rather than just information on the behaviours in a particular setting.

In terms of sensory problems, it is clear that the use of carefully designed experimental paradigms coupled with electrophysiological assessments is important in elucidating mechanisms responsible for the atypicalities in the sensory processing observed in this population. However, the main aim of the first part of this thesis was to examine the relationship between repetitive behaviours, anxiety and sensory modulation problems in the population of children with autism, more precisely, to examine an earlier hypothesis by Leekam, Prior and Uljarevic (2011) that different types of repetitive behaviours are differently related to anxiety and sensory problems. Since this hypothesis has not been examined in the literature before, it was decided that this relationship should be first examined by using well validated questionnaire measures that provide detailed information on each of the problems and phenomena of interest. As will be discussed in the general

discussion chapter, it is planned to follow up these findings with experimental work. However, for this thesis, it was necessary to establish this relationship using measures that collect information on the presentation of these behaviours and problems across different contexts and situations rather than in one specific and not necessarily ecologically valid context.

2. 2. 1. Selection of the questionnaires

Parallel with the systematic search of literature on the repetitive behaviours, sensory modulation problems and anxiety in children with autism, and the literature on the anxiety, and coping in parents of children with autism but also on anxiety and related phenomena in general, all the papers were screened for the measures that were used. The following section (2. 2. 2.) will provide an overview of the questionnaires that were used for the studies presented in this thesis. Before describing the questionnaire chosen to assess behaviour/trait/phenomenon of interest, a short description of the selection process will be presented. In general, a similar set of criteria was used when choosing every measure: a.) that it was previously used in the population of children and adolescents with autism (or population of parents/or relevant research are-coping and anxiety in general population); b.) that it was validated in the relevant population; c.) that it was not time consuming yet providing comprehensive assessment of the behaviours/trait/phenomena of interest. It is important to note here that the selection process of the repetitive behaviours measurement is described in a detailed manner in Chapter 3 and the selection process of the anxiety measure is described in Chapter 4. Currently, there is only one questionnaire that measures intolerance of uncertainty-the Intolerance of Uncertainty Scale (IUS; Freeston et al., 1994; Buhr & Dugas, 2002; Carleton, Norton, & Asmundson, 2007) and therefore the selection process for this measure will not be described.

2. 2. 2. Description of questionnaires

2. 2. 2. 1. Questionnaires related to children's behaviours

(a) Repetitive Behaviours Measure

Selection process: as noted above, a detailed description of the selection process is provided in chapter 3, section 3. 2.

The Repetitive Behaviour Questionnaire-2 (RBQ-2; Leekam et al., 2007) is a 20 item parental questionnaire designed to record repetitive behaviours in both children with autism and in typical development. Details on the design of the RBQ-2 will be provided in Chapter 3. Parents are asked to rate the frequency with which their child has engaged in a range of RRBs over the last month. The RBQ-2 has previously been used in a population of typically developed children (Leekam et al., 2007; Arnott et al., 2010). In an exploratory factor analysis conducted on a sample of 590 typically developing 2-year olds, Leekam and colleagues identified that both 4 factor and 2 factor solutions provided good fit for the data. A four-factor model accounted for 51% of the variance with the following 4 factors: repetitive motor movements, rigidity/adherence to routine, preoccupations with restricted patterns of interest and unusual sensory interests. A two-factor model accounted for 39% of the variance with the following 2 factors: the Motor/Sensory and Rigidity/routines/preoccupation with restricted patterns of interests. It has been found that RBQ-2 has good psychometric properties in a population of 15 month old (Arnott et al., 2010) and 24 month old (Leekam et al., 2007) typically developing children. Internal consistency in the 15 month old sample was: .85 for the total RBQ-2 score; .81 for motor/sensory and .71 for rigid/routines/preoccupations subscales (2 Factor solution); .82 for repetitive motor movements, .74 for rigidity, .64 for preoccupations with restricted patterns of interest and .51 for unusual sensory interest subscale (4 factor solution). In the 24 month sample, internal consistency of RBQ-2 was: .85 for the total RBQ-2 score; .80 for repetitive motor movements, .75 for rigidity, .72 for preoccupations with restricted patterns of interest and .66 for unusual sensory interest subscale (4 factor solution).

(b) Sensory Processing Measure

Selection process: articles identified through the initial systematic literature search on the sensory processing in autism were screened for the measures used. The following sensory processing measures were identified: the Sensory Profile (SP; Dunn, 1999), the Short Sensory Profile (SSP; Dunn, 1999), the Sensory Experiences Questionnaire (SEQ; Baranek, Favid, Poe, Stone, & Watson, 2006), the Sensory Sensitivity Questionnaire-Revised (Talay-Ongan & Wood, 2000), the Diagnostic Interview for Social and Communication Disorders (DISCO; Wing et al., 2002; Leekam et al., 2007), and the Sensory Processing Measure (SPM; Glennon, Miller-Kuhaneck, Henry, Parham, & Ecker, 2007). The following selection criteria were applied: (a.) that a measure was previously used and validated in the population of

children and adolescents with autism and validated in this population; (b.) that it was appropriate for a wide age range that was used in this thesis; (c.) that it was not time consuming but that it provided a comprehensive assessment of the sensory processing in children and adolescents with autism. More precisely the instrument had to provide an assessment of sensory processing across different sensory modalities and also provide an assessment of sensory modulation problems as defined by the Interdisciplinary Council on Developmental and Learning Disorders (ICDL, 2005) i. e. sensory hyper-, hypo-sensitivity and sensory seeking; (d.) that the instrument was norm referenced. The evaluation of the identified instruments against the set criteria is presented in the table 2. 1. As can be seen, although the Sensory Profile can be considered to be a time-consuming instrument (125 items), it was the only questionnaire that met other criteria and therefore it was chosen for this thesis. It is important to note that since studies that are included in this PhD started in June 2010, it was not possible to consider the inclusion of questionnaires that were published later. One such questionnaire is for example the Glasgow Sensory Questionnaire (Robertson & Simmons, 2013). This questionnaire provides information on sensory processing across different modalities and also provides sensory modulation scores, however, the norms have not been published yet.

Table 2. 1. Selection of the sensory processing measures

	Used & validated in Population with autism	Wide age range	Not time consuming	Comprehensive assessment	Norm referenced
Sensory Profile (SP; Dunn, 1999)	✓	✓	X	✓	✓
Short Sensory Profile (SSP; Dunn, 1999)	✓	✓	✓	X	✓
Sensory Experiences Questionnaire (SEQ; Baranek, Favid et al., 2006)	✓	X	✓	✓	X
Sensory Sensitivity Questionnaire- Revised (Talay- Ongan & Wood, 2000)	✓	X	✓	X	X
DISCO (Leekam et al., 2007)	✓	✓	X	✓	X
Sensory Processing Measure (SPM; Glennon et al., 2007)	✓X	X	✓	X	✓

The Sensory Profile (SP; Dunn, 1999). SP is a 125 item, norm-referenced, parental questionnaire. SP is divided into three main sections: sensory processing section (further subdivided into 6 subsections that assess sensory processing across different modalities e. g. auditory, touch processing), modulation section (further subdivided into 5 subsections) and behavioural and emotional section (further subdivided into three subsections reflecting emotional and behavioural responses to sensory stimuli). Dunn (1997; 1999) also conducted PCA and identified the following 10 factors: Sensory Seeking, Emotionally Reactive, Low Endurance/Tone, Oral Sensitivity, Inattention/Distractibility, Poor Registration (or hypo-sensitivity), Sensory Sensitivity, Sedentary, Fine Motor/Perceptual, and Other.

Also, Dunn proposed a model which suggests that the pattern of individual's sensory responsiveness can be classified based on their neurological threshold and behavioural responses to incoming stimuli. The neurological thresholds continuum ranges from high (when high intensity stimuli is necessary in order to respond), to low (when low intense stimuli provokes a response). Based on the interaction between neurological thresholds and behavioural responses, Dunn's model classified the pattern of individual's sensory responsiveness into four quadrants: (1) *Sensation Seeking*; (2) *Sensation Avoiding*; (3) *Sensation Sensitivity*; and (4) *Low Registration* (or hypo-sensitivity).

(c) Anxiety Measure

Selection process: as noted above, a detailed description of the selection process is provided in the chapter 4, section 4. 1. 1.

The Spence Anxiety Scale-Child Version (SCAS-C) is a self-report questionnaire designed to measure anxiety symptoms in children and adolescents. It is a 44-item questionnaire and the frequency of each item is rated on a 4-point Likert scale (from 1= never to 4= always). The SCAS-C provides a total anxiety score and scores for the following 6 anxiety subscales: panic attacks and agoraphobia (9 items), separation anxiety (6 items), physical injury fears (5 items), social phobia (6 items), obsessive-compulsive (6 items), and generalized anxiety disorder (6 items). Factor analysis by Spence (1997) confirmed a 6 factor structure of SCAS-C. Subsequent studies (Spence, 1998; Spence et al., 2001; Essau et al., 2002) provided further support for proposed subscales, however, 6 factors did not receive support from studies that used the SCAS-C translations (the German translation: Essau, Sakano, Ishikawa, & Sasagawa, 2004; the Chinese: Essau, Leung, Conrardt, Cheung, &

Wong, 2008; the Japanese: Essau et al., 2004). SCAS-C has been shown to have good psychometric properties in the general population. Internal consistency is .92 for the total scale score and ranges from .60 to .82 (Spence, 1997). Six months test-retest reliability was found to be acceptable for both total scale and subscales (Spence, 1998).

The Spence Anxiety Scale-Parent Version (SCAS-P) consists of 38 items that correspond to the items contained in the child version. For each item, parents indicate the frequency of particular anxiety symptom on a 4-point scale (from 0= never to 3= always). SCAS-P provides total anxiety scores as well as 6 subscales scores that match subscales from the SCAS-C. SCAS-P has been shown to have good psychometric properties (Nauta et al., 2004).

The Spence Preschool Anxiety Scale (SPAS) is a parent-completed questionnaire. It provides total anxiety scores and scores for the following anxiety subscales: separation anxiety, physical injury fears, social phobia, obsessive compulsive disorder, and generalized anxiety disorder. It also contains an additional open-ended item which relates to the child's experience of a traumatic event meant for clinical practice but not scored. The SPAS has been shown to have good psychometric properties in general population (Spence et al., 2001).

Multidimensional Anxiety Scale for Children (MASC; March et al., 1997): MASC is a 45-item measure of anxiety for children 4–19 years of age. It has both parental and a child version. MASC provides the total anxiety score and also the scores for the following factors: somatic/panic, general anxiety, separation anxiety, social phobia, and school phobia. Internal consistency (Cronbach's alpha) for these four subscales ranged from .74 to .85 (March et al., 1997). It has been shown to have good psychometric properties in non-ASD populations (Wood et al., 2002). However, although it has been used in 6 studies examining anxiety in ASD (Bellini, 2004; 2006; Sze & Wood, 2008; White & Roberson-Nay, 2009; White et al., 2009; Wood et al., 2009) its psychometric properties in this population haven't been evaluated.

(d) Autism Severity Measure

Selection process: due to the nature of the studies for this thesis, only questionnaire measures were considered. Interview based assessments such as the Diagnostic Interview for Social and Communicative Disorders (Wing et al., 2002), the Autism Diagnostic Interview-Revised (Lord, Rutter, & Le Couteur, 1994), the Developmental, Dimensional and Diagnostic Interview (Skuse et al., 2004) and observational measures such as the Autism Diagnostic

Observation Schedule (Lord, et al, 2000) could not be conducted. The following autism screening questionnaires were identified through the systematic search of the literature: the Childhood Autism Rating Scale (Schopler, Reichler, DeVellis, & Daly, 1980), the Autism Spectrum Quotient (AQ; Baron-Cohen, Wheelwright, Skinner, Martin, & Clubley, 2001), the Social Communication Questionnaire (Rutter, Bailey, & Lord, 2003) the Social Responsiveness Questionnaire (Constantino et al., 2004), the Childhood Asperger Syndrome Test (Williams et al., 2005). The same set of selection criteria as described in the section 2. 2. 1. was used with an additional requirement that in addition to providing overall autism severity score, the instrument had to also provide subscores for the diagnostic triad described by DSM-IV and ICD-10 criteria i. e. qualitative impairments in social interaction, communication and the presence of restricted, repetitive and stereotyped patterns of behaviour. The only questionnaire measure meeting the last criteria was the Social Communication Questionnaire (SCQ; Rutter, Bailey, & Lord, 2003)

The Social Communication Questionnaire (SCQ; Rutter et al., 2003). SCQ (formerly Autism Screening Questionnaire, ASQ) is a 40-items, parental questionnaire developed from the Autism Diagnostic Interview-Revised (ADI-R; LeCouteur et al., 1989). SCQ provides scores for the following 3 subscales: problems in the social interaction, language and communication and repetitive behaviours. In the original study, Berument et al. (1999) reported 15 to be an optimal cut-off score for differentiating between individuals with and without autism (sensitivity= .85 and specificity= .75) and between Individuals with autism from individuals with intellectual disability but without autism (sensitivity= .96 and specificity= .67). However, subsequent studies suggested 11 as a more sensitive cut-off score that improves discriminant validity (Allen et al., 2007; Corselo et al., 2007; Lee et al., 2007; Snow & Lecavalier, 2008).

2. 2. 2. 2. Questionnaires related to parents' behaviours and traits

In this section, I report the selection of measures against a set of general criteria.

(a) Anxiety Measure

Selection Process: Only questionnaires designed to specifically assess anxiety (and preferably depression as well) were eligible for consideration due to a more detailed assessment offered by these instruments. Therefore the general instruments such as the

General Health Questionnaire (GHQ; Goldberg et al., 1978) were not eligible. Furthermore, interview assessments such as the Anxiety Disorders Interview Schedule (Brown, DiNardo, & Barlow, 1994; 2001) were also not eligible. The following anxiety questionnaires were identified through the systematic literature search: the Hamilton Anxiety Scale (Hamilton, 1969), the Clinical Anxiety Scale (Snaith, Baugh, Clayden Husain, & Sipple, 1982), the Spielberger State Trait Anxiety Inventory (Spielberger, Gorrusch, Lushene, Vagg, & Jacobs, 1983), the Hospital Anxiety and Depression Scale (Zigmond & Snaith, 1983) and the Beck Anxiety Inventory (Beck, Epstein, Brown, & Steer, 1988). Identified questionnaires were evaluated against the following criteria: (a.) that it was widely used and validated in the general and various clinical populations, across different age groups; (b.) that it was also in a population of parents of children with autism (or at least other neurodevelopmental disorder); (c.) that it was not time consuming and (d.) that it preferably assessed depression in addition to anxiety. As can be seen from Table 2. 2., only the Hospital Anxiety and Depression scale satisfied all of the criteria as none of the other measures offered assessment of depression in addition to anxiety. Although Beck Inventory in addition to anxiety has also a depression inventory version, this is a separate questionnaire whose addition would make the completion of two questionnaires time consuming.

Table 2. 2. Selection of the parents' anxiety measure

	Used & validated in general and clinical populations	Used in population of parents of children with autism	Not time consuming	Assessing depression in addition to anxiety
Hamilton Anxiety Scale (Hamilton, 1969)	✓	✓	✓	X
Clinical Anxiety Scale (Snaith et al., 1982)	✓	X	✓	X
Hospital Anxiety and Depression Scale (Zigmond & Snaith, 1983)	✓	✓	✓	✓
Beck Anxiety Inventory (Beck & Steer, 1993)	✓	✓	✓	X

Hospital Anxiety and Depression Scale (HADS, Zigmond & Snaith, 1983). HADS is a 14-item self-report norm-referenced questionnaire. HADS provides anxiety and depression scores (each of subscales consists of 7 items). The items related to symptoms that might have physical cause (such as weight loss and insomnia) are not included and HADS is considered to be unaffected by potential co-morbid conditions (Snaith & Taylor, 1985). HADS has been shown to have good psychometric properties. (Herman et al., 1997) and to have a good agreement with other self-report measures of psychological distress and with clinical diagnosis (Bjelland, Dahl, Haug, & Neckelmann, 2002). HADS provides cut-off scores for classifying individuals into those who do not exhibit symptoms of anxiety/depression, those who have elevated levels of anxiety/depressions and those with clinically significant levels of anxiety/depression.

(b) Coping Measure

Selection Process: A systematic search of coping instruments that have been used in research on coping in parents of children with autism as well as parents of children with other neurodevelopmental conditions and various chronic neurological and somatic disorders was conducted. The COPE Inventory (Carver, Scheier, & Weintraub, 1989) and the Ways of Coping Checklist-Revised (Vitaliano, Kusso, Carr, Maiuro, & Becker, 1985; Folkman & Lazarus, 1988) were the only two coping instruments that were widely used in the coping research in the above listed populations. The Cope Inventory had an advantage in terms of number of items when compared to the Ways of Coping Checklist revised (28 vs 66 items). However, the Ways of Coping Checklist was chosen as it has been validated more extensively and used much more widely in a general population and in individuals with anxiety and depression.

The Ways of Coping Checklist-Revised (WCC-R; Vitaliano et al., 1985; Folkman & Lazarus, 1988). The WCC-R is a 66 items, self-report questionnaire. The WCC-R provides detailed assessment of escape-avoidance coping referring to individual's efforts to escape/avoid problem by either engaging in wishful thinking and/or active behavioural efforts to escape the problem; and problem-solving coping referring to individual's effort on solving the problem by careful analysis of the problem/situation and generation of possible ways to address/solve the problem/situation). The WCC-R has been used with various non-clinical as well as with clinical populations e. g. with individuals with cancer (Wonghongkul, Moore, Musil, Schneider, & Deimling, 2000; Rosberg, Edgar, Collet, & Fournier, 2002;) multiple

sclerosis (Jean, Paul, & Beatty, 1999; Lundqvist & Ahlström, 2006) Parkinson's disease (Sanders, Dewey, Mullins, Chaney, 2001), fibromyalgia (Cronan, Serber, Walen, & Jaffe, 2002), stroke (Rochette & Desrosiers, 2002; King, Zeldow, Carlson, Feldman, & Philip, 2002), chronic fatigue syndrome (Ax, 1999), spinal cord injury (Winemann, Durand, & McCulloch, 1994; Change, Lee & Lieh-Mak, 2000), muscular dystrophies and postpolio syndrome (Lundqvist & Ahlström, 2006) and their families.

(c) Intolerance of Uncertainty

The construct of intolerance of uncertainty, its relation to anxiety and other psychopathology and relevance to this thesis is discussed in chapters 7-9.

The Intolerance of Uncertainty Scale (IUS; Freeston et al., 1994; Buhr & Dugas, 2002). IUS is a 27-item, self-report questionnaire originally developed by Freeston et al. (1994). Freeston and colleagues originally found that IUS had a 5 factor structure: (1) uncertainty must be avoided, (2) being uncertain negatively influences the person, (3) uncertainties are frustrating, (4) uncertainties are stressful and anxiety provoking, and (5) uncertainty prevents one from acting. However, subsequent studies failed to find a support for the 5 factor structure of IUS. A recent factor analysis of the IUS (Carleton et al., 2007) suggested that reduced version of the scale (consisting of 12 items) had better psychometric properties than the original 27-item scales. McEnvoy and Mahoney (2011) recently reviewed factor analytic studies of the IUS conducted thus far, and found that the Carleton et al. (2007) two factor model had the best properties. Due to these reasons this thesis will use Carleton et al.'s 12 item modification of the Freeston et al. (1994) scale. Several factor analytic studies (Carleton et al., 2007; McEvoy & Mahoney, 2011; Helsen, van den Bussche, Vlaeyen, & Goubert, 2013) found that IU scale consists of two factors-the first termed as Desire for Predictability and the second termed as Uncertainty Paralysis and these factors will be explored in this thesis.

(d) Sensory Processing Measures

Selection Process: There are currently only two questionnaires suitable for assessing sensory processing in adults-the Adult Sensory Profile (ASP; Brown & Dunn, 2002) and the Highly Sensitive Person Scale (HSP; Aron & Aron, 1997). Since sensory processing in parents of children with autism has never been assessed before and also due to the fact that

these two instruments differ in terms of their focus, it was decided that both questionnaires would be used for this thesis.

The Highly Sensitive Person scale (HSP; Aron & Aron, 1997). HSP is a 20 item self-report questionnaire designed to measure sensitivity to various stimuli, situations and contexts. The measure has been found to have excellent psychometric properties (Aron & Aron, 1997; Smolewska, McCabe, & Woody, 2006; Liss et al., 2007). Aron & Aron (1997) originally proposed that the concept of highly sensitive person should be viewed as unidimensional. However, in contrast to Aron and Aron's finding that the scale is unidimensional, three recent factor analyses (Smolewska et al., 2006; Evans & Rothbart, 2008; Liss et al., 2010) found the following three factors: Aesthetic Sensitivity (AES), Low Sensory Threshold (LST), and Ease of Excitation (EOE). It was hypothesized that LST and EOE represent negative aspects of sensory processing while AES represents positive aspects.

Adults Sensory Profile (ASP; Brown & Dunn, 2002). ASP is a 60-item self-report questionnaire. ASP is based on the Sensory Profile and it provides the following 4 quadrant scores: poor sensory registration (or hypo-sensitivity), sensory seeking, sensory sensitivity and sensory avoidance that correspond to quadrant scores provided by Sensory Profile. It has been found to have good psychometric properties.

(e) Empathy Traits Measure (proxy for Broader Autism Phenotype Measure)

Selection Process: rather than using the Autism Spectrum Quotient (AQ; Baron-Cohen et al. 2001), the Social Responsiveness Scale-Adult (SRS-A; Constantino & Todd 2005) or the Broad Autism Phenotype Questionnaire (BAPQ; Hurley et al. 2007) as well as the established measures of the BAP traits, the decision was made to concentrate on the cognitive and affective empathy as proxy for the BAP traits. The reasons for this were twofold. Firstly, impairments in cognitive and affective empathy are considered to be a part of the BAP (Sucksmith, Allison, Baron-Cohen, Chakrabati, & Hoekstra, 2013) and more importantly, they have been previously found to be associated with anxiety in general population. Thus, there were strong theoretical reasons to concentrate on cognitive and affective empathy as features of BAP that might be associated with anxiety in parents. The second reason for concentrating on cognitive and affective empathy rather than using above mentioned measures was a pragmatic one as initial feedback received from parent support groups suggested that parents were not likely to complete classic BAP screening measure and that in some cases it might cause additional stress in parents. The Interpersonal Reactivity

Index (IRI; Davis, 1994) was chosen as a well established measure of various aspects of empathy.

The Interpersonal Reactivity Index (IRI; Davis, 1993). IRI is a 28 item, self-reported questionnaire. IRI provides total scores as well as scores for the following 4 subscales: “Perspective Taking” or Cognitive Theory of Mind, “Empathic Concern” or Affective Theory of Mind, “Personal Distress” (defined as “tendency to experience distress or discomfort in response to extreme distress in others;”) and “Fantasy” (defined as “the tendency to imaginatively transpose oneself into fictional situations”). Higher score in a subscale represents a higher functioning in each aspect of empathy. IRI subscales have been shown to have good psychometric properties (Davis, 1980; Siu & Shek, 2005).

2. 2. 2. 3. Background questionnaire

In addition to the above described questionnaires, a background questionnaire was also used. It collected data on the type of child’s diagnosis, date of the diagnosis and the diagnostic centre that established the diagnosis, the presence of co-morbid conditions in children, type of treatment that children receive for their autism related problems and also about the type of treatment that children receive for co-morbid condition, whether the child has a statement of the special education needs and whether the child receives extra support at school, the level of expressive language of child and whether child uses reciprocal social communication or not. The background questionnaire also collected information on parents: their educational level, nationality and language used at home. The background questionnaire is provided in the appendix 4.

As Expressive Language measure from the Background Questionnaire will be used frequently throughout this thesis, a short discussion on this measure will be provided below.

Parents completed this item as a part of the Background Questionnaire. Please see Table 2.3 for the overview of the Expressive Language measure.

Table 2. 3. Expressive language measure adapted from DISCO

What kind of language does your child use?	Score
No speech or sounds	0
Gives the names of some people or things when asked	1
Spontaneously say names of several familiar objects for some purpose	2
Says phrases not longer than three words	3
Say some longer phrases (longer than 3 words), missing out the small linking words (e.g. “when time go on holiday?”)	4
Talk in spontaneous sentences using small linking words	5
Use past, present and future tenses in complex sentences (e.g. “perhaps I will go out tomorrow if it has stopped raining?”)	6

The Expressive Language measure was taken from the Diagnostic Interview for Social and Communication Disorders (DISCO; Wing et al., 2002; Leekam et al., 2002). The DISCO itself has been validated for the use in research (Wing et al., 2002; Leekam et al., 2002; Kent, Carrington et al., 2013). Although this expressive language subscale has been independently used in published research (Honey et al., 2007) and also in unpublished PhD thesis (Honey, 2007), it has not been validated prior to this thesis. The current work provides an initial validation of the Expressive Language DISCO subscale as this scale was significantly correlated with WASI Verbal IQ ($r = .558$, $p = .047$), WASI full IQ ($r = .589$, $p = .044$), Mullen’s Scale of Early Learning Expressive Language Score ($r = .868$, $p = .005$) and Mullen’s Receptive Scale Score ($r = .787$, $p = .021$) in the subsample of children and adolescents with autism from the primary sample (please see page 83 for detailed description of this sample). However, the sample size of children that were tested using cognitive assessments was small with 12 children tested on WASI and 8 children tested on Mullen’s assessments. An independent published dataset of DISCO data ($N = 88$) was consulted in order to try to validate the language expressive scale used in this thesis. This analysis used the dataset from Kent, Carrington et al. (2013) and this particular analysis is reported in Kent, 2013 (PhD thesis). Using this independent dataset (children of the same age range selected), the expressive language scale was compared with the WASI verbal IQ score. A correlation of $r = .467$, $p = .000$ was found. Although in this thesis I used the expressive language measure alone, this result indicates that it is comparable to other measures (more on the limitations related to cognitive testing will be presented at the end of this chapter). Therefore, despite the

initial validation, the use of this scale will present an important limitation and all analysis where this scale is used are exploratory in nature and results should be considered as preliminary.

2. 3. Recruitment process

2. 3. 1. Ethics

Ethical approval for all the studies using the primary sample (South Wales) was given by the Research Ethics Committee of the School of Psychology, Cardiff University and by two local NHS health boards.

2. 3. 2. Recruitment Procedure

Families were recruited through local schools with specialist autism provision and parent support groups. Recruitment centred on the Cardiff and Vale area of South Wales. The recruitment period continued across a period of two years.

Head teachers of schools and co-ordinators of autism parent support groups were initially contacted about the project via email or post. An email or letter was sent that provided the background of the study, outlined the procedures for data collection and also included a copy of questionnaire booklet that families would receive. Head teachers and parent support group co-ordinators who expressed interest to help with the recruitment for the research project were contacted by telephone to discuss the research further. In cases where schools organized parents' meetings, a talk was given to a mixed group of parents and teachers, outlining the background of the research project. Talks were also given to parent support groups outside of school settings.

After learning more about the study, families were provided with leaflets that, in addition to brief description of the study, contained contact details of the PhD student, and told to contact the PhD student in case they were interested in taking part. Upon contacting, potential participants were again provided with a brief description of what the study involves and in case the participants were still interested in taking part in the study, an initial phone interview was scheduled. During a brief phone interview (approximately 5 minutes per participant), families were screened for eligibility of their child to take part in the study. Inclusion criteria were the following: the child, having received a formal diagnosis of autism by a professional (e.g., psychologists, psychiatrists, neurologists with experience working with individuals with autism), aged from 2 to 17 years, and absence of epilepsy, brain injury, cerebral palsy, any neuro/musculo/skeletal disorder/malformation that would seriously limit

ability to walk without help or a known genetic condition (e.g., Fragile X, Down syndrome). The child's language comprehension was also screened in order to choose the appropriate anxiety measure. After the phone screening, parents were then sent Booklet 1 containing 5 questionnaires. Please see tables 2. 4. for a list of questionnaires included.

Schematic representation of the whole recruitment process and the steps involved in the distribution of questionnaires and phone interviews is summarized in the figure 2. 1. below.

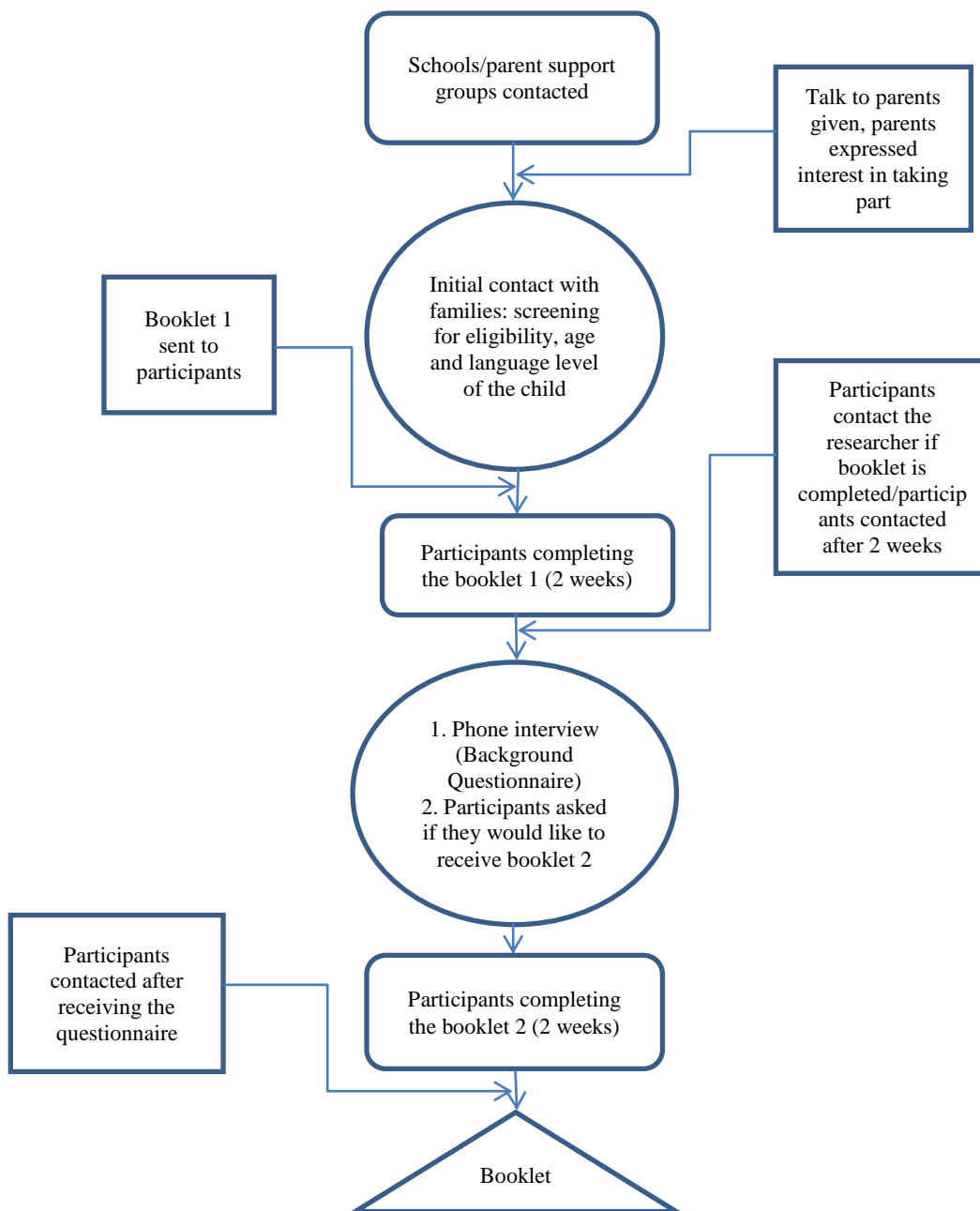


Figure 2. 1. Schematic representation of the recruitment process

Table 2. 4. Questionnaires (about the child) included in the Booklet 1

Behaviour/Trait Measured	Questionnaire (completed by)
Repetitive Behaviours	Repetitive Behaviour Questionnaire-2 (Parent)
Sensory Processing	Sensory Profile (Parent)
Anxiety	Spence Anxiety Scales (Parent and child) Multidimensional Anxiety Scale for Children (Parent)
Autism Severity	Social Communication Questionnaire (Parent)

During their initial phone call, parents were asked to contact the researcher within two weeks after receiving a questionnaire and also they were asked for a permission to be contacted by the researcher if he didn't hear from them. During the second phone call with parents, the researcher went through the background questionnaire with parents (described in the measures subsection) and asked if they would like to receive the booklet 2 containing additional 6 questionnaires. Please see Table 2. 5. for a list of questionnaires included. The second phone interview lasted between 15 and 20 minutes. Booklet 2 was sent to families who agreed to continue with the research.

Table 2. 5. Questionnaires (about parent) included in the Booklet 2

Behaviour/Trait Measured	Questionnaire (completed by)
Anxiety	Hospital Anxiety and Depression Scale (Parent)
Empathy traits	Interpersonal Reactivity Index (Parent)
Coping	Ways of Coping Scale (Parent)
Intolerance of Uncertainty	Intolerance of Uncertainty Scale
Sensory Processing	Adult Sensory Profile Highly Sensitive Person Scale

It is important to note here that this PhD research was a part of a larger research project conducted by the Wales Autism Research Centre. In addition to research questions

that were specific to this PhD, this project also explored various aspects of social and communication difficulties in children and adolescents with autism and also explored repetitive behaviours from a functional analytic perspective and due to this, additional questionnaires that were not used in this PhD project were also collected (9 additional questionnaires). As a result of this, parents had to complete 20 questionnaires in total and a phone interview. This required at least 3 hours of their time and because of the time consuming nature of this project and necessity to conduct an interview over the phone, it was not possible to collect data on-line. It was also felt that because parents had to complete a series of questionnaires about their own anxiety, coping strategies and other, potentially sensitive and stress inducing topics, it was necessary to present details about the study to parents in the form of a talk given by PhD student and allow parents to ask any question/express concerns related to the project. This also precluded online recruitment and data collection.

2. 4. Description of the primary sample (South Wales Sample)

Although parents also provided some data about themselves for this section, the results are predominantly based on their children's behaviours and characteristics. Therefore, the group characteristics presented are about children, as reported by their parents. Sixty three parents of children with DSM-IV-TR diagnoses of ASD returned completed Questionnaire Booklet 1 and 59 returned booklet Questionnaire 2. Data for the Group with autism was primarily provided by biological mothers, although two biological fathers provided data. In this section I will provide descriptive data for the whole sample of children in terms of their chronological age, expressive language levels, co-morbid diagnosis, nationality, their parents' educational level and language spoken at home. Data for all the studies presented in this thesis will be drawn from this sample. Descriptives for the specific relevant samples will be presented in the relevant chapters. Before providing descriptives for the whole sample it is important to note that from the whole sample (N= 63), 59 provided data on RBQ-2 questionnaire and this subsample was used in Chapter 3. As the goal of Chapters 4, 5 and 6 was to examine interrelationship between repetitive behaviours, sensory problems and anxiety, only individuals for whom data were available for all three measures were included in analyses conducted for these 3 chapters. As both the Sensory Profile and the Spence Anxiety Scales are norm-referenced measures, they do not allow for any missing data which reduced the size of sample used in Chapters 4, 5 and 6 to N= 49. Finally, aim of chapters 8

and 9 was to examine frequency and risk factors for anxiety in mothers of children with autism. As the Hospital Anxiety and Depression scale and the Adult Sensory Profile are both norm-referenced instruments, they also do not allow for any missing data and size of the sample for Chapters 8 and 9 was $N=50$ (out of those 50 cases 43 overlapped with sample used in Chapter 4, 5 and 6; this was a result of a combination of missing data from sample used in Chapters 4, 5 and 6 in terms of mother's anxiety and sensory questionnaires and data missing from sample used in Chapters 8 and 9 in terms of children's anxiety scales which resulted in 7 non-overlapping cases).

Diagnosis: Each child had a clinical diagnosis. As this was a questionnaire study and the time commitment for each participant was already high, it was decided not to reconfirm diagnosis for each child by the use of diagnosis tools such as the ADI-R (Lord, Rutter, & LeCouteur, 1994; Lord et al., 2000), ADOS (reference) or DISCO (Leekam et al., 2002; Wing et al., 2002). However, clinicians from two health boards (Cardiff and Vale and Aneurin Bevan), involved in diagnosing children, collaborated on this project. They confirmed the use of DSM-IV-TR diagnosis and the routine use of ADOS (following the ICD-10 criteria) by these services. Parents reported details of their child's diagnosis. 25.4 % of children had a clinical diagnosis of Asperger's Syndrome, 42.4% of Autism, 25.4% of Autism Disorder and 5.1% of High Functioning Autism. Social Communication Scores (SCQ; Rutter et al., 2003) were available for 43 children who had a developmental level in the appropriate range for this scale. The mean score was 27.63 ($SD=5.82$), range was 14-37. Three children scored 14, one scored 16 and 39 scored 21 or above. Several studies (e.g. Eaves, Winger, Ho, & Mickelson, 2006; Allen, Williams, & Hutchins, 2007; Lee, David, Rusyniak, Landa, & Newschaffer, 2007; Wiggins, Bakeman, Adamson, & Robins, 2007) report SCQ scores of 11 or 12 as a valid cut off point to indicate ASD when using this parent completed checklist.

Age and language ability: The mean age of children was 9 years 11 months ($SD=4$ years 4 months; range: 2 years 5 months to 17 years 9 months; 54 males). Expressive language level estimates were based on a language questionnaire item taken from the Diagnostic Interview for Social and Communication Disorders (Wing et al., 2002) used by Honey, Leekam, Turner, & McConachie (2007). Parents were asked to report if their child had no words, single words, 2-3 words, longer phrases, spontaneous sentences or complex sentences with past, present and future tense. 62.7% of parents reported that their child used complex grammatical speech or spontaneous sentences, 16.6% that their child used phrase

speech and 21.7% that their child used single words or no speech. 42.2% of children did not have any co-morbid conditions, 22% had one, 22% had two, 10.2% three and 1.6% had 4 co-morbid conditions.

Parents were asked to indicate if their child had any other health or learning difficulties in addition to autism. Table 2. 6. shows the frequency of specific conditions in this sample of children.

Table 2. 6. Health Difficulties in children

Health Difficulty	Number of children
Sleep Problems	12
Gastro-intestinal problems	11
Asthma	8
Alergies	5
Dyspraxia	5
ADHD	5
OCD	1
Dyslexia	1

Parent education: SES data were not available but data on educational level showed that 32% of parents had postgraduate qualifications, a further 30% had undergraduate or vocational qualifications and 25% did not have post-school qualifications (12.3% missing data).

2. 5. Description of the secondary data sets

2. 5. 1. Secondary Data Set 1

Data set 2 consisted of 61 children with autism. This sample consists of archival data of participants recruited from Cambridge University. The original purpose of this project was to assess the relationship between head circumference and cognitive/behavioural correlates related to autism. Only RBQ-2 data were used from this dataset with the full permission from the first author working on this project Dr Jillian Sullivan and her PhD supervisor Professor Simon Baron-Cohen. The mean age of the sample was 5 years 4 months, (SD= 1 year 2

months; range 2 years 9 months to 8 years 5 months; 56 males). All had a clinical diagnosis of an ASD, established according to ICD-10 criteria.

Data set 2 was used only in the first study presented in this thesis. RBQ-2 data from this sample were merged with RBQ-2 data from South Wales sample in order to increase power for the factor analysis of RBQ-2.

2. 5. 2. Secondary Data Set 2

This data set included longitudinal data from 88 typically developing children whose parents completed RBQ-2 questionnaire at when children had 15, 24 and 72 months. The children were part of the Tees Valley Baby Study (Durham University), an opportunity sampled community sample from the North-East of England, studied prospectively via parent questionnaires, observation and interview techniques. The majority of the children were White and British. SES using Hollingshead's (1975) scale represented the full range of deprived to affluent. Ethical permission was granted from Local Research Ethics committees and University Ethics Committees and parents provided written consent for their child to participate in the study.

Secondary data set 2 was only used in the first study presented in this thesis with the goal of examining developmental changes of repetitive behaviours in a typically developing general population child sample. The data set is used with the full permission of principal investigator Professor Sue Leekam and the researcher who worked on this study.

2. 6. Methodological Limitations of this thesis

This PhD was the first study conducted in the Wales Autism Research Centre and because of this no recruitment register existed and no connections with local schools and parent support groups were established before the commencement of this PhD project. Prior to any data collection it was first necessary to establish connections with both local schools and support groups and this involved series of meetings and talks to both teachers and various support and interest groups, more than 25 in total. As a result, recruitment and data collection process was very slow, and spanned over a period of two years. Although it was initially planned to recruit only from Cardiff area, due to slow response it was necessary to widen the recruitment to the whole of Wales. As described above, it was not possible to recruit and collect data online. This resulted in two significant limitations of this PhD. The first limitation, the consequence of a recruitment process, is a relatively small sample size. Because of the nature of recruitment (participants spread all over Wales), only questionnaire

measures were suitable together with phone interviews. As a consequence of this, it was not possible to conduct cognitive testing on the major portion of the sample (only 12 children were tested on WASI and 8 on Mullen's scale), therefore, there is a lack of a proper index of developmental level in this thesis. Also, as questionnaires were returned by post, there were missing data that rendered norm-referenced questionnaire unusable and further reduced sample size, and although in some cases it was possible to contact parents again regarding the missing data, in majority of cases, due to geographical constraints this was not possible.

