An empirical investigation into autistic catatonia:

The development of the Autistic Catatonia Questionnaire (ACQ)

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ABSTRACT: “An empirical investigation into autistic catatonia: The development of the Autistic Catatonia Questionnaire” - A thesis submitted to the University of Manchester for the degree of MPhil in the Faculty of Medical and Human Sciences (School of Psychological Sciences) in 2014 by Jennifer Breen.

Introduction: Research indicates that a small proportion of young people with Autistic Spectrum Disorder (ASD) experience onset of catatonic-like symptoms in adolescence, termed ‘autistic catatonia’. Autistic catatonia is an under-researched neurologically-based condition, and little is known about the presentation and variation of symptoms. The current study aims to empirically investigate ‘autistic catatonia’ in children and adolescents with ASD via a systematic examination of the prevalence and presentation of symptoms associated with autistic catatonia. A 34-item Autistic Catatonia Questionnaire (ACQ) was developed which could be used in clinical practice and for research purposes. A secondary aim of the study is to complete preliminary investigations into the usefulness of the ACQ as a clinical measure.

Method: Caregivers or parents (n=99) provided information about the presentation of symptoms in a young person they care for via the online completion of the ACQ and two established clinical measures (the Repetitive Behaviour Questionnaire and Carer Supplement to the Glasgow Depression Scale for people with Learning Disability).

Results: Catatonic symptoms are relatively common in young people with ASD and the prevalence in the current study is much higher than has been found elsewhere, with 20.2% having an existing diagnosis of autistic catatonia. The number of core symptoms provided key information about the presentation of autistic catatonia, with the data indicating an autistic catatonia continuum and a potential clinical cut-off for diagnosis when three or more core symptoms are present. The results indicate that the ACQ is a workable clinical measure in this population with a degree of discriminant validity. Statistical analysis reduced the ACQ from 34 to 28 items, comprising six core symptom items and 22 supplementary items. There is also evidence of a relationship between the presentation of autistic catatonia and measures of depression and repetitive and restricted behaviours, although a causal relationship was not determined.

Conclusion: Preliminary investigations into the utility of the ACQ as a clinical measure and research tool are promising. Future researchers must further investigate autistic catatonia empirically and finalise diagnostic criteria to allow the progression of theoretical understanding, enable the development of safe and effective evidence-based treatments and raise the profile of autistic catatonia in mainstream ASD research.
DECLARATION

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CHAPTER 1: INTRODUCTION

1.0 Aim: This chapter aims to review the literature on catatonia and Autism Spectrum Disorder (ASD) and introduce the concept of Autistic Catatonia. The limitations of the current knowledge base will also be discussed, along with the aims of the current research project.

1.1 Overview of current study:

Many individuals with Autistic Spectrum Disorder (ASD) display catatonic symptoms in isolation but some experience a catatonic-like deterioration termed autistic catatonia. Autistic catatonia has a serious impact on an individual’s quality of life and the level of care they require. Typically, individuals affected are adolescents who find it increasingly more difficult to maintain their existing independence due to the onset of a range of chronic motor symptoms. Autistic catatonia is an under-researched and poorly understood neurological condition, and little is known about the presentation and variation of symptoms.

The current study focuses on an empirical analysis of the phenomenology of autistic catatonia by more clearly operationalising the definition and developing a clinical research measure based on this definition. The current study was not primarily focussed on aetiology or interventions for autistic catatonia, but will support future work in these areas.
The introduction to this research study will examine the concept of autistic catatonia by critically reviewing literature on this topic before focussing on the current understanding of catatonia in individuals with autism, including diagnosis and treatment options. The current study and its aims will be outlined at the end of this chapter.

1.2 Conceptualising Autistic Catatonia:

1.2.1 Catatonia:

Catatonia is a psychiatric syndrome characterised by abnormal hyper or hypokinetic movements and behavioural alterations (Fink, 1994; Bräunig, Krüler, Shuger, Höfler & Börner, 2000). Coined by Karl Ludwig Kahlbaum (1828-1899), a German Psychiatrist working in Berlin in the nineteenth century, the core symptoms associated with catatonia have remained fairly consistent and can be recognised by clinicians and non-clinicians. Kahlbaum’s description of individuals who were totally immobile as if frozen and who had seemingly lost the will or ability to interact with their environments (Fink & Taylor, 2003; Starkstein et al., 1996) continues to define the syndrome.