Chapter 3: Repetitive Behaviours in Autism and Typical Development

Chapter Plan:

In the first part of this chapter, a brief overview of the literature on repetitive behaviours in both autism and typical development will be provided followed by a summary of the existing instruments for measurement of repetitive behaviours. After this, empirical work is provided for both autism and TD. The first aim is to examine the structure and psychometric properties of the RBQ-2 questionnaire in a population of children and adolescents with autism. The second aim is to examine the relationship between repetitive behaviours, chronological age, other core autism symptoms, expressive language and adaptive behaviours. Finally, the third aim is to examine the developmental trajectory of repetitive behaviours in a sample of typically developing children. The results will be discussed in the light of the existing literature.

3. 1. Introduction

As discussed in more detail in Chapter 1, findings from the literature on the development of repetitive behaviours in autism seem to support the suggestion put forward by Prior & Macmillan (1973) and Turner (1999) that repetitive sensory and motor behaviours can be conceptualized as “lower level” behaviours as they are generally found in younger children with autism and Individuals with autism with lower developmental level (Moore & Goodson, 2003; Fecteau et al. 2003; Esbensen et al., 2009). However, it is important to note that both sensory and motor behaviours continue to be seen in high functioning groups (South, Ozonoff, & McMahon, 2005). The conceptualization of IS behaviours as higher level behaviours is less straightforward as several studies failed to find association between this factor and developmental level. For example, Lam, Bodfish, & Piven (2008) found that insistence on sameness behaviours were not associated with either age nor verbal IQ in a group of 316 Individuals with autism (mean age= 9.02 years). Also, a study by Murphy, Beadle-Brown, Wing, Gould, Shah, and Holmes (2005) found that RRBs characterized by resistance to change did not change over time. Part of the conflicting results might be the fact that studies mentioned previously were cross-sectional. A longitudinal study by Richler, Huerta, Bishop & Lord (2010) used the ADI-R to track longitudinal change in RRBs in children with ASD over a 7 year period (RRBs were assessed when children were 2, 3, 5, and 9 years of age). Results showed that RSM behaviours remained relatively stable over a 7 year

period but RSM behaviours did decrease in children with higher NVIQs. Unlike RSM behaviours, IS behaviours, gradually (moderately) increased over time. In addition to older chronological age, higher IS scores were also associated with milder social and communication impairments. However, Richler et al. (2010) did not include typically developing group in their study and also, they did not conduct an analysis of the predictive effect of early RSM behaviours on higher level IS behaviours at later ages.

It is of crucial importance to consider findings on the change in repetitive behaviours in autism against what we know about the structure and change of repetitive behaviours during typical development. Several studies have used cross-sectional data to examine the developmental change of RRBs in typical development. For example, Evans et al. (1997) examined the developmental trajectory of 'just right' behaviours, and 'repetitive behaviours & insistence on sameness' using Childhood Routines Inventory (CRI; Evans et al., 1997). Evans and colleagues reported that 'just right' behaviours occurred significantly more in children aged 24 to 48 months than in children aged 12 months and 72 months, while 'repetitive behaviours and insistence on sameness' (e. g. preference for the same daily routine) were present to a similar degree as 'just right' behaviour, although these behaviours emerged earlier and had a steeper increase over time. Arnott et al. (2010) using the RBQ-2 found that the most common behaviour type at the age of 15 months were motor and sensory behaviours. Arnott and colleagues also reported that the total RRB score, especially for the motor total scores and sensory subtype was higher at 15 months than that reported for 2-year-olds. However, to date, there has been no published longitudinal study of RRBs in a typical community sample that describes the development of the range of RRBs and their subtypes across multiple age points.

Another issue that has not been addressed in detail is the question of the nature of the relationship between repetitive behaviours and core autism features. Although it is usually assumed that repetitive behaviours are related to the impairments in reciprocal social interaction and deficits in communication, several authors have questioned this assumption (Constantino et al., 2004; Happe, Ronald, & Plomin, 2006; Happe & Ronald, 2008). For example, Constantino et al. (2004), argued that unitary, rather than the three-factor solution provides the best explanation for the clustering of autism symptoms. On the other hand, Happe et al. (2006) and Happe and Ronald (2008) suggested that the association found between the social and non-social impairments is purely coincidental and that these impairments have distinctive genetic etiologies. Studies that examined the relationship

between repetitive behaviours and social/communication impairments have, in general found that higher RRBs scores were associated with more social/communication problems (Charman et al., 2005; Gabriels et al., 2005; Lam, Bodfish, & Piven, 2008; Ray-Subramanian & Weismer, 2012) For example, Lam et al. (2008) used ADI-R in a sample of 316 children with autism (mean age: 9 years) to look at the relationship between different RRBs subtypes and social/communication deficits. They found that both higher repetitive motor behaviours and insistence on sameness were significantly associated with more social/communication problems. However, no association between circumscribed interests and social/communication deficits was found.

To summarize, studies conducted thus far have provided some support for the two factor structure of repetitive behaviours originally proposed by Prior & Macmillan (1975), although several studies have found that 3 or even 5 factors provide a better description of the heterogeneous class of repetitive behaviours. Studies that examined the influence of chronological and developmental age on the expression of repetitive behaviours in autism have provided some support for conceptualizing repetitive sensory-motor behaviours as “low-level” behaviours as these behaviours seem to be more frequent in younger children and individuals of lower developmental level. However, it has also been found that RSM behaviours are frequent in older and high functioning individuals with autism. The results regarding the conceptualization of insistence on sameness behaviours as “higher-level” behaviours have been even more conflicting. At present, it is not clear whether RSM and IS behaviours are different classes of behaviours or whether, RSM behaviours might predate IS behaviours in developmental terms. Longitudinal studies on RRBs in typical development might provide an answer to this question, however, all the studies that looked at these two classes of behaviours have been cross-sectional. Based on this short review of the literature, it is clear that it is necessary to examine how different classes of repetitive behaviours are related to the chronological age and other core autism features in a systematic way and more importantly, to look at how RRBs change over time during the typical development. This chapter will use secondary RRBs data in order to address such questions.

However, as argued in Chapter 1, the type of measure that is used to assess RRBs sets the boundaries for the phenomenon under focus and necessarily influences the conceptualization of RRBs and, before examining any of the above mentioned issues, it is necessary to choose a measure that will allow a detailed and systematic assessment of repetitive behaviours, both in autism and TD population.

3. 1. 1. Measurement of Repetitive Behaviours

As reviewed above and in the Chapter 1, repetitive behaviours are a very heterogeneous group of behaviours that are present in the population with autism but also in other neurodevelopmental and psychiatric conditions, and during early development. Hence, it is necessary that every assessment tool must be comprehensive enough in order to gather information on a wide range of behaviours. Because of the above mentioned reasons, the instrument must also go beyond behaviours which may be exclusive to the Population with autism. This would also help to avoid the circularity of simply re-describing diagnostic criteria. However, it is also important to highlight the fact that including a large number of questions may not be suitable for the inclusion in research projects where participants are asked to complete other measures because of the time constraints. Therefore it is important to strike a balance between the need to provide a detailed and comprehensive assessment of the behaviours in question and the reduction in time associated with completion of the measure. To briefly summarize, a strong measure should be: with sound psychometric properties, allow examination of a wide range of repetitive behaviours, both the ones present in autism and the ones present during typical development and in various other conditions, allow the examination of repetitive behaviours across all ages, and finally, be easily administered and not too time consuming.

Articles that were identified in a systematic search of the literature for Chapter 1 were also screened for the type of questionnaire/interview measure that was used to assess the repetitive behaviours. Twenty eight questionnaire/interviews were initially identified. However, only the instruments that assess at least two behaviours from each of the four categories of repetitive behaviours described in the current diagnostic criteria for ASD are reviewed here. Despite the fact that Maudsley Item Sheet used by Carcani-Rathwell, Rabe-Hasketh, & Santos (2006) met the initial inclusion criteria, this instrument was not evaluated as it was not possible to obtain the full description of the repetitive behaviours part of the instrument from the published literature. In the end, a total of 12 instruments met the initial inclusion criteria. A detailed description of instruments is provided in Appendix 2 and evaluation of instruments is provided below.

3. 1. 1. 1. Evaluation of the reviewed instruments against the previously set criteria

As can be seen from Table 3. 1., RBS/RBS-R questionnaires provide a detailed assessment of various types of repetitive behaviours, however, the time needed for the instrument to be completed might not make it an optimal instrument for the research projects with multiple measures. Similar can be said for DISCO that, as mentioned, is a very detailed assessment interview, however, it is very time consuming. The ADI-R was developed around the DSM-IV criteria for autism which makes it less than a perfect instrument for researching repetitive behaviours across different conditions. Similar can be said about the SCQ that was modelled around ADI-R algorithm items. The CRI and especially Y-BOCS were developed to assess what can be considered as higher order repetitive behaviours and as such do not represent the full spectrum of all types of repetitive behaviours that individuals might show.

Table 3. 1. Questionnaire/Interview Evaluation

Measure	Psychometric Properties	Suitability across		Easily administered/not time consuming
		Ages	Diagnostic groups	
SCQ	✓	X	X	✓
DISCO	✓	✓	✓	X
ADI-R	✓	✓	X	X
Y-BOCS/CY- BOCS	✓	✓	X	X
RBS/RBS-R	✓	✓	✓	X
RBI/RBQ	✓	✓	✓	✓
RBQ-2	✓	✓	✓ (TD)	✓
CRI	✓	✓	X	✓

Only 3 questionnaires (RBI/RBQ/RBQ-2) met all the criteria. I decided to use the RBQ-2 that was developed by our group (Leekam et al., 2007) as this instrument, as mentioned above, provides a detailed assessment of repetitive behaviours, items are applicable across conditions and chronological ages, and since it is a 20 item questionnaire it is not time consuming. In addition, unlike CRI, it covers a wider range of behaviours, while the CRI is more appropriate for the assessment of IS types of repetitive behaviours. Also, the

RBQ-2 is more applicable across different age range and developmental level than RBS-R which is more applicable to younger children and/or individuals of lower developmental levels.

The first objective of this chapter (Study 1) is to explore the developmental trajectory of repetitive behaviours in typical development. The second objective (Study 2) is to assess the factor structure and the psychometric properties of the RBQ-2 in a sample of Individuals with autism with autism spectrum disorders. It is hypothesized that the two or four structure of the RBQ-2 will be confirmed via exploratory factor analysis. The third objective is to examine if repetitive behaviours are moderated by any of the following variables: chronological age, the presence of functional language, communication and social impairments.

3. 2. Study 1 Developmental change in RRBs in typical development

Methods

Subjects

Characteristics of the typically developing children were described in more details in Chapter 2¹. Briefly, parents of 88 children completed RBQ-2 at 3 age points: when their children were approximately 15 months (age range: 14-16 months, mean age: 14.86), 24 months and approximately 72 months (age range: 73-83 months, mean age: 77.4) old.

Measures

The Repetitive Behaviour Questionnaire-2 secondary data were used.

Results

Mean RBQ-2 scores broken down by age are presented in Table 3. 2. Scores for RSM and IS behaviours at both 15 and 24 months have previously been published (Arnott et al., 2010) and are provided here to enable comparison with 6 year old (unpublished) data collected from parents of the same children. RSM and IS subscales were calculated following the procedure used by Leekam et al. (2007) and Arnott et al., 2010).

¹ As described in the Chapter 2, this was a secondary data set 2.

Table 3. 2. Mean RBQ-2 scores at 15, 24 and 72 months

Age	RSM	IS	RBQ-2 Total Score
15 months	1.83 (SD= .43)	1.37 (SD= .32)	1.62 (SD= .30)
24 months	1.52 (SD= .40)	1.54 (SD= .42)	1.55(SD= .33)
72 months	1.31 (SD= .36)	1.39 (SD= .37)	1.40 (SD= .31)

From Table 3. 2. it is clear that RSM and total RRBs steadily decreased with age while IS behaviour increased between 15 and 24 months and then decreased by 72 months (although mean score was still higher than at 15 months).

It is important to note that as most of RBQ-2 subscale scores were skewed, analyses were run with logarithmically transformed data.

A series of paired t-tests were performed in order to examine whether there were significant differences between mean RBQ-2 scores at 15, 24 and 72 months. Mean RSM score at 15 months was significantly higher than at both 24 ($p=.000$; Cohen's $d=.72$) and 72 months ($p=.000$; Cohen's $d=1.31$) and that mean RSM score at 24 months was significantly higher than at 72 months ($p=.000$; Cohen's $d=.55$). Mean IS score at 24 months was higher than at both 15 months ($p=.000$; Cohen's $d=.49$) and at 72 months ($p=.01$; Cohen's $d=.37$). Mean RBQ-2 total score at 72 months was significantly lower than at both 15 ($p=.000$; Cohen's $d=.75$) and at 24 months ($p=.004$; Cohen's $d=.45$). There was no difference between RBQ-2 total scores at 15 and 24 months (Cohen's $d=.22$).

A series of paired T tests was conducted to compare RSM and IS scores at 15, 24 and 72 months². It was found that at 15 months, RSM score was significantly higher than IS score ($t=9.977$, $p=.000$; Cohen's $d=1.21$), at 24 months there were no significant differences between RSM and IS scores ($t=.28$, $p=.779$; Cohen's $d=.05$) nor were there differences at 72 months after adjusting significance level to avoid Type I error ($t=2.085$, $p=.04$; Cohen's $d=.22$).

Mean IS, RSM and total RBQ-2 scores at three times points, broken down by gender are shown in table 3. 3. A series of mixed RRBs type x gender ANOVAs were performed for

² Comparison between IS and RSM scores at 15 months was already performed by Arnott et al. (2010) in a larger sample ($N=139$) from this data set. These comparison are performed here for the sake of comprehensiveness. However, comparison at other ages nor across agers were not performed previously in any of the published papers and that is an original contribution of this thesis using the secondary dataset.

15, 24 and 72 months of age. Although, as can be seen from the table 3. 6, males had higher mean RSM, IS and total RBQ-2 scores at all ages, the only differences that reach statistical significance were for RSM ($p=.014$; Cohen's $d= .52$) at 72 months and total RBQ-2 scores ($p= .013$; Cohen's $d= .55$) at 72 months. As noted above, analyses were run with logarithmically transformed scores.

Table 3. 3. Mean RBQ-2 scores at 15, 24 and 72 months for each gender

Age	RSM			IS			RBQ-2 Total Score		
	Male	Female	P	Male	Female	P	Male	Female	P
	(N= 49)	(N= 39)		(N= 49)	(N= 39)		(N= 49)	(N= 39)	
15 months	1.89 (.47)	1.76 (.38)	.169	1.39 (.37)	1.34 (.26)	.44	1.66 (.32)	1.56(.27)	.135
24 months	1.54 (.44)	1.50 (.36)	.654	1.59 (.44)	1.47 (.38)	.201	1.59 (.37)	1.49 (.28)	.125
72 months	1.39 (.41)	1.21 (.26)	.014	1.44 (.43)	1.33 (.27)	.163	1.47 (.33)	1.31 (.25)	.013

Linear regression analyses were used to examine the predictive effect of RSM and IS behaviours on the same behaviours at later ages. RSM behaviours at 15 months predicted RSM at both 24 months ($F(1, 86)= 47.871$; Adjusted $R^2= .350$; $t= 6.919$, $p= .000$, $\beta= .622$) and at 72 months ($F(1, 86)= 13.185$; Adjusted $R^2= .123$; $t= 3.631$, $p= .000$, $\beta= .362$). When both RSM scores at 15 and 24 months were entered as predictors of RSM scores at 72 months, RSM behaviours at 15 months were no longer significant predictors ($t= .550$, $p= .583$, $\beta= .061$) but at 24 months were ($t= 4.453$, $p= .000$, $\beta= .505$). The whole model accounted for 28% of variance. The same procedure was repeated for IS behaviours. IS behaviours at 15 months were significant predictors of IS behaviours at both 24 ($F(1, 86)= 26.241$; Adjusted $R^2= .225$; $t= 5.123$, $p= .000$, $\beta= .581$) and 72 months ($F(1, 86)= 27.853$; Adjusted $R^2= .236$; $t= 5.278$, $p= .000$, $\beta= .495$). When both IS scores at 15 and 24 months were entered as predictors of IS scores at 72 months, both IS behaviours at 15 months ($t= 3.112$, $p= .003$, $\beta= .309$) and at 24 months ($t= 3.863$, $p= .000$, $\beta= .384$) were significant predictors. The whole model accounted for 34.2% of variance.

Next, a set of regressions was performed to examine the relative influence of RSM and IS behaviours as predictors of later outcome. When both RSM and IS behaviours at 15 months were examined as predictors of IS behaviours at 24 months, only IS behaviours were significant predictors ($t= 4.205$, $p= .000$, $\beta= .427$) while RSM behaviours were not ($t= 1.446$, $p= .152$, $\beta= .147$). The whole model accounted for 30.6% of variance. When RSM and IS

behaviours at both 15 and 24 months were entered as predictors of IS behaviours and 72 months, IS behaviours at both 15 ($t= 2.379$, $p= .02$, $\beta= .242$) and 24 months ($t= 3.239$, $p= .002$, $\beta= .332$) were significant predictors while RSM behaviours at either 15 ($t= 1.611$, $p= .111$, $\beta= .176$) or 24 months ($t= .623$, $p= .535$, $\beta= .070$) were not. The whole model accounted for 36.8% of variance. Similar set of regressions was performed with RSM behaviours as dependent variable. When both RSM and IS behaviours at 15 months were examined as predictors of RSM behaviours at 24 months, only RSM behaviours were significant predictors ($t= 5.823$, $p= .000$, $\beta= .540$) while IS behaviours were not ($t= 1.634$, $p= .106$, $\beta= .152$). The whole model accounted for 36.3% of variance. The whole model accounted for 36.3% of variance. When RSM and IS behaviours at both 15 and 24 months were entered as predictors of IS behaviours and 72 months, only RSM behaviours at 24 months were significant independent predictor ($t= 3.935$, $p= .000$, $\beta= .475$) while RSM behaviours at 12 months ($t= .477$, $p= .635$, $\beta= .056$) not IS behaviours at 15 ($t= .015$, $p= .988$, $\beta= .002$) and 24 months ($t= .731$, $p= .467$, $\beta= .082$) were not with the whole model accounting for 26.9% of variance. These regression analyses suggest that IS and RSM behaviours are relatively independent classes of behaviours in developmental terms.

3. 3. Study 2 Repetitive Behaviours in Autism

Methods

Subjects

Parents of 120 children with Autism Spectrum Disorders (ASD), with ages ranging from 2 years 5 months to 17 years 9 months (mean 7 years 7 months, SD 3 years 10 months) participated as part of their involvement in a research study being carried out in two different parts of the UK (South Wales, $n= 59^3$, and the South East of England, $n = 61^4$) investigating the association between repetitive behaviour and other factors. The South Wales sample was aged 2 years 5 months to 17 years 9 months (mean age= 9 years 11 months, SD 4 years 4 months; 54 males) and South East England sample was aged 2 years 9 months to 8 years 5 months; (mean age= 5 years 4 months, SD 1 year 2 months; 56 males). As described in Chapter 2, all had a community multidisciplinary team assessment leading to a best estimate

³ Primary Data Set, please see Chapter 2 for details

⁴ Secondary Data Set, please see Chapter 2 for details

clinical diagnosis of an ASD (including autism and Asperger syndrome) according to DSM-IV (American Psychiatric Association, 1994) and ICD-10 (World Health Organization, 1993) criteria.

Measures

The RBQ-2 questionnaire was used.

Results

Initial analysis using t-tests revealed that the South Wales and the South East of England samples did not differ in their Total RBQ-2 Score ($M = 1.99$ ($SD = .38$) vs $M = 1.92$ ($SD = .44$); $t(118) = .986$, $p = .33$; Cohen's $d = .19$). Furthermore, although the two groups differed in age, the Total RBQ-2 score was not associated with age ($r = -.018$, $p = .84$).

Factor Analysis

Before conducting a factor analysis, the rate of item endorsement was examined in order to check if any of the items had less than 10% of frequency (in other words, for every item, the 'occasional' to 'marked' response range had to be endorsed by at least 10% or more of the sample). The endorsement rates can be seen in the table 3. 4.

Table 3. 4. Response frequencies for each RBQ-2 item

Does your child:	Never or	One or more	15 or more	Mean (SD)
	Rarely 1 (%)	times daily 2 (%)	times daily (or at least once an hour) 3 (%)	
1. Arrange toys or other items in rows or patterns?	47.5	48.3	4.2	1.57 (.58)
2. Repetitively fiddle with toys or other items?	32.2	32.2	35.6	2.03 (.83)
3. Spin him/herself around and around?	59.3	26.3	14.4	1.56 (.73)
4. Rock backwards and forwards, or side to side, either when sitting or when standing?	58.5	22.9	18.6	1.6 (.79)
5. Pace or move around repetitively?	41.5	28	30.5	1.89 (.85)
6. Make repetitive hand and/or finger	32.2	30.5	37.3	2.05 (.84)

movements?				
7. Have a fascination with specific objects	9.3	35.6	55.1	2.46 (.66)
8. Like to look at objects from particular or unusual angles?	29.7	38.1	32.2	2.02 (.79)
9. Have a special interest in the smell of people or objects?	63.6	15.3	21.2	1.58 (.82)
10. Have a special interest in the feel of different surfaces?	39	33	28	1.89 (.81)
11. Have any special objects he/she likes to carry around?	42.4	27.1	30.5	1.88 (.85)
12. Collect or hoard items of any sort?	42.4	22.9	34.7	1.92 (.88)
13. Insist on things at home remaining the same?	33	39	28	1.95 (.78)
14. Get upset about minor changes to objects	39	37.3	23.7	1.85 (.78)
15. Insist that aspects of daily routine must remain the same?	23.7	32.2	44.1	2.2 (.80)
16. Insist on doing things in a certain way or re-doing things until they are “just right”?	28	32.2	39.8	2.12 (.82)
17. Play the same music, game or video, or read the same book repeatedly?	10.2	37.3	52.5	2.42 (.67)
18. Insist on wearing the same clothes or refuse to wear new clothes?	46.6	33.9	19.5	1.73 (.77)
19. Insist on eating the same foods, or a very small range of foods, at every meal?	30.5	26.3	43.2	2.13 (.85)

Since none of the items had frequency of less than 10%, the next step was to conduct a factor analysis. The principal component analysis (PCA) with varimax rotation was chosen following the same methods used by other researchers working in this field (Evans et al., 1997; Szatmari et al., 2006; Leekam et al., 2007; Honey et al., 2012). Item 20, the non-quantitative response item, was excluded. Initial screening showed that assumptions of non-multicollinearity, sampling adequacy, and factorability were all met (The Kaiser-Meyer-Olkin (KMO) measure verified the sampling adequacy for the analysis, $KMO = .779$. Bartlett's test of sphericity, $\chi^2 (171) = 657.038$, $p < .001$, indicated that correlations were sufficiently large for PCA).

An initial analysis was run to obtain eigenvalues for each component in the data. Five components had eigenvalues over Kaiser's criterion of 1 and in combination explained 60% of the variance. However, it has been pointed out that this method has a tendency to retain more factors than is appropriate (Hayton et al., 2004). The other commonly used rule is to use the Scree Test (Figure 3. 1.).

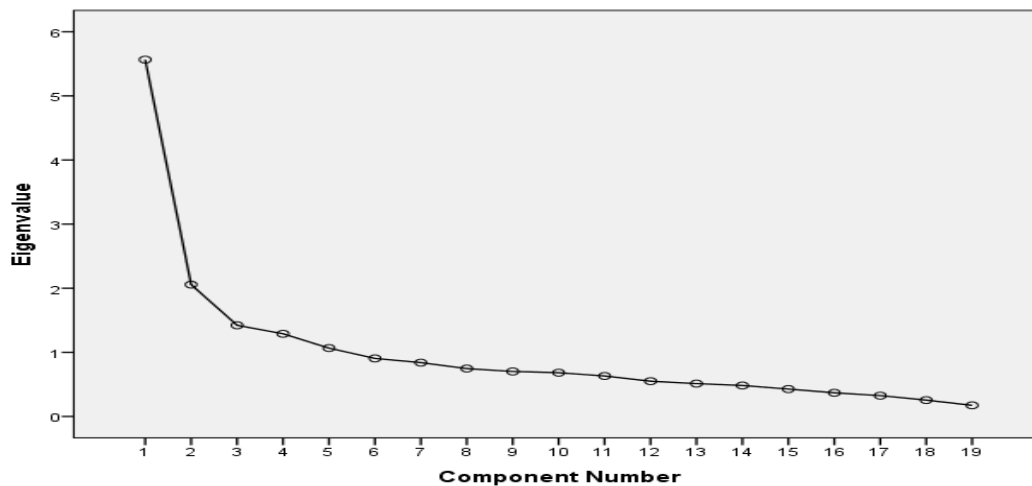


Figure 3. 1. Scree Plot

However, the criticism of basing the decision on a number of factors that should be retained on just the Scree Plot is that the decision is not robust and reproducible, therefore we used Horn's parallel analysis (PA; Horn, 1965) which is based on the Monte Carlo simulation of random production of Eigen values to determine the number of components that should be extracted. The [Parallel Analysis Program](#) was used to run parallel analysis and it was shown that factors 3 onwards have Eigen values less than that from simulations and 2 factor solution should be retained in the final analysis. The PCA with varimax rotation was rerun specifying a two-factor solution. In combination, these two factors accounted for 40.3% of the variance. Factor loadings for items were set at .38 following Comrey & Lee (1992, cited in Tabachik & Fidell, 2001). Table 3. 5. shows the final 2-factor solution (RSM factor accounted for 11.1% and IS 29.2% of the variance).

Table 3. 5. Factor structure of the RBQ-2 questionnaire

Questionnaire items within each factor	Item factor loading
Factor 1: Motor/sensory behaviours – 11.1% variance	
2. Repetitively fiddle with toys etc.	.683
3. Spin self around and around	.639
4. Rock backwards and forwards	.617
5. Pace/move around repetitively	.697
6. Repetitive hand/finger movements	.660
8. Looks at objects from particular/unusual angles	.571
10. Special interest in feel of different surfaces	.565
19. Insists on eating same foods or small range of foods	.447
Factor 2: Insistence on sameness– 29.2% variance	
7. Fascination with specific objects	.39
9. Special interest in smell of people/objects	.484
12. Collect or hoard items of any sort	.579
13. Insists on things (e.g. in house) remaining the same	.777
14. Gets upset about minor changes to objects	.716
15. Insists on aspects of routine remaining the same	.741
16. Insists on doing or re-doing things in a certain way	.829
17. Plays same music, game, video, book repeatedly	.594
18. Insists on wearing same clothes/refuses new clothes	.569

None of the items loaded on both factors. Only Item 1, arranging objects into patterns or rows, and item 11, has special objects that likes to carry around, had loadings of .36 or lower both factors. A further two items loaded in the opposite way than they had for typically developing children. One of these items that loaded on the RSM factor for typically developing children (interest in smell), loaded on the IS factor for the autism sample, and another item (eating same/small range of foods) which loaded onto the IS factor for typically developing children, loaded onto the RSM factor for Children with autism. Apart from these two items all others from the original factor analysis with typical children (Leekam et al., 2007) loaded in the same way for children with autism. There were two additional RBQ-2 items, that had not loaded for the original sample of typically developing children (hoarding objects and fascination with specific objects), both of which loaded for the children with autism onto the IS factor and were retained.

The internal consistency of identified RBQ-2 scores was examined using Cronbach's alpha. Cronbach's alpha showed that internal consistency was high for the Total RBQ-2 scale (Cronbach's alpha $\alpha = .86$ for 20 items and $.86$ for 19 items) and for each RSM and IS subscale separately; RSM $\alpha = .79$; IS $\alpha = .83$). As an additional way of validating the two factor solution, item-total correlations were calculated. The mean item-correlation for Factor 1 was $.63$, $SD = .05$, range $.56$ to $.71$, and for Factor 2 the mean item-correlation was $.67$, $SD = .11$, range $.50$ to $.82$, please see Table 3. 6. for the full list of correlations.

Table 3. 6. Correlated item-total correlations for the two factor of the RBQ-2

RBQ-2 Items	Factors	
	RSM	IS
RSM factor Items		
2. Repetitively fiddle with toys or other items	.677***	.252**
3. Spin self around and around	.627***	.193*
4. Rock backwards and forwards	.606***	.235*
5. Pace/move around repetitively	.709***	.279**
6. Repetitive hand/finger movements	.676***	.291**
8. Looks at objects from particular/unusual angles	.573***	.226*
10. Special interest in feel of different surfaces	.635***	.407***
19. Insists on eating same foods or small range of foods	.563***	.336***
IS Factor Items		
7. Fascination with specific objects	.379***	.503***
12. Collect or hoard items of any sort	.153	.606***
13. Insists on things (e.g. in house) remaining the same	.372***	.779***
14. Gets upset about minor changes to objects	.318***	.716***
15. Insists on aspects of routine remaining the same	.312**	.739***
16. Insists on doing or re-doing things in a certain way	.325***	.820***
17. Plays same music, game, video, book repeatedly	.330***	.610***
18. Insists on wearing same clothes/refuses new clothes	.231*	.584***

Note: * = $p < .05$; ** = $p < .01$; *** = $p < .001$

Relationship between repetitive behaviours and other children's variables

Subjects

Only children from the South Wales sample (N= 59) were used for this analysis.

Measures

In addition to the RBQ-2 questionnaire, parents completed the following measures: The Social Communication Questionnaire (SCQ; Rutter et al., 2003) and DISCO expressive language measure.

Results

Descriptive statistics presented in the table 3. 7.

Table 3. 7. Descriptive statistics

Variables	Mean	SD	Range	Skewness (SE)
Chronological age	9.7 years	4.5	2.2 – 17.8	-.057 (.311)
DISCO Expressive Language	4.25	2.26	0– 6	-1.101 (.306)
RBQ-2 RSM Score	1.92	.57	1– 3	-.110 (.311)
RBQ-2 IS Score	2.04	.47	1– 3	.035 (.311)
SCQ Social Interaction score	10.60	2.46	3– 15	-.728 (.354)
SCQ Communication Score	9.58	2.33	4– 13	-.554(.354)

The relationship between repetitive behaviours and chronological age, SCQ, communication and reciprocal interaction scores were examined using Pearson's correlations. The significance level was set at .01 because of multiple correlations. As can be seen from Table 3. 8. RSM behaviours were significantly negatively associated with chronological age and expressive language scores. It is important to note that association between RSM behaviours with expressive language fell below the significance level after controlling for

chronological age ($p = .118$). IS behaviours were not significantly associated with any of the variables.

Table 3. 8. Relationship between repetitive behaviours and other children's variables

	CA	Expressive Language	SCQ Communication	SCQ Social Interaction
RSM	-.490**	-.533**	.071	.258
IS	.092	.288	.088	.326

3. 4. Discussion

3. 4. 1. Developmental trajectory of RRBs in TD children

The first aim of this chapter was to explore the developmental trajectory of RRBs in a sample of 88 TD children whose parents completed RBQ-2 questionnaire at 3 time points: when children were 15, 24 and 72 months old.

It was found that RSM behaviours scores were significantly higher than IS scores at 15 months. After this, RSM behaviours steadily decreased, and scores at 24 months were significantly lower than scores at 15 months, also scores at 72 months were significantly lower than at both 15 and 24 months. On the other hand, IS behaviours significantly increased between 15 and 24 months and then decreased by 72 months, however, still stayed at higher levels than at 15 months. These results are in line with the findings from cross-sectional studies that have looked at similar types of behaviours in typical development. For example, in a study that examined the developmental trajectory of two components of 'compulsive-like' behaviours: 'just right' behaviours, and 'repetitive behaviours & insistence on sameness', Evans et al. (1997) found that 'just right' behaviours occurred significantly more in children aged 24 to 48 months than in children aged 12 months and 72 months who displayed similar lower levels of these behaviours. 'Repetitive behaviours and insistence on sameness' were found to be present to a similar degree as 'just right' behaviour, although these behaviours emerged earlier and had a steeper increase over time.

However, none of the previous studies have addressed the question of whether RSM behaviours are precursors or an earlier developmental form of the same type of behaviours that is then replaced by the IS behaviours or whether, these two behaviours represent

independent classes of behaviours. When both RSM and IS behaviours at 15 months were concurrently examined as predictors of IS behaviours at later ages, it was found that for IS behaviours at 24 months, IS behaviours at 15 months were significant predictors while RSM behaviours were not. Also, for IS behaviours at 72 months, when RSM and IS behaviours at both 15 and 24 months were entered as predictors IS behaviours at both 15 and 24 months were significant predictors while RSM behaviours at either 15 or 24 months were not. A similar picture emerged when RSM behaviours were considered as dependent variable with only RSM behaviours at 15 months predicting RSM behaviours at 24 months while IS behaviours at 15 months were not significant predictors and only RSM behaviours at 24 months predicting RSM behaviours at 6 years while IS behaviours at neither 15 nor 24 months were significant predictors. These series of regression analysis seem to suggest that IS and RSM behaviours represent relatively independent classes of behaviours. In summary, findings from this thesis indicate that there is an independence of IS and RSM behaviours in terms of their developmental trajectory in a sample of TD children. Theoretical and clinical implications of these findings will be discussed in more detail in Chapter 10.

The second aim of this chapter was to examine the factor structure and psychometric properties of RBQ-2 questionnaire, a measure developed by our group (Leekam et al., 2007). The RBQ-2 was originally designed to provide a brief and comprehensive assessment of RRBs in Population with autism. A distinctive contribution of this instrument is that it has been previously used and validated in a population of 15 and 24 month old TD children.

The results from this chapter indicate that the RBQ-2 provides a highly consistent measure of repetitive behaviours for children with autism aged from 2 to 17 years. A two-factor structure best represented the data in Population with autism. The first factor is best described as a repetitive motor and sensory behaviour (RSM) and the second factor is best described as the insistence on sameness (IS). The structure of RRBs found in the sample of children and adolescents with autism was very similar to young typically developing children by Leekam et al. (2007) and Arnott et al. (2010). Of the 19 items entered in the factor analysis, only a minority did not load in the same way as for the original factor analysis carried out with typically developing children (Leekam et al., 2007). This might be explained in terms of the developmental level of each sample, given that the autism sample was older than the original published sample. For example, two items (carrying around objects and arranging objects) commonly seen in typical toddlers and infants, did not load sufficiently highly to be included for the autism sample, while two other items (hoarding objects and

fascination with specific objects), which are common in older children, loaded only on the factor analysis for Children with autism but not in the original published study. Apart from these four items that might be explained by their developmental appropriateness, only two further items loaded onto different factors for the two samples, although both might be considered to have a mixed interpretation relevant to both sensory and restricted interest element. Item 19 (insists on eating same foods or small range of foods) which loaded onto the IS factor in TD children, loaded onto the RSM factor in the sample of children and adolescents with autism. Although at first surprising, this finding can be explained by recent findings which suggest that restricted choice of foods in the population of children with autism is tied to the sensory properties or sensations caused by different types of food, mostly due to the texture of food (Cermak, Curtin, & Bandini, 2010; Nadon, Ehrmann Feldman, Dunn, & Gisel, 2011). Having this in mind, it was decided to retain the item 19 and accept its new loading. Similarly, item 9 (special interest in the smell of people/objects) loaded onto the RSM factor in TD children and in the sample of children and adolescents with autism used in this thesis, item 9 loaded onto the IS factor 9.

Internal consistency was high for the Total RBQ-2 scale (Cronbach's alpha $\alpha = .86$ for 20 items and $.86$ for 19 items) and for each RSM and IS subscale separately; RSM $\alpha = .79$; IS $\alpha = .83$).

3. 4. 2. Moderators of repetitive behaviours in autism

It has been suggested that RSM behaviours can be conceptualized as “lower-level” behaviours, in other words as behaviours associated with younger age and lower developmental level, while IS behaviours are “higher-level” behaviours, seen in older and higher functioning Individuals with autism (Turner, 1999). Previous research has provided some support for the proposal that RSM behaviours are more frequent in younger and/or lower functioning Individuals with autism (Moore & Goodson, 2003; Fecteau et al., 2003; Esbensen et al., 2009; Richler et al., 2010). However, some studies have failed to support such findings. For example, South et al. (2005) found that severity scores of RSM behaviours were not associated with the age in a group of 21 HFA individuals (age range: 8-20 years) and 19 Individuals with autism (age range: 8-19 years).

The results from this chapter seem to support the conceptualization of RSM behaviours proposed by Turner (1999) as RSM behaviours were significantly negatively correlated with CA and expressive language level. However, the relationship between RSM

behaviours and expressive language was mediated by chronological age. IS behaviours were not associated with neither chronological age nor expressive language levels. The results regarding the IS behaviours are in line with the findings by South et al. (2005), Lam et al. (2008) who also showed a relative independence of IS behaviours of chronological age. However, due to the already noted limitations in term of the measure of developmental level and also due to the cross-sectional nature of the sample used in this thesis these analyses were exploratory in nature and should be considered preliminary at this point.

In addition to exploring the relationship between RRBs, chronological age and expressive language, this chapter has also looked at the relationship between the RRBs and other core autism features. It is important to emphasize the fact that although the issue of the relationship between RRBs and other core features is very important, it has not been explored in much detail. In fact, only a few studies have previously looked at the association between the core autism features in a systematic way. The results presented in this chapter found that RSM behaviours were not significantly associated with the impairments in social interaction and problems in communication subscales of SCQ. These findings are not in line with other studies that found that higher levels of RRBs are strongly associated with more social and communication problems (Charman et al., 2005; Gabriels et al., 2005; Lam et al., 2008; Ray-Subramanian & Weismer, 2012). For example, in a recent study, Ray-Subramanian & Weismer (2012) showed that by the age of 3, RRBs were significantly negatively correlated with both receptive and expressive language, and NVIQ in a sample of 115 children with autism. Furthermore, Ray-Subramanian and Weismer reported that decrease in RRBs was predicted by the increase in the receptive and expressive language from age 2 to 3. The results from this thesis found that IS behaviours were also not associated with communication and social interaction subscales of SCQ. Although IS behaviours were not associated with SCQ Social Interaction scores after correcting significance level due to multiple comparisons, IS behaviours were positively associated with social interaction problems at $p = .02$ level which is a finding in line with findings by Lam et al. (2008) who reported that IS behaviours were significantly associated with more social problems. However, due to the already emphasized limitations, these results should be considered exploratory and preliminary in nature.

3. 4. 3. Limitations

The most important limitation of this chapter is a sample size for both the study that examined the factor analysis of the RBQ-2 and the study that looked at the developmental trajectory of RRBs in typical development. Regarding the factor analysis, although it is considered that 5 participants per each item are sufficient for conducting the exploratory factor analysis, it has been suggested that a figure of 10 participants per item is more optimal (Tabachnik & Fidell, 2001). Sample size for the factor analysis consisted of 120 participants, thus with 6.31 participants per item falling below the optimal criteria set by Tabachnik & Fidell, (2001). Although, as mentioned above, this sample size can be considered as sufficient for the exploratory factor analysis it will be important to replicate the structure of the RBQ-2 found in this study on a larger sample size using the confirmatory factor analysis. Also, although sample size of typically developing children assessed with RBQ-2 at 15 and 24 months was significantly higher, due to the lower response rate and missing data, the final longitudinal sample consisted of 88 TD children whose parents provided RBQ-2 data at all three time points. It is important to consider this sample size when interpreting the results. Another very important limitation, as already pointed out in Chapter 2 and throughout this chapter is the fact that it was not possible to conduct a cognitive assessment for the whole sample and that DISCO items were used as a proxy for the developmental level. Although DISCO items were used previously by Honey (2007) and Honey, Leekam, Turner and McConachie (2007) they were not validated prior to this thesis. As has been reported in Chapter 2, DISCO scores did significantly correlate with both Mullen's Expressive Language scores and WASI Verbal IQ scores, however, the use of DISCO scores present limitation and all the analysis that used these scores should be considered exploratory and results preliminary in nature.

3. 4. 4. Summary

This chapter has provided a validation of the RBQ-2 as a measure of repetitive behaviours in a population of children and adolescents with autism. The two factor structure with RSM and IS factors previously found in TD population was largely replicated. Furthermore, both total score as well as IS and RSM factors were found to have a good to excellent internal consistency. This chapter also examined the relationship between IS and RSM factors and, chronological age and core autism features. It was found that RSM behaviours were negatively associated with lower chronological age, and lower expressive

language levels. IS factor was not associated with any of the moderating variables. Finally, this chapter has provided evidence for relatively independent developmental trajectories of IS and RSM behaviours in a population of 88 TD children.

Chapters 4 and 5: Anxiety and Sensory Problems in children and adolescents with autism

The main goal of the first part of this thesis was to examine the relationship between repetitive behaviours, sensory problems and anxiety in children and adolescents with autism. The hypothesis put forward was that anxiety and sensory problems are the triggers for repetitive behaviours. However, as noted in the introduction section of this thesis, in order to fully understand any association that may exist between these phenomena, it is necessary to first explore and understand these behaviours as separate entities. The previous chapter (Chapter 3) provided a validation and examined the factor structure of the Repetitive Behaviour Questionnaire-2 (RBQ-2) in the population of children and adolescents with autism and also explored the moderators of repetitive behaviours in autism. The following two chapters (Chapters 4 and 5) will explore anxiety and sensory problems in children and adolescents with autism. Limitations and summary will be provided for both chapters together.

Chapter 4: Anxiety in children and adolescents with autism

Chapter Plan:

This chapter is divided into two subsections. In the first subsection, key findings from the literature on anxiety in autism will be very briefly summarized (an in-depth review was provided in chapter 1, section 1. 2.). Second, a review and evaluation of questionnaire measurements that have been used to investigate anxiety in the population with autism will be provided. Then, the psychometric properties of the chosen anxiety questionnaires will be assessed and findings on frequency and moderators of anxiety will be presented. Relevant findings will be discussed in the light of the existing literature.

4. 1. Introduction

The existing research literature has consistently shown that children and adolescents with autism exhibit higher levels of anxiety than both typically developed individuals and individuals from other clinical groups, such as Down Syndrome, Conduct Disorder, and Specific Language Impairment (Evans et al., 2005; White et al., 2009). However, as the in

depth review provided in Chapter 1 showed, the results of studies which examined both prevalence and moderators of anxiety in autism have been very inconsistent.

In terms of the exact prevalence of anxiety in children and adolescents with autism, different studies reported prevalence of anxiety in autism ranging from 11% to 84% (White et al., 2009; MacNeil et al., 2009). Similarly, results regarding the prevalence of specific anxiety disorders have also been mixed, though specific phobias, generalized anxiety disorder and separation anxiety seem to be the most frequent types of anxiety observed in the autism population (Muris et al., 1998; Evans et al., 2005; Mazefsky et al., 2011).

The role of chronological age, developmental level, language and communication in the expression of anxiety in the autism population is not currently clear. For example, while the results of Lecavalier (2006) and Kuusikko et al. (2008) suggest that higher functioning and older individuals with autism show higher levels of anxiety, several other studies found no relationship between age, developmental level and overall levels of anxiety (Meyer et al., 2006; Strang et al., 2012). Davis et al. (2011) reported that autism individuals with better language and communication abilities had higher levels of anxiety, however, results of several other studies did not support these findings (Sukhodolsky et al., 2008; Strang et al., 2012). Furthermore, it has been suggested that language and communication problems, as well as the level of cognitive functioning in general, affect expressed levels rather than experienced levels of anxiety (Tsai, 1996; Strang et al., 2012).

It has been suggested that one of the key reasons for the conflicting findings in the literature on anxiety in ASC is the inconsistent way in which anxiety is assessed across studies (White et al., 2009; MacNeill et al., 2009; Wood et al., 2010; Reaven, 2010; van Steensel et al., 2011). There are several measures that have been developed to assess a broad range of psychiatric conditions (and among them anxiety) in individuals with developmental conditions but currently, no instruments developed specifically for the assessment of anxiety in individuals with autism exist. However, as discussed in Chapter 1, a more important issue than the lack of autism specific anxiety measure is the fact that the majority of studies reviewed here and in Chapter 1 have used the anxiety assessments that have not been validated even in general population. As pointed out by Wood et al. (2009), a strong case can be made that it is first necessary to try and understand anxiety in autism in the context of what is already known about anxiety disorders in general population, rather than looking at anxiety problems in autism in isolation. In order to achieve this, it is of crucial importance to choose a measure or a set of measures that will enable us to look at anxiety across conditions.

The second issue in the anxiety assessment relates to the question of informant or in other words, on whose report (child's or caregiver's) to rely on. This issue is not specific to autism. Indeed, agreement between children's and parents' reports of anxiety has been found to be low to moderate in general population (Achenbach, McConaughy, & Howell, 1987; Krain & Kendall, 2000; Nauta, Scholing, Rapee, Abbott, Spence, & Waters, 2004). Numerous factors such as the age of children (Achenbach et al., 1987), gender (Sourander, Helstela, Helenius, & Piha, 2000), the children's desire to present themselves in a more desirable way as well as the fact that parents themselves might suffer from anxiety leading them to appraise their children's behaviours in more negative way (Achenbach et al., 1987), have all been suggested to contribute to the inconsistencies found in the non-autism population. These inconsistencies may also be present in the population with autism. Indeed, that would not be surprising if we consider that individuals with autism are presented with the impairments in communication and problems with emotional self-awareness (Baron-Cohen, 2002; Silani, Bird, Brindley, Singer, Frith, & Frith, 2008). Also, parents of children with autism show more anxiety themselves when compared to parents of TD children (Mazefsky, Conner, & Oswald, 2010), so the issue of interpreting children's behaviours in a negative way may be even more pronounced than is the case with parents of children without autism.

Having all this in mind, it is obvious that without assessing anxiety in autism population in a standardized, consistent manner, our understanding of this problem will not go further than noting its prevalence. Therefore, one of the main aims of this chapter was to identify the appropriate measure of anxiety in autism through an in-depth literature search and then evaluate its psychometric properties in a population of children and adolescents with ASC

4. 1. 1. Assessment of anxiety in Autism

Articles identified through a systematic search of the literature on anxiety in autism population for the preparation of the review chapter were also screened for the type of anxiety measure that was used. A detailed summary of identified instruments is provided in Appendix 3. Identified anxiety measures were evaluated against the following criteria: (1) measure had to be specifically designed to assess anxiety (because of the comprehensiveness that these instruments provide when compared to instruments that were designed to assess a broad range of pathologies); (2) to provide total anxiety scores and also to assess anxiety subtypes described by DSM-IV and ICD-10 criteria; (3) have both a parent and a child

version; (4) can be used for both young children and adolescents; (5) used widely in the general and/or clinical populations, across different age groups; (6) had published psychometric in general and/or clinical population; (7) have already been used with population with autism before and (8) not lengthy. Evaluation of identified anxiety instruments against these criteria is summarized below.

4. 1. 2. Evaluation of the anxiety instruments

Evaluation of the summarized instruments against the set criteria is presented in the Table 4. 1. As can be seen, each instrument has a unique set of strengths and weaknesses. Only ADIS, Spence Anxiety Scales, SCARED and MASC were specifically created to assess anxiety. Most of the instruments were used widely in the general population and had acceptable psychometric properties. Apart from DASH-II and BISCUIT, all other instruments were applicable to the wide age range. Apart from ADIS and DICS, other instruments were easy to administer in the research context and were not too time consuming. As can be seen from Table 4. 1., the instruments that fit all the criteria were the Spence Anxiety Scales and SCARED. Spence Anxiety Scales was chosen as a primary anxiety measure rather than SCARED because Spence Scales have been used most widely in the population with autism, have been widely used in other populations and have detailed norms for different age groups published. As mentioned in the description of the measure, MASC is considered as a gold standard for the assessment of anxiety in TD population and therefore it was also used.

Table 4. 1. Anxiety Instruments Evaluation

Measure	Specifically designed to assess anxiety	Assesses total anxiety and anxiety subtypes	Both a parent and a child version	Applicable for both young children and adolescents	Widely used in general population/other conditions	Acceptable Psychometric Properties	Easily administered/not time consuming
ADIS	✓	✓	✓	✓	✓	✓	x
Spence	✓	✓	✓	✓	✓	✓	✓
SCARED	✓	✓	✓	✓	✓	✓	✓
ECI-4	x	✓	x	✓	✓	✓	✓
MASC	✓	x	✓	✓	✓	✓	✓
DISC	x	✓	x	✓	✓	✓	x
CBCL	x	x	✓	✓	✓	✓	✓
DASH-II	x	x	x	x	✓	✓	✓
AUTISM-CC	x	x	x	✓	✓	✓	✓
BISCUIT	x	x	x	x	✓	✓	✓
BASC-2	x	x	✓	✓	✓	✓	✓

4. 1. 3. Objectives

The first and main objective was to establish the psychometric properties of the different versions of SCAS and compare it to the MASC. The parent-child agreement on the total SCAS anxiety scores, as well as on the subtypes scores will be examined, as well as the issue of whether parents' own levels of anxiety might be related to the higher levels of anxiety in their children. The second objective was to establish the frequency of the overall anxiety, as well as the frequency of anxiety subtypes. The third objective was to explore the influence of chronological age on the expression of anxiety, to examine the association between anxiety, and impairments in communication and social interaction and expressive language levels.

4. 2. Methods

4. 2. 1. Subjects

Participants were a subset of the South Wales parents described in Chapter 2. The sample for this chapter comprised 49 children and adolescents (45 boys), aged 3 years to 17 years 9 months (mean age= 10 years 7 months, SD= 3 years 10 months). All children had a clinical diagnosis of an ASD, established according to ICD-10 criteria. Exclusion criteria were the same as the ones described in Chapter 2.

4. 2. 2. Measures

Anxiety Measures

The Spence Anxiety Scales (Spence, 1997; 1998): The Spence Preschool Anxiety Scale was designed to provide both the total anxiety score and scores for the following 5 types of anxiety: generalized anxiety, social anxiety, obsessive compulsive disorder, physical injury fears and separation anxiety. The scale consists of 28 items which are scored and one open-ended item assessing whether the child has experienced a traumatic event.

The Spence Children's Anxiety Scale assesses six domains of anxiety including generalized anxiety, panic/agoraphobia, social phobia, separation anxiety, obsessive compulsive disorder and physical injury fears. There is parent, as well as child version of this scale. According to the Spence Anxiety Scales instructions (see http://www.scaswebsite.com/1_1.html). Total scores from the SPAS and SCAS-C and SCAS-P can be combined by first transferring raw scores into the standardized T scores. Subscale scores from the different versions of the Spence Anxiety Scales can also be combined and compared to each other by first converting raw scores into the standardized T scores (by following the instructions provided in the manual).

Multidimensional Anxiety Scale for Children (MASC; March et al., 1997): MASC is a 45-item measure of anxiety for children 4–19 years of age. It has both parental and a child version. MASC provides the total anxiety score and also the scores for the following factors: somatic/panic, general anxiety, separation anxiety, social phobia, and school phobia.

Additional measures

As mentioned above, one of the objectives of this chapter was to explore the influence of chronological age, communication and social interaction impairments, and language levels on the expression of anxiety. The Social Communication Questionnaire (SCQ) was used to assess impairments in communication and social interaction, and the DISCO items were used to assess expressive language. A detailed description of these measures and associated limitations of some of them is provided in Chapter 2.

4. 3. Results

4. 3. 1. Psychometric Properties of Anxiety Instruments

Internal Consistency

Table 4. 2. Internal Consistency of Spence Children's Anxiety Scale Preschool Version

Scales	Cronbach's alpha
Total Anxiety	.764
Social Anxiety	.715
Separation Anxiety	.468
Physical Injury	.347
Fears	
OCD	.297
Generalized anxiety	.779

Table 4. 3. Internal Consistency of Spence Children's Anxiety Scale School age Version

Scales	Parent Report	Child's Report
	Cronbach's alpha	
Total Anxiety	.908	.855
Social Anxiety	.772	.602
Separation Anxiety	.756	.776
Physical Injury	.628	.649
Fears		
OCD	.668	.657
Generalized Anxiety	.804	.805
Panic/Agoraphobia	.750	.832

Table 4. 4. Internal Consistency of Multidimensional Anxiety Scale for Children

Scales	Cronbach's alpha
Total Anxiety	.836
Social Anxiety	.645
Separation Anxiety	.676
Harm Avoidance	.582
Somatic Symptoms	.718

As can be seen from the tables, internal consistency of total scores for all anxiety measures was in either good or excellent range with SCAS-P having Cronbach's alpha of .908, SCAS-C of .855, MASC of .836 and PAS of .764. Questionnaires performed less well

in terms of internal consistency of anxiety subscales. The best performing was SCAS-P with alphas in the good or excellent range for all subscales apart from the Physical Injury Fears and OCD subscales whose alphas were in the fair range. SCAS-C had alphas in excellent or good range for the Separation, Generalized and Panic/Agoraphobia anxiety and in fair range for Social Anxiety, OCD and Physical Injury Fears. MASC subscales were in good or fair range, however, internal consistency of Harm Avoidance subscale was unacceptable. The worst performing measure in terms of internal consistency of its subscales was the PAS. Internal consistency of Generalized and Social anxiety was good, however, Separation Anxiety, OCD and Physical Injury Fears had very low internal consistency.

Construct Validity of Spence Anxiety Scales and MASC

Construct validity was examined by using Spearman's correlations between the Spence Anxiety Scales and MASC total scores as well as between the subscale scores that two scales have in common. Correlations are presented in the table 4. 5. Total SCAS and MASC scores were highly correlated ($r = .540$, $p < .001$), Social Subscales of both instruments were also highly correlated ($r = .566$, $p < .001$) and Separation Anxiety Subscales showed moderate correlations ($r = .404$, $p = .04$).

Table 4. 5. Construct Validity of the Spence Anxiety Scale

Scales	MASC Total	MASC Separation Anxiety	MASC Social Anxiety
Spence Anxiety total	.540***	.559***	.509**
Spence Separation Anxiety	.182	.404*	.279
Spence Social Anxiety	.575***	.402*	.566***

Note: * = $p < .05$; ** = $p < .01$; *** = $p < .001$

Agreement between Self- and Parent-Report of anxiety and the relationship between anxiety in children and their mothers

34 parents completed SCAS-P, however, only 18 children and adolescents completed SCAS-C. So in total there were 18 children-parent pairs that completed relevant versions of

SCAS questionnaire. Due to the small sample size, only total anxiety scores were compared between parent and child report. The mean total anxiety score for parents' report was 59.5 (SD= 10.24; range: 30-77) and for children's report was 58.56 (SD= 9.68; range: 36-77). Paired t test showed that there were no significant differences between the total anxiety scores ($p = .774$; Cohen's $d = .09$). Based on parents' reports, 10 of 18 (55.6%) children met the cut-off criteria for elevated total anxiety and based on children's report 7 of 18 (38.9%) met the cut-off criteria,

As reviewed in the introductory section of this chapter, it has been suggested that parents own increased levels of anxiety, if present, might lead them to appraise their children as more anxious (Achenbach et al., 1997), association between levels of anxiety in children (according to parents' report) and the anxiety levels in parents (HADS scale, more details are provided in Chapter 2 and Chapter 8) was explored. However, no significant association was found ($N = 42$; $r = .282$, $p = .08$).

4. 3. 2. Prevalence of Anxiety

Overall anxiety

The mean total anxiety score was 57.71 (SD= 10.26, range: 30-77). 24 of 49 children (49%) met the cut-off criteria for elevated anxiety on Spence Anxiety Scales.

Anxiety Subtypes

Descriptive statistics for 6 SCASC-P anxiety subscales are presented in Table 4. 6.

Table 4. 6. Descriptive statistics for SCAS-P anxiety subscales (N= 32 children)

Variables	Mean	SD	Range	Skewness (SE)
Social Anxiety	5.26	4.6	0-14	.316 (.403)
Separation Anxiety	7.36	4.2	0-18	.293(.403)
Physical Injury Fears	6.85	3.8	0-16	.069 (.403)
OCD	4.94	9.41	0-13	.670 (.403)
Generalized Anxiety	5.76	3.7	0-13	.156 (.403)
Panic/Agoraphobia	6.71	3.8	0-15	.789 (.403)

The most prevalent subtype of anxiety was separation anxiety with 62.5% of children meeting the cut-off criteria for elevated separation anxiety, it was closely followed by physical injury fears (59.4% of children). 43.8% of children met the cut-off criteria for elevated panic/agoraphobia, 40.3% for social anxiety, 34.4% for generalized anxiety and 21.9% for OCD.

4. 3. 3. Relationship between anxiety levels and other children's variables

Pearson's correlations were used to examine the relationship between the total anxiety and chronological age, social interaction and communication problems. As can be seen from Table 4. 7., no significant associations were found. As noted throughout this thesis, due to the lack of cognitive assessments and limitations of some of the measures, in particular DISCO items, these analyses should be considered exploratory and results as preliminary at this point.

Table 4. 7. Relationship between anxiety levels and other children's variables

	CA	Expressive Language	SCQ Communication	SCQ Reciprocal Social Interaction
Total Anxiety	.157	.161	.08	.157

4. 4. Discussion

The first aim of this chapter was to examine the psychometric properties of the anxiety measure identified through the systematic search of the literature. Eleven different instruments were identified and only the Spence Anxiety Scales met the following criteria suitable for this research: (1) specifically designed for the assessment of anxiety disorders; (2) providing total anxiety scores as well as scores for specific DSM-IV and ICD-10 anxiety disorders; (3) having both parent and child versions (4) being applicable to the wide age range; (5) widely used in the literature on anxiety in general population and/or anxiety conditions other than autism and (6) being evaluated for the psychometric properties in these populations; (7) established in previous studies on anxiety in autism, (8) not being time consuming and/or complicated to administer in the research context. In addition to the Spence Anxiety scales, the Multidimensional Anxiety Scale for Children (MASC) was also used. Although this measure did not satisfy all of the above mentioned criteria (it does not provide

assessment of all the specific anxiety disorders defined by current diagnostic systems), it is currently considered as one of the gold standard assessments for the total anxiety in general population (Silverman et al., 2004).

Internal consistency of total anxiety scores provided by all three versions of the Spence Anxiety Scales was either excellent (SCAS-P had Cronbach's alpha of .908; SCAS-C of .855) or good (SPAS had Cronbach's alpha of .764). MASC total score also had excellent internal consistency (alpha = .836). The construct validity of total Spence Anxiety scales was also excellent. However, internal consistency of anxiety subscales provided by Spence Anxiety Scales was less convincing. Still, SCAS-P subscales were in the good to excellent range for Social, Separation, Generalized and Panic/Agoraphobia anxiety and fair for Physical Injury fears and OCD. SCAS-C had internal consistency in good to excellent range for the Separation, Generalized anxiety and Panic/Agoraphobia anxiety, and fair for Social Anxiety, OCD and Physical Injury Fears. The preschool version of Spence Anxiety scales had good internal consistency for Generalized and Social anxiety, however, internal consistency for other subscales was very poor. Social, Separation and Somatic Anxiety subscales from the MASC had a good internal consistency, however, internal consistency for the Harm Avoidance symptoms was low. The construct validity of the Separation and Social Anxiety subscales of Spence Anxiety Scales was good.

Agreement on total anxiety scores between children's and parents' reports was good. Although parents reported slightly higher levels of total anxiety than children ($M = 59.5$ ($SD = 10.24$) vs 58.86 ($SD = 9.68$), this difference in scores was not statistically significant ($t(34) = .290$, $p = .774$; Cohen's $d = .09$). This finding is surprising in the light of findings from the general anxiety literature where agreement between children's and parents' reports on anxiety levels tends to be moderate or low (Achenbach et al., 1987; Nauta et al., 2004). Also, due to the language impairments and limited emotional awareness in autism, it would not be surprising if children with autism had problems in interpreting and reporting their levels of anxiety. It is important to highlight that the issue of agreement between children's and parent's anxiety reports has not been thoroughly addressed in the existing literature. A study by Russell & Sofronoff (2005), using also Spence anxiety scales, found that parents reported significantly higher levels of overall anxiety as well as higher scores for OCD and physical injury symptoms than their AS children. However, results presented in this chapter are necessarily limited by having only 18 children's and parents' reports for the comparison. It is possible that the comparison between different subscale scores would yield differences

between self and parents' reports as some types of anxiety e. g. Social anxiety require more introspection and higher levels of social awareness than other types such as specific phobias for example (Silverman, & Eisen, 1992). The suggestion put forward by several authors (please see Achenbach et al., 1987) that parents own levels of anxiety might lead them to appraise their children's behaviours in a more negative way and judge them as more anxious was not supported as no significant association between levels of anxiety in children (according to parents' report) and anxiety levels in parents was found.

The second aim of this chapter was to establish the frequency of overall anxiety, as well as the frequency of anxiety subtypes in the population of children and adolescents with autism. The finding that 49% of children and adolescents with autism had clinically significant levels of anxiety confirmed the findings from the existing literature that suggest that between 33 and 55% of autism individuals have elevated levels of anxiety. For example, findings from this chapter are identical to findings by Rodgers et al. (2012) who also found that 49% of children from their study met the criteria for elevated anxiety. Rodgers and colleagues also used the Spence Anxiety scales.

In terms of the frequency of anxiety subtypes, the most frequent was separation anxiety with 62.5% of children meeting the cut-off criteria. Separation anxiety was closely followed by physical injury fears (59.4% of children). 43.8% of children met the cut-off criteria for elevated panic/agoraphobia, 40.3% for social anxiety, 34.4% for generalized anxiety and 21.9% for OCD. These findings are in line with some of the previous reports (Muris et al., 1998; Evans et al., 2005; Gadow et al., 2005; Weisbrot et al., 2005; de Bruin et al., 2006; Gillot et al., 2007; Sukhodolsky et al., 2008; Mazefsky et al., 2011). It is also interesting to consider these findings in the light of what we know about the developmental trajectory of different anxiety subtypes in typically developing children and adolescents. As discussed in more details in Chapter 1, the separation anxiety and phobias of animals is the most frequent type of anxiety in children between the ages of 6 and 9 years, generalized anxiety and phobias related to danger and death in children aged 10-13 years, and social anxiety in adolescents between the ages of 14 and 17 years (Warren & Sroufe, 2004).

The finding from this thesis that separation anxiety and specific phobias were the most prevalent subtypes of anxiety suggests that children and adolescents with autism tend to express more immature types of anxiety. Low incidence of OCD behaviours may be surprising considering the fact that RRBs are the diagnostic features of autism. However, OCD subscale of all the anxiety measures used in this thesis had the lowest internal

consistency suggesting that items related to OCD should be revised. Also, compulsions and obsessions in autism have been shown to be somewhat different than what is usually seen in OCD which might influence the way that parents evaluate OCD related items from the anxiety measures. For example, Zandt, Prior & Kyrios (2007) examined repetitive behaviours in 19 children with autism age and IQ matched children with OCD. They found that children with autism and OCD also differed in the types of compulsions and obsessions which were found to be less sophisticated in children with autism. Similar findings were reported by McDougle et al. (1995) in adult population.

The third objective of this chapter was to explore the influence of chronological age on the expression of anxiety, to examine the association between anxiety, and impairments in communication and social interaction and expressive language levels. Anxiety was not significantly associated with any of the moderating variables. These results are not surprising as several studies found no relationship between age, developmental level and overall levels of anxiety (Sukhodolsky et al., 2008; White & Roberson Nay, 2009; Strang et al., 2012) and it has also been suggested that language and communication problems, and the level of cognitive functioning in general, affect expressed levels rather than experienced levels of anxiety (Tsai, 1996; Strang et al., 2012). However, due to the cross-sectional design of this study, it was not possible to look at the developmental change of overall anxiety as well as different anxiety subtypes and it will be important to conduct such study in the future.

Chapter 5: Sensory Processing Problems in Autism

Chapter Plan:

In the first part of this chapter the key findings from the literature on sensory processing problems in autism will be very briefly summarized (an in-depth review was provided in chapter 1, section 1. 3.). Then, the findings on the frequency of sensory processing problems and on the mutual relationship between different types of sensory responsiveness in a sample of children and adolescents with autism will be presented. The findings on the relationship between sensory processing problems and chronological age, language level and core autism symptomatology will also be reported. These findings will be discussed in the light of the existing literature.

5. 1. Introduction

Evidence coming from autobiographical accounts, retrospective video studies, experimental laboratory protocols and questionnaire studies suggests that sensory processing problems are very prevalent in population with autism. The questionnaire studies report prevalence ranging from 30%-100% (Rogers & Ozonoff, 2005; Leekam et al., 2007; Kern et al., 2007 ; Ben-Sasson et al., 2009). The dysfunction affects all primary sensory modalities although it seems that auditory and touch modality are affected more than other modalities (Dunn et al., 2002; Ashburner, Ziviani, & Rodger, 2008).

Some reports have indicated that hypo-sensitivity to sensory stimuli is characteristic of autism (Rogers & Ozonoff, 2005). Indeed, several studies have shown that Individuals with autism show higher levels of sensory hypo-sensitivity when compared to TD individuals (Baranek et al., 2006; Dunn, Smith-Myles & Orr, 2002). However, other studies found no difference between the group with autism and the individuals with various developmental delays, ADHD and TD individuals (Ermer & Dunn, 1998; Watling et al., 2001; Baranek et al., 2006). It has been suggested that sensory hyper-sensitivity, and sensory seeking are not distinctive of autism and that these sensory modulation problems should be considered as a consequence of younger age and lower developmental level. Similarly, sensory avoidance has not been suggested to be distinctive of autism.

As discussed in Chapter 1, several studies have found the high prevalence of sensory hyper- and hypo-responsiveness, sensory avoidance and sensory seeking in the same individuals (Baranek et al. 2006; Ben-Sasson et al., 2007; Tomchek & Dunn, 2007) and it is possible that what distinguishes individuals with autism from both TD individuals and from other clinical groups is a frequency of sensory problems, rather than their form. This simultaneous occurrence of sensory hyper-, hypo-responsiveness, avoidance and sensory seeking has not been well researched in the past. As discussed in more details in Chapter 1, one account suggests that hyper-responsiveness predates sensory hypo-responsiveness, in other words sensory hyper-sensitivity is characteristic of autism toddlers and younger children, however, due to the constant heightened responsiveness and excitotoxic damage, over time, physiological systems gradually change their profile of responding and individuals change their pattern of responding from hyper- to hypo-responsiveness (Markram et al., 2010; McEwen, 1998). If this account is correct, it is expected that sensory hyper-sensitivity will be more prevalent in young children with autism and that sensory hypo-sensitivity will be more prevalent in adolescents and adults. An alternative account suggests a more circular relationship between sensory hyper- and hypo-responsiveness (Lane et al., 2011). More specifically, it has been theorized that autism individuals have problems in arousal modulation and due to these problems, they fluctuate between the states of over-arousal and hyper-responsiveness, and states of under-arousal and hypo-responsiveness. This theory further suggests that sensory seeking and sensory avoidance behaviours might represent an attempt to modulate the current state of arousal, in other words, sensory seeking might represent a soothing mechanism when an individual is in the state of over-arousal, and for raising stimulation in cases of under-arousal; and sensory avoidance might represent a mechanism for coping with the state of over-arousal and consequent over-stimulation (Lane, 2011). However, these competing theories have not been formally tested before and one of the aims of this chapter will be to test both the mutual relationship between different types of sensory responsiveness and test previously summarized theories.

Moderators of sensory problems in autism: as reviewed in more detail in Chapter 1, section 1. 3., the findings of studies that examined moderators of sensory processing problems in autism have been inconsistent. Several studies showed that sensory hyper-sensitivity and sensory seeking are the consequence of a lower developmental level and younger chronological age and not characteristic of autism per se (Rogers & Ozonoff, 2005). However, other studies seem to indicate that older children with autism exhibited more

seeking and hyper-sensitive behaviours when compared to young children with autism (Ben-Sasson et al. 2009; Liss et al. 2006). Also, it seems that CA is not related with an overall level of SP dysfunction in autism (Adamson et al. 2006) and that sensory problems in visual modality improve over time in children with autism (Leekam et al. 2007). The results of the studies that examined the relationship between sensory problems and communication and social interaction problems have also been mixed. Hilton, Graver and LaVesser (2007) found significant correlations between the Social Responsiveness Scale total scores and all four sensory profile quadrant scores in a sample of 36 children aged 3-6 years. However, other studies have found that only hypo-responsiveness and hyper-responsiveness were associated with communication and social problems (Watson et al., 2012). Finally, sensory problems have been found to be associated with lower levels of adaptive behaviours. For example, Baker et al. (2008) used the Short Sensory profile and VABS with 22 children with autism (mean chronological age= 5.5 years) and found that higher total SSP total scores predicted lower VABS scores.

5. 1. 1. Objectives

The goal of this chapter is three fold. Firstly, the detailed description of sensory processing in a sample of children and adolescents with autism will be provided. Secondly, mutual relationship between sensory hyper-, hypo-sensitivity, sensory avoidance and sensory seeking types of sensory responsiveness will be examined. Thirdly, the role of chronological age, language level and core autism symptomatology will be examined. Also, the influence of sensory processing difficulties on adaptive abilities will be examined.

Based on the reviewed literature, it is expected that sensory processing problems will be highly prevalent in a sample of children and adolescents with autism. It is also expected that difficulties will be multimodal and that individuals will show a mixed type of sensory responding i. e. both hyper-, hypo-sensitivity, sensory seeking and sensory avoidance. In terms of mutual relationship between different sensory responsiveness types, as discussed in the introduction to this chapter and also in the section 1. 3., two alternative possibilities exist. According to one account sensory hyper-sensitivity predates sensory hypo-sensitivity, in other words sensory hyper-sensitivity is characteristic of toddlers and younger children with autism, however, over time, physiological systems gradually change their profile of responding and individuals make transition from hyper- to hypo-responsiveness. If this account is to be supported, it is expected that sensory hyper-sensitivity will be associated

with lower chronological age (CA) and that sensory hypo-sensitivity will be associated with older CA. Alternative account suggests circular relationship between sensory hyper- and hypo-sensitivity, in other words, relationship between these two quadrants is mediated by sensory seeking and sensory avoidance. More precisely, sensory seeking serves to either increase stimulation in the cases of under-arousal and hypo-responsiveness or decrease stimulation in the case of over-arousal and hyper-responsiveness. Sensory avoidance will, according to this account serve to reduce stimulation when individual is over-aroused and finds environment over-stimulating. A series of mediation models will be used to test these accounts.

Regarding the influence of chronological age, the results of the literature have been conflicting thus far and it is difficult to predict the direction of results. Regarding the relationship between sensory problems, and social deficits, communication impairments and overall autism severity, results have been mixed, and similarly like with the influence of chronological age, it is difficult to predict the results.

5. 2. Methods

5. 2. 1. Subjects

The data collected from the same participants (N= 49) whose characteristics were described in the section 4. 2. 1. will be used here.

5. 2. 2. Measures

The Sensory Profile (SP; Dunn, 1999) was used to examine sensory processing in autism sample in this thesis. A detailed description of the Sensory Profile is provided in Chapter 2.

Additional measures

As mentioned above, one of the objectives of this chapter was to explore the relationship between sensory processing problems and, chronological age, impairments in communication and social interaction and language levels. The Social Communication Questionnaire (SCQ) was used to assess impairments in communication and social interaction, and DISCO items were used to assess expressive language. A detailed description of these measures is provided in the Methods chapter.

5. 3. Results

5. 3. 1. Frequency of Sensory Processing problems across different SP categories

Descriptive statistics for sensory modality and sensory quadrant scores are shown in Table 5. 1.

Table 5. 1. Descriptive statistics for Sensory Modalities and Sensory Quadrants

Variables	Mean	SD	Range	Skewness (SE)
Auditory	22.1	7	10-38	.418 (.340)
Visual	29.75	6.37	19-44	.130 (.340)
Vestibular	42.12	7.07	23-55	-.381 (.340)
Touch	59.24	11.36	33-82	-.013 (.340)
Gustatory	40.31	12.5	16-60	-.217 (.340)
Multisensory	20.71	5.19	10-32	-.145 (.340)
Poor Registration (Hypo-Sensitivity)	49.35	10.96	23-71	-.480 (.340)
Sensory Seeking	89.19	17.27	43-123	-.132 (.340)
Sensory Sensitivity	63.53	14.72	30-89	-.252 (.340)
Sensory Avoidance	86.22	15.71	53-116	.013 (.340)

Sensory Dysfunction Across Sensory Processing Modalities

The Sensory Profile is a norm-referenced questionnaire. The manual provides cut scores derived from a large normative sample of individuals without disabilities. Based on scores individual can be classified as showing:

- (a) Typical performance which corresponds to scores that are at or above the point 1SD below the mean for TD individuals;
- (b) Probable difference which corresponds to scores at or above the point 2 SD below the mean but also lower than 1SD below the mean for TD individuals;
- (c) Definite difference which corresponds to score below the point 2SD below the mean for TD individuals.

As can be seen from Figure 5. 1., the majority of individuals with autism had scores in the range of probable or definite differences across modalities. The most prevalent were problems with touch processing with 71.4% of children and adolescents with autism having scores in the definite difference (DD) range. They were closely followed by the problems in auditory processing (69.4% had DD scores), multisensory processing (67.3%), vestibular processing (65.3%), gustative processing (40.8%) and visual processing (32.7%).

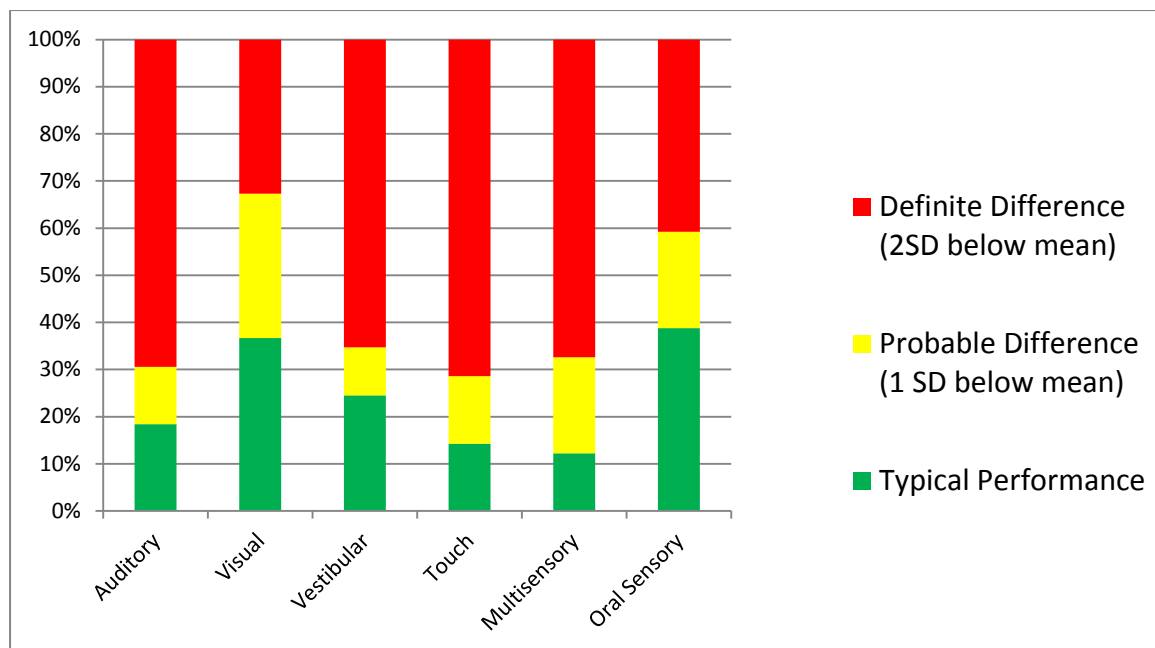


Figure 5. 1. Sensory Profile Scores Across Six Sensory Processing Modalities

It is important to highlight the fact that 20 children (39.2%) had scores in the probable and definite differences for all 5 primary sensory modalities (auditory, visual, vestibular, tactile and gustatory modality), 13 children (25.5%) for 4 primary sensory modalities, 4 (7.8%) for 3, 7 (13.7%) for 2 and 2 children for only single sensory modality. Only one child had scores in the typical range for all sensory modalities.

Sensory Dysfunction Across Sensory Quadrant Scores

As can be seen from Figure 5. 2, 79.6% of Children with autism had sensory avoidance scores in the DD range, 75.5% for sensory registration (hypo-sensitivity), 73.5% of sensory hyper-sensitivity and 55.1% for sensory seeking.

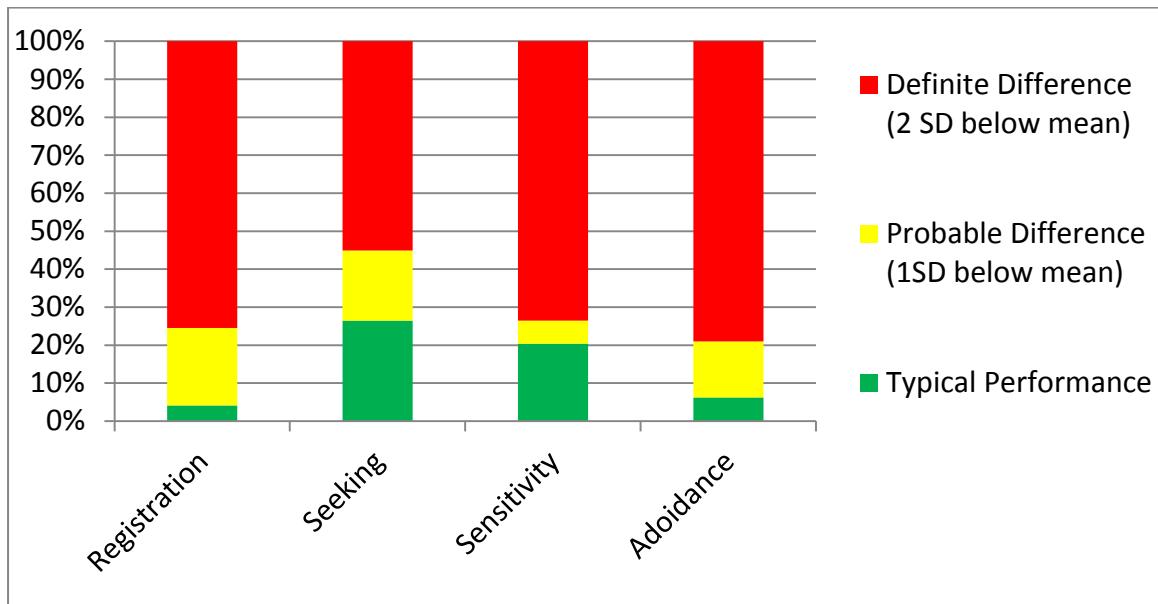


Figure 5. 2. Sensory Quadrant Scores

32 children (65.3%) had scores in the probable (PD) or definite difference (DD) range for all four Sensory Profile Quadrants. 11 children (25.58%) had scores in the PD or DD range for 3 out of 4 quadrants (5 children (10.2%) for registration (hypo-sensitivity) + seeking +avoidance and 6 children (12.24%) for registration (hypo-sensitivity) + sensitivity +avoidance combination). 3 children (6.12%) had scores in the PD or DD range for registration (hypo-sensitivity) and avoidance. 1 child (2%) had scores in DD range for only registration quadrant, with typical scores for other quadrants and only 2 children (4%) had typical scores for all four quadrants.

5. 3. 2. Relationship between the sensory quadrants

Pearson's correlations were first used to examine the mutual relationship between the sensory quadrants (Table 5. 2.).

Table 5. 2. Association between sensory quadrants

	Registration	Seeking	Sensitivity	Avoidance
Registration	1			
Seeking	.459**	1		
Sensitivity	.723***	.587***	1	
Avoidance	.598***	.253	.742***	1

Note: *= $p < .05$; **= $p < .01$; ***= $p < .001$

As can be seen from Table 5. 2., apart from Sensory Seeking and Sensory Avoidance quadrants, all other quadrants were highly associated.

The next step in the analysis was to explore the mutual relationship between sensory quadrants. As previously discussed, according to one theory, sensory hyper-sensitivity, sensitivity is characteristic of toddlers and younger children with autism, however, over time, there is a transition from hyper- to hypo-responsiveness. However, CA was not significantly associated with either hypo-sensitivity ($r = .055$, $p = .707$) nor hyper-sensitivity ($r = .069$, $p = .639$) thus not supporting this account. Alternatively, as previously discussed, the relationship between sensory hyper- and hypo-sensitivity is mediated by sensory seeking and sensory avoidance. A series of mediation models was first used to examine the mutual relationship between sensory hypo-, hyper-sensitivity and sensory avoidance, following with a series of mediation models examining the mutual relationship between sensory hypo-, hyper-sensitivity and sensory seeking.

Generally speaking, mediation relationships between independent (IV), dependent (DV) and mediation (MV) variables can be represented graphically as in the Figure 5. 3.:

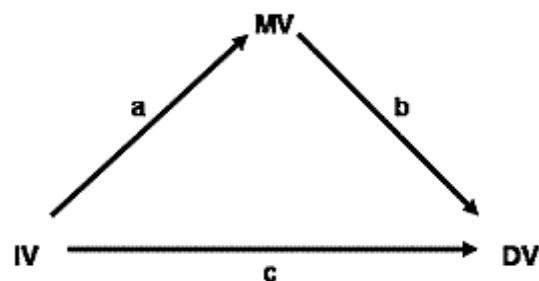


Figure 5. 3. Graphical representation of the mediated relationship

According to Barron and Kenny (1986) testing for mediation can be performed via three separate regression analyses.

The first regression tests whether there is a significant association between the IV and DV variables (path c from the figure 5.3). In order for the hypothesised MV to be a mediator, this association needs to be significant.

The second regression tests whether there is a significant association between the IV and MV variables (path a from the figure 5.3). Similarly as above, in order for the hypothesised MV to be a mediator, this association needs to be significant.

Thirdly, a two step hierarchical regression analysis is performed. The first step is to explore whether there is a significant association between the MV and DV variables (path b from the figure 5.3) and this step needs to be significant in order for the hypothesised MV to be a mediator. The second step explores path c. After controlling for the effects of the MV on the DV, the effects of the IV should no longer be significant for the hypothesised MV to be a mediator.

The relationship between Sensory Hyper-Sensitivity, Hyper-Sensitivity and Avoidance

Mediation analysis 1 examined the model where sensory hyper-sensitivity would lead to avoidance which in turn, would lead to hypo-sensitivity. Model is graphically presented in Figure 5. 4.