Catatonia is diagnosed on the presentation of both positive (i.e. a change in behaviour) and negative (i.e. a deterioration in functioning) symptoms including mutism, catalepsy, facial grimaces, catalepsy, echolalia and akinesia (International Classification of Diseases 10th Edition (ICD-10), World Health Organization (WHO), 1992; Diagnostic and Statistical Manual of Mental Health Disorders 4th Edition (DSM-IV); American Psychiatric Association (APA), 1994). In severe cases, the individual experiences a sudden and dramatic loss of functional skills that impairs their quality of life and
independence. Sufferers typically experience difficulties with independent personal care, expressive communication and engagement in activities. There are also health concerns associated with the development of catatonia as individuals often eat and exercise significantly less. Overall, the development of catatonia is very distressing both for the individual and those who are close to them.

Catatonia is rarely identified in isolation and is usually diagnosed as a co-morbid syndrome of psychological disorders such as schizophrenia (Morrison, 1976) and depression (Barnes, Saunders, Walls, Saunders & Kirk, 1986; Starkstein et al., 1996) or medical conditions such as encephalitis (Shill & Stacy, 2000) and epilepsy (Primavera et al., 1994). Catatonia is also increasingly evident in populations with developmental disorders such as autism (Wing & Shah, 2000) or Prader-Willi syndrome (Dhossche & Bouman, 1997).

The history of catatonia as a clinical syndrome is complex. Kahlbaum envisioned catatonia as a unitary disease that followed a set course in line with his classification system based on symptomatology and prognosis (Fink, Shorter & Taylor, 2010; Sienhart, Rooseleer & De Fruyt, 2011) and rejected a physiological aetiology (Fink & Taylor, 2003). Kahlbaum also noted the affective alterations associated with catatonia (Northoff et al., 1999; Carroll et al., 2008) and proposed manic-depressive illness as a factor in the development of catatonia (Barnes et al., 1986).

Emil Kraeplin (1856-1926), an eminent German Psychiatrist who postulated a biological basis to mental illness, developed a classification system that continues to influence contemporary diagnostic systems such as ICD and DSM. By focussing on classifying
accordin to symptomatology, Kraeplin believed that a discrete number of illnesses would be delineated allowing researchers to focus on aetiology and neuropathy (Bentall & Beck, 2003). Kraeplin assigned Kahlbaum’s catatonia as a basic clinical trajectory of ‘dementia praecox’ (Fink & Taylor, 2003; Shorter, 2012), focussing primarily on the motor abnormalities described by Kahlbaum (Northoff et al., 1999). Over time and influenced by protagonists such as Eugen Bleuler (1857-1939), Kahlbaum’s concept of ‘dementia praecox’ (translates as ‘senility of the young’) became relabelled ‘schizophrenia’ (Francis, Divadeenam, Bush & Petrides, 1997; Bentall & Beck, 2003).