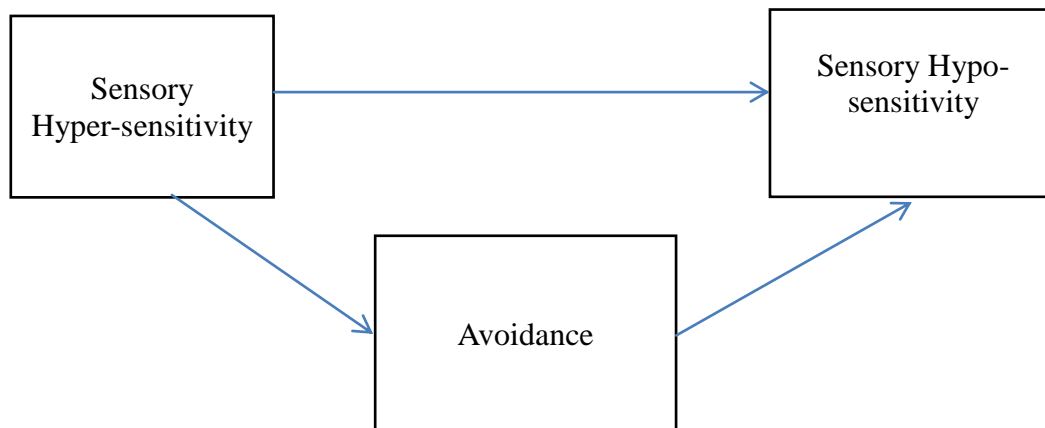


Figure 5. 4. Graphic representation of the Mediation Model 1

The first two regressions showed that Sensory Hypo-Sensitivity was predicted by hyper-sensitivity ($R^2 = .512$, $p = .000$) and that avoidance was predicted by sensory hyper-sensitivity ($R^2 = .541$, $p = .000$) satisfying first two conditions proposed by Baron & Kenny. In the first step of hierarchical regression, Sensory Hypo-Sensitivity was predicted by avoidance ($R^2 = .344$, $p = .000$), and when controlling for the effects of avoidance, the relationship between sensory hypo-sensitivity and sensory hyper-sensitivity was still significant ($R^2 = .510$, $R^2 \text{ change} = .173$, $p = .000$) indicating that the model where Sensory

Hyper-sensitivity leads to avoidance which, in turn, leads to hypo-sensitivity, was not confirmed. Steps of the Mediation Analysis 1 are presented in Table 5. 3.

Table 5. 3. Mediation Analysis 1

	R2	R2 Change	Beta
Regression One:			
Sensory Hypo- on Sensory Hyper-sensitivity	.512***		.723
Regression Two:			
Avoidance on Hyper-sensitivity	.541***		.742
Regression Three:			
Step 1: Hypo-sensitivity on avoidance	.344***		.138
Step 2: Hypo- on Hyper-sensitivity	.510***	.173***	.620

Note: *= p<.05; **= p<.01; ***= p<.001

Mediation analysis 2 examined the model where sensory hyper-sensitivity would lead to hyper-sensitivity which in turn, would lead to sensory avoidance. Model is graphically presented in Figure 5. 5.

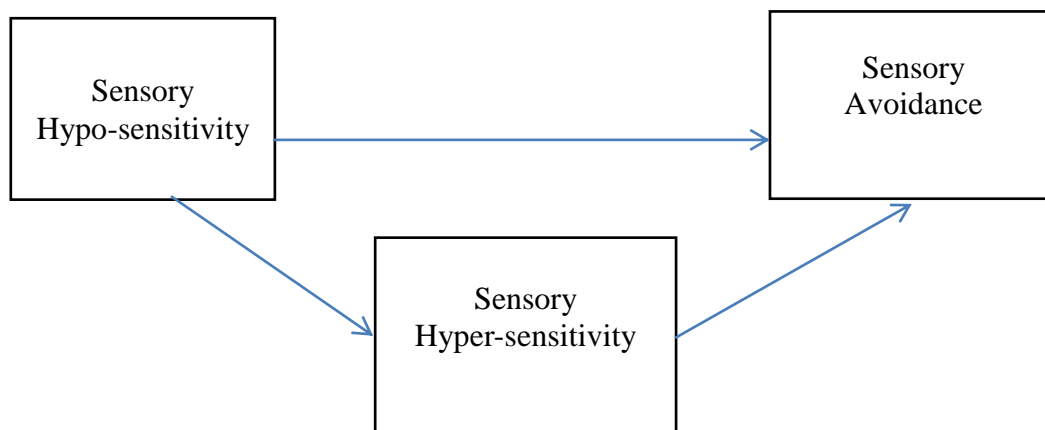


Figure 5. 5. Graphic representation of the Mediation Model 2

As can be seen from Table 5. 4., this model was confirmed.

Table 5. 4. Mediation Analysis 2

	R2	R2 Change	Beta
Regression One:			
Sensory Avoidance on Sensory Hypo-sensitivity	.344***		.598
Regression Two:			
Hyper-sensitivity on Hypo-Sensitivity	.512***		.723
Regression Three:			
Step 1: Avoidance on Hyper-sensitivity	.541***		.649
Step 2: Avoidance on Hypo-sensitivity	.539	.008	.129

In addition to 2 theoretically driven mediation models described above, the following “dummy” mediation models were also tested: (a) model where sensory hyper-sensitivity leads to hypo-sensitivity which in turn leads to avoidance; (b) model where avoidance leads to hypo-sensitivity which in turns leads to hyper-sensitivity; and (c) model where avoidance leads to hyper-sensitivity which in turn, leads to hypo-sensitivity. None of these models was supported.

The relationship between Sensory Hypo-Sensitivity, Hyper-Sensitivity and Seeking

Firstly, a mediation model where sensory seeking mediates the relationship between sensory hypo- and hyper-sensitivity was tested. As can be seen from Table 5. 5., this model was not supported.

Table 5. 5. Seeking as a mediating variable between hypo- and hyper-responsiveness

	R2	R2 Change	Beta
Regression One:			
Hyper- on Hypo-sensitivity	.521***		.722
Regression Two:			
Seeking on Hypo-sensitivity	.209**		.457
Regression Three:			
Step 1: Hyper-sensitivity on Seeking	.341***		.335
Step 2: Hyper- on Hypo-sensitivity	.594***	.245***	.569

Secondly, a mediation model where the relationship between sensory hyper-sensitivity and hypo-sensitivity is mediated by sensory seeking was examined. As can be seen from the table 5. 6., this model was not supported.

Table 5. 6. Seeking as a mediating variable between hyper- and hypo-responsiveness

	R2	R2 Change	Beta
Regression One:			
Hypo- on Hyper-sensitivity	.521***		.722
Regression Two:			
Seeking on Hyper-sensitivity	.354***		.595
Regression Three:			
Step 1: Hypo-sensitivity on Seeking	.193**		.457
Step 2: Hypo- on Hyper-sensitivity	.502***	.313***	.696

Thirdly, a mediation model where the relationship between sensory hypo-sensitivity and sensory seeking is mediated by sensory hyper-sensitivity was examined. As can be seen from Table 5. 7. this model was supported.

Table 5. 7. Hyper-sensitivity as a mediator between hypo-sensitivity and seeking

	R2	R2 Change	Beta
Regression One:			
Seeking on Hypo-sensitivity	.209**		.457
Regression Two:			
Hyper on Hypo-sensitivity	.521***		.722
Regression Three:			
Step 1: Seeking on Hyper-sensitivity	.354***		.595
Step 2: Seeking on hypo-sensitivity	.356	.002	.058

Finally a mediation model where sensory hypo-sensitivity mediates the relationship between sensory hyper-sensitivity and sensory seeking was examined. As can be seen from Table 5. 8., this model was not supported.

Table 5. 8. Hypo-sensitivity as a mediator between hyper-sensitivity and seeking

	R2	R2 Change	Beta
Regression One:			
Seeking on Hyper-sensitivity	.354***		.595
Regression Two:			
Hypo on Hyper-sensitivity	.521***		.722
Regression Three:			
Step 1: Seeking on Hypo-sensitivity	.209**		.457
Step 2: Seeking on hyper-sensitivity	.356***	.147	.553

5. 3. 3. Moderators of Sensory Modulation Problems

Pearson's Correlations were used to examine associations between sensory processing data and chronological age, communication problems and reciprocal social interactions. Due to multiple correlations significance level was adjusted to .01. As can be seen from Table 5.9., sensory seeking was significantly negatively correlated with chronological age, as lower scores on SP suggest more problems, this implied that younger children exhibited more sensory seeking behaviours. Already noted constraints of the data collected to measure language warrants these analyses to be considered exploratory and results as preliminary at this point.

Table 5. 9. Moderators of Sensory Modulation Problems

	CA	Expressive Language	SCQ Communication	SCQ Reciprocal Social Interaction
Poor Registration	.055	-.040	-.337	-.172
Sensory Seeking	.380*	.187	-.150	-.154
Sensory Sensitivity	.069	.029	-.323	-.185
Sensory Avoiding	-.194	.019	-.205	-.203

5. 4. Discussion

The first aim was to examine the frequency of sensory problems in children and adolescents with autism. Results confirmed previous findings from the literature regarding the pervasive and multimodal nature of sensory problems in autism.

Almost 40% of children (of N= 49) had problems simultaneously across all five primary sensory modalities, 13 children (25.5%) across four, 4 (7.8%) across three and 7 (13.7%) in two primary sensory modalities. Only 2 children had problems in a single sensory modality and one child had scores in the typical range across all sensory modalities. When looking at individual sensory modalities, the most common were problems related to touch, with 71.4% of children scoring in the definite difference range for this modality, problems with audition and vestibular modalities were also very common. Problems with gustatory and visual modalities were somewhat less frequent with 40.8% of children having scores in the definite differences range for gustatory and 32.7% of children for visual modality. These results are in line with the previous literature that showed problems with touch and auditory stimuli being the most prevalent sensory problems in autism and that sensory problems are usually multimodal (Kientz & Dunn, 1997; Talay-Ongan & Wood, 1999; Dunn et al., 2002; Rogers et al., 2003; Kern et al., 2006; Leekam et al., 2007; Tomchek & Dunn, 2007; Baker et al., 2008; Schoen et al., 2009; Wiggins et al., 2009; Hilton et al., 2010; Klintwall et al., 2010).

As discussed in more detail in introduction, one of the main topics in sensory literature in autism is which of the sensory modulation patterns is distinctive of autism. Many authors have suggested that sensory hypo-sensitivity (or poor registration in Dunn's terminology) is the most common sensory problem in autism and further suggested that while sensory seeking and sensory hyper-sensitivity might be also common in this population, those problems are a consequence of lower developmental level and younger chronological age rather than being specific to autism (Ermer & Dunn, 1998; Watling et al., 2001; Baranek et al., 2006; Dunn, 2002; Dunn et al., 2002; Watling et al., 2001; Crane et al., 2009).

The findings from this thesis do not seem to support the suggestions that sensory hypo-sensitivity is the most prevalent sensory modulation problem in children and adolescents with autism. The most frequent sensory modulation problem was sensory avoidance with 79.6% of Children with autism having sensory avoidance scores in the definite difference (DD) range. Sensory hypo- and hyper-sensitivity were almost equally

frequent with 75.5% children showing DD scores for sensory hypo-sensitivity and 73.5% of children for sensory hyper-sensitivity. Around half of the children (55.1%) had DD scores for sensory seeking. More importantly, the findings from this thesis show that almost 91% of the children and the adolescent showed mixed type of sensory modulation problems with 65.3% of the children having scores for all four sensory quadrants in either the probable (PD) or definite difference (DD) range, 25.6% of children for 3 out of 4 quadrants (10.2% for registration (hypo-sensitivity) + seeking +avoidance 12.24% for registration (hypo-sensitivity) + sensitivity +avoidance combination). Only one child showed problems in a single quadrant i. e. sensory hypo-sensitivity quadrant.

These results are in line with other research that also found that a significant percentage of individuals with autism show a mixed type of sensory modulation problems (Baranek et al., 2006; Ben-Sasson et al., 2007; Tomchek & Dunn, 2007). These findings also seem to lend support for the suggestion that was put forward in the introductory section of this chapter that it is a frequency of sensory problems and mixed type of sensory responsiveness that is a distinguishing feature of sensory processing problems in autism rather than any specific type of sensory problems. Indeed it has been consistently found that the individuals from other clinical groups tend to show consistent patterns of sensory modulation problems, for example sensory hyper-sensitivity in individuals with ADHD.

The second aim of this chapter was to explore the mutual relationship between four sensory quadrants i. e. sensory hyper-sensitivity, sensory hypo-sensitivity, sensory seeking and sensory avoidance. The results from this thesis showed that the majority of children and adolescents with autism (91%) show problems across at least 3 sensory quadrants. As discussed earlier, two explanations of why ASC individuals might show a mixed type of sensory responsiveness were put forward. According to the first explanation, sensory hyper-responsiveness (hyper-sensitivity) is characteristic of toddlers and young children with autism, in the beginning over-stimulation and over-arousal lead to the adaptive response from the hypothalamic-pituitary-adrenal axis (HTPA axis) and target physiological systems, however, constant state of physiological stress will eventually lead to excito-toxic damage, and over time, physiological systems gradually change their profile of responding and individuals change their response pattern from hyper- to hypo-responsiveness (Markram et al., 2010; McEwen, 1998). These changes have been noted in depression (McEwen & Wingfield, 2010) and also, findings on the developmental changes in the structure and function of amygdala (Juraneck, Filipek, Berenji, Modahl, Osann, & Spence, 2006) also seem

to lend support to this account. In terms of sensory responsiveness, if this account is true, it is expected that sensory hyper-sensitivity will be more prevalent in young children with autism and that sensory hypo-sensitivity will be more prevalent in adolescents and adults. However, findings from this thesis did not support this pattern. According to the second hypothesis, individuals with autism have problems in maintaining homeostatic state of arousal or in other words, that they fluctuate between the states of hypo-arousal and sensory hypo-responsiveness, and over-arousal and subsequent sensory hyper-responsiveness. In addition, sensory avoidance and sensory seeking are compensatory behaviours that are used by individuals to regulate their state of arousal. A series of mediation analyses supported this second explanation of the mixed pattern of sensory responsiveness in autism. Mediation analyses suggested that children and adolescents with autism fluctuate between the states of hypo- and hyper-responsiveness and that avoidance behaviours are compensatory strategies related to being over-stimulated. Mediation analyses also suggested that seeking behaviours are more related to being over-stimulated rather than hypo-stimulated. It is important to note that although the age range for this study was quite wide (from 3 to 17 years), it is possible that the transition from sensory hyper-responsiveness to hypo-responsiveness over time, would have been supported if adults were recruited. Also, it is possible that the two accounts summarized above are not mutually exclusive. However, conclusions are necessarily limited by the cross-sectional design of the study and will be important to test the allostatic account in a developmental study with addition of relevant endocrine and physiological measures.

In terms of moderators of sensory modulation problems, chronological age was only correlated with sensory seeking behaviours ($r = .380$, $p = .007$) suggesting that younger children showed more sensory seeking behaviours (lower scores on sensory quadrants imply more problems). Results regarding sensory seeking are in accordance with the findings of meta-analysis by Ben-Sasson et al. (2009) who reported that sensory seeking was more frequent in younger than in older children. However, they also reported that the same trend existed for the sensory hyper-sensitivity which was not found in this thesis. Ben-Sasson et al. (2009) reported that the results from studies that looked at the change of sensory hypo-sensitivity over time were very inconsistent between the studies. The results from this thesis, similar like all other studies that looked at the change of sensory modulation problems over-time are cross-sectional, and not sensitive enough to look at the developmental course of sensory problems in autism. A cross-sectional study by Kern et al. (2006) reported that sensory problems in group with autism improved over time, while study by Leekam et al.

(2007) showed that only visual sensitivity improves over time. However, age range of their participants was much wider than the sample used for this thesis (age range was 3-56 years in the study by Kern et al). Future work will need to examine the development of sensory problems by employing the prospective study design.

Relatively few studies have addressed the question of how different sensory modulation patterns relate to core autism symptoms. This is surprising considering the fact that the research in other clinical groups has shown that early sensory atypicalities in infancy and toddlerhood predict poorer language and social interaction abilities at later ages (DeGangi, Breinbauer, Roosevelt, Porges, & Greenspan, 2000) suggesting that sensory problems might be developmental precursors of social, communication, and language problems in infants subsequently diagnosed with autism. Studies that examined the relationship between sensory problem and core autism symptoms found that overall sensory problems are associated with the autism severity (Adamson et al., 2006; Kern et al., 2006; Ben-Sasson et al., 2008), and that either problems in all four sensory quadrants (Hilton et al., 2007) or in hypo-sensitivity, avoidance and seeking (Watson et al., 2011) were associated with more social, communication and language problems. The results from the sample of children and adolescents from this thesis show moderate correlations between both hypo-sensitivity and sensory sensitivity with SCQ communication scores at .05 level ($p = .035$ and $p = .027$ respectively) which was in line with the findings from the literature reviewed above. However, it is important to point out that after adjusting significance level to .01 for multiple correlations, associations between SCQ communication scores, hypo-sensitivity and sensory sensitivity fell below the significance level.

5. 5. Summary of the findings on Anxiety and Sensory Processing in Autism

The findings in chapters 4 and 5 point out the pervasive nature of both sensory problems and anxiety in the population of children with autism. An important contribution of this chapter was the validation of the Spence Anxiety Scale for the use in the population of children and adolescents with autism. Also, findings point out on how it will be possible to improve the scale for the use in population with autism in the future. For example, it will be important to revise the OCD subscale of the Spence Anxiety Scales. It was also found that around 90% of the children and adolescents with autism have mixed types of sensory responsiveness, showing sensory hyper- and hypo-responsiveness at the same time. This, as discussed in Chapter 1 and throughout this chapter, suggests that children and adolescents

with autism have problem in arousal modulation and due to this, they fluctuate between the states of over-arousal and hyper-responsiveness, and states of under-arousal and hypo-responsiveness. However, as findings from this thesis are based on the use of questionnaire data, the use of physiological measures in the future work will be necessary to further explore preliminary findings from this thesis regarding the problems in arousal modulation in population with autism. Finally, this chapter addressed the relationship between different sensory modulation problems in children with autism.

5. 6. Limitations of the findings on anxiety and sensory processing in autism

The first limitation was the sample size. This was mainly relevant for the comparison between children's and parents' reports on anxiety as there were only 18 overlapping reports for anxiety. This necessarily limits the generalizability of these findings. Furthermore, due to only 18 overlapping reports, it was not possible to compare the reports on separate anxiety subtypes. The second limitation related to the analysis that considered the relationship between anxiety, sensory problems and other children's characteristics, in particular language levels, was that DISCO items, although used previously by Honey (2007) and Honey et al. (2007) were not validated prior to this thesis. As has been reported in Chapter 2, DISCO scores did significantly correlate with both Mullen's Expressive Language scores and WASI Verbal IQ scores, however, as pointed out earlier, the use of the DISCO scores presents limitation and all the analyses that used these scores should be considered exploratory and results preliminary in nature.

Thirdly, this study would be much improved if the third party reports, in particular teachers' reports, were available on children's anxiety levels and their sensory problems. It will be important to use a multi-informant assessment of sensory processing and anxiety in future studies. The cross-sectional nature of this study was a limiting factor in being able to make stronger inferences regarding the relationship between age, developmental level and language and the presence of anxiety and sensory problems. A longitudinal research will be of crucial importance to tease apart these influences.

Chapter 6: Repetitive Behaviours, Anxiety and Sensory Problems in Autism

Chapter Plan:

As already described in Chapter 1, one of the main aims of this thesis was to examine the interrelationship between repetitive behaviours, anxiety and sensory problems. However, as noted throughout this thesis, before addressing this aim, it was first necessary to explore and understand each of these behaviours as separate entities. Chapter 3 explored repetitive behaviours and showed that repetitive behaviours formed two separate classes of behaviours: repetitive sensory-motor (RSM) behaviours such as rocking, hand-flapping and spinning of objects and insistence on sameness (IS) behaviours such as narrow interests, rigid routines and rituals. Chapter 4 explored anxiety in children and adolescents with autism and demonstrated that anxiety was prevalent in children and adolescents with autism, with 49% of the sample meeting the criteria for clinically significant anxiety. Finally, Chapter 5 explored sensory processing problems and indicated that all children and adolescents showed some form of sensory modulation problems.

Early autism researchers (Hutt, Hutt, Lee, & Ounsted, 1964; Ornitz & Ritvo, 1968; Kinsbourne, 1980) have theorised that sensory problems and anxiety are the two key triggers for the development and maintenance of repetitive behaviours in individuals with autism. It was suggested that, due to the faulty modulation of arousal, individuals with autism oscillate between the states of hyper- and hypo-arousal. When in the state of hyper-arousal, individuals with autism would be hyper-sensitive to any external sensory stimulation and experience distress and anxiety. On the other hand, in the state of hypo-arousal, individuals with autism would be under-stimulated and under-responsive to their environment. Repetitive behaviours were proposed to serve as coping mechanisms for maintaining a homeostatic state of arousal, in other words, to increase stimulation and arousal in cases of under-stimulation and reduce arousal when a person is over-stimulated and distressed. In a recent review, we further developed these early proposals by suggesting the possibility that different types of repetitive behaviours might be differently associated with sensory symptoms and anxiety. However, the relationship between repetitive behaviours, anxiety and sensory problems has not been fully examined in children and adolescents with autism. Given both theoretical and clinical importance of addressing the relationship between these factors, the aim of this

chapter will be to describe the inter-relationship between different types of repetitive behaviours, sensory problems and anxiety.

6. 1. Introduction

6. 1. 1. Problem in arousal modulation as a key factor for the repetitive behaviours

As discussed in more detailed manner in Chapter 1, several authors have proposed that individuals with autism have problems in regulating arousal and maintaining their levels of arousal within optimal limits. Based on their EEG findings, Hutt, Hutt, Lee, & Ounsted (1964; also Hutt & Hutt, 1965) hypothesised that the activity of the reticular activating system (RAS) is constantly at a high level in individuals with autism, and that individuals with autism engage in motor movements and stereotypies in order to block further sensory input and reduce levels of arousal. Hutt and colleagues also proposed that due to the constant high activity of the RAS, individuals with autism find any novel and/or unexpected situation as arousing and distressing, which leads to the development of insistence on sameness behaviours in order to avoid any such situation. Ornitz and Ritvo (1968) and later Kinsbourne (1980) refined the hypothesis proposed by Hutt and colleagues, by suggesting that rather than being chronically over-aroused, individuals with autism have a very narrow range of optimal arousal, and in addition to this had problems in regulating their arousal levels. This would lead to constant fluctuation between the states of over- and under-arousal and repetitive behaviours would have alternative function, depending on the current state of individual with autism. In the state of over-arousal, repetitive behaviours would have a soothing function by providing predictable stimulation and reducing overall levels of arousal. Alternatively, if an individual was in the state of under-arousal, repetitive behaviours would provide additional stimulation and increase current levels of arousal.

Based on arousal theories reviewed above, and also in Chapter 1, sensory symptoms and anxiety would serve as triggers for repetitive behaviours. Furthermore, the contribution of anxiety to the development and maintenance of repetitive behaviours would be dependent on, or mediated by, sensory symptoms.

6. 1. 2. Sensory problems and anxiety as triggers for repetitive behaviours

As discussed in detail in previous chapters, both anxiety and sensory problems are highly prevalent in population with autism. Previous studies have found that individuals with autism show significantly higher levels of anxiety than both typical controls (Kim et al.,

2000; Bellini, 2004; Bradley et al., 2004; Gadow et al., 2005) and individuals with other clinical conditions such as Conduct Disorder, Williams Syndrome and Specific Language Impairment (Green et al., 2000; Evans et al. 2005; Rodgers et al., 2012a) and similar levels of anxiety when compared to individuals diagnosed with anxiety disorder (Russell & Sofronoff, 2005; Farugia & Hudson, 2006). Although the reported rates of anxiety are highly variable, most of the studies found the prevalence of anxiety to be between 35 and 55% (Bellini, 2004; Bradley et al., 2004; Gadow et al., 2005; de Bruin et al., 2006; Leyfer et al., 2006; Rodgers et al., 2012b). Majority of studies have found that at least 90% of individuals with autism show either hyper-, hypo-sensitivity or sensory seeking and that quite often, a single individual can exhibit all of those sensory problems together (Leekam et al., 2007; Ben-Sasson et al., 2009; Lane et al., 2010).

The potential relationship between sensory problems and anxiety was addressed in 4 recent studies (Green et al., 2012; Ben-Sasson et al., 2008; Mazurek et al., 2012; Pfeiffer et al., 2005). For example, in a study that examined the relationship between the sensory processing problems and anxiety in 2973 children with autism aged 2-17 years, Mazurek et al. (2012) found that anxiety and sensory hyper-sensitivity were highly associated. Other studies have reported similar findings, supporting the claims of arousal theories that sensory sensitivity is experienced as distressing and anxiety provoking.

Several studies have addressed the potential relationship between sensory problems and repetitive behaviours. For example, Boyd et al. (2009) used RBS-R and Sensory Questionnaire to examine repetitive behaviours and sensory processing in 61 children with HFA aged 6-17 years with 64 typically developing children. Not surprisingly, children with HFA had more repetitive behaviours and sensory problems than individuals in the TD group. Furthermore, a significant, positive association was found between the total repetitive behaviours scores and overall sensory problems and this relationship remained significant after controlling for IQ. However, Boyd et al. (2009), similarly like the studies by Gabriels et al. (2008) and Chen, Rodgers & McConachie (2008), did not explore whether there was a differential association between the different subtypes of repetitive behaviours are different sensory problems. Studies that have addressed this issue, have supported suggestions by Ornitz & Ritvo (1968) that repetitive behaviours might be associated with both sensory hyper- and hypo-sensitivity. For example, Boyd et al. (2010) used the RBS-R to measure repetitive behaviours and a total of 4 different measures (Sensory Experiences Questionnaire, Sensory Profile, Sensory Processing Assessment for Young Children and the Tactile

Defensiveness and Discrimination Test—Revised (TDDT-R; Baranek, 1998) to assess sensory processing problems in 67 children with autism (mean chronological age= 51.69 months) and 42 children with developmental delay (mean chronological age= 49.45 months). It was found that children with autism, when compared to DD children, had significantly higher sensory scores on all three sensory constructs (sensory hypo-, hyper-sensitivity and sensory seeking), for total RBS-R total scores and all RBS-R subscales apart from the self-injury subscale. Regression analysis showed that high levels of hyper-responsive behaviours predicted high levels of repetitive behaviours. More specifically, significant correlations were found between hyper-responsiveness and the total RBS-R scores and the following subscales: stereotypies, compulsions, and rituals/sameness behaviours. These relationships remained significant after controlling for mental age. However, hypo-responsiveness, sensory seeking, and repetitive behaviours were not significantly associated with the exception of sensory seeking-ritualistic/sameness behaviours association. In the study that examined the relationship between stereotyped motor behaviours and sensory processing problems in 56 children with autism (mean chronological age= 9.71) and four other groups of children: TD children, DD children, children with visual, and children with hearing impairments, Gal, Dyck & Passmore (2010) found that repetitive behaviours were associated with both sensory hypo- and hyper sensitivity as well as with sensory seeking. In general, studies that explored relationship between sensory problems and repetitive behaviours provided support for the arousal theories of repetitive behaviours in autism.

Finally, studies on anxiety have consistently found that repetitive behaviours were associated with higher levels of anxiety (Sukhodolsky et al., 2008; Rodgers et al., 2012a; Rodgers et al., 2012b; Gotham et al., 2013). Sukhodolsky et al. (2008) examined the frequency of anxiety (by using the Child and Adolescent Symptom Inventory (CASI) and its relation to the core autism features in a group of 172 individuals with autism, aged 5-14 years. They found that 43% of their sample met the cut-off criteria for at least one anxiety disorder and that, in addition to being associated with higher language levels and greater impairment in reciprocal social interaction, higher anxiety levels were also associated with higher levels of stereotyped behaviours (as assessed by ADI-R). Also, in a study that examined the relationship between repetitive behaviours and anxiety in 34 individuals with autism (age range: 8-16 years) and 20 individuals with Williams Syndrome (WS; age range: 6-15 years), Rodgers et al. (2012a) found that, while in the group with autism, higher levels of anxiety were associated with higher levels of repetitive behaviours, this was not the case in

Williams Syndrome (WS) group. Anxiety levels were significantly higher in the group with autism (mean SCAS-P score= 35) than in WS group (mean SCAS-P score= 26).

Only one study has addressed the question of how are different subtypes of repetitive behaviours are associated with anxiety. Rodgers et al. (2012b) used the Spence Children's Anxiety Scale-Parent version (SCAS-P, Spence, 1998) and the Repetitive Behaviour Questionnaire (RBQ; Turner, 1995) with 67 HFA children and adolescents aged 8 to 16 years. They found that the total anxiety score and insistence on sameness behaviours (including rigid routines and restricted interests) were significantly associated and this association was particularly high in children that met the criteria for clinically significant anxiety. Repetitive sensory and motor behaviours were not significantly associated with anxiety. Repetitive motor behaviours such as pacing or rocking may help to soothe anxiety (Rodgers et al., 2012b) while obsessive insistence on sameness may serve to create and maintain anxiety. Finally, a recent study by Gotham et al. (2013) used the anxiety score of the Child Behaviour Checklist (Achenbach & Rescorla, 2001) and an IS score from the Autism Diagnostic Interview-Revised (ADI-R, Rutter, Le Couteur, & Lord, 2003; IS factor was comprised of 6 items) in a very large sample of 1429 children aged 5 ½ to 18 years in order to explore the relationship between anxiety and insistence on sameness. Similarly like Rodgers and colleagues (Rodgers et al., 2012b), Gotham et al. found statistically significant relation between IS and anxiety. Unfortunately, unlike Rodgers et al., Gotham and colleagues did not explore a potential relationship between anxiety and RSM.

6. 1. 3. Summary and aims

The results of the studies reviewed above seem to provide some support for the arousal theory of repetitive behaviours in that repetitive behaviours have consistently been found to be associated with sensory symptoms, e. g. either hyper- or hypo-sensitivity and that several studies have found an association between repetitive behaviours and anxiety. Furthermore, sensory hyper-sensitivity was found to be associated with anxiety in all studies that explored the relationship between anxiety and sensory processing. However, none of the studies have looked at the three-way relationship between repetitive behaviours, sensory problems and anxiety. The aim of this chapter will be to look at the inter-relationship between these behaviours. Furthermore, this chapter will examine our proposal that different subtypes of repetitive behaviours are differently associated with anxiety and sensory problems. Based on the reviewed literature and arousal theories of repetitive behaviours, it is hypothesized that

both subtypes of repetitive behaviours e. g. IS and RM behaviours will be associated with sensory difficulties. Following Rodgers and colleagues, it is further hypothesized that RM behaviours will be primarily associated with sensory problems and not anxiety while IS behaviours will be associated with both sensory problems and anxiety. Furthermore, the interrelationship between IS behaviours, sensory problems and anxiety will be explored in a detailed manner. According to one account (Green & Ben-Sasson, 2010), sensory hyper-sensitivity leads to anxiety while, according to other account, anxiety might be the contributing factor to sensory hyper-sensitivity (Craske, Rauch, Ursano, Prenoveau, Pine, & Zinbarg, 2009; Niles, Mesri, Burklund, Lieberman, & Craske, 2013). Although, in both accounts IS behaviours would serve as a means of reducing sensory problems and anxiety, in one case the relationship between IS and anxiety would be mediated by sensory symptoms and in other, the relationship between IS behaviours and sensory problems would be mediated by anxiety. This issue will be addressed by using a series of mediation analyses.

It is important to note that, in order to avoid artificially inflating the relationship between IS and RM factors with the Sensory Profile Quadrants, sensory items from the RBQ-2 were removed and additional factor analysis was run. This factor analysis will be reported at the beginning of the results section and IS and RM from this factor analysis will be used in all of the subsequent analyses presented in this chapter.

6. 2. Methods

6. 2. 1. Subjects

The same participants described in Chapter 2, and used in Chapter 4 and 5 (N= 49), provided the data reported here. In short, only parents whose children were aged 3.0 years or more were included (as the Sensory Profile is not valid for 2-year-olds) and the sample comprised 49 children and adolescents (45 boys) with a clinical diagnosis of autism, established according to ICD-10 criteria. Children were aged 3 years to 17 years 9 months (mean age= 10 years 7 months, SD= 3 years 10 months). Language items from the background questionnaire were used to establish an expressive language estimate (more details are provided in chapter 2). Of 48 parents who responded to this item, 36 (75%) reported that their child used complex grammatical speech or spontaneous sentences, 8 (17%) that their child used phrase speech and 4 that their child used single words or no speech (8%).

6. 2. 2. Measures

Repetitive Behaviour Questionnaire-2 (RBQ-2). Parents of all children completed the RBQ-2. As noted above, in order to avoid sensory items within the RBQ-2 artificially inflating relations with sensory symptoms, the following sensory RBQ-2 items were excluded: item number 8 (angles), 9 (smell), 10 (feel), 18 (clothes) and also item 7 (fascination) and 19 (foods).

Sensory Profile. All parents completed the Sensory Profile (Dunn, 1999), a caregiver-report measure of a child's/young person's responses to everyday sensory experiences. Poor registration (sensory hypo-sensitivity), Sensory Sensitivity (hyper-sensitivity), Sensory Seeking and Sensory Avoidance quadrants were used in this chapter.

Spence Anxiety Scales. As noted in the previous empirical chapter, 34 parents of 7- to 17-year-olds completed the Spence Children's Anxiety Scale-Parent Version (SCAS-P, Spence et al., 1998) and 15 parents of younger children completed the preschool version (Preschool Anxiety Questionnaire, PAS, Spence et al., 2001). As described in the previous chapter, raw scores were converted into standardized T (total) scores following the procedure described here: http://www.scaswebsite.com/1_1.html.

6. 3. Results

RBQ-2 factor analysis with sensory items excluded

After removing sensory items, the 2-factor solution was retained with Repetitive Motor (RM) factor explaining 17.05% of variance and Insistence on Sameness (IS) factor explaining 36.85% of variance (see Table 6. 1. For details). The Cronbach's alpha for RM factor was .76 and .83 for IS factor.

Table 6. 1. Factor structure of RBQ-2 questionnaire items resulting from 2-factor analysis with sensory items excluded

Questionnaire items within each factor	Item factor loading
Factor 1: Motor/sensory behaviours – 17.05% variance	
2. Repetitively fiddle with toys etc.	.687
3. Spin self around and around	.607
4. Rock backwards and forwards	.721
5. Pace/move around repetitively	.785
6. Repetitive hand/finger movements	.706
Factor 2: Insistence on sameness– 36.85% variance	
12. Collect or hoard items of any sort	.550
13. Insists on things (e.g. in house) remaining the same	.816
14. Gets upset about minor changes to objects	.730
15. Insists on aspects of routine remaining the same	.785
16. Insists on doing or re-doing things in a certain way	.860
17. Plays same music, game, video, book repeatedly	.616

The relationship between repetitive behaviours, sensory problems and anxiety

The relationship between variables was examined in two ways. First, Pearson's correlations were used to examine the relationship between IS, RMB, sensory problems and anxiety. Table 6. 2. shows the significant 3-way associations between anxiety scores, repetitive behaviours and sensory symptoms. Alpha level was adjusted to .01 to reduce Type 1 error. Repetitive motor behaviours (RMB) did not correlate with anxiety but did correlate with sensory seeking, and sensory avoidance at the .01 level. The insistence on sameness (IS) was associated with all sensory symptoms and with anxiety. Anxiety was associated with poor registration (hypo-sensitivity), sensory sensitivity and sensory avoidance

Three way relations were found between anxiety and IS and three of the four sensory quadrants (poor registration (hypo-sensitivity), sensory sensitivity and sensory avoiding).

Table 6. 2. Correlations between RBQ-2, Sensory Profile Quadrants and Anxiety

	Repetitive motor behaviours	Insistence on sameness	Anxiety
Poor registration	-.21	-.38**	-.40**
Sensory seeking	-.42**	-.49**	-.34
Sensory sensitivity	-.31	-.43**	-.61**
Sensory avoiding	-.42**	-.49**	-.71**
Anxiety	.24	.46**	1

* $p < .01$, ** $p < .001$

Secondly, following Rodgers et al.'s (2012b) analysis, children were first divided into anxious and non-anxious group depending on their anxiety scores. Twenty-four children (49%) scored above the clinical cut-off, a proportion equivalent to that found by Rogers et al. ($n = 33$, 49%). The two groups did not differ in age ($p = .26$) or language level ($p = .10$). As can be seen from Table 6. 3., the anxious group had significantly higher scores for the IS factor (Mean IS score = 2.37, $SD = 0.43$), than the non-anxious group (Mean IS score = 2.03, $SD = 0.42$), $t(47) = 2.79$, $p = .008$. However, there was no significant difference between groups for the RMB factor (clinically anxious group: Mean RMB score = 1.86, $SD = 0.43$; non-anxious group: Mean RMB score = 1.83, $SD = 0.42$, $p = .87$). Anxious and non-anxious groups differed significantly on two of the four sensory quadrants (sensitivity and avoiding) with high effect sizes. However, the difference for registration (hypo-sensitivity) and seeking did not reach significance.

Table 6. 3. Differences between groups meeting clinical cut-off on the Spence Anxiety Scale

	Anxious group (<i>n</i> = 24)	Non-anxious group (<i>n</i> = 25)	<i>t</i> test		Cohen's <i>d</i>
	<i>M</i> (<i>SD</i>)	<i>M</i> (<i>SD</i>)	<i>t</i>	<i>p</i>	
Age (months)	135.65 (43.04)	120.59 (49.43)	1.14	.26	0.32
Language Level	5.35 (.89)	4.56 (2.1)	1.72	.10	0.49
Repetitive motor behaviours	1.86 (0.55)	1.83 (0.54)	0.17	.87	0.05
Insistence on sameness	2.37 (0.43)	2.03 (0.42)	2.79	.008	0.80
Registration (hypo-sensitivity)	46.33 (11.62)	52.56 (9.97)	-2.02	.05	0.57
Seeking	86.37 (19.05)	93.20 (14.92)	-1.40	.17	0.40
Sensitivity	57.37 (15.51)	70.12 (11.31)	-3.28	.002	0.94
Avoidance	77.87 (14.48)	94.24 (12.48)	-4.24	.000	1.21

In order to further examine the three way relations found between the insistence on sameness, anxiety, sensory hyper-sensitivity and sensory avoidance, two series of mediation analyses were performed. The first series had the aim to examine the relationship between IS, anxiety and sensory hyper-sensitivity. The second series of mediation analyses examined the mutual relationship between IS, anxiety and sensory avoidance.

The relationship between Insistence on Sameness, Sensory Hyper-Sensitivity and Anxiety

Mediation analysis 1 examined the model where sensory hyper-sensitivity would lead to anxiety which in turn, would lead to IS behaviours. As can be seen from Table 6.4 , this model was supported.

Table 6. 4. Mediation Analysis 1

	R2	R2 Change	Beta
Regression One:			
IS on Sensory Hyper-sensitivity	.183**		.427
Regression Two:			
Anxiety on Hyper-sensitivity	.361***		.612
Regression Three:			
Step 1: IS on Anxiety	.196**		.319
Step 2: IS on Sensory Hyper-sensitivity	.214	.034	.232

Note: *= p<.05; **= p<.01; ***= p<.001

Mediation analysis 2 examined the model where anxiety would lead to sensory hyper-sensitivity which in turn, would lead to IS behaviours. As can be seen from Table 6.5, this mediation model was supported.

Table 6. 5. Mediation Analysis 2

	R2	R2 Change	Beta
Regression One:			
IS on Anxiety	.196**		.461
Regression Two:			
Hyper-sensitivity on Anxiety	.361***		.612
Regression Three:			
Step 1: IS on Sensory Hyper-sensitivity	.183**		.232
Step 2: IS on Anxiety	.214	.064	.319

It has been suggested by Rodgers et al. (2012b) that IS behaviours, although efficient in the short term in reducing anxiety, might in the long term reinforce it. As it has been shown in the above model that anxiety is leading to sensory hyper-sensitivity, a model where IS behaviours would lead to anxiety which in turn would lead to sensory hyper-sensitivity was explored. As can be seen from the table 6. 6., this model was supported.

Table 6. 6. Mediation Analysis 3

	R2	R2 Change	Beta
Regression One:			
Hyper-sensitivity on IS	.183**		.427
Regression Two:			
Anxiety on IS	.196**		.461
Regression Three:			
Step 1: Hyper-sensitivity on Anxiety	.361***		.527
Step 2: Hyper-sensitivity on IS	.375	.027	.184

The relationship between IS, anxiety and sensory avoidance

Mediation analysis 4 examined the model where anxiety would lead to sensory avoidance which in turn, would lead to IS behaviours. As can be seen from Table 6. 7., this model was also supported.

Table 6. 7. Mediation Analysis 4

	R2	R2 Change	Beta
Regression One:			
IS on Anxiety	.196**		.461
Regression Two:			
Sensory Avoidance on Anxiety	.488***		.706
Regression Three:			
Step 1: IS on Sensory Avoidance	.229***		.337
Step 2: IS on Anxiety	.238	.025	.223

Mediation analysis 5 examined the model where IS would lead to Anxiety which in turn, would lead to Sensory Avoidance behaviours. As can be seen from Table 6. 8., this model was also supported.

Table 6. 8. Mediation Analysis 5

	R2	R2 Change	Beta
Regression One:			
Sensory Avoidance on IS	.245***		.495
Regression Two:			
Anxiety on IS	.196**		.461
Regression Three:			
Step 1: Sensory Avoidance on Anxiety	.488***		.607
Step 2: Sensory Avoidance on IS	.515	.036	.215

Mediation analysis 6 examined the model where IS would lead to Avoidance which in turn, would lead to Anxiety. As can be seen from Table 6.9, this model was supported.

Table 6.9. Mediation analysis 6

	R2	R2 Change	Beta
Regression One:			
Anxiety on IS	.196**		.461
Regression Two:			
Avoidance on IS	.245***		.495
Regression Three:			
Step 1: Anxiety on Avoidance	.488***		.633
Step 2: Anxiety on IS	.494	.017	.148

Mediation analysis 7 examined the model where Avoidance would lead to Anxiety which in turn, would lead to IS behaviours. As can be seen from Table 6. 10., this model was supported.

Table 6. 10. Mediation analysis 7

	R2	R2 Change	Beta
Regression One:			
IS on Sensory Avoidance	.245***		.495
Regression Two:			
Anxiety on Avoidance	.488***		.706
Regression Three:			
Step 1: IS on Anxiety	.196**		.223
Step 2: IS on Avoidance	.238	.057	.337

In addition to theoretically driven mediation models described above, the following “dummy” mediation models were also tested: (a) a model where anxiety would lead to IS behaviours which in turn, would lead to sensory hyper-sensitivity; (b) a model where IS would lead to sensory hyper-sensitivity which in turn, would lead to Anxiety; (c) a model where Avoidance would lead to IS which in turn, would lead to Anxiety; (d) a model where Anxiety would lead to IS which in turn, would lead to Avoidance. None of these models was supported.

6. 4. Discussion

The aim of this chapter was to examine the relationship between repetitive behaviours, anxiety and sensory problems in children and adolescents with autism and to test our earlier proposal that different RRBs subtypes are differently associated with anxiety and sensory problems. Correlational analysis found that the insistence on sameness behaviours were highly correlated with anxiety ($r = .461, p < .001$). This finding is consistent with previous findings by Sukhodolsky et al. (2008), Joosten, Bundy and Einfeld (2009), Rodgers et al. (2012a), Rodgers et al. (2012b) and Spiker, Lin, Van Dyke and Wood (2012) who showed that increased levels of anxiety are associated with higher repetitive behaviours scores, in particular with rigid/insistence on sameness behaviours. For example, Spiker et al. (2012) found that restricted interests, a form of insistence on sameness behaviours, serve as a coping mechanism for anxiety. On the other hand, repetitive motor behaviours were not significantly associated with anxiety ($r = .237, p = .101$). These findings were in line with the findings by Rodgers et al. (2012b) who also found no evidence for the association between

motor stereotypies and anxiety in a sample of 67 children and adolescents with HFA. The findings from this chapter, together with the findings by Rodgers and colleagues therefore lend support to our proposal for the selective role of certain types of repetitive behaviours in relation to anxiety.

The findings from the correlational analysis suggest that both the insistence on sameness behaviours and the repetitive motor behaviours are related to sensory problems. More specifically, the insistence on sameness behaviours were significantly correlated with all four subtypes of sensory modulation problems e. g. sensory hypo-sensitivity, sensory hyper-sensitivity, sensory seeking and sensory avoidance. The repetitive motor behaviours were associated with all sensory modulation problems apart from sensory hypo-sensitivity. These findings are in line with the findings by Boyd et al. (2010), Gabriels et al. (2005) and Gal et al. (2010). Also, these findings lend support to the hypothesis put forward by Ornitz and Ritvo (1968; also Ornitz, 1974) and Kinsbourne (1980) who suggested that Individuals with autism, due to problems in the regulation of arousal constantly fluctuate between the states of under-arousal and hypo-stimulation, and over-arousal and hyper-stimulation, and that repetitive behaviours are used as a form of coping mechanism. In the case of over-arousal and hyper-sensitivity; repetitive behaviours provide a soothing function by either avoiding stimulation or providing predictable stimulation that reduces arousal; on the other hand, in cases of under-arousal and hypo-stimulation, repetitive behaviours serve to provide additional stimulation and increase arousal to the optimal level. Suggestions regarding the problems in the regulation of arousal are also supported by the findings from previous chapter that demonstrated that around 90% of children and adolescents showed atypical scores on at least three out of four sensory quadrant subscales, implying fluctuation between the states of over- and under-arousal.

Finally, anxiety was also highly associated with the sensory hyper-sensitivity and sensory avoidance, and moderately with sensory hypo-sensitivity, replicating the findings from previous studies that showed an association between anxiety and sensory problems in Individuals with autism (Green et al., 2012; Ben-Sasson et al., 2008; Mazurek et al., 2012; Pfeiffer et al., 2005).

The next step was to examine the three-way relationship between the insistence on sameness, anxiety and three sensory quadrants that were associated with both IS and anxiety e. g. sensory hyper-sensitivity, sensory avoidance and sensory hypo-sensitivity.

The first set of mediation analyses examined the mutual relationship between the insistence on sameness, anxiety and sensory sensitivity. According to the arousal theory, sensory hyper-sensitivity leads to anxiety and also to IS that serve as means of reducing sensory both hyper-stimulation and consequent anxiety. This theory is similar to the theoretical proposal put forward by Green and Ben-Sasson (2010; see also Green et al., 2012). They suggested that sensory hyper-responsiveness is a causal factor for the development of anxiety by means of classical conditioning. Based on these proposals, a mediation model where sensory hyper-sensitivity would lead to anxiety which would lead to repetitive behaviours was examined. The results confirmed that this model was significant. However, it has also been suggested that anxiety might be a contributing factor to sensory hyper-sensitivity (Craske et al., 2009; Niles et al., 2013). According to this account, anxious individuals are characterized by hyper-arousal and hyper-vigilance to specific types of sensory stimuli, in other words, anxious children constantly scan their environment and are more likely to detect unpleasant and arousing stimuli, which, together with the threat-based affective regulation will lead children to find environmental stimulation as highly aversive and become hyper-responsive to various types of stimuli. In order to test this second account, a second mediation model that examined the possibility that anxiety leads to the sensory hyper-responsiveness which, then in turn leads to the insistence on sameness behaviours was performed. The results confirmed that this second mediation model was significant as well.

Although the fact that both mediation models were significant might seem counterintuitive at first, it is important to have in mind the cross-sectional design of this study. The majority of children were older than 4 and it was not possible to assess whether, in terms of developmental trajectory, sensory problems predated anxiety. A recent study by Green et al. (2012) examined the development of anxiety and sensory over-responsivity (SOR) and the extent to which anxiety might predict changes in SOR and vice versa over a 1-year period. Parents of 149 PDD-NOS toddlers (mean chronological age: 28.3 months) completed the Infant-Toddler Social and Emotional Assessment (ITSEA; Carter & Briggs-Gowan, 2005), Beck Anxiety Inventory (BAI; Beck et al., 1988), and the Center for Epidemiological Studies Depression Scale (CES-D; Radloff, 1977) at two annual time points. At initial assessment, a significantly higher percentage of children met the cut-off criteria for SOR when compared to anxiety (22.6% vs 8.3%). Within 12 months, the percentage of children who met the SOR cut-off criteria significantly increased with 13.5% of children showing anxiety at the follow-up assessment. On the other hand, the percentage of children

who showed SOR has only slightly increased to 23.6% at the follow-up assessment (increase was not statistically significant). Anxiety and SOR were significantly correlated at both time 1 and time 2. The result of a cross-lag analysis suggested that SOR appears earlier than anxiety, and support the theory that SOR may increase the risk of developing anxiety or exacerbate a predisposition for anxiety. It is possible to hypothesise a mechanism where unpleasant sensory stimulation (as the unconditioned stimuli) would become associated with particular objects, situations of wider contexts which would then become conditioned stimuli, capable themselves to cause anxious reaction, in the absence of the sensory stimulation. According to this account, although sensory stimulation would initially be the cause of anxiety, over time, these two problems would exacerbate each other, which would explain significant findings for both of mediation models. Furthermore, both sensory problems and anxiety might be a consequence of common underlying neuropathology such as amygdala dysregulation for example, which would, as a consequence, lead to the dysfunction in the arousal modulation and over-focused attention. In this case, sensory problems would be a developmentally earlier manifestation of such dysfunction, while anxiety would be a later manifestation. However, a prospective study is necessary to examine these possibilities.

The second set of mediation analyses examined the mutual relationship between the insistence on sameness, anxiety and sensory avoidance. Although sensory avoidance has been suggested by Dunn (1997) as a sensory phenomenon, it is, according to this account defined primarily in behavioural terms. Behavioural avoidance is considered as one of the key consequences of anxiety (Rachman, Craske, Tallman, & Solyom, 1986; Craske, Sanderson, & Barlow, 1987; Muris, van Brakel, Arntz, & Schouten, 2011), and it is possible that anxiety is the primary cause of sensory avoidance. Highly significant correlations certainly imply such possibility. A mediation model that examined the possibility that anxiety leads to sensory avoidance which is in turn related to the insistence on sameness behaviours was performed. The results showed that this model was significant. This model supports the proposals from the literature that suggest that anxious individuals avoid unpleasant or anxiety provoking situations and develop a rigid type of behaviours in order to further limit the possibility of encountering anxiety provoking situations. However, Rodgers et al. (2012b) drawing on the cognitive models of anxiety, suggested that although sameness types of behaviour, might reduce anxiety in the short term by means of constraining the environment, this pattern would lead to the development of positive beliefs about these behaviours rather than to the development of more adaptive, flexible behaviours and strategies, and would, in

the long term, perpetuate the negative anxiety cycle. Thus, a second mediation model examining the possibility that the insistence on sameness behaviours lead to the anxiety and in turn, to more avoidance was performed. The results showed that this model was indeed significant. Interpretation of these findings is again constrained by the cross-sectional design of the study, however, results suggest that the possibility that, the insistence on sameness behaviours might originally serve a function of reducing anxiety, however, as suggested by Rodgers et al. (2012b), in the long term, this strategy might actually perpetuate anxiety cycle rather than serving adaptive function of warding off anxiety. This possibility is indeed likely if we have in mind the literature on the relationship between the insistence on sameness behaviours and fears and anxieties during typical development. For example, Evans et al. (2005) have shown that in young children, rigid type of behaviours are increased during the times of transitions which are associated with specific fears and serve to ward off these fears and anxieties, however, as children develop more sophisticated cognitive and affective regulation mechanisms these behaviours begin to decrease. However, a longitudinal study is needed to examine whether there is a lack of such shift in disorders characterized by anxiety and repetitiveness such as OCD and autism, and if so, what is the reason for the absence of such developmental progression.

6. 4. 1. Summary and limitations

Similar limitations that were listed in chapter 4 apply here. In brief, the report by a third party, especially by teachers on repetitive behaviours, sensory problems and anxiety would make the results reported here more generalizable to various settings and contexts. Also, as discussed in Chapter 2, the inferences on the problems in arousal modulation made here are necessarily indirect because of the use of questionnaires to assess sensory modulation in children and adolescents with autism. Although sensory modulation quadrants from the sensory profile have received validation in terms of physiological measures (McIntosh et al., 1999), it will be of crucial importance to follow up the findings from this chapter with experimental work.

This chapter replicated findings from the previous literature on the relationship between repetitive behaviours and sensory problems, between sensory problems and anxiety and between anxiety and repetitive behaviours. However, this chapter, to the best of my knowledge, provided the first examination in the autism literature of the three way relationship between two subtypes of repetitive behaviours (IS and RSM behaviours), anxiety

and the sensory modulation disorders. Theoretical and clinical implications of the findings will be discussed in Chapter 10.

Chapter 7: Anxiety in Parents of Children with Autism: Prevalence and Risk factors

“My child has very severe autism, is extremely anxious, not happy and distressed most of the time, I personally do not see any good in seeing my daughter like that day in day out. I hate it, I feel as a parent a failure, I want to comfort my child but she will fight me and push me away. I am not in a relationship as a mother to her, I am no different to anyone else. I feel isolated and criticised frequently by others, my own confidence has taken a battering, I have been anxious and depressed for years but only got help now, so thank goodness for that.”

Parenting a child with autism is a complex situation of chronic stress for parents, and parents of children with autism have been found to have higher levels of stress and higher prevalence of affective disorders when compared to both parents of typically developing children and parents of children with other disabilities. As autism is increasingly being recognized as one of the most common childhood disorders, and most of the interventions have one or both parents as co-therapists increasing their responsibilities even more, a systematic study of the factors that lead to the development of anxiety and depression in parents is needed in order to help parent to successfully adapt to the challenges related to parenting an individuals with autism. This review will first summarize the studies that have looked at the prevalence of anxiety in parents of children with autism and then consider the contributing factors to the development and maintenance of anxiety in parents.

7. 1. Prevalence of anxiety in parents of children with autism

Parents of children with autism have consistently been found to report more stress and higher levels of anxiety when compared to both parents of typically developing (TD) children and parents of children with other developmental disabilities including Down’s Syndrome, Fragile X syndrome and cystic fibrosis (Bouma & Schweitzer, 1990; Bolton, Pickles, Murphy, & Rutter, 1998; Piven & Palmer, 1999; Abbeduto, Seltzer, Shattuck, Krauss, Orsmond, & Murphy, 2004; DeLong, 2004; Mazefsky, Folstein, & Lainhart, 2008; Kuusikko-Gauffin et al., 2013). For example, Kuusikko-Gauffin et al. (2013) using the Social Phobia and Anxiety Inventory (SPAI; Turner et al. 2004) found that both mothers and fathers of children with autism had significantly higher total SPAI scores as well as subscale scores

when compared with mothers and fathers of TD children. Piven and Palmer (1999) found that parents of the children with autism had significantly higher rates of social phobia, major depressive disorder and OCD symptoms when compared to parents of the children with Down's syndrome. Abbeduto et al. (2004) also found that parents of children with autism had lower levels of well-being than parents of children with Down's syndrome. In addition to this, Abbeduto and colleagues also included a group of parents of children with Fragile X syndrome and found that parents of children with autism had more problems across different measures of well-being including elevated rates of anxiety and depression, and more pessimism. In terms of specific rates of anxiety problems, results between studies are highly variable, ranging from 15.6% to 50% (Bitska & Sharpley, 2004; Mazefsky et al., 2008; Kuusikko-Gauffin et al., 2012). For example, in a study that looked at the incidence and contributing factors to anxiety, depression and stress in parents of 107 children with autism, Bitska and Sharpley (2004) found that nearly half of the participants were severely anxious and nearly two thirds were clinically depressed. Slightly lower rates were reported by Mazefsky et al. (2008) who found that 29% of parents of children with autism from their sample (N= 19 families) met the criteria for elevated anxiety on the Schedule of Affective Disorders and Schizophrenia – Lifetime Version (SADS-L; Spitzer & Endicott, 1978).

Despite the inconsistencies in terms of specific rates of affective disorders reported in various studies which can, at least partially, be attributed to the use of different anxiety assessments, it is clear that anxiety and stress are significantly more prevalent in parents of children with autism than in parents or children without any disorder and parents of children with various neurodevelopmental and somatic disorders. Research to date has mainly considered stress and anxiety in parents as being a consequence of raising a child with autism. However, it has been suggested that more comprehensive, multidimensional models of parental stress and anxiety that consider a range of positive as well as negative factors that might have an impact on well being of parents are necessary (Lazarus, 1993; Mash & Johnston, 1990; Kuhn & Carter, 2006). For example, Mash and Johnston (1990) proposed that parental stress and anxiety are influenced by the following three types of factors: characteristics of the child, characteristics of the parent, and environmental characteristics. Firstly, existing research that has looked at these three factors and their impact on parental anxiety will be summarized and the aims of the first empirical chapter on the anxiety in parents of children with autism will be defined. After this, in the second part of this review chapter, the intolerance of uncertainty and sensory processing problems, the factors that are

currently considered to be the major contributors for the development and maintenance of anxiety in general population, but have not been explored before in the literature on parents of children with autism will be considered and the goals of second empirical chapter on parents will be defined.

7. 2. Characteristics of children with autism as a contributing factor to anxiety in their parents

Previous studies have shown that impairments in communication and reciprocal social interaction, and the presence of restricted and repetitive behaviours are associated with increased stress in parents (Bebko, Konstantareas, & Springer, 1987; Kasari & Sigman, 1997; Hastings & Johnson, 2001; Beck, Hastings, & Daley, 2004; Konstantareas & Papageorgiou, 2006). For example, Bebko et al. (1987) explored a potential impact of various autism symptoms in 20 children with autism on the well-being of their mothers and fathers. Mothers and fathers independently rated the severity of autism symptoms in their child by completing the Childhood Autism Rating Scale (CARS; Schopler et al., 1980) and also evaluated how stressful they found each symptom listed in CARS (on 1 to 4 scale). In addition to parents themselves, the levels of parental stress and characteristics of children that were most likely to have a negative effect were also estimated by the therapists. It was found that all informants reported children's language and cognitive impairment as the most stressful. It is important to note that teachers reported that parents had higher levels of stress than parents did themselves. Hastings & Johnson (2001) investigated predictors of parental stress in 141 families who took part in an intensive home-based behavioural intervention for young children with autism (mean age: 4.9 years) and found that overall levels of autism symptomatology in children (as assessed by the Autism Behaviour Checklist (ABC; Krug, Arick, & Almond, 1980) were associated with higher levels of stress in parents. Problems in social interaction skills as a possible cause of stress in parents were explored by Beck et al. (2004) and Kasari and Sigman (1997). Both studies found that impairments in reciprocal social interaction in children were predictive of the levels of stress in their mothers. Further to this, Kasari and Sigman (1997) found that parents whose children were more socially responsive reported lowest levels of stress.

With regards to the potential influence of repetitive behaviours, Lecavalier, Leone, and Wiltz (2006) found that hyperactivity, stereotypy and repetitive behaviours of children with autism were associated with anxiety levels in their mothers. Similarly, Bishop, Richler, Cain and Lord (2007) interviewed mothers of 110 children with autism (mean age of children

was 9 years) in order to examine predictors of their stress levels. It was found that higher repetitive behaviour scores (using ADI-R) were significant predictors of higher stress levels.

In addition to the diagnostic triad of symptoms, individuals with autism also exhibit deficits in adaptive behaviours (e.g., toileting, getting dressed, taking public transit), numerous “problem” behaviours and co-morbid conditions. Deficits in adaptive behaviours and “problem” behaviours have been reported to contribute to maternal stress, anxiety and depression levels (Konstantareas & Homatidis, 1989; Bishop et al., 2007; Tomanik, Harris, & Hawkins, 2004). For example, Hastings (2003) found that behavioural problems of children with autism (N= 18, mean age: 11.8 years) were associated with higher levels of anxiety and depression in their parents. Fitzgerald, Birkbeck, and Matthews (2002) found that lower levels of adaptive behaviours in children with ASD were predictive of elevated stress levels and reduced well being in their mothers. However, other studies failed to find association between stress and anxiety in parents and adaptive skills in their children (Beck et al., 2004; Lecavalier et al., 2006; Estes, Munson, Dawson, Koehler, Zhou, & Abbott, 2006; Hastings et al., 2005). In general population, sleep problems, restricted diet and epilepsy in children have consistently been found to be associated with more stress and anxiety in parents (Richdale, Gavidia-Payne, Francis, & Cotton, 2000; Doo & Wing, 2006; Cushner-Weinstein et al., 2008). However, there has been little research on the association between these co-morbid problems in children with autism and stress and anxiety in their parents. This is surprising considering the fact that these co-morbid conditions are very prevalent in autism (Rzepecka, McKenzie, McClure, & Murphy, 2011; Wang, Tancredi, & Thomas, 2011; Geier, Kern, & Geier, 2013). One of the rare exceptions is a study by Wiggs and Stores (2001) that reported that stress levels in mothers reduced after successful sleep intervention with their children. Furthermore, mothers of children in the treatment group had lower stress levels when compared to mothers of ASD children who did not participate in sleep intervention.

According to the wear-and-tear hypothesis of care giving for an ill or disabled family member (Townsend, Noelker, Deimling, & Bass, 1989) long term care providing care will lead to the reduction in personal well-being and the development of anxious and depressive symptoms. However, studies that have explored the impact of child’s chronological age on parent’s anxiety and well being have mostly found the opposite trend with parents of older children with autism reporting less stress and anxiety than parents of younger children (Seltzer & Krauss, 2001; Tomanik et al., 2004; Seltzer, Floyd, Song, Greenberg, & Hong, 2011). For example, in one of the earliest studies that considered a possible impact of

children's chronological age on the levels of stress in mothers, Koegel et al. (1992) found that mothers of children who were younger than 7 years reported higher levels of stress than mothers whose children were older. In a study that examined the change in well-being of mother of adolescent and young adult children with autism, Lounds, Seltzer, Greenberg and Shattuck (2007) found that mothers' well-being improved over time. However, it is important to note that despite improvements, mothers still had elevated levels of anxious and depressive symptoms when compared with mothers of children without autism. When considering the impact of child's age on the well-being of parents it is important to stress the fact that it is unlikely that anxiety would change linearly over time but rather that certain periods in the life of a child might be more stressful than others. Studies in the general population suggest that adolescence is a period of reduction of well-being in parents (Putnick, Bornstein, Hendricks, Painter, Suwalsky, & Collins, 2010). These findings in general population were mirrored by findings of Bishop et al. (2007) who indeed found that parents of adolescents with autism had higher levels of anxiety than parents of younger children.

In summary, the findings from the studies that explored the relationship between the characteristics of children and anxiety in parents have been somewhat conflicting. Numerous studies found the evidence to support the hypothesis that characteristics of children such as core autism symptoms, cognitive and adaptive level, chronological age, co-morbid condition and various "problem behaviours" are associated with higher levels of anxiety in parents. However, as the literature review shows, a significant number of studies failed to find the relationship between the anxiety in parents and the specific children's characteristics.

7. 3. Characteristics of parents as a contributing factor to their own levels of anxiety

Although caring for a child with disability is challenging, it has been shown that these challenges are not only possible to overcome but that the experience of having a child with a disability may strengthen families, expand their social network and lead to increased personal growth in parents and other family members (Turnbull, Behr, & Tollefson, 1986; King, Zwaigenbaum, Bates, Baxter, & Rosenbaum, 2012). From the brief review of literature on the prevalence of anxiety in parents provided at the beginning of this chapter, it is clear that at least half of the parents do not show elevated levels of anxiety and it is plausible that certain parents' own personal characteristics might on one hand put them at increased risk for developing affective problems, and on the other, protect them from anxiety despite the problems they face in their everyday lives. Two parents' characteristics that have been addressed in the existing research as a potential risk or protective factors for the development of anxiety are: the presence of the Broader Autism Phenotype (BAP) and strategies that parents employ to cope with problems and stress that caring for a child with autism brings.

7. 3. 1. Broader Autism Phenotype (BAP) as a risk factor for the development of anxiety in parents

BAP has been defined as the presence of autistic traits, most commonly the impairments in social interaction and communication skills in the family members of individuals with autism, but exhibited to a lesser degree. It has been suggested that BAP may act as a risk factor for the development of the affective disturbances, either directly (affective disorders being a feature of BAP and sharing genetic liability with BAP) or indirectly (BAP traits may interfere with various aspects of functioning and therefore put individuals at risk for the development of the affective disturbances). Findings from the two studies by Ingersoll and colleagues suggest that affective disorders in parents of children with autism can indeed be considered as a consequence of the BAP traits. In the first study (Ingersoll, Meyer, & Becker, 2010), 71 mothers with an autistic child and 94 mothers with a child without autism and without developmental delay completed the Family Impact Questionnaire (FIQ; Donenberg & Baker, 1993), the Center for Epidemiological Studies-Depression Scales (Radloff, 1977) and the Autism Spectrum Quotient (AQ, Baron-Cohen et al., 2001). Mothers of children with autism had significantly more depressive symptoms, higher stress levels, and BAP traits than mothers of children without autism. Furthermore, they found that the

depression scores of mothers of children with autism were predicted by BAP traits, even after controlling for the characteristics of their children. In the second study, Ingersoll and Hambrick (2011) found that positive association between both the children's symptom severity and maternal BAP traits with the presence of depression and elevated stress levels in mothers. Based on these findings, Ingersoll and colleagues concluded BAP traits place parents of children with autism at increased risk for affective disorders.

However, these results are not supported by findings of 5 other studies (Bolton et al., 1998; Piven & Palmer, 1999; Murphy, Bolton, Pickles, Fombone, Piven, & Rutter, 2000; Micali, Chakrabarti, & Fombone, 2004; Kuusikko-Gauffin et al., 2012). Bolton et al. (1998) used Maudsley version of the SADS-L and Family History Interview (FHI) to assess affective and other psychiatric disorders in 218 first-degree relatives of 99 children with autism, and in 87 first degree relatives of children with Down's syndrome, and found that affective disorders, in addition to motor tics and OCD were significantly more frequent in relatives of children with autism. Bolton et al. reported that OCD symptoms were associated with BAP (measure by the Personality Assessment Schedule (Tyrer & Alexander, 1979), but found no evidence to support BAP-affective disorders association. Since the majority of parents of children with autism reported the first episode of affective disorder prior to the birth of a child, Bolton et al. (1998) concluded that affective disorders were not likely to be a consequence of difficulties that parents might face in raising a child with autism. Similarly, Piven and Palmer (1999) found no significant association between BAP and either major depressive disorder or social phobia in parents of children with autism and also, that a substantial proportion of the parents reported the onset of depression and anxiety prior to the birth of the child with autism.

Finally, Murphy et al. (2000) used an adapted version of the Personality Assessment Schedule (Tyrer & Alexander, 1979) as a measure of BAP and Maudsley version of the SADS-L and Family History Interview (FHI) as assessment of affective and other psychiatric disorders to a sample of adult relatives of 99 children with autism and adult relatives of 36 children with Down's syndrome. Murphy and colleagues found evidence of increased levels of anxious, impulsive, aloof, shy, over-sensitive, irritable and eccentric traits in relatives of children with autism when compared to relatives of children with Down's syndrome. Murphy and colleagues conducted a factor analysis and found that identified traits in relatives of children with autism grouped into 3 broad groups of traits which they named 'withdrawn', 'difficult' and 'tense'. The 'withdrawn' and 'difficult' group of traits appeared to reflect

problems in social functioning, were more frequent in male relatives and were associated with a measure of BAP. The third group of traits named ‘tense’ appeared to be anxiety-related traits. These traits were more frequent in female relatives and did not show a significant association with BAP. Findings of no association between BAP and affective problems in parents were also replicated by Micali et al. (2004) who used a semi-structured questionnaire (designed by the authors) to gather information on developmental, medical and psychiatric history of parents and other family members as well as information on parents’ current medical and psychiatric problems.

In summary, findings from the literature that explored the relationship between the presence of BAP in parents and levels of anxiety are highly inconsistent, with Ingersoll et al. (2010) and Ingersoll and Hambrick (2011) reporting the evidence to support such relationship and other studies not being able to replicate these findings (Bolton et al., 1998; Piven & Palmer, 1999; Murphy et al., 2000; Micali et al., 2004; Kuusikko-Gauffin et al., 2012).

7. 3. 2. Coping in parents of children with autism as a protective/risk factor for the development of anxiety

Coping has been defined as “the cognitive and behavioural efforts used to manage a stressful situation and to manage the negative emotions that result from the stressful situation” (Lazarus & Folkman, 1984) and a distinction is usually made between the problem solving/engagement (approach) and avoidant/disengagement styles of coping (Folkman & Lazarus, 1980; Lazarus & Folkman, 1984; Folkman & Lazarus, 1988; Lazarus, 1993; Holahan, Moos, Holahan, Brennan, & Schutte, 2005). While problem solving or approach coping is aimed at dealing with the stressor and involves changing the troubled person-environment relationship by instrumental actions, avoidant/disengagement coping is aimed at escaping or avoiding the stress provoking situation, rather than actively changing the source of stress. In general, high problem-focused coping and low emotion-focused coping is suggested to serve as a protective factor against the development of affective disorders (Seltzer & Krauss, 2001; Seltzer et al., 2011). More specifically, it has been found that parents of children with autism, when compared to parents of both TD children and parents of children with other neuro-developmental disorders, used more avoidant coping strategies. Furthermore, it was also found that a higher use of escape-avoidance was associated with higher levels of anxiety (Sivberg, 2002; Hastings et al., 2005; Dabrowska & Pisula, 2010).

For example, Hastings et al. (2005) examined the strategies that 89 parents of preschool and 46 parents of school-aged children with autism used to cope with the stresses related to caring for their child. It was found that parents who adopted the escape-avoidance as a coping strategy reported more stress and mental health problems; in contrast, those using the positive reframing strategies reported less stress. Sivberg (2002) compared the coping behaviours of 66 parents of children with autism to those of 66 parents with non-autistic children. Parents with a child with autism showed more distancing and escape coping strategies, while the parents in the control group showed more self-control and problem solving coping strategies. Dabrowska and Pisula (2010) found higher levels of stress in mothers and fathers of children with autism when compared to stress levels in parents of TD and children with Down's syndrome and that emotion-oriented coping (the Coping Inventory for Stressful Situations (Endler & Parker, 1999) was used as a coping measures) was associated with higher stress levels in the parents of children with autism and Down syndrome. Similar findings were also reported by Smith et al. (2008).

Although the studies reviewed above clearly suggest that certain types of coping such as the escape-avoidance are a risk factor for anxiety, the question of why parents use this particular coping style has not been addressed in any detail. The only factor that has been explored as a potential contributor to the style of coping that parents might use is the age of their child. Gray (2002) suggested that parents of very young children with autism are motivated to use problem-solving strategies as they expect that their child will make substantial progress as he/she gets older. However, as the majority of children, although showing some improvements, do not "grow out of autism", parents' motivation to use problem solving strategies might reduce over time and they might shift towards using emotion coping strategies in order to manage their own negative feelings. Another reason that parents of younger children with autism might use more problem-focused styles of coping when compared to parents of adolescents and adults is the fact that currently, much more support exists for very young children (Vismara & Rogers, 2010). Only one study has tested the hypothesis that the age of children, or in other words, the changes in the cycle of caring have influence on the type of coping used by parents (Hastings et al., 2005). However, Hastings and colleagues found no relationship between the age of children and the coping style in parents.

In summary, several studies have shown that the use of emotion-focused coping strategies, especially the escape-avoidant style of coping, are associated with higher stress and anxiety levels in parents. However, despite these findings, the question of why parents of children with autism resort to using coping strategies that have negative impact on their well-being, has not been addressed in the literature.

7. 4. Characteristics of the environment as contributing factors to the development of anxiety

The term environment refers to all factors other than characteristics of children with autism and characteristics of parents themselves that might influence anxiety in parents. One of the most commonly researched environmental factors is social support (Boyd et al., 2002). Literature distinguishes between the two main types of social support: formal support defined as any form of assistance that is offered or provided through an organized group, institution or agency, either for free or in exchange for a fee; and informal social support that refers to the support network that may include spouses/partners, immediate or extended family, friends and other parents that have children with either autism or other conditions (Schopler & Mesibov, 1984). In one of the first studies that looked at the relationship between social support and anxiety in parents of children with autism, Bristol (1984) found that parents who had the lowest levels of anxiety received the greatest levels of both informal (support from their spouses and relatives) and formal support (in terms of intervention services). A systematic review by Boyd (2002) confirmed that a low level of social support was one of the most important predictors of stress, anxiety and depression in parents of children with autism and numerous other studies conducted since have also reported that lower levels of both formal and informal support are associated with higher levels of stress and anxiety in parents of children with autism (Dunn, Burbine, Bowers, & Tantleff-Dunn, 2001; Hastings et al., 2005; Pakenham, Samios, & Sofronoff, 2005).

Another environmental factor that has been explored is the number of siblings of the child with autism. There are two elements at play here. Firstly, there is strong evidence for the increased risk of autism and BAP in siblings of children with autism (Gamliel, Yirmiya, & Sigman, 2007; Losh, Childress, Lam, & Piven, 2008), and they also exhibit various problem behaviours at higher rates than siblings of children without autism (Rodrigue, Geffken, & Morgan, 1993). It would not be surprising then that parents experience increased stress and anxiety due to caring for multiple children with autism or for a child with autism

and one or more children that exhibit autism-related features, cognitive impairments and various problem behaviours. Secondly, even if siblings of children with autism are typically developing, parents might still blame themselves for concentrating most of their parenting efforts on the child with autism and giving less attention to TD sibling, which can have a negative impact on parents' well-being. However, the results of the studies have been conflicting so far, with Bishop et al. (2007) finding that mothers with fewer children reported higher levels of anxiety and Tomblin et al. (2003) finding the opposite pattern.

A literature review on the environment as a factor in parental anxiety suggests that a lack of social support, be it formal or informal, is associated with the higher levels of anxiety in parents. As noted above, it is still not clear what the relationship is between the number of siblings of children with autism and the levels of anxiety in their parents.

The aim of Chapter 8 is to first examine the frequency of anxiety in parents of children and adolescents with autism and then to examine the influence of children's, parents' and environmental characteristics on the anxiety in parents. Characteristics that will be evaluated in this first chapter are the characteristics that have been previously evaluated in the literature. On the part of the children, the characteristics that will be evaluated are: chronological age, core autism characteristics, and the presence of co-morbid conditions. On the part of the parents, the relationship between anxiety and the type of coping style that is used by parents and the presence of BAP will be looked at. In terms of environmental characteristics, this chapter will concentrate on the social support and the number of siblings and their possible relationship with the levels of anxiety in parents will be explored. A particular emphasis in this chapter will be on exploring the structure of coping strategies.

As reviewed above, although parental coping styles and the presence of broader autism phenotype traits have been considered as potential risk factors for anxiety in parents of children with autism, risk factors that have been associated with affective disorders in general population have not been explored. After conducting an exhaustive literature review on risk factors for affective problems in general population, Intolerance of Uncertainty and Sensory Sensitivity were identified as major risk factors for the development and maintenance of affective problems. This literature will be briefly summarized below.

7. 5. Intolerance of Uncertainty

Intolerance of uncertainty (IU) is defined as a “dispositional tendency of an individual to consider the possibility of a negative event occurring as both unacceptable and threatening, irrespective of the actual probability of its occurrence” (Freeston, Rhéaume, Letarte, Dugas, & Ladouceur, 1994; see also Buhr & Dugas, 2002; Ladouceur, Gosselin, & Dugas, 2000; Buhr & Dugas, 2002; Korener & Dugas, 2006;).

IU was initially considered to be a specific risk factor for the Generalized Anxiety Disorder (GAD; Freeston et al., 1994; Dugas, Gagnon, Ladouceur, & Freeston, 1998; Dugas & Koerner, 2005). For example, both Ladouceur et al. (1995) and Dugas et al. (2005) reported that, when compared with individuals who had phobic disorder, social anxiety, and depression, individuals with GAD had significantly higher IU scores. However, recent evidence suggests that rather than only being associated with GAD, IU is risk factor for other types of anxiety disorders, for OCD and depression. For example, Tolin, Abramowitz, Brigidi, & Foa (2003) found that compulsive checkers and repeaters had higher IU scores than obsessive-compulsive non-checkers and non-anxious individuals. Oglesby, Medley, Norr, Capron, Korte and Schmidt (2013) found that IU was a unique predictor of hoarding behaviours. Also, Holaway, Heimberg, and Coles (2006) found that individuals with GAD and OCD showed comparable levels of IU and significantly higher IU scores when compared to non-anxious individuals. Furthermore, Steketee, Frost, and Cohen (1998) found that IU was the unique predictor OCD symptoms above both the mood and worry.

IU has also been reported as a risk factor for social anxiety. Both Boelen and Reijntjes (2009) and Carleton, Collimore and Asmundson (2010) reported that IU explained a unique variance in the symptoms of social anxiety, even after controlling for fear of negative evaluation, anxiety sensitivity, positive and negative affectivity, low self-esteem, worry, and neuroticism. There is accumulating evidence that IU is also associated with depression (van der Heiden, Melchoir, Muris, Bouwmeester, Bos, & van der Molen, 2010). Indeed, IU is currently considered to be a “transdiagnostic” factor – it crosses clinical diagnostic categories of both anxiety disorders and depression (Boelen & Reijntjes, 2009; McEvoy & Mahoney, 2011). In addition to the studies reviewed above, this hypothesis is supported by the findings that IU is the main contributing factor for anxiety and depression even after controlling for various other risk factors such as neuroticism (Boelen & Reijntjes, 2009), anxiety sensitivity (Boelen & Reijntjes, 2009, Carleton et al., 2010, Dugas, Gosselin, & Ladouceur., 2001), fear

of anxiety (Buhr & Dugas, 2009), meta beliefs about worry (de Bruin, Rassin, & Muris, 2007; Dugas et al., 2007), and positive and negative affectivity (Carleton et al., 2010).

Despite the reviewed findings that suggest the key role of the intolerance of uncertainty in the development of anxiety in general population, so far no studies have considered the contribution of intolerance of uncertainty to anxiety in parents of children with autism. As noted in the first section, although parents of children with autism are faced with similar problems that are undoubtedly stressful, only a subset of parents develops clinically significant levels of anxiety and depression. Although stress and anxiety are related concepts, increased levels of stress do not necessarily lead to the development of anxiety and depression. According to the diathesis-stress theories, in general, individual differences are the deciding factor in whether negative and stressful life events will lead to the development of psychiatric symptoms or not (Dohrenwend & Dohrenwend, 1981; Monroe & Simons, 1991). As uncertainty abounds in everyday lives of parents of children with autism, IU would certainly present a significant risk factor for developing anxiety and/or depression. Parents with pronounced intolerance of uncertainty trait would be less able to successfully adapt to the stressful life events and would have greater number of anxiety symptoms than parents who do not exhibit pronounced intolerance of uncertainty traits. The support for this hypothesis comes from two longitudinal studies which have shown that what distinguished individuals who developed anxiety from individuals who had the same stressors in their everyday lives but didn't develop anxiety was the presence of IU (Ciarrochi, Said, & Deane, 2005; Chen & Hong, 2010).

The Relationship between intolerance of uncertainty and anxiety in parents of children with autism will be explored in Chapter 9. Based on the literature reviewed in this section, it is expected that parents who are anxious will show higher levels of intolerance of uncertainty. As mentioned above, sensory processing problems are another factor that is, in addition to the intolerance of uncertainty, considered to put individuals at risk for anxiety. However, the potential presence of sensory processing problems in parents of children with autism has not been explored before. The literature that suggests why it is important to consider sensory processing in parents, and especially in relation to anxiety, coping styles and intolerance of uncertainty will be briefly presented below.

7. 6. Sensory Processing in Parents of Children with Autism

Research has shown that sensory problems are not specific to autism but are also prevalent in various other neurodevelopmental and psychiatric conditions such as ADHD, Fragile X syndrome and schizophrenia (Mangeot et al., 2001; Brown, Cromwell, Filion, Dunn, & Tollefson, 2002). Importantly, sensory problems also occur in individuals without any formal diagnosis as well as in around 15% of general population (Aron & Aron, 1997). Additionally, findings from the general literature show that sensory processing problems are some of the main contributing factors for the development of affective disorders in typical population (Aron & Aron, 1997; Ahn, Miller, Milberger, & McIntosh, 2004; Goldsmith, van Hulle, Arneson, Schreiber & Gernsbacher, 2006; Ben-Sasson, Carter, & Briggs-Gowan, 2009). For example, Hofman and Bitran (2008) used the Highly Sensitive Person Scale (HSPS; Aron & Aron, 1997) to examine the sensory processing sensitivity in 89 adults (mean age: 30 years) with social anxiety disorder and found that sensory sensitivity is associated with both harm and agoraphobic avoidance. Liss, Mailloux, and Erchull (2008) found that, in a sample of 201 psychology students (142 females and 59 men), sensory processing sensitivity was associated with anxiety and depression. Kinnealey and Filipek (1999) examined the levels of anxiety and depression in two groups of adults: one group of 15 individuals who showed sensory defensiveness (mean age: 32.60 years) and the other group of 16 individuals without sensory problems (mean age: 33.10 years). They found that anxiety and depression symptoms were significantly higher in sensory defensive adults. However, despite the evidence of association between anxiety and sensory sensitivity in general population, in addition to the existence of such relationship in autism, ADHD and other developmental disorders, so far no studies have explored sensory problems in parents of children with autism or a potential relationship between these problems and anxiety.

The presence of sensory problems in parents of children with autism will be explored in Chapter 9. Further to this, the potential relationship between sensory problems and anxiety in parents will also be explored. Based on the reviewed literature, it is expected that anxiety will be associated with sensory problems. As previously mentioned, the presence of sensory problems in parents of children with autism has not been explored before; however, based on the findings that sensory problems have a genetic component (Goldsmith, Buss & Lemery, 1997; Zawdaski, Strelau, Wlodzimierz, Riemann, & Angleitner, 2001; Goldsmith et al., 2006), it is expected that an increased frequency of atypical sensory processing will be found.

7. 7. Relationship between Intolerance of Uncertainty, Sensory Sensitivity and Coping Styles

As noted in the above, at present, it is not clear what are the contributing factors to the choice of coping strategies in parents of children with autism. There is a strong reason to explore the role of intolerance of uncertainty and sensory sensitivity in the choice of coping strategies that are employed by parents of children with autism.