Catatonia is not currently recognised as a separate syndrome in DSM-IV (APA, 1994) and continues to be associated with schizophrenia, which in turn remains the subject of much debate (Fink, Shorter & Taylor, 2010; Heckers, Tandon & Bustillo, 2010). The notion of schizophrenia encompassing catatonia has become an established ‘hangover’ in clinical practice despite an increasing evidence base suggesting that catatonia is more prevalent in other populations (Barnes et al., 1986; Fink, Shorter & Taylor, 2010) and that the majority of individuals diagnosed with schizophrenia do not experience catatonic symptoms (Bush, Fink, Petrides, Dowling & Francis, 1996; Heckers, Tandon & Bustillo, 2010). Many challenge this association and demand that catatonia is reclassified as an independent syndrome (Fink & Taylor, 2003; Fink, Taylor & Ghaziuddin, 2006; Rosebush & Mazurek, 2010). A range of catatonic presentations and different thresholds exist for defining catatonia, leading some authors to suggest the existence of a ‘catatonic spectrum’ (Fink & Taylor, 2003).
A biological aetiology for the majority of instances of catatonia is now commonly accepted with research indicating dysfunction in a number of brain areas associated with motor control, including the front parental cortex (Northoff et al., 1999), basal ganglia (McKenna, Lund, Mortimer & Biggins, 1991; Stoppelbein et al., 2006; Brunoni, Nakata, Tung & Busatto, 2009; Russowsky, Nakata, Tung & Busatto, 2009) and medial frontal abnormalities (Joseph, 1999). Considering the specific example of catatonia in individuals with autistic spectrum disorder, aetiology is suggested to be common neural circuitry in the cerebellum and frontal lobes (Dhossche, Carroll and Carroll, 2006; Bozkurt & Mukaddes, 2010; Barnhill, 2012), dysregultion of gamma-aminobutyric acid (GABA) (Dhossche, 2004; Dhossche & Rout, 2006; Dhossche, Shah & Wing, 2006), common genetic abnormalities on chromosome 15 (Chagnon, 2006) and brain structural abnormalities (Fink et al., 2006). Similarly, side effects or synergism of psychotropic medications such as fluphenazine, haloperidol, risperidone, and clozapine can also cause catatonic symptoms (Mahendra, 1981; Duggal & Singh, 2005).

Estimates of the prevalence of catatonia vary, usually in the range of 7-14% depending on clinical population (Bush et al., 1996; Bräunig et al., 2000; Wing & Shah, 2000), but other studies have reported higher frequencies (e.g. Chasalasani, Healy & Morriss, 2005; Ghazuiddin, Dhossche & Marcotte, 2012). Starkstein et al. (1996) found that 20% of a sample of psychiatric patients with a diagnosis of depression also displayed catatonia and there is other evidence linking specific factors that might trigger catatonia, notably traumatic experiences (Dhossche, Ross & Stoppelbein, 2012) and a common co-morbidity with depression (Fink & Taylor, 2003). While assessing individuals presenting with potential catatonia, it is important that clinicians rule out --
existing medication as a causal factor before attempting additional intervention (Dhossche et al., 2006).

Catatonia rating scales are an important clinical tool for diagnosis and to examine any changes in symptom presentation over time, which is particularly important for evaluating the effectiveness of clinical interventions. In order to assess catatonia more accurately, a number of catatonia rating scales have been developed including the widely used Bush-Francis Catatonia Rating Scale (Bush et al., 1996), the Northoff Catatonia Scale (Northoff et al., 1999) and the Bräunig Catatonia Rating Scale (Bräunig et al., 2000). Although inter-rater reliability and test-retest reliability is high for these instruments (Sienhart et al., 2011), the lack of a ‘gold standard’ definition of catatonia (Dhossche et al., 2006, p272) has restricted the development of catatonia rating scales and consequently negatively impacted the field of research (Sienhart et al., 2011). Additionally, Carroll et al. (2008) propose that individual rating scales are required for the different populations of catatonic patients. There are several inconsistencies in the extant catatonia rating scales, including inconsistent item definitions (Bräunig et al., 2000), the number of items included in each rating scale varying from 21 [Bräunig Catatonia Rating Scale (Bräunig et al., 2000)] to a possible 54 [Modified Rogers Scale (Lund, Mortimer, Rogers & McKenna, 1991)] and the threshold for diagnosis varies between two symptoms [Bush-Francis Catatonia Rating Scale (Bush et al., 1996)], at least one symptom per subdomain [Northoff Catatonia Scale (Northoff et al., 1999)] and an undetermined number [Kanner Scale (Carrol et al., 2008)]. Affective symptoms are evident in catatonic patients (Abrams & Taylor 1976), but most catatonia rating scales fail to consider these (Northoff et al. 1999). Furthermore, although a number include a measurement of severity - usually
on a 0-2 or 0-3 Likert scale - only the Bräunig Catatonia Rating Scale (Bräunig et al., 2000) allows measurement of symptoms in terms of frequency and severity, which is crucial for providing a true reflection of the longitudinal course of catatonia and the effectiveness of clinical intervention.