It has been suggested that coping has two elements: appraisal of the situation followed by the choice and use of coping style deemed as appropriate for the situation based on the appraisal (Lazarus & Folkman, 1984; Volrath & Torgersen, 2000). Furthermore, it was suggested by Lazarus (1993) that personality determines coping style that people use to deal with stress by influencing the appraisal of stressful situation. Taking this into account, there is a strong reason to explore the role of intolerance of uncertainty and sensory sensitivity in the choice of coping strategies that are employed by parents of children with autism. A high level of intolerance of uncertainty has been shown to affect both the way in which an individual perceives information in stressful situations and the manner in which they respond to uncertain information. For individuals with pronounced intolerance of uncertainty traits, even the minimal uncertain elements of a stressful event are perceived, experienced as threatening and the negative effect of those elements is amplified (Dugas & Koerner, 2005; Koerner and Dugas, 2008). Due to perceiving various problems as threatening, individuals with pronounced intolerance of uncertainty have been shown to be poor problem-solvers as they lack confidence and the sense of control (Dugas et al., 1998; Behar, DiMarco, Hekler, Mohlman, & Staples, 2009). This can lead to cognitive and emotional avoidance that are efficient in reducing stress and anxiety in a short term, however, these avoidant strategies will in the long term result in more emotion and cognitive avoidance instead of efficient problem orientation and the use of appropriate coping strategies. Also, being easily overwhelmed with environmental stimulation can be associated with avoidant behaviours and less flexibility in dealing with complex situations. Kinnealey, Oliver and Wilbarger (1995) found that in order to cope with the discomfort of being sensory sensitive, individuals from their sample most frequently employed strategies that included avoidance of situations where they could be easily overwhelmed by environmental stimulation and maintaining very predictable routines.

Chapter 9 will explore the relationship between the intolerance of uncertainty and anxiety in parents of children with autism. It will also explore the prevalence of sensory problems and the potential relationship between sensory sensitivity and anxiety in parents.

Finally, this chapter will explore the relationship between both intolerance of uncertainty and sensory sensitivity, and the dominant coping strategy that parents might use.

Chapter 8: Prevalence of Anxiety in Parents of children with autism and contribution of children's, parents' and environmental characteristics

Chapter Plan:

This chapter will provide a brief overview of the key findings from the literature on the prevalence of anxiety in parents of children with autism and summarize the literature that has explored potential contributing factors to the development and the maintenance of anxiety in parents of children with autism. The literature overview on the contributing factors will be divided into the following subsections: children's characteristics, parents' characteristics and factors related to the environment. Then, the data from this thesis that examined the influence of the above mentioned factors on the presence of anxiety in parents will be presented and these results will be discussed in the light of the existing literature.

8. 1. Introduction

8. 1. 1. Prevalence of anxiety in parents of children with autism

There is a marked heterogeneity in terms of the exact frequency of anxiety between studies, with reported rates ranging from 15 to 50% (Bitska & Sharpley, 2004; Mazefsky, Folstein, & Lainhart, 2008; Kuusikko-Gauffin et al., 2012); however, it is clear that anxiety is prevalent in the population of parents with Children with autism and that they show higher levels of anxious symptoms when compared to parents of either TD children or children with various neurodevelopmental and somatic disorders (DeLong, 2004; Mazefsky et al., 2008; Kuusikko-Gauffin et al., 2012).

The first aim of this chapter will be to examine the frequency of anxiety in parents of children with autism. Based on the findings from the literature, it is expected that anxiety will be a prevalent problem among parents.

8. 1. 2. Risk factors for anxiety in parents of children with autism

Research to date has mainly considered the impact that individuals with autism have on the levels of stress and affective disorders in their parents, and several studies have also focused on some of the characteristics of parents e. g. coping strategies used to manage stressful events and the presence of Broader Autism Phenotype (BAP), as well as other factors such as social support and family environment. The second main aim of this chapter is

to explore the contribution of these factors to the levels of anxiety in parents of children with autism. A summary of these characteristics will be briefly presented below. A detailed overview of this literature was provided in the previous chapter (Chapter 7), and only a very brief summary will be provided here.

8. 1. 2. 1. Characteristics of children with autism as contributing factors to the anxiety in their parents

The majority of studies have focused on the possible contribution of the impairments in social interaction and communication in children to stress and anxiety in parents and in general, found that parents who experienced higher levels of stress and reported higher anxiety levels had children who were less socially responsive and had more difficulties in communication (Bebko et al., 1987; Kasari & Sigman, 1997; Hastings & Johnson, 2001; Beck et al., 2004; Konstantareas & Papageorgiou, 2006). Although clinicians report repetitive behaviours as the most difficult and stressful autistic feature (Bishop et al., 2007), only a few studies have examined the relationship between restricted, repetitive behaviours in children and anxiety in parents and found that the increased frequency and severity of repetitive behaviours in children were associated with higher levels of anxiety in mothers (Lecavalier, Leone, & Wiltz, 2006; Bishop et al., 2007). Several studies have found that lower levels of adaptive behaviours in children were associated with higher levels of stress and anxiety in their parents (Fitzgerald, Birkbeck, & Matthews, 2002; Hastings, 2003; Hall & Graff, 2010). However, it is important to highlight the fact that other studies failed to find an association between elevated stress levels in parents and adaptive skills in their children (Beck et al., 2004; Lecavalier, Leone & Wiltz, 2006; Hastings et al., 2005). In terms of co-morbid problems, gastrointestinal problems and sleep problems have been found to be associated with the increased levels of stress and anxiety (Wiggs & Stores, 2001).

The second aim of this chapter will be to evaluate the relationship between anxiety in parents and the following characteristics of their children: chronological age, problems in communication, social interaction, the presence of repetitive behaviours, and the presence of co-morbid problems. Although some of the findings from the literature have been inconsistent, it is expected that core autism problems, especially repetitive behaviours in children, will be associated with the increased levels of anxiety in parents. It is also expected that the presence of co-morbid problems and younger chronological age of children will also be related to the anxiety levels in parents

8. 1. 2. 2. Characteristics of parents as a contributing factor to their own levels of anxiety

It has been suggested that anxiety problems commonly seen among parents of children with autism, might be, at least partially, considered as a consequence of their own Broader Autism Phenotype (BAP) traits. In two studies (Ingersoll, Meyer, & Becker, 2010; Ingersoll & Hambrick, 2011), Ingersoll and colleagues found that BAP traits (social and communication subscales of AQ) uniquely predicted the number of affective symptoms for mothers of children with autism, even after controlling for child autism severity. However, several other studies failed to find support for the relationship between BAP traits and anxiety in parents (Bolton et al., 1998; Piven & Palmer, 1999; Murphy et al., 2000; Micali, Chakrabarti, & Fombone, 2004).

Coping strategies and their relationship with anxiety have also been explored in the population of parents of children with autism and it has been found that parents of children with autism, when compared to parents of both TD children and parents of children with other neuro-developmental disorders, used more emotion focused strategies, in particular escape-avoidance style of coping. Furthermore, it was also found that a higher use of escape-avoidance was associated with higher levels of anxiety (Sivberg, 2002; Hastings et al., 2005; Smith et al., 2008; Dabrowska & Pisula, 2010). However, despite these findings that suggest that emotion style of coping is a risk factor for anxiety in parents of children with autism, studies conducted thus far have not addressed the question of why parents use particular styles of coping in any detail. As discussed in more detail in Chapter 7, the only factor that has been considered in relation to the choice of a particular coping style is the age of children with autism, however, no relationship between the age of children and coping style in parents was found (Hastings et al., 2005).

The third aim of this chapter will be to examine the relationship between anxiety in parents and the empathy traits (as proxy for BAP traits). Based on the results of previous studies, it was predicted that no strong association will be found between BAP and parents' anxiety and that escape-avoidance will be the primary coping strategy used by anxious parents of children with autism. This chapter will also examine the hypothesis that a younger age of children might be associated with the higher use of problem-focused coping strategies and that an older age of children will be associated with the higher use of escape-avoidance style of coping. Based on the fact that only Hastings et al. (2005) has examined this issue, it is difficult to make any predictions regarding the results. Also, although this question has not been previously addressed in the literature on the coping in parents of children with autism,

this chapter will also examine the association between BAP traits in parents and their style of coping.

8. 1. 2. 3. Characteristics of the environment as contributing factors to the development of anxiety

Numerous studies have consistently found that insufficient social support is a risk factor for elevated levels of stress and anxiety in parents of children with autism (Boyd et al., 2002; Hastings et al., 2005; Pakenham, Samios, & Sofronoff, 2005). Another environmental factor that has been explored is the number of siblings that a child with autism has. However, findings have been inconsistent with Bishop et al. (2007) finding that mothers with fewer children reported higher levels of anxiety and Tomblin et al. (2003) finding the opposite pattern.

The fourth aim of this chapter will be to examine the relationship between anxiety in parents and the support they receive. Also, the relationship between parents' level of education and their anxiety will be examined. Finally, the potential relationship between the number of siblings and anxiety will also be addressed. Based on the findings from the literature, it was expected that higher levels of support will be associated with lower levels of anxiety in parents.

8. 1. 3. Summary of the chapter aims

This chapter will examine the prevalence of anxiety in parents of children with autism. Also, as the literature review has identified the key characteristics of children, characteristics of parents and environmental factors that might serve as risk factors for the anxiety in parents, this chapter will evaluate the contribution of these factors.

8. 2. Methods

8. 2. 1. Subjects

A detailed description of participants was provided in Chapter 2. In brief, the sample for this chapter comprised 50 mothers of children and adolescents with ASD, the mean age of children was 10.6 years ($SD= 4.1$; range 3.0 to 17.7 years). All children had a clinical diagnosis of an ASD, established according to ICD-10 criteria. Exclusion criteria were the

same as the ones described in the Chapter 2. Descriptives of the sample are provided in Table 8.1.

Table 8. 1. Characteristics of children and their mothers

Children's Variables	
Expressive Language	-34 (68%) complex grammatical speech or spontaneous sentences -8 (16%) phrase speech -7 (14%) single words or no speech (11.9%)
Reciprocal Social Communication	-11 (22.9%) none/communicating own needs -24 (50%) communicating only own needs or interests or interests -13 (27.1%) able to share other people's interests -Data missing for 2 cases
Co-morbid conditions	-18 (36%) no co-morbid conditions -32 (64%) co-morbid condition present
Mothers' variables	
Educational Level of parents	-17 (37.8%) postgraduate qualifications -18 (40%) undergraduate or vocational qualifications -10 (22.2%) did not have post-school qualifications -Data missing for 5 cases
Marital Status	-38 (76%) married -12 (24%) divorced/not married
No of children	-30 (60%) only one (child with autism) -20 (40%) more than one

8. 2. 2. Measures

A detailed overview of the measures is provided in Chapter 2.

8. 2. 2. 1. Measures related to parents' characteristics

Hospital Anxiety and Depression Scale (HADS, Zigmond & Snaith, 1983) was used to assess anxiety and depression.

The Ways of Coping Checklist-Revised (WCC-R; Vitaliano et al., 1985; Folkman & Lazarus, 1988) was used to explore the avoidant and problem-solving coping strategies.

The Interpersonal Reactivity Index (IRI; Davis, 1994) was used to examine cognitive and affective empathy.

8. 2. 2. 2. Measures related to children's characteristics

The measures of children's characteristics have already been described in Chapter 2 and also in the relevant empirical chapters in the first part of this thesis and for this reason the measures will only be listed here. The Repetitive Behaviour Questionnaire-2 (RBQ-2, Leekam et al., 2007) was used to assess repetitive behaviours and the Social Communication Questionnaire (SCQ; Rutter et al., 2003) was used to assess impairments in social interaction and communication. As already described above, the items from the DISCO were used to assess expressive language and reciprocal social communication. Finally, the data from the background questionnaire were used to examine the presence of co-morbid conditions in children.

8. 3. Results

Descriptive statistics for the parents' and children's data are presented in table 8. 2.

Table 8. 2. Descriptive characteristics of parents' and children's variables.

Variables	Mean	SD	Range	Skewness (SE)
Children				
RBQ-2 Total Score	2.04	.40	1.30-2.75	-.224 (.337)
SCQ Communication	9.47	2.19	5-13	-.406 (.361)
SCQ Social Interaction	10.28	2.39	5-14	-.481 (.361)
Parents				
Anxiety	10.82	4.26	1-21	.113 (.337)
Depression	6.66	4.07	1-18	.707 (.337)
Escape/Avoidance Coping	7.27	4.98	0-17	.398 (.347)
Problem Solving Coping	9.38	3.81	0-16	-.491 (.347)
Cognitive Empathy	18.68	5.07	6-27	-.512 (.369)
Emotional Empathy	19.71	4.85	7-27	-.271 (.369)

8. 3. 1. Frequency of anxiety in parents

Psychometric properties of the HADS-A subscales were first addressed by examining the internal consistency. It was found that the anxiety subscale had excellent internal consistency (Cronbach's alpha= .876). Internal consistency of the depression subscale was .830.

23 parents (46%) had anxiety scores of 12 or more meeting the cut-off criteria for clinically significant anxiety. It is important to note that 7 parents that met the cut-off criteria for anxiety also met the criteria for clinically significant depression.

8. 3. 2. Relationship between anxiety and children's variables

Pearson's correlations were used to examine a potential association between anxiety in parents and the following children's variables: chronological age, overall autism severity (SCQ total score), impairments in communication and social interaction (SCQ social and communication scores), repetitive behaviours (RBQ-2 total scores), and expressive language. Correlations are shown in table 8. 3.

Table 8.3. Association between Anxiety in parents and children's variables

Children's variables	Anxiety in parent R
Chronological Age	.06
Repetitive Behaviours	.03
SCQ Communication	.015
SCQ Socialization	.228
Expressive Language	-.104

As can be seen from table 8.3., no significant correlations were found.

T test was used to examine whether there was a difference between the levels of anxiety in a group of parents whose children engaged in only one-way communication e. g. only communicated their own needs and/or own interests and a group of parents whose children listened to and shared other people's interests. Although anxiety scores were slightly higher for parents of children who engaged in only one-way communication than for parents whose children shared other people's interest (M= 11.29 (SD= 3.75) vs M= 11.09 (SD= 4.15), the difference was not statistically significant (p= .87; Cohen's d= .05). Similarly, although a group of parents whose children had one or more co-morbid conditions had slightly higher levels of anxiety when compared to a group of parents whose children did not

have any co-morbid conditions ($M= 11.28$ ($SD= 3.30$) vs $M= 10.0$ ($SD= 5.58$), the difference was not statistically significant ($p= .312$; Cohen's $d= .28$).

T test was also used to examine whether there were any differences between the anxious group of parents ($N= 23$) and non-anxious parents ($N= 27$) in terms of their children's characteristics. No differences between the two groups of parents were found in terms of their children's chronological age ($p= .992$; Cohen's $d= .02$), expressive language ($p= .420$; Cohen's $d= .23$), SCQ socialization ($p= .203$; Cohen's $d= .39$), SCQ communication scores ($p= .148$; Cohen's $d= .45$), RBQ-2 total scores ($p= .719$; Cohen's $d= .10$).

8. 3. 3. Relationship between parent's own characteristics and their levels of anxiety

Before the analysis, the psychometric properties of the measures used to assess BAP and coping styles were examined by looking at their internal consistency. Internal consistency (Cronbach's alpha) for IRI and Ways of Coping Scale-Revised subscales is presented in Table 8. 4.

Table 8.4. Internal Consistency of IRI and WOC-R

Scales	Cronbach's alpha
IRI Affective Empathy	.750
IRI Cognitive Empathy	.716
Escape-Avoidance Coping	.776
Problem Solving Coping	.666

As can be seen from table 8.3., IRI subscales had good internal consistency. Internal consistency for WOC-R subscales was in a fair to good range.

Pearson's correlations were used to examine a potential association between the anxiety in parents, BAP traits and coping styles. The full list of correlations is presented in table 8. 5. The only variable that was associated with the anxiety in parents was Escape-Avoidance style of coping. T test confirmed that the anxious group of parents had significantly higher escape-avoidance coping scores than the non-anxious group of parent, no other differences between the groups were found. Please see Table 8. 6. for the full list of comparisons.

Table 8.5. Associations between the anxiety in parents, BAP traits and coping styles

	Anxiety r
IRI Affective Empathy	.075
IRI Cognitive Empathy	-.35
Escape-Avoidance Coping	.460**
Problem Solving Coping	-.110

Note: * = $p < .01$; ** = $p < .001$

Table 8.6. Comparison between anxious and non-anxious group of parents in terms of their coping style and BAP traits

	Anxious group ($n=23$)	Non-anxious group ($n=27$)	<i>t</i> test		Cohen's <i>d</i>
	<i>M</i> (<i>SD</i>)	<i>M</i> (<i>SD</i>)	<i>t</i>	<i>p</i>	
IRI Affective Empathy	19.57 (4.23)	19.85 (5.54)	.182	.857	.06
IRI Cognitive Empathy	17.55 (5.06)	19.76 (4.97)	1.41	.166	.44
Escape-Avoidance Coping	9.52 (5.13)	5.46 (4.11)	3.01	.001	1.08
Problem Solving Coping	9.10 (4.17)	9.62 (3.55)	.462	.647	.13

The next step was to examine whether there was a relationship between the age of children and the coping strategy that parents used. A potential relationship between BAP traits and WOC-R subscales was also addressed. As can be seen from Table 8.7, no significant relationship was found.

Table 8. 7. Associations between coping styles, BAP and chronological age of children

	IRI Affective Empathy	IRI Cognitive Empathy	Chronological age of children
Escape-Avoidance Coping	.071	-.061	-.189
Problem Solving Coping	.083	.072	-.212

8. 3. 4. Relationship between environmental characteristics and anxiety in parents

A potential relationship between the support that parents receive and anxiety in parents was examined. As noted in the method section, parents had to be enrolled/finished Early Bird or Early Bird Plus programme, be active members of parent support groups and their children had to receive an extra support at school in order to be classified into the 'receive support' group. 32 parents were classified as receiving support and 18 parents did not receive support. Although a group of parents who did not receive support had higher anxiety scores (mean anxiety score= 11.83, SD= 4.39) than parents who did receive support (mean anxiety score= 10.42, SD= 4.43), the difference was not significant ($p = .352$; Cohen's $d = .31$).

As noted in the method section, 38 parents were married and 12 were single parents. Parents who were married had higher levels of anxiety than parents who were divorced ($M = 11.05$ (SD= 4.63) vs $M = 10.08$ (SD= 2.78)); however, the difference was not statistically significant ($p = .497$; Cohen's $d = .25$). 30 parents had only one child and 20 more than one child. Parents who had more than one child had higher anxiety scores when compared to parents who had only one child ($M = 11.70$ (SD= 3.76) vs $M = 10.23$ (SD= 4.87)); however, the difference was not statistically significant ($p = .236$; Cohen's $d = .34$).

Finally, a potential relationship between the parents' level of education and their levels of anxiety was examined. As noted above, of 45 parents that provided information on their level of education, 17 parents had postgraduate qualifications, 18 undergraduate or vocational qualifications and 10 did not have post-school qualifications. Parents who had postgraduate qualifications (Group 1) had the lowest anxiety scores ($M = 10.35$; SD= 2.57), parents with undergraduate or vocational qualifications (Group 2) had slightly higher anxiety scores ($M = 10.94$; SD= 4.08) and parents who did not have post-school qualifications (Group 3) had the highest anxiety scores ($M = 13.70$; SD= 4.60). However, when ANOVA was used

the examine the potential difference in the anxiety levels between these 3 groups, no differences were statistically significant ($F= 2.723$, $p= .077$). Cohen's d s for comparisons are shown in Table 8.8.

Table 8. 8. Effect Sizes

	Group 1 vs Group 2	Group 1 vs Group 3	Group 2 vs Group 2
Cohen's d	.17	.88	.63

8. 4. Discussion

The first aim of this chapter was to examine the frequency of anxiety in a group of parents of children and adolescents with autism. The finding was that 46.5% of parents met the criteria for the clinically significant anxiety. Furthermore, 30% of anxious parents also met the criteria for clinically significant depression. These results are in line with the findings from literature suggesting that anxiety, and affective disorders more generally, are indeed a prevalent problem among the parents of children with autism.

The frequency of anxiety problems in the population of parents of children with autism found in this thesis is considerably higher than the frequency of anxiety found in general, adult population. For example, Somers, Goldner, Waraich and Hsu (2006) did a systematic review of the prevalence and incidence studies of anxiety in general population published between 1980 and 2004 (46 studies in total). Pooled 1-year prevalence for total anxiety was 10.6% while the lifetime prevalence for total anxiety was 16.6%. The prevalence of anxiety subtypes was highly variable. Also, in a study that looked at the prevalence of DSM-IV psychiatric disorders in a large ($n = 21425$) random sample of non-institutionalized adult population from the following countries: Belgium, France, Germany, Italy, Netherlands and Spain), Alonso et al. (2004) found that 1-year prevalence of anxiety was 6.4% and lifetime prevalence was 13.6%. The rates of anxiety reported in this thesis were also higher than the rates found in parents of children with various neurodevelopmental and somatic disorders. For example, Merrill et al. (2007) reported that the frequency of anxiety in parents of children that were undergoing cancer workup was around 37%.

The second step was to examine the potential contributing factor to the high rates of anxiety among parents found in this thesis. Three potential sources were examined: characteristics of children, characteristics of parents and environmental characteristics.

The previous literature has mainly concentrated on the characteristics of children as a potential cause of elevated anxiety in their parents. Core autism features, the presence of co-morbid conditions, the chronological age of children were all reported to be related to anxiety in parents although, as reviewed in the introduction section of this chapter, and also in more detail in Chapter 7, results have been somewhat conflicting. One of the goals of this chapter was to examine the relationship of these characteristics of children and anxiety in parents in a systematic way, using validated measures. No significant association was found between anxiety in parents and the following characteristics of children: children's chronological age, social and communication impairments, expressive language levels, repetitive behaviours, and anxiety in parents. Furthermore, the relationship between the presence of co-morbid condition in children and anxiety in parents was also explored; however, no significant association was found.

It would be tempting to conclude, based on these results, that the hypothesis that anxiety in parents is a consequence of raising a child with autism is not a viable explanation. However, this conclusion would not be valid. Although the finding from this chapter that anxiety in parents was not associated with any of the children's characteristics might seem surprising in the context of findings from the previous studies, it is important to highlight the fact that the majority of the previous work has used terms *stress* and *anxiety* interchangeably. However, stress and anxiety, although related, are not the same concepts (Monroe & Peterman, 1988). As discussed in Chapter 7, prolonged elevated levels of stress can lead to anxiety; however, individual differences significantly influence whether negative events will lead to anxiety and other psychiatric problems or not (Dohrenwend & Dohrenwend, 1981; Monroe & Peterman, 1988). It is without a doubt that stress abounds in everyday life of parents of children with autism, and, previous studies have indeed found that the levels of stress in parents are related to various characteristics of Children with autism. However, previous studies have reported that only a portion of parents develop anxiety. This is confirmed by the finding from this thesis that 53.5% of parents did not have elevated levels of anxiety.

Furthermore, the comparison between the anxious and non-anxious parents did not reveal significant differences between the groups in terms of children's characteristics. All these findings do point out that although children's characteristics are stressful for parents and are putting parents at risk for the development of anxiety, it is of crucial importance to consider characteristics of parents and also characteristics of their environment as factors that

might either serve as a buffer against stress and protect parents from developing anxiety or might, on the other hand, put parents at even increased risk of developing anxiety by making them less able to deal with the stress that they inevitably encounter on a daily basis.

Two of parents' characteristics that were examined in this chapter are the coping strategies and the empathy traits (as proxy for BAP traits). These characteristics were chosen based on the review of literature that identified them as the only two characteristics of parents of children with autism that were addressed in the previous literature as correlates of anxiety. As reviewed in more details in the introduction section and Chapter 7, the findings from the studies that examined the relationship between the presence of BAP characteristics in the parents of children with autism and their levels of anxiety have been mixed. Studies by Ingersoll et al. (2010) and Ingersoll and Hambrick (2011) suggested that the presence of BAP traits, more specifically, the social aspects of BAP place the parents of children with autism at an increased risk of experiencing affective disorders. However, findings from this chapter suggest that BAP traits of parents (more specifically, affective and cognitive empathy measured by IRI) are not associated with their levels of anxiety. These results are supported by the findings from Bolton et al. (1998) Piven and Palmer (1999) Murphy et al. (2000) and Micaliet al. (2004) who also did not find support for the hypothesis that anxiety in parents is a consequence of their BAP traits.

The second parents' characteristic that was addressed is the coping strategies that they used to deal with stressful events. The Ways of Coping Scale-Revised (Folkman & Lazarus, 1988), a well validated instrument that provides a detailed assessment of coping and has been used with various non-clinical as well as with clinical populations e. g. with individuals with cancer (Wonghongkul, Moore, Musil, Schneider, & Deimling, 2000; Rosberg, Edgar, Collet, & Fournier, 2002) multiple sclerosis (Jean, Paul, & Beatty, 1999; Lundqvist & Ahlström, 2006), fibromyalgia (Cronan, Serber, Walen, & Jaffe, 2002), stroke (Rochette & Desrosiers, 2002; King, Zeldow, Carlson, Feldman, & Philip, 2002), chronic fatigue syndrome (Ax, 1999), spinal cord injury (Winemann, Durand, & McCulloch, 1994), muscular dystrophies and postpolio syndrome (Lundqvist & Ahlström, 2006) and their families. It was found that the parents of children with autism who met the criteria for clinically significant levels of anxiety used the escape-avoidance style of coping significantly more than the non-anxious parents. No differences between the two groups were found on their use of problem solving coping. These results were in line with the findings from both the literature on coping in parents of children with autism and the general literature on coping which suggests that the

use of escape-avoidance as a dominant style of coping is an important risk factor for the development of affective disorders (Carver, & Connor-Smith, 2010).

As already discussed, the previous literature on autism has not addressed the key issue of why the parents of children with autism tend to use the escape-avoidance as preferable strategy. The only factor that has been considered is the age of children, based on the fact that parents of younger children with autism might be more pro-active and motivated in terms of using problem-solving strategies as they still have hope that their children's impairments might significantly improve and also due to the simple fact that currently, there is more support for parents of young children with autism when compared to the level of support that is available to adolescents and especially adults with autism. However, no significant association between the age of a child and the type of coping strategy that the parents used was found in this chapter, replicating similar findings reported by Hastings et al. (2005). No association was found between BAP characteristics and the coping strategies used by parents.

Finally, this chapter has looked at the support that parents receive and its relationship to their levels of anxiety. The previous studies have found that the lack of support is a significant contributing factor to both parents' stress and levels of anxiety (Boyd, 2002). However, it is important to emphasize the fact that the previous studies have looked at the parents' perception of the level of support that they receive rather than looking at the actual levels of support. This might explain the fact that this thesis failed to find any significant differences in terms of anxiety levels between the parents who were classified as the "receive support" group and the ones who were classified into the "does not receive support" group. Also, no significant differences were found between parents who were married and parents who were single parents. However, the parents' perception of the quality of their marriage and the actual level of support that they receive from their spouses was not assessed.

8. 4. 1. Limitations

Considering the small sample used in this study it is possible that the frequency of anxiety found might not be generalizable to the wider population of parents of children with autism. Also, of self-selection of parents who were anxious for the study is possible. In addition to the sample size, it is important to highlight several measurement issues as a potential limitation of the findings reported in this chapter. Firstly, rather than using the Autism Spectrum Quotient (AQ; Baron-Cohen et al. 2001), the Social Responsiveness Scale-Adult (SRS-A; Constantino and Todd 2005) or the Broad Autism Phenotype Questionnaire

(BAPQ; Hurley et al. 2007) this thesis has used cognitive and affective empathy subscales of IRI as a proxy of social aspects of BAP. Impairments in cognitive and affective empathy are considered to be a part of BAP (Sucksmith, Allison, Baron-Cohen, Chakrabati, & Hoekstra, 2013) and more importantly, they have been previously found to be associated with anxiety in general population (Fernandez-Berrocal, Alcaide, & Extremera, 2006, Karukivi et al., 2010). Thus, there were strong reasons to concentrate on cognitive and affective empathy as features of BAP that might be associated with anxiety in parents and furthermore use the IRI as a well established measure of various aspects of empathy (a detailed review of IRI is provided in Chapter 2). Secondly, as already noted above, this chapter concentrated on the actual support that parents and their children received, rather than on their own perceptions of the support. Furthermore, this thesis has not explored the perception of married parents about the quality of their marriage, the level of support that they receive from their spouses, or from their family and friends, and how having a child with autism has impacted on their marriage. Finally, the parents' perception of the levels of support, their experiences with the diagnostic process, the impact that having a child with autism has on marriage and family structure are all important avenues for future research.

8. 4. 2. Summary

In summary, this chapter had several aims. The first aim was to examine the frequency of anxiety problems in parents of children with autism. It was found that anxiety was a prevalent problem in this population with 46.5% parents meeting the cut-off criteria for clinically elevated anxiety. The second aim was to examine in a systematic way the relationship between the anxiety in parents and the key characteristics of children, parents and environment that were identified through a systematic search of the literature. It was found that the only factor that distinguished the anxious group of parents from the non-anxious parents was the higher use of escape-avoidance coping by parents who were anxious. Finally, the relationship between the age of children, BAP traits of parents and the coping strategies used by parents was examined; however, no significant relationship was found.

Chapter 9: Intolerance of Uncertainty and Sensory Problems in Children with Autism and their Contribution to Anxiety Levels in Parents

Chapter Plan:

The findings from Chapter 8 suggest that characteristics of parents were the most important contributing factor to their own levels of anxiety, more specifically, the parents who had clinically significant levels of anxiety used the escape-avoidance coping strategy significantly more than the non-anxious parents. The main aim of this chapter is to go beyond the existing literature in autism and examine the contribution of the two characteristics that have been shown to be the major contributing factors in the development and maintenance of anxiety in general population. This chapter will provide a brief overview of the key findings from the literature on the concepts of the intolerance of uncertainty and sensory sensitivity and their contribution to the development and maintenance of anxiety in general population. An overview of the literature that suggests the importance of these concepts for the adoption of certain coping styles will also be provided. The importance of examining these factors in the population of parents of children with autism will be highlighted. Then, the data from this thesis that examined the influence of the intolerance of uncertainty and sensory sensitivity on the presence of anxiety in parents and on the choice of their coping strategies will be presented. The results will be discussed in the light of the existing literature, and the limitations will be addressed.

9. 1. Introduction

9. 1. 1. Intolerance of uncertainty and anxiety

Intolerance of uncertainty (IU) is a dispositional characteristic of an individual that leads that individual to experience any uncertainty as stressful and upsetting (Freeston et al. 1994; Ladouceur, Gosselin, & Dugas, 2000). Uncertainty in this context is defined as the subjective perception that the possibility of a negative event or a negative outcome occurring is high, regardless of how low or unlikely the actual possibility might be (Buhr & Dugas, 2002; Koerner & Dugaes, 2008).

When originally described and defined by Freeston and colleagues, IU was considered to be a specific risk factor for the development of Generalized Anxiety Disorder (GAD). Indeed, several studies have found that the individuals with GAD have significantly higher

levels of IU than the individuals with some other affective disorder such as phobic disorder, social anxiety, and depression (Ladouceur et al., 1995; Dugas & Koerner, 2005).

However, more recent research seems to suggest that IU is a risk factor for various affective disorders, rather than being specifically associated with GAD. Several studies found IU to be the main predictor of social anxiety (Boelen & Reijntjes, 2009; Carleton et al., 2010), depression (McEvoy & Mahoney, 2011) and certain aspects of OCD (Steketee, Frost, & Cohen, 1998; Tolin et al., 2003; Holaway, Heimberg, & Coles, 2006; Oglesby et al., 2012). For example, in terms of the relationship between IU and OCD, Tolin, et al. (2003) reported that the individuals diagnostically classified as compulsive checkers and repeaters had higher IU scores than the typical, non-anxious controls. Furthermore, it was reported that the individuals with OCD had significantly higher levels of IU than the TD controls. Regarding the role of IU as a risk factor for social anxiety and depression, Boelen and Reijntjes (2009) found that, even after controlling for the contribution of various other risk factors such as: fear for negative evaluation, anxiety sensitivity, positive and negative affectivity, low self-esteem, worry, and neuroticism, IU was the strongest predictor of social anxiety symptoms. Finally, Carleton et al. (2010) reported that the depression symptoms in their sample were predicted by IU, even after controlling for neuroticism, meta beliefs about worry and positive and negative affectivity.

As the previous chapter has demonstrated, anxious and non-anxious parents of children with autism did not differ in terms of the characteristics of their children. As pointed out, this is not to say that these behaviours are not stressful but that some parents seem to manage their stress levels better than others. Findings from Chapter 8 suggest that the use of escape-avoidance coping seems to put parents at higher risk for the development of anxiety. However, it is surprising that, considering the convincing evidence which points out to IU as a significant factor for the development and maintenance of anxiety in general population, the importance of this factor has not been considered before in the population of parents of children with autism. Uncertainty is undoubtedly prevalent in everyday lives of parents of children with autism and being intolerant towards uncertain situations would certainly put parents at increased risk for the development of anxiety.

The first aim of this chapter is to examine the relationship between the IU and anxiety in parents of children and adolescents with autism. Based on the findings from the reviewed literature, it is hypothesized that IU will be associated with anxiety and that the parents who are anxious will have higher IU scores when compared to the non-anxious parents.

9. 1. 2. Sensory atypicalities and their relationship with anxiety

Sensory problems are not specific to autism (Bröring, Rommelse, Sergeant, & Schreder, 2008) and they have been found to be prevalent in both general population (Aron & Aron, 1997; Ahn et al., 2004; Goldsmith, et al., 2006; Ben-Sasson et al., 2009) and various other neurodevelopmental and psychiatric conditions such as ADHD, Fragile X syndrome and schizophrenia (Bröring et al., 2008; Mangeot et al., 2001; Brown et al., 2002). For example, Ahn et al. (2004) and Ben-Sasson et al. (2009) estimated prevalence of sensory atypicalities in general population to be around 13% and 16.5% respectively.

Sensory problems have been found to be important contributing factors to the development and maintenance of affective disorders in non-Population with autism (Aron & Aron, 1997; Kinnealey & Fuiiek, 1999; Neal, Edelmann, & Glachan, 2002; Hofman & Bitran, 2007; Liss, Mailloux, & Erchull, 2008). For example, in a study that used the Highly Sensitive Person Scale (HSPS; Aron & Aron, 1997) to examine the sensory processing sensitivity in 89 adults with social anxiety disorder, Hofman & Bitran (2007) found that sensory sensitivity was significantly associated with various aspects of social anxiety. Also, Kinnealey and Filipek (1999) found that both anxiety and depression symptoms were significantly higher in sensory defensive adults when compared to individuals without sensory atypicalities.

Similarly, like intolerance of uncertainty, despite the strong evidence of association between anxiety and sensory sensitivity in general population, autism, ADHD and other neurodevelopmental and psychiatric disorders, the presence of sensory atypicalities and their potential relationship with anxiety in the population of parents of children with autism has not been explored before. The second aim of this chapter is to examine the frequency of sensory problems in parents of children with autism and also to examine the relationship between sensory problems and anxiety. Based on the findings from the studies that showed that certain sensory atypicalities have a genetic component (Goldsmith, Buss, & Lemery, 1997; Zawdaski et al., 2001; Goldsmith et al., 2006), it is hypothesized that sensory problems will be prevalent among parents of children with autism and furthermore, that they will be associated with parents' levels of anxiety. The relationship between sensory atypicalities in children and their parents will also be explored.

9. 1. 3. Intolerance of uncertainty, sensory atypicalities and coping

As discussed in more detail in Chapters 7 and 8, despite the fact that the literature on anxiety in parents of children with autism has consistently shown that certain types of coping strategies used by parents might put them at increased risk for developing anxiety, the question of why parents of children with autism resort to using these particular strategies, rather than more adaptive ones, has not been addressed. The previous chapter looked at the relationship between the chronological age of children, BAP traits of parents and the coping strategies used; however, no significant relationship was found.

Several lines of evidence suggest that both IU and sensory problems might have an important influence on the choice of coping strategies used by parents of children with autism. Individuals who show IU as a personality trait were found to have poor confidence in their problem-solving abilities, low appraisals of control, and to show both cognitive and behavioural avoidance (Dugas & Koerner, 2005; Koerner & Dugas, 2008), and it has been hypothesised that as a result of this, those individuals would use avoidant coping strategies (Dugas et al., 1998; Ladouceur, Gosselin, & Dugas, 2000; Behar et al., 2009). Furthermore, it has been found that sensory sensitive individuals employ emotionally and behaviourally avoidant strategies as dominant modes of coping (Kinnealey, Oliver, & Wilbarger, 1995). However, neither of these hypotheses has been formally tested in general population and it certainly has not been tested in the population of parents of children with autism.

The third aim of this chapter is to examine the relationship between IU, sensory sensitivity and coping strategies used by parents of children with autism. It is hypothesised that the escape-avoidance style of coping will be associated with higher IU scores and sensory sensitivity.

9. 1. 4. Notes on the measurement of Sensory Problems and Intolerance of uncertainty

As pointed out in Chapter 2, currently there are only two questionnaires that are suitable for assessing sensory processing in adults-the Adult Sensory Profile (ASP; Brown & Dunn, 2002) and the Highly Sensitive Person Scale (HSP; Aron & Aron, 1997). These questionnaires differ in terms of their focus. ASP was developed from the Sensory Profile (SP; Dunn, 1999) and it provides scores for 4 different sensory patterns: hypo-sensitivity (registration), seeking, hyper-sensitivity and avoidance. However, it is important to emphasize the fact that ASP, like SP, provides data on individual's response to basic sensory stimuli across different modalities such as response to noises, intense visual and tactile

stimuli for example. HSP scale on the other hand, assesses individual's responsiveness to a wider range of stimuli from the environment. HSP scale factor-Low Sensory Threshold (LST) is in its concept similar to ASP in that it measures response to more basic sensory stimuli from the environment (albeit not providing as detailed assessment as ASP). Ease of Excitation (EOE) is the second HSP factor which refers to being easily overwhelmed in response to demands of environment and is more similar to temperamental feature of behavioural inhibition. Although both of these scales have been used in general population, the relationship between these measures has not been explored before. Since sensory processing in parents of children with autism has never been assessed before and also due to the above mentioned difference between the two measures, it was decided that both ASP and HSP scale would be used for this thesis.

Currently there is only one questionnaire that measures intolerance of uncertainty-the Intolerance of Uncertainty Scale (27 item version-IUS27; Freeston et al., 1994; Buhr & Dugas, 2002; and 12 items version-IUS12: Carleton et al., 2007). Although majority of studies reviewed in this chapter and Chapter 7 have considered intolerance of uncertainty as a unitary construct, several factor analytic studies (Carleton et al., 2007; McEvoy & Mahoney, 2011; Helsen, van den Bussche, Vlaeyen, & Goubert, 2013) found that IU scale consists of two factors-the first termed as Desire for Predictability and the second termed as Uncertainty Paralysis. Furthermore, it has been suggested that these two factors are differently associated with anxiety and depression and further that these two factors have different behavioural consequences, in other words that they represent approach and avoidance responses to uncertainty (Meares & Freeston, 2008; Birrell, Meares, Wilkinson, & Freeston, 2011; Helsen et al., 2013). Having this in mind, it was decided that rather than looking at intolerance of uncertainty as a unitary construct, the two factor approach with Desire for Predictability and Uncertainty Paralysis factors would be used

9. 1. 5. Summary of the chapter aims

This chapter will explore: the relationship between intolerance of uncertainty and anxiety in parents of children with autism; the prevalence of sensory problems and the potential relationship between sensory sensitivity and anxiety in parents; the relationship between both intolerance of uncertainty and sensory sensitivity, and the dominant coping strategy that parents might use. The potential relationship between sensory atypicalities in children and their parents will be also explored. Finally, as the previous chapter has shown

that the escape-avoidance style of coping was related to anxiety in parents, relative contribution of escape-avoidance, intolerance of uncertainty and sensory processing to the prediction of anxiety in parents will be explored.

9. 2. Methods

9. 2. 1. Subjects

Participants were the same 50 parents of children and adolescents with ASD described in the method section of the previous chapter.

9. 2. 2. Measures

A detailed overview of the measures is provided in Chapter 2.

The Intolerance of Uncertainty Scale 12 (IUS12; Carleton et al., 2007) was used to assess the trait intolerance of uncertainty.

The Adult Sensory Profile (AASP; Brown & Dunn, 2002) and the Highly Sensitive Person scale (HSP; Aron&Aron, 1997) were used to explore various aspects of sensory processing in parents of children with autism.

The Hospital Anxiety and Depression Scale (HADS, Zigmond & Snaith, 1983) was used to assess anxiety and the Ways of Coping Checklist-Revised (WCC-R; Vitaliano et al., 1985; Folkman & Lazarus, 1988) was used to assess escape-avoidance and problem solving coping strategies

9. 3. Results

Prior to conducting any of the planned analyses, the psychometric properties of the questionnaire were first addressed by examining the internal consistency, and, as can be seen from Table 9. 1., all measures had good to excellent internal consistency.

Table 9. 1. Internal Consistency of measures

Scales	Cronbach's alpha
Desire for Predictability IU Factor (DP IU)	.744
Uncertainty Paralysis IU Factor (UP IU)	.85
Ease of Excitation HSP Factor (EOE HSP)	.845
Low sensory Threshold HSP Factor (LST HSP)	.799
ASP–Low Registration Quadrant	.796
ASP –Sensory Seeking Quadrant	.701
ASP–Sensory Sensitivity Quadrant	.795
ASP–Sensory Avoidance Quadrant	.866

Descriptive statistics are presented in table 9. 2. Data were screened for outliers, no outliers were found.

Table 9. 2. Descriptive statistics for IUS, HSP and AASP scales

Variables	Mean	SD	Range	Skewness (SE)
DP IU	16.48	5.14	7-28	.147 (.337)
UP IU	9.4	4.65	5-20	.895 (.337)
EOE HSP	48.38	14.68	23-79	.306 (.337)
LST HSP	19.28	8.66	8-37	.665 (.337)
Low Registration	38.84	10.24	19-58	.433 (.337)
Sensory Seeking	40.32	8.65	24-67	.582 (.337)
Sensory Sensitivity	41.60	11.70	15-65	.088 (.337)
Sensory Avoidance	41.35	12.08	19-63	.162 (.337)

9. 3. 1. Frequency of sensory problems in parents of children with autism

Adult Sensory Profile (ASP) is a norm referenced instrument that provides cut off scores. The manual provides scores from a large normative sample of individuals without disabilities. Based on those scores, an individual's performance can be classified in 5 following categories (shows particular type of responses):

- Much Less Than Other People which corresponds to scores at or above the point 2SD below the mean but also lower than 1SD below the mean for TD individuals;
- Less Than Other People which corresponds to scores below the point 2 SD below the mean for TD individuals;
- Similar to Other People (Typical Performance) which corresponds to scores that are at or above the point 1SD below the mean for TD individuals;
- More than Most People opposite to less than other people cut off score;
- Much More than Most people opposite to Much less than other people cut off score.

The performance of parents of children with autism across four sensory quadrants is shown on Table 9. 3.

Table 9. 3. Performance of parents across four sensory quadrants

Quadrants	Response Classification				
	Much Less than Other People	Less than Other People	Typical Performance	More than Most People	Much More than Most People
Low Registration	0	1 (2%)	18 (36%)	18 (36%)	13 (26%)
Sensation Seeking	15 (30%)	15 (30%)	18 (36%)	1 (2%)	1 (2%)
Sensory Sensitivity	2 (4%)	1 (2%)	25 (50%)	7 (14%)	15 (30%)
Sensation Avoiding	0	7 (14%)	19 (38%)	11 (22%)	13 (26%)

As can be seen, 62% of parents had higher sensory hypo-sensitivity (registration) scores than TD norms, 44 % had higher scores for sensory sensitivity and 48% had higher scores for sensory avoidance. Interestingly, 60% of parents had lower sensory seeking scores than TD norms.

9. 3. 2. The relationship between Sensory problems in children and mothers

The relationship between sensory atypicalities in children and their mothers is shown in table 9.4.

Table 9. 4. Relationship between sensory atypicalities

Parents	Children			
	Low Registration	Sensation Seeking	Sensory Sensitivity	Sensation Avoiding
Low Registration	.496*	.199	.626**	.452*
Sensation Seeking	.007	.115	.118	.243
Sensory Sensitivity	.345	.214	.588**	.372
Sensation Avoiding	.287	.076	.587**	.524**

9. 3. 3. Relationship between sensory atypicalities and anxiety

Before addressing the issue of the relationship between sensory processing in parents of children with autism, the correlations between two sensory measures: Highly Sensitive Person Scale (HSP) and Adult Sensory Profile (ASP), were examined.

Table 9. 5. Construct Validity of Highly Sensitive Person Scale and Adult Sensory Profile

	Low Registration	Sensation Seeking	Sensory Sensitivity	Sensation Avoiding
EOE HSP	.230	-.059	.248	.130
LST HSP	.415*	.088	.383*	.292

Note: *= p<.01; **= p<.001

As can be seen from Table 9. 5., and as hypothesised in the introduction, the Ease of Excitation factor of HSP scale did not show a significant correlation with any of the quadrants of the Adult Sensory Profile. On the other hand, the Low Sensory Threshold factor of the HSP scale was significantly associated with Low Registration (p= .003) and Sensory Sensitivity Quadrant (p= .006) of the Adult Sensory Profile.

9. 3. 4. The relationship between anxiety, intolerance of uncertainty and sensory problems in mothers of children with autism

Pearsons correlations were used to explore the association between anxiety, intolerance of uncertainty and sensory problems. The significance level was set at $p < .01$ due to multiple correlations. As can be seen from Table 9. 6., anxiety was significantly associated with the Desire for Predictability factor of IU scale and with the Ease of Excitation factor of the HSP scale. It is important to note that Uncertainty was correlated with anxiety at $p < .05$ level ($p = .013$) and the case was similar with the Low Sensory Threshold factor of HSP scale ($p = .022$).

Table 9. 6. Relationship between anxiety, intolerance of uncertainty and sensory problems

	Anxiety R
Desire for Predictability IU Factor	.510**
Uncertainty Paralysis IU Factor	.354
Ease of Excitation HSP Factor	.389*
Low sensory Threshold HSP Factor	.324
Low Registration Quadrant	.107
Sensory Seeking Quadrant	-.229
Sensory Sensitivity Quadrant	.037
Sensory Avoidance Quadrant	.087

Note: * = $p < .01$; ** = $p < .001$

T test was used to compare anxious and non-anxious groups of mothers in terms of their sensory processing and intolerance of uncertainty scores. The significance level was set at .01 level due to multiple comparisons. As can be seen from Table 9.7, anxious and non-anxious groups of mother significantly differed in terms of the Desire for Predictability IU factor ($p = .002$; Cohen's $d = .92$) and the Ease of Excitation HSP factor ($p = .009$; Cohen's $d = .77$).

Table 9. 7. Differences between anxious and non-anxious groups of mothers

	Anxious Group	Non-anxious group	t test		Cohen's d
	(n=23)	(n=27)	t	p	
	M(SD)	M(SD)			
Desire for Predictability IU Factor	18.83 (5.0)	14.48 (4.43)	3.255	.002	.92
Uncertainty Paralysis IU Factor	10.91 (4.94)	8.11 (4.03)	2.207	.032	.62
Ease of Excitation HSP Factor	54.17 (14.54)	43.44 (13.12)	2.742	.009	.77
Low sensory Threshold HSP Factor	21.3 (9.04)	17.56 (8.09)	1.546	.129	.44
Low Registration Quadrant	39.96 (9.58)	37.85 (10.87)	.717	.477	.21
Sensory Seeking Quadrant	38.48 (7.3)	41.89 (9.49)	1.404	.167	.40
Sensory Sensitivity Quadrant	42.3 (11.71)	41.0 (11.88)	.389	.699	.11
Sensory Avoidance Quadrant	42.32 (11.38)	40.56 (12.79)	.504	.617	.15

Chapter 8 found that anxiety in mothers was significantly associated with escape-avoidance style of coping ($r = .460, p = .000$). A hierarchical regression was performed in order to examine a relative contribution of the escape-avoidance style of coping, the desire for predictability IU factor and the ease of excitation HSP factor to anxiety in mothers. The escape avoidance coping was entered in the first step, significantly predicting anxiety ($p = .001$) and accounting for 19.4% of total variance. The desire for predictability was entered in the second step, accounting for the additional 16.1% of variance. Finally, in the third step, the ease of excitation was entered. When the final model was considered, both the escape avoidance and prospective anxiety scores were significant predictors of anxiety in mothers, while the ease of excitation was not. Regression is shown in the table 9.8.

Table 9. 8. Predictors of anxiety in mothers

	R2	R2 Change	Beta
Step one:			
Escape-avoidance coping	.194**		.460
Step two:			
Escape-avoidance coping			.344
Prospective anxiety	.355**	.171**	.430
Step three:			
Escape-avoidance coping			.333
Prospective anxiety			.413
Ease of Excitation	.334	.001	.04

Note: * = $p < .05$; ** = $p < .01$

9. 3. 5. Predictors of escape-avoidance coping in mothers of children with autism

As can be seen from Table 9. 9., only the Ease of Excitation was correlated with Avoidance coping. As individual predictor, the ease of excitation significantly predicted avoidance coping at $p = .005$ level and account for 16.4% of variance (Beta = .405).

Table 9. 9. Predictors of escape-avoidance coping

	Escape-Avoidance Coping
	R
Desire for Predictability IU Factor	.269
Uncertainty Paralysis IU Factor	.280
Ease of Excitation HSP Factor	.405*
Low sensory Threshold HSP Factor	.260
Low Registration Quadrant	.177
Sensory Seeking Quadrant	-.209
Sensory Sensitivity Quadrant	-.075
Sensory Avoidance Quadrant	.033

Note: * = $p < .01$; ** = $p < .001$

9. 3. 6. Interrelationship between avoidance coping, anxiety and ease of excitation

As anxiety, the avoidance coping and the ease of excitation were highly significantly intercorrelated, a mediation analysis was performed in order to explore the nature of this relationship.

Mediation analysis 1 examined the model where the ease of excitation would lead to the avoidance coping which in turn, would lead to anxiety.

The first two regressions showed that anxiety was predicted by the ease of excitation ($R^2 = .133$, $p = .005$) and that the avoidance coping was also predicted by the ease of excitation ($R^2 = .146$, $p = .004$) satisfying first two conditions proposed by Baron & Kenny. In the first step of hierarchical regression, anxiety were predicted by avoidance coping ($R^2 = .194$, $p = .001$), however, when controlling for the effects of ease of excitation, the relationship between anxiety and avoidance coping was not significant ($R^2 = .225$, R^2 change = $.047$, $p = .102$) indicating that the model where ease of excitation would lead to avoidance coping which in turn, would lead to Anxiety was significant (or confirmed). Steps of the Mediation Analysis 1 are presented in Table 9. 10.

Table 9. 10. Mediation Model 1

	R2	R2 Change	Beta
Regression One:			
Anxiety on Ease of Excitation	.133**		.389
Regression Two:			
Avoidance Coping on Ease of Excitation	.146**		.405
Regression Three:			
Step 1: Anxiety on Avoidance Coping	.194**		.364
Step 2: Anxiety on Ease of Excitation	.225	.047	.237

Note: * = $p < .05$; ** = $p < .01$; *** = $p < .001$

Mediation analysis 2 examined the model where the ease of excitation would lead to anxiety which in turn, would lead to the avoidance coping. The first two regressions showed that Avoidance coping was predicted by the ease of excitation ($R^2 = .146$, $p = .004$) and that anxiety was also predicted by the ease of excitation ($R^2 = .133$, $p = .005$) satisfying first two conditions proposed by Baron & Kenny. In the first step of hierarchical regression, the

avoidance coping were predicted by anxiety ($R^2 = .194$, $p = .001$), when controlling for the effects of the ease of excitation, the relationship between the avoidance coping and anxiety did not remain significant ($R^2 = .240$, R^2 change = $.061$, $p = .06$) indicating that the model where the ease of excitation would lead to anxiety which in turn, would lead to the avoidance coping was supported. Steps of the Mediation Analysis 2 are presented in Table 9.11.

Table 9. 11. Mediation Model 2

	R2	R2 Change	Beta
Regression One:			
Avoidance Coping on Ease of Excitation	.146**		.405
Regression Two:			
Anxiety on Ease of Excitation	.133**		.389
Regression Three:			
Step 1: Avoidance Coping on Anxiety	.194**		.357
Step 2: Avoidance Coping on Ease of Excitation	.240	.061	.268

Note: * = $p < .05$; ** = $p < .01$; *** = $p < .001$

9. 4. Discussion

This chapter reports on the relationship between the intolerance of uncertainty (IU) and anxiety in parents. It was found that the anxious parents had significantly higher levels of the intolerance of uncertainty when compared to the non-anxious parents. These findings are consistent with a large body of work which suggests that the intolerance of uncertainty is one of the key contributing factors in the development, maintenance and exacerbation of anxiety in general population. This is the first time that this relationship has been addressed in the literature on anxiety in parents of children with autism. It is particularly important to point out that it was the Desire for Predictability factor of Intolerance of Uncertainty rather than the Uncertainty Paralysis factor that distinguished anxious from non-anxious mothers. The Desire for Predictability which is also termed as Prospective anxiety by some authors is a dimension of IU that relates to “fear and anxiety in anticipation of uncertainty” (McEvoy & Mahoney, 2011). It is considered to be an active dimension of IU in the sense that it represents an attempt to make a situation more predictable by seeking to obtain sufficient

information in order to judge particular situation as predictable and in this way reduce anxiety (Meares & Freeston, 2008). The finding that prospective anxiety was highly correlated with anxiety in mothers of children with autism is very relevant in terms of support that parents need. This issue will be addressed in more detail in General Discussion Chapter.

Secondly, this chapter aimed to address the potential presence of sensory atypicalities in parents of children with autism. It was found that sensory problems were indeed prevalent in parents, with 62% of the parents scoring higher on sensory hypo-sensitivity quadrant than the TD norms, 44% of parents scoring higher for sensory sensitivity quadrant and 48% higher for sensory avoidance quadrant. Finally, 60% of the parents had lower sensory seeking scores than the TD norms. These results are not surprising, considering the previous literature on sensory processing in both general population and various clinical populations. As noted previously, sensory atypicalities are widely distributed among non-clinical populations and highly prevalent among individuals with neurodevelopmental disorders and various psychiatric conditions. The literature that examined sensory processing in children and adolescents with autism has consistently found that more than 90% of individuals with autism show various sensory problems (Leekam et al., 2007; Ben-Sasson et al., 2007; 2009). Furthermore, results presented in Chapter 4, 79.6% of Children with autism had elevated scores for sensory avoidance quadrant, 75.5% for sensory registration (hypo-sensitivity), 73.5% for sensory hyper-sensitivity and 55.1% for sensory seeking.

It has been demonstrated that some aspects of sensory sensitivity show a degree of heritability. For example, Goldsmith et al. (2006) evaluated tactile and auditory defensiveness, temperament, and behaviour in a population-based sample of 1,394 toddler-aged twins. They found that both the tactile and auditory defensiveness showed moderate genetic influences. Goldsmith, Buss, and Lemery (1997) estimated twin similarity on a perceptual sensitivity scale from the Children's Behavior Questionnaire (Rothbart & Ahadi, 1994). Identical twin correlations were .58 and and fraternal twin correlations were .37.

Considering the findings from this chapter and all the findings reviewed above that suggest: the presence of sensory problems in general, non-clinical population, high frequency of such problems in clinical populations, in particular autism, and finally, the evidence of heritability of some of sensory problems, it is surprising that sensory problems have not been evaluated previously in parents of children with autism. One recent study by De la Marche, Steyaert and Noens (2012), used the Adult Sensory Profile to examine sensory processing in

80 individuals with autism, 56 non-affected adolescent siblings and 33 adolescent controls. They found that the only difference between autism siblings and typical controls was that the non-affected siblings of individuals with autism exhibited significantly less sensory seeking behaviours than the TD controls; there were no differences in terms of hypo-sensitivity, sensory sensitivity or sensory avoidance quadrants. De la Marche and colleagues have not assessed sensory problems in parents of children with autism so it is difficult to draw any comparisons between their findings and the findings presented in this thesis. However, it is clear that parents of children with autism exhibit more atypicalities than the siblings from De la Marche et al.'s study. This might be due to several reasons. Firstly, children with autism from this sample had significantly more sensory problems than the individuals with autism from De la Marche et al. study and considering the potential genetic component of sensory problems, these results are not surprising. However, it is important to point out that unlike this thesis, De la Marche et al. used TD controls rather than norms from the manual which might also have impacted the differences between the studies. Based on their results, De la Marche et al. suggested that sensory atypicalities could be considered as candidate intermediate phenotype, since they meet some of the criteria proposed by Bearden and Freimer (2006), more specifically, that the trait should co-occur with the condition of interest, that it should co-segregate with the disorder in families, and that the trait should be expressed at a higher level in non-affected family members than in general population (De la Marche et al., 2012). The results from this chapter also meet these criteria, and furthermore suggest a strong relationship between sensory problems in children with autism and sensory problems in their mothers. However, although the association found between sensory problems in children and their mothers is intriguing, this thesis was not designed to test whether sensory problems are the intermediate phenotype(s) and further research will need to be conducted to examine this possibility. For example, experimental work coupled with questionnaire measures from multiple informants is a direction to take these findings forward, this will be addressed in more details in General Discussion Chapter.

As already pointed out in Chapter 2 and introductory section of this chapter, two currently dominant sensory processing measures for adult population - the Adult Sensory Profile (ASP; Brown & Dunn, 2002) and the Highly Sensitive Person Scale (HSP; Aron & Aron, 1997) differ in terms of their focus and the way they conceptualize sensory processing. While ASP, provides data on individual's response to basic sensory stimuli, HSP scale provides data on individual's responsiveness to basic sensory stimuli (Low Sensory

Threshold Factor) and also data on whether a person is being easily overwhelmed in response to the demands of the environment, akin to the temperamental feature of behavioural inhibition (Ease of Excitation). Surprisingly, the relationship between ASP and HSP measures has not been explored before. The results from this thesis contribute to the general literature on sensory processing by showing that while the Ease of Excitation factor of the HSP scale did not show a significant correlation with any of the quadrants of the Adult Sensory Profile, the Low Sensory Threshold factor of the HSP scale was significantly associated with Low Registration (hypo-sensitivity) and Sensory Sensitivity Quadrant of the Adult Sensory Profile.

Different concepts behind the two sensory measures are particularly important when results on the relationship between anxiety and sensory problems in mothers of children with autism from this thesis are considered. The previous findings that suggested the relationship between sensory sensitivity and anxiety in general population were not replicated when the Adult Sensory Profile or Low Sensory Threshold factor of the Highly Sensitive Person scale were used as measures of sensory processing. However, the ease of excitation factor was highly associated with anxiety in mothers of children with autism. As already pointed out, the ease of excitation is closely related to the concept of behavioural inhibition and being easily overwhelmed by various demands from the environment and novel situations and is conceptually different from being hyper-responsive to basic sensory stimuli which is what ASP and Low Sensory Threshold factor of HSP measure. These findings are also important to consider in the light of the findings reported in Chapter 6 of this thesis that show that in children with autism, hyper-sensitivity to basic sensory stimuli was highly related to children's levels of anxiety. It has been shown that children and adolescents with autism have problems in emotion processing (Uljarevic & Hamilton, 2012) and emotion regulation (Mazefsky et al., 2013) which might make them unable to cope with basic sensory stimuli from their environment. Parents, on the other hand, due to more developed emotion regulation are able to suppress negative reactions to basic sensory stimuli. Intriguing hypothesis is that hyper-responsiveness to basic sensory stimuli from the environment is a developmental precursor to behavioural inhibition which then, rather than basic sensory stimuli serves as a risk factor and reinforces anxiety. However, future work will need to provide a support to this hypothesis and it remains highly speculative at this point.

The hypothesis that sensory problems and intolerance of uncertainty would be related to the higher use of escape-avoidance style of coping was partially supported. As discussed in

the introduction section of this chapter and Chapter 7, several lines of evidence seemed to suggest that both IU and sensory sensitivity would lead to the adoption of avoidant coping style. For example, high levels of the intolerance of uncertainty were previously shown to affect the way in which information is perceived in stressful situations and also affects the way that individuals will respond to uncertain information. In situations of stress, uncertain elements of a problem would become more salient to an intolerant individual and the problem would be interpreted as threatening. Because of perceiving various problems as threatening, individuals with high IU would show poor confidence in their problem-solving abilities and low appraisals of control that impedes their problem-solving ability. In turn, this would lead to the cognitive and emotional avoidance as a coping strategy (Dugas et al., 1998; Dugas & Koerner, 2005; Koerner & Dugas, 2008; Behar et al., 2009). However, as already highlighted, this hypothesis has not been formally tested before. The results from this chapter do not lend support to the relationship between IU and coping strategies. It is important to highlight the fact that a questionnaire assessment might not be the best approach to test this hypothesis. Indeed, the use of tasks that would manipulate individuals' levels of uncertainty while monitoring their performance on either decision making tasks or the coping in hypothetical scenarios will be the more fruitful approach to disentangling the relationship between these concepts. Recent studies have started to conduct similar experimental work with general population (Leijenhorst, Westenberg, & Crone, 2008; Drechsler, Rizzo, & Steinhausen, 2010) and it would be important to follow-up and extend this work with parents of children with autism.

The hypothesis that sensory problems might be related to escape-avoidant coping strategy was supported. Furthermore, mediation models have shown that Ease of Excitation, escape-avoidance coping and anxiety reinforced each other. A longitudinal approach will be needed to explore this interrelationship further.

Finally, the relative contribution of the desire for predictability factor of intolerance of uncertainty, the ease of excitation and the escape-avoidance coping to the levels of anxiety in parents of children with autism was examined. Hierarchical regression showed the escape-avoidance coping style and the desire for predictability were significant predictors of anxiety. The whole model accounted for 33.4% of variance.

9. 4. 1. Limitations

The limitations of this study were addressed throughout the discussion section. One additional limitation that should be emphasized is that the data rely on the use of self-report measures and it would be important in the future to also analyse the third-party reports in terms of sensory problems in parents of children with autism.

9. 4. 2. Summary

This chapter has several important contributions. Firstly, this is the first time that the concept of intolerance of uncertainty and its relationship with anxiety has been explored in the population of parents of children with autism. IU was found to be highly associated with anxiety. Secondly, this is the first study that has examined sensory problems in parents of children with autism. Sensory problems were indeed found to be very prevalent among parents, however, they were not related with the levels of anxiety. Finally, it has been demonstrated that both the escape-avoidance style of coping and the intolerance of uncertainty independently predicted levels of anxiety in parents. Future directions for this line of research have been highlighted throughout this discussion section and will be addressed in more details in the Conclusion chapter.

Chapter 10: General Discussion

This thesis had two main goals. The first goal was to examine the interrelationship between repetitive behaviours, sensory problems and anxiety in children and adolescents with autism and test the hypothesis that anxiety and sensory problems are the triggers for repetitive behaviours. The second goal was to examine the correlates of anxiety in parents. Findings from a series of questionnaire studies that tried to address two main goals of this programme of research were presented in this thesis. This final chapter will summarize the main findings of this research before considering implications and future directions.

10. 1. Interrelationship between repetitive behaviours, sensory problems and anxiety

10. 1. 1. Why is the interrelationship between repetitive behaviours, sensory problems and anxiety an important area of research?

Restricted and Repetitive behaviours (RRBs), anxiety and sensory problems each create a number of difficulties for individuals with autism and their families. RRBs often interfere with a child's ability to learn, engage in toy play and to attend to academic instructions (Nuzzolo-Gomez et al. 2002), and also create social stigma that further reduces opportunities for interaction with peers. In addition, RRBs present management challenges and interfere with family functioning and have been cited among the most stressful behaviours for parents (Bishop et al., 2007). Similarly, both anxiety and sensory problems have been reported by parents to have a more negative impact on both child's and family's functioning than the autism itself (Ozsivadijan, Knott, & Magiati, 2012; Ben-Sasson, Soto, Martinez-Pedraza, & Carter, 2013). It is not surprising then that sensory features, repetitive behaviours and anxiety each represent targets for autism interventions. However, understanding of the complex inter-relationships between these constructs is a necessary step in further enhancing the specificity of interventions as well as the sensitivity of outcome measures. Understanding the interrelationship between these constructs is also necessary to further our theoretical understanding of each of these problems, which, as discussed throughout this thesis, have until recently received considerably less attention than other aspects of autism.

10. 1. 2. What were the aims and what has this programme of research achieved?

As discussed throughout this thesis, early autism researchers (Hutt et al., 1964; Ornitz & Ritvo, 1968; Kinsbourne, 1980) suggested that RRBs serve as coping mechanisms for maintaining a homeostatic state of arousal by increasing stimulation and arousal in cases of under-stimulation and reducing arousal when a person is over-stimulated and distressed. In other words, repetitive behaviours were hypothesised to play a role in controlling sensory unpredictability and warding off anxiety. These early proposals were further developed in our recent review (Leekam, Prior, & Uljarevic, 2011) by suggesting the possibility that different types of repetitive behaviours might be differently associated with sensory symptoms and anxiety. Since this association has not been fully examined in children and adolescents with autism, the main goal of the first part of this thesis was to describe the inter-relationship between different types of repetitive behaviours, sensory problems and anxiety.

Before addressing this goal, it was first necessary to explore and understand each of these behaviours as separate entities with Chapter 3 exploring repetitive behaviours, Chapter 4 anxiety and Chapter 5 sensory problems.

As the type of measure that is used to assess RRBs influences the conceptualization of RRBs first main aim of Chapter 3 was to identify and validate a measure of RRBs that would allow detailed and systematic assessment of repetitive behaviours, both in autism and TD population. This chapter provided a validation and examined the factor structure of the Repetitive Behaviour Questionnaire-2 (RBQ-2; Leekam et al., 2007). The results indicated that the RBQ-2 provides a highly reliable measure of repetitive behaviours for children with autism aged from 2 to 17 years. A two-factor structure resulted; the first factor best described as repetitive motor and sensory behaviour (RSM) and the second factor is best described as the insistence on sameness (IS), which best represented the data in population with autism. Internal consistency was high for both the total RBQ-2 scale and for each RSM and IS subscale separately. As discussed throughout this thesis, at present, it is not clear whether RSM behaviours are precursors (earlier developmental form) of the same type of behaviours that is then replaced by the IS behaviours or whether these two behaviours represent independent classes of behaviours. The second main aim of Chapter 3 was to use the secondary longitudinal repetitive behaviours data to address this issue. Longitudinal secondary TD data collected on RRBs when children were 15, 24 and 72 months old, suggested that IS and RSM behaviours represent relatively independent classes of behaviours.

As it has been pointed out, inconsistencies in the research on anxiety in autism are in a large part due to the issues of how to best assess the anxiety in this population. Two major issues related to the assessment of anxiety that were identified are: the choice of appropriate measure and the informant issue. Therefore, the main aim of Chapter 4 was to identify and validate an appropriate anxiety measure, explore the issue of potential discrepancies between informants and then to use identified measure to explore frequency and moderators of anxiety in a population of children and adolescents with autism. This provided validation for different versions of Spence Anxiety Scales (which were identified through a systematic literature search as the most appropriate measure to be used with a population of children and adolescents with autism). Internal consistency of total anxiety scores provided by all three versions of the Spence Anxiety Scales was either excellent or good. Although internal consistency for anxiety subscales was lower, in general it was satisfactory with a few exceptions such as OCD and physical injury fears subscales of the Spence Preschool Scale. The analysis on the issue of potential discrepancies between informants found a good agreement on total anxiety scores between children's and parents' reports. 49% of children met the criteria for the significantly elevated total anxiety. When anxiety subtypes were considered, it was found that separation anxiety and physical injury fears were the most prevalent subtypes suggesting that children and adolescents with autism tended to express more immature types of anxiety. The analysis on the mediators of anxiety suggested that anxiety was not associated with chronological age, impairments in communication and social interaction and expressive language levels.

Although it is well established that sensory modulation problems are very prevalent in autism, it is still not clear which (if any) of the modulation problems are specific to population with autism. As discussed in Chapter 1, several studies have found presence of various sensory modulation problems in the same individuals i. e. being hypo- and hyper-responsive at the same time. The simultaneous presence of hypothetically opposite sensory response patterns (hyper- and hypo-responsiveness) in autism is an interesting paradox that surprisingly had not been properly looked into before this thesis. The main aim of Chapter 5 was therefore, in addition to exploration of how frequent sensory problems are in population with autism to examine the mutual relationship between sensory hyper-, hypo-sensitivity, sensory avoidance and sensory seeking types of sensory responsiveness. Chapter five provided evidence that sensory problems in children and adolescents with autism are both multisensory and multimodal in nature. More precisely, only 2 children had problems in a

single sensory modality while, on the other hand, 40% of children had problems simultaneously across all five primary sensory modalities. Almost 91% of children and adolescent showed a mixed type of sensory modulation problems with 65.3% of children having problems in all four sensory quadrants and 25.6% of children for 3 out of 4 quadrants (10.2% for registration + seeking + avoidance 12.24% for registration + sensitivity + avoidance combination). The second aim of this chapter was to explore the mutual relationship between four sensory quadrants i. e. sensory hyper-sensitivity, sensory hypo-sensitivity, sensory seeking and sensory avoidance. The results suggested that children with autism and adolescents fluctuate between the states of hypo- and hyper-responsiveness and that avoidance behaviours are compensatory strategies related to being over-stimulated. Mediation analyses also suggested that seeking behaviours are more related to being over-stimulated rather than hypo-stimulated providing comfort/soothing from sensations. Younger children exhibited more sensory seeking behaviours. Sensory sensitivity and poor registration were moderately associated with SCQ communication scores.

Finally, chapter 6 examined the interrelationship between repetitive behaviours, anxiety and sensory problems. It was found that while both IS and repetitive motor (RM) behaviours were associated with sensory problems, only IS behaviours were associated with anxiety while RM behaviours were not, supporting our (Leekam, Prior, & Uljarevic, 2011) hypothesis that different types of RRBs were differently associated with anxiety and sensory problems. Mediation analyses showed that sensory sensitivity and anxiety were reinforcing each other's relationship with IS behaviours and implied that, although IS behaviours might be mechanisms for warding off anxiety in a short term, that in the long term they actually reinforce anxiety, supporting proposal put forward by Rodgers et al. (2012b).

10. 2. Correlates of anxiety in parents

10. 2. 1. Why is it important to understand anxiety problems in parents of children with autism?

Although the main effects of autism are on the individual who is on the autism spectrum, families of Individuals with autism, in particular their parents, are greatly affected by this condition as well. Indeed parents of children with autism have been found to have higher levels of stress and higher prevalence of anxiety when compared to both parents of typically developing (TD) children and parents of children with other disabilities.

Understanding the factors that are associated with anxiety in parents is necessary in order to design efficient parent support programmes.

10. 2. 2. What were the aims and what has this programme of research achieved?

As noted above, and throughout this thesis, it is established that anxiety is a frequent problem in parents of children with autism. It has been suggested that parental anxiety is influenced by the following three types of factors: characteristics of the child, characteristics of the parent, and environmental characteristics. However, an exhaustive literature review conducted for this thesis identified that research to date has mainly considered anxiety in parents to be a consequence of a burden caused by raising a child with autism and that parents' own characteristics that might serve as risk for their anxiety have been under-researched. Therefore, the second major goal of this thesis was to systematically evaluate the relative contribution of children's characteristics, environment characteristics and parental own characteristics to parents' anxiety levels.

The aim of Chapter 8 was to explore the frequency of anxiety in parents of children with autism, the relationship between parents' anxiety levels and the following variables: children's age and core autism features, environmental characteristics such as support, marital status and number of children and coping strategies that parents use to deal with stress as well as parental empathy traits (as a proxy for BAP traits). The findings from this chapter confirmed that anxiety is very prevalent among mothers of children with autism with 46% of mothers meeting the cut-off criteria for clinically significant anxiety. None of children's or environmental characteristics were associated with mothers' anxiety levels. It was found that higher levels of anxiety in mothers were associated with higher use of avoidant style of coping.

Chapter 9 had the goal of examining the relationship between parental anxiety and the levels of intolerance of uncertainty and sensory problems, two of individual traits that have been identified to serve as major risk factors for anxiety in general population but have not been evaluated in a population of parents with autism. In addition to this, this chapter aimed to provide data on the factors that influence parents' choice of coping strategies. Results from Chapter 9 suggest that, in addition to the use of escape-avoidance coping as a dominant coping style, what distinguished the anxious from the non-anxious group of mothers were higher levels of intolerance of uncertainty, and higher levels of sensory over-sensitivity. Furthermore, the hypothesis that sensory sensitivity might be related to escape-avoidant

coping strategy was supported and the mediation analysis suggested that sensory sensitivity, escape-avoidance coping and anxiety reinforced each other. Also, sensory problems were found to be very prevalent in mothers, with 62% of the mothers scoring higher on sensory hypo-sensitivity quadrant than the TD norms, 44% of parents scoring higher for sensory sensitivity quadrant and 48% higher for sensory avoidance quadrant and 60% of the parents had lower sensory seeking scores than the TD norms. Finally, a strong association between sensory problems in mother and their children were found. All of the above mentioned results represent a unique contribution of this programme of work to the literature on anxiety in parents of children with autism since, at the time of the writing none of these concepts have been addressed in the existing literature. The findings from this thesis also showed that parental characters had more important contribution to mothers own anxiety level than characteristics of children and characteristics of the environment.

10. 3. Limitations

The work of this thesis was limited by a number of factors which constrain its conclusions. These have been referred to in each chapter and the main limitations are reviewed again here.

The first major limitation of this thesis, was sample size which was the consequence of a recruitment process (please see section 2.6 for more details). The sample size in particular limited the analysis of the relationship between children's and parents' reports on anxiety presented in Chapter four, as there were only 18 overlapping parent-child reports for anxiety. This necessarily limits the generalizability of that particular finding. Furthermore, due to only 18 overlapping reports, it was not possible to compare the reports on separate anxiety subtypes. However, it is important to point out that, as suggested in Chapter 3, the sample size was sufficient for conducting exploratory factor analysis as with the sample size of 120 participants, analysis had 6.31 participants per item. Furthermore, all the analyses throughout the thesis were supplemented with effect sizes that in all cases suggested that results were not likely to change in a larger sample.

The second major limitation was the fact that it was not possible to conduct cognitive assessments for the whole sample and that DISCO items were used as a proxy for developmental level. Although DISCO items were used previously by Honey (2007) and Honey, Leekam, Turner and McConachie (2007) as an index of language level they were not validated prior to this thesis. As has been reported in Chapter 2, DISCO scores did

significantly correlate with both Mullen's Expressive Language scores and WASI Verbal IQ scores from a subsample of children and adolescents with autism from the primary data set. Also, as already discussed, an independent published dataset of DISCO data ($N = 88$) was consulted in order to try to validate the language expressive scale used in this thesis. Using this independent dataset (children of the same age range selected), the expressive language scale was compared with WASI verbal IQ score. A high correlation of $r = .467$, $p = .000$ was found. This analysis used the dataset from Kent, Carrington et al. (2013) and this particular analysis is reported in Kent, 2013 (PhD thesis). Although in this thesis I used the expressive language measure alone, this result indicates that it is comparable to other measures. However, as noted throughout this thesis, the lack of cognitive assessments for majority of children and the use of DISCO scores does present significant limitation and all the analysis that used these scores should be considered exploratory and results preliminary in nature.

The third limitation was reliance on parent reported measures only and the lack of third-party informants such as teachers or other members of family. This issue, although by no means specific to the work presented in this thesis should be taken into account when results are interpreted. Furthermore, as discussed throughout this thesis, the use of questionnaire data and cross-sectional research design is tied to clear limitations in terms of identifying underlying neurophysiological mechanisms, identifying causal relations and developmental trajectory of the investigated phenomena. However, results from the questionnaire data provide solid starting point for future studies as will be discussed in next subsection.

Finally, although as described in Chapter 2, families were recruited through different recruitment channels, the issue of self-selection of parents who were anxious one hand, and the possibility that high anxiety or depression precluded some parents from taking part on the other, should be taken into account.

10. 4. Implications and Future Directions

Findings from this thesis showed relative independence of IS and RSM behaviours in terms of their developmental trajectory in a sample of TD children and suggests that these two classes of behaviours might indeed differ in terms of their genetic and neurobiological basis. In terms of clinical relevance, the importance of early interventions has been highlighted by several authors (Dawson, 2008; Vismara & Rogers, 2010) and RRBs are indeed an important early intervention target as these behaviours create significant difficulties

by themselves but can also interfere with the development of social and communication skills and reduce the efficacy of interventions that aim to improve social and communication functioning in children with autism (Ben-Itzhak & Zachor, 2009; Hudry et al., 2013). The finding that RSM and IS behaviours are relatively independent is an important consideration as it suggests that targeting RSM behaviours which emerge earlier, will not have an effect on IS behaviours and that separate strategies should be employed for the reduction of IS behaviours as well.

The results from the RBQ-2 factor analysis have clinical implications for diagnostic assessment. These findings indicate that RBQ-2 is a suitable measure of RRBs not only for very young typically developing children but also for children and adolescents with autism. As discussed in Chapters 1 and 3, 18 of its 20 RBQ-2 items come from the DISCO, a comprehensive semi-structured diagnostic interview. Furthermore, these RRB items are part of the DISCO diagnostic algorithms for ICD-10 and DSM-5 (Kent, Carrington et al., 2013). It is therefore possible that the RBQ-2, due to the fact that it consists of items drawn from a diagnostic interview, could function as a stand-alone supplement to the diagnostic process.

However, before the use of the RBQ-2 in clinical settings could be recommended, further comparative research with females with autism instead of the predominantly male sample included in thesis, research across all developmental levels and other clinical groups should be conducted. Analysis of convergent validity of the RBQ-2 is also needed, comparing it with questionnaire measures such as CRI and RBS-R, parent interviews such as DISCO and ADI-R and observational assessments (e.g. ADOS). Meanwhile, the findings from this thesis indicate the value of collecting information on RRBs through parent questionnaires.

This thesis explored the interrelationship between RRBs, anxiety and sensory problems and found that RM and IS behaviours showed different associations with anxiety and sensory problems. Furthermore, a series of mediation analyses showed that anxiety and sensory sensitivity and sensory avoidance were reinforcing each other's relationship with IS behaviours. However, due to the cross-sectional nature of the research presented in this thesis, it was not possible to assess whether, in terms of developmental trajectory, sensory problems predated anxiety as suggested by Green and Ben-Sasson (2010) or whether anxiety might be the contributing factor in development of sensory problems (Niles et al., 2013). Furthermore, although one explanation is that IS behaviours are successful in reducing anxiety in the short term but in the long term they actually reinforce anxiety (Rodgers et al.,

2012), the cross sectional design limits us in drawing this conclusion here. Future work should employ longitudinal design in order to further develop findings from this thesis and answer questions raised above and in Chapter 6.

As already discussed in chapter 6, an interesting possibility is that both sensory problems and anxiety might be a consequence of a common underlying mechanism which would lead to the dysfunction in the arousal modulation and over-focused attention. In this case, sensory problems would be developmentally earlier manifestation of such dysfunction, while anxiety would be a later manifestation. Indeed, at present, neurophysiological mechanisms underlying atypical sensory problems, RRBs and anxiety are not completely understood. One possibility is that the underlying mechanism might be an exaggerated perception of even the slightest changes in the environment, and failure to habituate to these changes leads to sensory overstimulation, distress and the perception of the environment as highly unpredictable. This proposal builds on findings from Kootz, Marinelli and Cohen (1982) that problems in filtering of environmental stimulation and modulation of response to novelty might be an important feature of autism. Furthermore, a series of recent studies conducted in TD child and adult population as well as population at risk for developing of anxiety seems to support this possibility. For example, research has shown that increased and sustained orientation to novelty is a feature of behaviourally inhibited infants and toddlers, that this attentional profile is an important obstacle in the development of effective emotion regulation to novel and unpredictable situations (Fox, Russo, Bowles, & Dutton, 2001; Fox, Russo, & Georgiou, 2005), that it is associated with the preference for routines and sameness and that it represents risk for development of anxiety in later life (Reeb-Sutherland et al., 2009). Evans and Maliken (2011) recorded P300 ERP response to oddball task that was designed to assess sensitivity to asymmetry in twelve TD children and found that repetitive behaviours (assessed via CRI) were strongly associated with more sensitivity of asymmetric stimuli. Furthermore, in a study with TD adults, Herry et al. (2007) used a series of irregular auditory stimuli to examine neural and behavioural responses to sensory unpredictability. They found that exposure to unpredictable auditory stimuli resulted in prolonged activation of amygdala and in performance on attentional bias task that was consistent with performance of anxious individuals. Finally, a study by Gomot, Belmonte, Bullmore, Bernard and Baron-Cohen (2008) reported that a group of children with autism (CA range: 10-15 years) were faster in detecting auditory novelty than IQ, age and gender matched TD children. Gomot and colleagues also found higher activation of the right prefrontal-premotor and the left inferior

parietal regions in children with autism than in controls during the task and that interestingly, the activation of prefrontal regions showed significant positive correlation with AQ scores in both TD and children with autism.

From this literature it seems possible that a candidate mechanism described as intolerance of sensory unpredictability lies behind sensory sensitivity, anxiety and preference for sameness in individuals with autism. Future work should test this hypothesis experimentally by varying degree of temporal predictability of stimuli presented across different modalities and monitoring neurophysiological responses in individuals with autism. It would be also important to include other clinical groups that show sensory problems, anxiety and RRBs such as ADHD and OCD in order to examine the extent to which this mechanism might be common across different disorders.

The results of the thesis showed that mother's own characteristics contributed to their anxiety levels. These characteristics included intolerance of uncertainty, sensory sensitivity and escape-avoidance coping and made a greater contribution to their anxiety than characteristics of their children. This suggests that on one hand, these characteristics should be explored more in the research literature and also that parents need proper support in raising children with autism.

There are several ways that this could translate into practical support services. Parents often have to wait for long periods of time to be seen for diagnosis. In addition to this wait, parents also report diagnostic process to be confusing and sometimes frustrating experience with parents usually seeing an average of 4.5 professionals before the diagnosis is established (Siklos & Kerns, 2007). At least half of parents report not to be satisfied with the diagnostic process (Chamak et al., 2011). In particular, one of the major sources of parental dissatisfaction were difficulties in obtaining relevant information from professionals during the diagnostic process and especially after. For example, an online survey conducted by Rhoades, Scarpa and Salley (2007) showed that only 40% of professionals provided additional information about autism to parents after the diagnosis was established, only 15–34% provided advice on relevant intervention programs, and only 6% of professionals referred parents to an autism specialist following diagnosis. Findings from this thesis that desire for predictability was highly correlated with anxiety in mothers of children with autism is very relevant here. It suggests that lack of adequate support during diagnostic process and lack of relevant information linked to diagnosis might be particularly distressing. Putting adequate support in terms of streamlined information packages for parents is of great

importance as it will reduce uncertainty and save parents from being overburdened with unnecessary activities.

Furthermore, the finding that anxiety in parents was predicted by the escape-avoidance coping style and desire for predictability factor of intolerance of uncertainty points to possible directions for the development of support programmes for parents. For example, approaches such as the Unified Protocol for the Transdiagnostic Treatment of Emotional Disorders (Barlow, Allen, & Choate, 2004) incorporate elements from several evidence based treatments and was developed for dealing anxiety and emotion-related disorders. This protocol is well-suited to address both intolerance of uncertainty and coping because it alters faulty antecedent cognitive appraisals, modifies avoidant behavioural tendencies, prevents emotional avoidance and facilitates emotional exposure. It has been shown that this approach reduces intolerance of uncertainty, both behavioural and emotional avoidance and reduces symptoms of anxiety and depression in groups of patients diagnosed both with heterogeneous anxiety disorders and depression (Boswell, Thompson-Hollands, Farchione, & Barlow, 2013).

Research has shown that higher parenting stress, anxiety and depression are ubiquitous factors in poorer outcomes across a range of child mental health interventions (Bögels & Phares, 2008). Furthermore, several studies have shown that elevated levels of stress and anxiety in parents of children with autism, prior to the beginning of intervention, were associated with poorer intervention outcomes for their children (Osborne, McHugh, Saunders, & Reed, 2008; Perry et al., 2008; Shine & Perry, 2010). For example, in a study of different types of teaching interventions Osborne et al. (2008) reported that high levels of pre-intervention parenting stress reduced the intervention gains in intellectual functioning, adaptive and social skills in their children, particularly in the case of high intensity interventions. As parents are expected to act as co-therapists in majority in comprehensive intervention packages (National Research Council, 2001), it is clear that preventing anxiety and reducing levels of stress in parents is of crucial importance for a good intervention outcome of their children as well.

This thesis has not explored levels and correlates of anxiety in fathers of children with autism. Research on parenting in typically developing population has clearly demonstrated the important role of positive involvement of fathers in better social, emotional and cognitive development in their young children and in better self-esteem, academic achievement and mental health when their children reach adolescence and adulthood (Lamb, 2000; Lamb &

Lewis, 2004; Tamis-LeMonda & Cabera, 2002; Bögels & Phares, 2008; Möller, Majdandžić, Vriends, & Bögels, 2013). Furthermore, research in the field of developmental psychopathology has shown that anxiety and depression in fathers are associated with a range of externalizing and internalizing problems in their children (Phares, Lopez, Fields, Kamboukos, & Duhig, 2005; Phares, Fields, & Binities, 2006; Bögels, Stevens, & Majdandžić, 2011; Verhoeven, Bögels, & Van der Bruggen, 2012). As currently the knowledge on influence of having a child with autism has on fathers' well-being is very limited, this is an important avenue of future research.

Research has shown that well-functioning marital relationship is a protective factor against the stress related to parenting for parents of both typically developing (Belsky, 1984; Benzies, Harrison, & Magill-Evens, 2004) and children with intellectual disability (Essex & Hong, 2005; Kersh, Hauser-Cram, & Warfield, 2006). Considering the fact that rate of divorce in parents of children with autism has been found to be twice as high as in families of TD children (Freedman, Kalb, Zaboltsky, & Stuart, 2012), research on marital relationships in parents of children with autism is of particular importance. Marital relationship is also important to study as parents form a dynamic system in the way they influence their children and the quality of marital relationship can influence parent-child relationship (McHale & Rasmussen, 1998; Katz & Low, 2004; Bögels et al., 2011). For example, a study by Bögels, Barmelis and van der Bruggen (2008) found that in families where father had elevated levels of anxiety, fathers showed more controlling parenting and both mothers and fathers were more rejecting towards their anxious child than in families where none of parents were anxious. Furthermore, fathers dominated the conversation and interaction with child and were less supportive of their spouse.

It is clear that the burden related to raising a child with autism can have a negative impact on parental well-being, especially in parents who are more susceptible to negative effects of stress, and that consequent stress and anxiety in parents can negatively influence family system and have negative effects on child with autism itself. However, cross-sectional designs are unable to capture these dynamic processes that develop and unfold over time and it is clear that carefully designed longitudinal study combining the use of standardized questionnaires, structured or semi-structured interviews, in-depth qualitative interviews with thematic analysis, electronic diaries, observational designs and the use of physiological measures and supported by the use of advanced statistical approaches such as structural equation modelling, multilevel modelling, sequential and path analysis is necessary to answer

some of the questions and themes that were raised by findings from this thesis and summarized in this discussion section.

10. 5. Conclusions

In conclusion, despite some of the limitations, this thesis provided significant contributions to the literature on repetitive behaviours, anxiety and sensory problems in autism and to the literature on anxiety in parents of children with autism. Unique contributions of this thesis were: (a) validation of a questionnaire that provides detailed assessment of repetitive behaviours (Repetitive Behaviours Questionnaire-2) in those with autism, addressing the need for such a measure in a literature dominated by the use of ADI-R and RBS-R, instruments whose limitations were addressed in chapter 3; (b) reporting for the first time on the longitudinal trajectory of two subtypes of repetitive behaviours – repetitive sensory and motor behaviours and insistence on sameness behaviours, and finding that they are relatively independent in terms of their development; (c) validation of the Spence Anxiety Scales for the measurement of anxiety in the population of children and adolescents with autism and providing original data on how can these scales be modified for the use with this population; (d) characterizing the inter-relationship between different sensory processing response patterns (sensory quadrants) and the inter-relationship between repetitive behaviours, sensory problems and anxiety in children with autism; (e) for the first time in the literature on the anxiety in parents of children with autism considering intolerance of uncertainty and sensory problems as possible risk factors for parental anxiety and also as a contributing factors to the use of escape-avoidance coping as a dominant coping strategy.

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Appendices

Appendix 1 – The Relationship between RRBs and other characteristics in individuals with autism

Table 1. Relationship between Repetitive behaviours and other characteristics in individuals with Autism

Authors	Subjects	RRBs Measure	Chronological Age	Developmental Level	Other Variables (Social/Communication; Adaptive level)
Piven et al., 1996	N= 38 HFA adolescents and adults (age range: 13 -28 yr)	ADI-R	No significant change in RRBs from age 5 to the current age. 55% of individuals improved in RRBs, none of the subject showed RRBs increase.		
Cox et al., 1999	N= 50 children prospectively identified with autism or PDDs at the age of 20 and 42 months	ADI-R	Minority of children with autism and PDDs showed definite RRBs abnormality at 20 months. At 42 months, more children showed abnormalities on: hand and finger, and complex mannerisms, and repetitive use of objects		
Liss et al., 2001	N= 35 HFA children (mean CA= 9yr) and N=31 age matched children with developmental language disorder, and 40 LFA	Wing Autism Diagnostic Interview Checklist (Wing, 1985)			In HFA group, RRBs were significantly correlated with adaptive behaviour, there was no such correlation for the LFA group.

	children (CA= 9yr) with 17 age matched children with low IQ.				
Authors	Subjects	RRBs Measure	Chronological Age	Developmental Level	Other Variables (Social/Communication; Adaptive level)
Militerni et al., 2002	N= 121 ASD children divided into toddler (N= 75, mean CA= 3.4 yr, range: 2.4 – 4.1yr) and children (N= 46, mean CA= 8.9 yr, range: 7.2 – 11.4 yr)	Semi-structured questionnaire developed by authors	While toddler group showed significantly more motor behaviours than the child group, child group showed significantly more complex RRBs.	Low IQ group (<35) had more sensory behaviours. Medium (60-70) and High (> 70) IQ groups had more complex motoric sequences	
Cuccaro et al., 2003	N= 292 autistic individuals, CA range: 3-21 yr.	ADI-R			While the repetitive sensory-motor behaviours were negatively correlated with adaptive functioning. resistance to change was not.
Fecteau et al., 2003	N= 28 autistic individuals (mean CA=13yr, range 7-20.4yr; mean IQ=83.79, range 40-108).	ADI-R	When current ADI-R algorithm scores were compared with retrospective scores (for the 4- to 5-year age range), a significant reduction in RRBs was detected.	No significant correlation between FSIQ and the level of change in RRBs.	

Authors	Subjects	RRBs Measure	Chronological Age	Developmental Level	Other Variables (Social/Communication; Adaptive level)
Moore & Godson, 2003	N= 20 children with severe communication problems, assessed at 2 years and 10 months (ADI-R) and reassessed when aged between 4 and 5 years.	ADI-R	Number of repetitive behaviours between the ages of 2 and 4 years increased. At the age of 2, body mannerisms, repetitive use of objects and unusual sensory interests were the most frequent. Between 2 assessments circumscribed interests, unusual preoccupations, compulsions and rituals, hand and finger mannerisms and repetitive use of objects increased (non significant increase). Complex mannerisms decreased significantly.		
Charman et al., 2005	N= 26 children diagnosed with autism at age 2 re-assessed at ages 3 and 7 years	ADI-R	RRBs scores increased between ages 2 and 3 and then decreased by age 7.		Social and communication domain scores at age 3 were predictive of RRBs scores at age 7. However, repetitive behaviours at age 3 were not predictive of any outcome domain at age 7.

Authors	Subjects	RRBs Measure	Chronological Age	Developmental Level	Other Variables (Social/Communication; Adaptive level)
Gabriels et al., 2005	N= 14 ASD individuals, divided in high NVIQ group (N=8, mean CA=10.6 yrs NVIQ > 97,) and low NVIQ group (N= 6, mean CA=10.8 yrs, NVIQ < 56).	RBS-R	RRBs total scores and subscale scores were significantly higher in lower NVIQ group. When adjusted for multiple comparisons the groups differed on only on the Sameness scale.		Total adaptive scores negatively correlated with total RBS-R scores. Total RBS-R scores more highly correlated with communication than with social ability (VABS).
South Ozonoff, & McMahon, 2005	N= 21 HFA (mean CA=14.1yr), N=19 AS (mean CA=14.28) and N=21 TD individuals (mean CA= 13.34) matched on VIQ, PIQ and FSIQ.	Repetitive Behaviour Interview (RBI; Turner, 1997) Yale Special Interests Interview (YSII; South, Klin, & Ozonoff, 1999)	None of RRBs categories were significantly correlated with CA. Scores on object use, motor movements, and rigid behaviours were highest in the preschool years and then decreased. Circumscribed Interests showed gradual increases over time.		
Bishop, Richler & Lord, 2006	N=830 children with ASD (N=560 autism, N=268 PDD-NOS, 2 AS). Mean CA 58 months (range: 15 months – 11 years, 11 months).	ADI-R	CA was positively associated with the self-injury, sensitivity to noise, circumscribed interests, difficulties with change in routine, resistance to trivial changes in the environment, and compulsions and rituals and negatively with repetitive use of objects and unusual sensory interests	NVIQ was positively associated with circumscribed interests and negatively with self-injury, unusual preoccupations, repetitive use of objects, unusual sensory interests, hand/finger mannerisms, and complex mannerisms	

Authors	Subjects	RRBs Measure	Chronological Age	Developmental Level	Other Variables (Social/Communication; Adaptive level)
Szatmari et al., 2006	N= 339 individuals with autism, mean CA: 100.79 months. Mean IQ (Leiter) = 65.7 (SD = 28.7).	ADI-R			Repetitive sensory-motor behaviours negatively associated with Vineland and ADI-R communication scores. Insistence on sameness behaviours positively associated with communication scores.
Hus et al., 2007	N= 983 individuals (N= 663 with autism, N= 320 with ASD). All with mean CA= 7.75yr (range 4-52 yr).	ADI-R	IS behaviours not associated with either CA, IQ, or symptom domains	Lower functioning group showed higher levels of RSM behaviours.	
Richler et al., 2007	N= 165 ASD (CA < 3yr), N= 44 DD (13-35 months) and N= 65 TD children (CA < 3yr). Children were assessed when they were 2, 3, 5 and 9yr old.	ADI-R	RSM behaviours were very common in children with ASD, insistence on sameness behaviours were not.		
Honey et al., 2008	N= 104 children with ASD or language disorders, CA range: 24-48 months.	ADI-R	ADI-R repetitive behaviour algorithm scores increased over time.		Ability was related to the degree of RRBs. The only exception was a subgroup of relatively able children.
Lam, Bodfish & Piven, 2008	N= 316 ASD individuals. Mean CA=9.02 yr (range 20 months to 29 yr). Mean IQ=69.5 (range 20-133).	ADI-R	Higher RM behaviour scores associated with younger age. IS and CI scores were not.	Higher RM behaviours associated with lower VIQ. IS and CI scores were not	Higher RM behaviours associated greater social deficits, communication impairments and loss of skills. Higher scores associated with greater social deficits and communication impairments. CI were not associated with either.

Authors	Subjects	RRBs Measure	Chronological Age	Developmental Level	Other Variables (Social/Communication; Adaptive level)
Paul et al., 2008	N= 37 children with ASD, assessed at two time points: when they were 15-25 months old and 12 months later	ADOS			Lower RRBs at time 1 (together with better receptive language) were significant predictor of better expressive language 12 months later. VABS was used to assess both expressive and receptive language
Mirenda et al., 2010	N= 287 children with ASD, mean CA: 40.72 months	RBS-R		Developmental index standard scores (MPR) not correlated with any factors in either model.	Factors in both models negatively correlated with the VABS II total score, most strongly for RSB (Model III) and Stereotypy (Model V).
Morgan, Wetherby & Barber, 2008	N= 50 ASD, N= 25 DD and N=50 TD children. Mean CA at ADOS assessment= 44.18 months for ASD group and 47.33 months for DD group.	RSMS: companion to Communication and Symbolic Behaviour Scales (CSBS; Wetherby & Prizant, 2002)		No significant correlations between RSM with body and NVDQ and VDQ in ASD group. RSM with objects in 2nd year significantly predicted NVDQ and VDQ in the fourth year for the ASD group. Rate and restricted inventory of RSM with objects negatively correlated with NVDQ and VDQ.	
Watt et al., 2008	N= 50 ASD, N=25 DD and N=50 TD children, all with CA range: 18 - 24 months.	RRBs were coded from videotaped Behaviour Sample of the CSBS		Significant correlations between RRBs with objects and developmental level and between RRBs with objects and the social composite were found.	Significant correlations between RRBs with objects in the 2nd year and verbal and nonverbal DQ on the MSEL at 3 years.

Authors	Subjects	RRBs Measure	Chronological Age	Developmental Level	Other Variables (Social/Communication; Adaptive level)
Mooney et al., 2009	N= 137 DD children with PDD and N= 61 DD children without PDD, all with CA 20-55 months.	ADI-R	IS significantly positively associated with CA and developmental age.	RSM significantly negatively associated with developmental age. IS significantly positively associated with developmental age.	
Esbensen et al., 2009	N= 712 ASD individuals (62.2 % with comorbid diagnosis of ID). Mean CA= 19.6 years (range 2-62 yr).	RBS-R	Significant negative correlation between CA and all five subscales of RBS-R were found.	Individuals with co-morbid ID showed significantly more stereotyped movements and SIB than individuals with ASD alone. ID was not significantly correlated with the expression of ritualistic/sameness behaviours, compulsions and restricted interests.	
Chowdhury, Benson, & Hillier, 2010	N= 34 HFA adults (Mean CA= 22.5 yr, range: 19-28 yr)	RBS-R	RRBs were assessed at current age and retrospectively at age of 5. All RRBs with the exception of self-injurious behaviours improved. The highest proportion of participants showed improvements on the Compulsive Behaviour subscale (75%), and the lowest proportion on the Restricted Behaviour subscale (44.1%).		

Authors	Subjects	RRBs Measure	Chronological Age	Developmental Level	Other Variables (Social/Communication; Adaptive level)
Kim & lord, 2010	N= 121 children with autism, N= 71 children with PDD-NOS, N= 90 children with non-spectrum disorder and N= 173 TD children. Children divided into 6 age cohorts (18; 19-24; 25-30; 31-36; 37-42; and 43-56 months)	ADI-R	RRBs increased with age for children with autism and PDD-NOS. RRBs severity not associated with CA.	NVIQ was not a significant predictor of RRBs for children with autism but was for children with PDD-NOS.	
Richler et al., 2010	N= 192 children with ASD who were referred for a diagnosis when they were under the age 3 and followed up at the age of 3, 5 and 9.	ADI-R	Increasing CA was associated with decreasing RSM. Higher CA associated with increasing IS.	Children with higher NVIQ scores at age 2 showed higher decrease in RSM scores over time. NVIQ at age 2 was not predictive of change in IS.	Milder social/communicative impairments associated with positive change in IS.
Ray-Subramanian & Weismer, 2012	N= 115 ASD children assessed at two points (mean CA at visit 1= 31 months; at visit 2= 44 months).	ADOS			RRBs negatively correlated with receptive and expressive language, and non-verbal IQ. Increase in receptive and expressive language from age 2 to 3 predicted decrease in RRBs

Appendix 2 - Summary of the most widely used questionnaire and interview measures of repetitive behaviours

(a) The multi-assessment Instruments

The Autism Diagnostic Interview Revised (ADI-R; Rutter et al., 2003)

General Overview of the instrument: The ADI-R is a semi-structured interview that can be completed in about 90 to 180 minutes. It contains the following sections: early development, communication, social development and play, repetitive and restricted behaviours, and behaviour problems. The content of the interview was based on the descriptions of autism provided in DSM-IV ICD-10 criteria for Autism.

Overview of the instrument as a measure for repetitive behaviours: Within the ADI-R there are fourteen items which target repetitive behaviours, however, the following two of these items: circumscribed interests and repetitive language are not applicable to children with CA under 4 and/or children with low VIQ. Eight of the 14 ADI-R repetitive behaviour items are included in the algorithm score and are categorised into four subsections mirroring the ICD-10 and DSM-IV RRBs groups. The following items (behaviours) are not included in the algorithm: unusual fears, self-injury, difficulties with minor changes, resistance to change, abnormal idiosyncratic responses and unusual attachment to objects.

Limitations: Despite the sound psychometric properties of the total ADI-R scores and Social and Communication subscales, the psychometric characteristics of the Repetitive Behaviour domain have been found to be weak (Lecavalier et al. 2006). It has also been suggested that ADI-R undersamples repetitive behaviours (Esbensen et al., 2009) and as already pointed out, the use of diagnostic instruments in the research is necessarily tied to the problem of circularity

The Social Communication Questionnaire (SCQ; Berument, Rutter, Lord, Pickles, & Bailey, 1999; Rutter, Bailey, & Lord, 2003)

General Overview of the instrument: The SCQ is a parent/caregiver completed questionnaire which consists of 40 items. Items are based on the ADI-R. All items are presented in yes/no format and similar like ADI-R, the SCQ is not applicable to children with CA under 4 and/or children with low VIQ. SCQ assesses three core feature of autism: reciprocal social interaction (15 items), communication (13 items) and repetitive behaviours

(8 items). SCQ has been used extensively in the ASD literature and has been found to have good psychometric properties (Berument et al., 1999, Rutter et al., 2003).

Overview of the instrument as a measure for repetitive behaviours: The SCQ assesses the following types of repetitive behaviours: verbal rituals, compulsions and rituals, unusual preoccupations, repetitive use of objects, circumscribed interests, unusual sensory interests, hand and finger mannerisms, and complex body mannerisms. SCQ also includes a question about the presence self-injurious behaviours. Rutter et al. (2003) reported that the repetitive behaviours subscale of SCQ strongly correlated with both total ADI-R and all three ADI-R subscales scores.

Limitations: Since SCQ items were based on the ADI-R, the same limitations of the ADI-R repetitive behaviours subscale are applicable. Furthermore, yes/no response format only indicates whether certain behaviour is present or not but it is not possible to determine the frequency and the impact of the behaviour in question. Also, as the factor analysis of the SCQ repetitive subscale hasn't been conducted so far, only the total repetitive behaviour score can be extracted which, having in mind heterogeneity of repetitive behaviours limits the interpretation of the collected data. Finally, SCQ has never been used specifically to assess repetitive behaviours.

Diagnostic Interview for Social and Communication Disorders - 10 (Wing, Leekam, Libby, Gould & Larcombe, 2002)

General Overview of the instrument: The DISCO is a semi-structured parent interview which provides algorithms for a number of different diagnostic systems including ICD-10 and DSM-V. The DISCO enables clinicians to rate behaviours according to whether they are present, and their current level and development. The DISCO has good psychometric properties (Leekam, Libby, Wing, Gould, & Taylor, 2002; Billstedt, Gillberg, & Gillberg, 2007; Wing et al., 2002; Nygren, Hagberg, Billstedt, Skoglund, Gillberg, & Johansson, 2009).

Overview of the instrument as a measure for repetitive behaviours: The DISCO includes more than 50 items covering repetitive behaviours and sensory interests. Twenty eight of these repetitive behaviour items are included in the DISCO ICD-10 algorithm. Algorithm items are categorised into the following types of behaviours: limited interests, routines and rituals, motor stereotypies and interests in part objects. There are also a further

30 non-algorithm items that include additional information on special skills and responses to sensory stimuli.

Limitations: Although psychometric properties of the DISCO have been previously published (Leekam et al., 2002; Wing et al., 2002), no psychometric properties or factor analysis of the repetitive behaviours subscale have been published thus far. The other significant limitation of the DISCO for assessing the repetitive behaviours domain is the fact that it is a very time consuming instrument and it might not be easy to administer it in all the situations.

(b) Instruments specifically developed for the assessment of Repetitive Behaviours

The Yale-Brown Obsessive Compulsive Scale (Y-BOCS; Goodman et al., 1989) and the Child Yale-Brown Obsessive Compulsive Scale (CY-BOCS; Scahill et al., 1997)

Overview of the instrument: Y-BOCS and CY-BOCS were originally developed for the use with the Obsessive Compulsive Disorder (OCD) population and not for the use with the populations with developmental disabilities. Both Y-BOCS and CY-BOCS are interviews administered by the clinician assessing both the presence and severity of key compulsions and obsessions. Both interviews assess the following categories of obsessions: Contamination Obsessions, Aggressive Obsessions, Hoarding/Saving Obsessions, Health-Related Obsessions, Religious/Moral Obsessions, Magical Obsessions, Sexual Obsessions, and Miscellaneous Obsessions; and the following categories of compulsions Washing/Cleaning Compulsions, Checking Compulsions, Repeating Compulsions, Counting Compulsions, Arranging/Symmetry, Hoarding/Saving Compulsions, Excessive Games/Superstitious Behaviours, Rituals Involving Other Persons, and Miscellaneous Compulsions. After identifying the most prominent obsessions and compulsions, their severity is assessed using the following five items: Time Occupied, Interference, Distress Associated with Compulsions, Resistance Against Compulsions, and Degree of Control.

Use in the ASD population: both instruments have been used in ASD population, mainly in the psychopharmacological studies (Hollander et al., 2006; King et al., 2009; McDougle et al., 2005). One factor analysis of Y-BOCS in the autistic population has been conducted thus far. Anagnostou et al. (2011) identified the following four factors: obsessions, higher-order repetitive behaviours, lower-order repetitive behaviours, and hoarding.

Limitations: although both the CY-BOCS and Y-BOCS gather information about the presence of specific obsessions and compulsions, the severity ratings do not represent individual behaviours. Because of this, distinctly different RRBs are grouped together which significantly reduces the amount of information. Because of the communication deficits that are usually present in the ASD population, in most of the cases, only the compulsive subscale can be used. Also, to the best of my knowledge, psychometric properties of Y-BOCS and CY-BOCS in the ASD population have not been evaluated yet.

The Repetitive Behaviour Scale (RBS; Bodfish, Symons, & Lewis, 1999) and the Repetitive Behaviour Scale-Revised, RBS-R; Bodfish, Symons, & Lewis, 2000)

Overview of the instrument: RBS consists of the following 3 subscales: stereotypic behaviour, self-injurious behaviour, and compulsions subscales. Since the original RBS did not assess more complex RRBs such as ritualized behaviours, insistence on sameness, and restricted interests, Bodfish et al. (2000) designed the revised version of the scale. The revised version (RBS-R) provides scores for the following 6 subscales: stereotyped behaviour, self-injurious behaviour, compulsive behaviour, ritualistic behaviour, sameness behaviour, and restricted behaviours.

Use in the ASD population: it has been used in numerous studies (Boyd et al., 2009; 2010; Dichter, Lam, Turner-Brown, Holtzclaw, & Bodfish, 2009; Esbensen et al., 2009; Gabriels et al., 2005; 2008; Mirenda et al., 2010) that supported the proposed factor structure of the instrument and found the psychometric properties to be acceptable. For example, a factor analytic study by Lam et al. (2007) identified 5 factors that accounted for 47.5% of total variance in a group of 307 individuals with ASD aged 3-48 years. Identified factors mostly replicated original conceptualization of the scale with an exception of collapsing ritualistic and sameness behaviours into one factor.

Limitations: both original RBS and the revised RBS-R are time consuming instruments. Furthermore, a large proportion of the items are appropriate only for younger children or individuals with lower developmental level thus limiting its use with higher functioning ASD individuals and also its use in studying the whole range of RRBs in TD population.

The Repetitive Behaviour Interview (RBI; Turner, 1995)

Overview of the instrument: the RBI is a semi-structured parent-report interview which consists of 50 items. RBI provides the total score and the scores for the following 4 subscales: motor movements, object use, repetitive use of language, and insistence on rigid routines. Parent rates each behaviour on both the frequency and the duration of the behaviour in questions (the duration is assessed on a 5-point scale ranging from ‘less than 60 seconds’ to ‘more than 30 minutes’).

Use in the ASD population: The RBI has been used in several studies (Turner, 1995; South et al., 2005), however, its psychometric properties have not been evaluated.

Limitations: the presence of repetitive use of language subscale limits the population of the individuals with ASD to which RBI is applicable as it is not suitable for young children and individuals with limited language. Although the presence of over 50 items allows detailed assessment of wide variety of behaviours, this also makes it time consuming.

The Repetitive Behaviour Questionnaire

Overview of the instrument: The RBQ was developed from the RBI. It is a 33 item, parent-completed questionnaire that collects information about the severity, frequency and the nature of repetitive behaviours. Items are grouped into the following 4 subscales: repetitive movements, repetitive use of language, sameness behaviours and circumscribed interests.

Use in the ASD population: the RBQ was used in a number of studies of repetitive behaviour in ASD (Barrett, Prior, & Manjiviona, 2004; Ozonoff et al., 2000; Zandt et al., 2007; Zandt et al., 2009; Honey et al., 2012). In a recent factor analytic study, Honey et al. (2012) identified a reliable two factor solution. Two factors, sensory/motor behaviours and sameness/circumscribed interests accounted for 42% of total variance. Honey et al. (2012) also reported that RBQ showed good psychometric properties.

Limitations: RBQ has similar limitations like the RBI from which it was developed.

The Repetitive Behaviours Questionnaire-2 (RBQ-2, Leekam et al., 2007)

Overview of the instrument: The RBQ-2 is a 20 item parental questionnaire. It was developed from the RBQ and DISCO. RBQ-2 provides both total score and scores for the 2 factors (repetitive motor 4 sensory (RSM) factor and rigidity, routines, and preoccupations (IS) factor) and/or four factors (motor, rigidity, preoccupation the sensory factors).

Psychometric properties of RBQ-2 have been reported for typically developing children (Leekam et al., 2007; Arnott et al., 2010). It has been found that RBQ-2 has good psychometric properties with high internal consistency. With respect to the two factors solution, it has been shown that both factors have good reliability (Cronbach's alpha = 0.81 for the Factor 1 and 0.71 for the Factor 2; Arnott et al., 2010). For the four factor solution Arnott et al. (2010) found that internal consistency ranged from good (Cronbach's alpha = 0.82 for the Factor 1 and 0.74 for the Factor 2) to acceptable (Cronbach's alpha 0.64 for the Factor 3 and 0.51 for the Factor 4).

Use in the ASD population: the RBQ-2 has not been used in a published research thus far.

Limitations: although the instruments from which the RBQ-2 was developed have been used extensively in ASD populations with good psychometric properties, psychometric properties and factor structure of this instrument hasn't been evaluated in ASD population.

The Childhood Routines Inventory (CRI; 1997)

Overview of the instrument: the CRI is a 19 item parental completed questionnaire originally developed to assess the compulsive-like behaviours in TD children. Parents rate items/behaviours on the frequency/intensity, the onset of each of the behaviours and also whether the child presently engages in the behaviour in question. The CRI has been used in numerous studies with typically developing children (Evans et al., 1997; Evans & Gray, 2000; Evans et al., 2001) and children with various disabilities such as Down syndrome for example (Evans & Gray, 2000) and it has been found to have good psychometric properties. Evans et al. (1997) examined the factor structure of CRI in a population of 1492 children aged 8 to 72 months and found evidence for the following two factors: 'Just Right' factor (e.g., preference for particular ways of doing things), and repetitive behaviours (e.g., performing the same task repeatedly).

Use in the ASD population: The CRI has been used by Chen et al. (2009) and Greaves et al. (2006).

Limitations: The CRI collects data on a wide range of behaviours. However, it does not represent the range of autistic-like behaviours as there is a clear lack of items that assess sensory and motor repetitive behaviours.

Appendix 3 - Summary of the identified anxiety instruments

(a) Instruments originally designed for the assessment of anxiety in TD population:

Anxiety Disorders Interview Schedule for DSM-IV–Child and Parent Versions (ADIS; Silverman & Albano, 1996): The ADIS is a semi-structured interview appropriate for children and adolescents aged 6–17 years. It has both a parent and a child version. Anxiety symptoms are rated as either present or absent. The only difference between the child and the parent version is that the parent version also assesses externalizing disorders. The ADIS was based on the current version of the DSM. The interview is quite lengthy, taking between 60 and 90 min to administer. This instrument has been used widely in the general and various clinical populations (Silverman, Saavedra, & Pina, 2001; Wood, Piacentini, Bergman, McCracken, Barrios, 2002) and in 6 studies examining anxiety in ASD so far (Chalfant et al., 2007; Drahota et al., 20011; Sze & Wood, 2007; 2008; White et al., 2009; Wood et al., 2009). Psychometric properties of ADIS in ASD population haven't been evaluated.

The Spence Anxiety Scales (Spence, 1997; 1998): The Spence Preschool Anxiety Scale was designed to provide both the total anxiety score and scores for the following 5 types of anxiety: generalized anxiety, social anxiety, obsessive compulsive disorder, physical injury fears and separation anxiety. The scale consists of 28 items which are scored and one open-ended item assessing whether the child has experience a traumatic event.

The Spence Children's Anxiety Scale assesses six domains of anxiety including generalized anxiety, panic/agoraphobia, social phobia, separation anxiety, obsessive compulsive disorder and physical injury fears. There is parent, as well as child version of this scale. The psychometric properties of Spence Anxiety Scales have been reported to be fair to good. Spence (1997) and Essau, Muris, & Ederer (2002) found an alpha of .93 and .92 respectively for the full scale. Spence (1997) and Nauta et al. (2004) reported the following alpha coefficients for the anxiety subscales Panic Attack/ Agoraphobia (.61), Separation Anxiety Disorder (.74), Social Phobia (.74), Physical Injury Fears (.58), Obsessive-Compulsive Disorder (.74), and Generalized Anxiety Disorder-Overanxious Disorder (.67). Spence et al. (2003) reported that the test-retest reliability after 12 weeks was .63. SCAS scales have been used in 5 studies examining anxiety in ASD population (Murriss et al., 1998; Gillott, Furniss & Walter, 2001; Russell & Sofronoff, 2005; Chalfant et al., 2007; Greenway & Howlin, 2010). SCAS was used for both determining the prevalence of anxiety disorders in ASD population as well as outcome measure for CBT. Psychometric properties in ASD population haven't been evaluated.

Screening for Childhood Anxiety and Related Emotional Disorders (SCARED; Birmharer et al., 1997; 1999): SCARED consists of 41 items completed by parents and children and it takes around 15 minutes to complete. It provides a total score as well as the following subscales: somatic/panic, generalized anxiety, separation anxiety and social phobia. The five-factor structure has been independently confirmed for diverse populations (Hale et al., 2005).

Early Child Inventory-4 (ECI-4; Gadow & Sprafkin, 1997): ECI-4 is a 108 item scale designed to assesses psychiatric symptoms based on the DSM-IV classification. It has both a parent and a teacher version. The ECI-4 assesses the following symptoms: Attention Deficit Hyperactive Disorder, Oppositional Defiance Disorder, Conduct Disorder, Major Depressive Disorder, Dysthymic Disorder, PDD Symptoms, sleep problems, feeding problems, reactive attachment disorder, and tics. In terms of anxiety, the following types of anxiety are assessed: Generalized Anxiety Disorder, Separation Anxiety Disorder, Social Phobia, Posttraumatic Stress Disorder, Simple Phobia, OCD. It has been found to have satisfactory psychometric properties (Gadow, Sprafkin, & Nolan, 2001; Sprafkin, Volpe, Gadow, Nolan, & Kelly, 2002).

Multidimensional Anxiety Scale for Children (MASC; March et al., 1997): MASC is a 45-item measure of anxiety for children 4–19 years of age. It has both parental and a child version. MASC provides the total anxiety score and also the scores for the following factors: somatic/panic, general anxiety, separation anxiety, social phobia, and school phobia. Internal consistency (Cronbach's alpha) for these four subscales ranged from .74 to .85 (March et al., 1997). It has been shown to have good psychometric properties in non-ASD populations (Wood et al., 2002). However, although it has been used in 6 studies examining anxiety in ASD (Bellini, 2004; 2006; Sze & Wood, 2008; White & Roberson-Nay, 2009; White et al., 2009; Wood et al., 2009) its psychometric properties in this population haven't been evaluated.

Diagnostic Interview Schedule for Children IV (DISC IV; Shaffer, Fisher, Lucas, Dulcan, & Schwab-Stone, 2000): DISC is an interview designed to assess DSM-IV Axis I psychiatric disorders. It has both parental and child version. The DISC section on anxiety consists of 154 items and assesses the following types of anxiety: simple phobia, social phobia, agoraphobia, panic disorder, separation anxiety disorder, avoidant disorder of childhood or adolescence, overanxious disorder, and obsessive-compulsive disorder. It has been found to have adequate psychometric properties (Shaffer et al., 2000).

The Child Behaviour Checklist (CBCL; Achenbach & Rescorla, 2001): CBCL has is appropriate for children aged 1.5–18 years. It has parental, teacher, and child versions. CBCL consists of 118 items/questions and has the following 3 subscales: Problem Subscales; Syndrome Subscales; and DSM-Oriented Subscales. The CBCL anxiety assessment provides two different subscales: the Anxiety/Depressed Syndrome Subscale and the Anxiety Problems DSM-Oriented Subscale. It has been shown to have adequate psychometric properties in both general and ASD population (Pandolfi, Magyar, & Dill, 2009). CBCL has been used in several studies examining anxiety in ASD (Juranek et al., 2006; Kuusikko et al., 2008; White & Roberson-Nay, 2009).

(b) Instruments designed for individuals with DD (and ASD)

Diagnostic Assessment for the Severely Handicapped-II (DASH-II; Matson, 1996): DASH-II is a 84 item instrument designed to assess a wide range of psychopathology in children with severe DD. It has the following 13 subscales: impulse control, organic problems, anxiety, mood disorders, mania, pervasive developmental disorders/autism, schizophrenia, stereotypies, self-injurious behaviour, elimination disorders, eating disorders, sleep disorders, and sexual disorders. Each of the subscales provides three types of scores: frequency within the last two weeks, duration of the behaviour in question, and the severity of the behaviours in question within the last 2 weeks. It provides total anxiety score and not scores for the separate anxiety subtypes. It has been found to have good ipsychometric properties (Matson, 1996).

Autism Comorbidity Interview–Present and Lifetime Version (ACI-PL; Leyfer et al., 2006): ACI-PL is parental psychiatric parent interview developed to asses the presence of various types of psychopathology in children with ASD. It was developed from the Kiddie Schedule for Affective Disorders and Schizophrenia (K-SADS; Ambrosini, 2000). ACI-PL assesses the presence of the following disorders: depression, psychotic disorders, ADHD, disruptive disorders, oppositional adjustment disorders, and the following types of anxiety: panic disorders, separation anxiety, social phobia, specific phobia, generalized anxiety and OCD. ACI-PL has been used by three studies in ASD (Leyfer et al., 2006; Mazefsky et al., 2011; Mazefsky et al., 2012), however, its psychometric properties in ASD population haven't been evaluated.

Autism Spectrum Disorders–Comorbid for Children (ASD-CC; Matson & Gonzalez, 2007): ASD-CC is a 49 item instrument designed to evaluate eating problems, ADHD, conduct disorder, tic disorder, OCD, and specific phobia in children aged 3–16 years (Matson

et al., 2009). Matson & Wilkins (2008) conducted a factor and identified the following 7 factors: Tantrum Behaviour, Repetitive Behaviour, Worry/Depressed, Avoidant Behaviour, Under-Eating, Conduct, Over-Eating. In terms of anxiety assessment it only measures worry. ASD-CC was used by four studies (Davis et al., 2011a, b; Hess et al., 2010; Worley & Matson, 2011) that looked at levels of anxiety in ASD when compared to TD and non-ASD populations.

Baby and Infant Scale for Children with Autistic Traits (BISCUIT; Matson, Boisjoli, & Wilkins, 2007): BISCUIT was designed to assess socio-emotional problems in very young children aged between 17 and 37 months. Parts 1 and 3 of BISCUIT evaluate the symptoms of ASDs and externalizing symptoms respectively and part 2 assesses co-morbid psychopathology. The factor analysis conducted by Matson et al. (2009) indicated a 5 factor structure of the instrument consisting of: Tantrum/Conduct Problems, Inattention/Impulsivity, Avoidance Behaviour, Anxiety/Repetitive Behaviour, and Eating Problems/Sleeping. Matson et al. (2009) reported that Cronach's alpha for Anxiety/Repetitive Behaviour subscale was .81. Biscuit was used by 4 studies (Davis et al., 2010; Matson et al., 2010; Davis et al., 2011a; Fodstad et al., 2010,) that examined anxiety prevalence in ASD population.

Behavioural Assessment System for Children-2 (BASC-2; Reynolds & Kamphaus, 2004): was designed to be used with individuals aged between 2 and 21 years. It provides assessment of the problems: Attention Problems, Aggression, *Anxiety*, Conduct Problems, Hyperactivity, Depression, *Somatization*, *Withdrawal*, Learning Problems, Atypicality, Adaptability, Activities of Daily Living, Functional Communication, Leadership, Social Skills, and Study Skills. It has parental, child and teacher version. Achenbach et al. (2004) reported that BASC-2 had good to excellent psychometric properties. BASC-2 was used in several studies (Bellini, 2004; Burnette et al., 2005; Meyer et al., 2006; Solomon et al., 2008; Lopata et al., 2010) that explored anxiety in ASD.

Appendix 4 – Background Questionnaire

PLEASE NOTE: These questions are completed by the researcher when you meet or talk by telephone. The questions are provided below so you know what questions the researcher will ask but you do not need to fill in the form.

Questions about your child:

1. Does your child have any medical conditions (including accidents or operations, epilepsy etc.), illnesses or any behavioural, emotional, or psychological problems (including sleep problems and bowel problems) (apart from social and communication difficulties)?
Yes/No

If yes, please specify details (name of condition, age of onset, if your child is currently receiving treatment/type of the treatment, in case of accident/operation-type of accident/operation, age, length of stay in hospital)?

Name of the condition	When did condition begin	Treatment/Therapy and dates

2. Has your child been diagnosed with Autism? Yes/ No

If yes, please specify details (when was child diagnosed, who gave the diagnosis and at which centre)?

Date of diagnosis	Description of diagnosis (Autistic Disorder, Asperger syndrome, atypical autism, autism spectrum disorder etc.)	Which diagnosis centre / Name of clinician

3. Has your child received any treatment for social and communication difficulties?

Yes / No

If yes, please specify details (what type of treatment: Applied Behavioural Analysis, EarlyBird programme, TEACCH, Intensive Interaction, SPELL, PECS, The Son-Rise Program, any Dietary / Supplementary treatment, for how long, by whom, who recommended this type of treatment)?

Name of treatment/therapy	Dates when your child received treatment/therapy	Who recommended?

4. Does your child have any language? Yes / No

a) At what age did your child first start to use single words?

.....

b) When did your child first start to use to 2-3 word phrases?

.....

Does your child:

- c) Give the name of some people or things when asked Yes/No
- d) Spontaneously say names of several familiar objects for some purpose Yes/No
- e) Says phrases not longer than three words Yes/No
- f) Say some longer phrases (longer than 3 words), missing out the small linking words (e.g. “when time go on holiday?”) Yes/No
- g) Talk in spontaneous sentences using small linking words. Yes/No
- h) Use past, present and future tenses in complex sentences (e.g. “perhaps I will go out tomorrow if it has stopped raining”) Yes/No

What kind of communication does your child normally use? (Whether or not they use speech)

- a) No communication or communicates needs only _____
- b) Communicates own interests (never shares other’s interests) _____
- c) Listens, asks after and shares other peoples interests _____

5. (a) Is your child currently in a mainstream education school? Yes/No

If no, please specify what type of school your child attends, or if your child receives home schooling

.....

(b) Does your child have a statement of special education needs? Yes/No

If yes, please specify education needs

.....

6. Does your child receive extra support at school? Yes/No
 If yes, please specify support received

.....

Questions about you:

1. What is your nationality?

.....

2. What language do you speak at home in the presence of the child?

.....

3. Do you have any major medical conditions (including accidents or operations), illnesses or any behavioural, emotional, or psychological problems? Yes/No

If yes, please specify details (name of condition, age of onset, if you are currently receiving treatment/type of the treatment) and if any of your first-degree relatives (i.e. parent, sibling) have the same condition

.....

4. Please state at what age did you leave the school and what is your level of education (i.e. A levels, University Degree, etc)

.....