The uncertainty around clinical definition, lack of routine screening in high risk population groups and the low profile of catatonia may result in a potential high level of under-diagnosis (Heckers et al., 2010; Ghazuddin et al., 2012) and uncertainty around the true prevalence of catatonia. Several authors note that clinicians can find it difficult to identify catatonia in patient populations (Bräunig et al., 2000) and Fink et al. (2010) report on a Dutch research study where the research team identified nine times as many psychiatric inpatients with catatonia than the clinicians. It is clear that the knowledge base surrounding catatonia requires attention to ensure that the complexities of this syndrome, particularly factors relating to diagnostic criteria, detection and intervention, are elucidated for the benefit of those affected.

1.2.2 Treating catatonia:

Catatonia generally responds well to clinical intervention with symptoms alleviating (Fink et al., 2010; Sienhart et al., 2011) when it is identified and treated early (Carroll et al., 2008; Heckers et al., 2010). However, recurrent episodes of catatonia are not uncommon and symptom presentation is often uniform longitudinally (Francis et al., 1997). Common treatment options are medication (specifically benzodiazepines such as Lorazepam) and electroconvulsive therapy (ECT) (Francis et al., 1997; Takota & Takata, 2003; Fink & Taylor, 2003; Weiss, Allan & Greenaway, 2012; Dhossche et al., 2012). Usually, pharmacological intervention is attempted first with ECT only being
prescribed if medication has no effect or doses are dangerously high (Fink et al., 2006). There are many reports of dramatic response to pharmacological intervention within a period of a few hours (Northoff et al., 1995; Fink et al., 2006) but repeated, often daily, doses are often required to maintain the level of symptom abatement. Pharmacological approaches are inherently risky, particularly when high doses are prescribed as the likelihood of experiencing side effects increases (Medical Healthcare Regulations Agency (MHRA), 2010). There is evidence that for some patients, ECT alleviates some of the more distressing symptoms of catatonia on a short term basis (Shorter, 2012) but repeated treatment sessions are often required to maintain this effect (Kakooza-Mwesige et al., 2008; Dhossche, 2009; Wachtel et al., 2010). ECT is strongly advocated by some as a treatment for catatonia (e.g. Kakooza-Mwesige et al., 2008) but it remains controversial and there are significant ethical considerations, particularly when the patient is unable to consent to treatment due to lack of capacity or for minors aged below 18 years (Zaw et al., 1999; Dhossche et al., 2006; Wachtel, 2008; Kakooza-Mwesige et al., 2008), and legislation is in place to control the use of ECT in vulnerable groups (Dhossche et al., 2006; Ghaziuddin et al., 2010). Currently, there is little known about the mechanisms whereby ECT effectively treats catatonia.

However, it is important to note that outcomes are poorer for some patients with catatonia and experiencing more than one episode is not uncommon (Francis et al., 1997). Catatonia can be fatal if untreated or treated ineffectually (Francis et al., 1997), particularly if onset is rapid and the syndrome is acute (Barnes et al., 1986). Outcomes have been found to become more positive with age with older individuals found to exhibit fewer and milder symptoms, particularly for individuals with higher IQ (Wing & Shah, 2006) although it is unclear if this is due to symptoms decreasing with age or
that older individuals are less likely to develop catatonia. Prompt diagnosis and effective treatment are key to promoting positive outcomes (Shah & Wing, 2006; Wachtel, 2008) and response to treatment is reported to be slower after delayed clinical recognition (Dhossche, 2009).

The notion of a catatonic spectrum with potentially distinct subtypes of catatonia – which could be aetiological divergent – raises the possibility of different treatments being optimally effective for different catatonic subtypes on the catatonic spectrum (Northoff et al., 1995). In this regard, Rosebush and Mazurek (2010) postulate that benzodiazepines are less effective for individuals with schizophrenia and catatonia compared to other patient groups.

### 1.2.3 Autistic Spectrum Disorder:

Autism is a pervasive neurodevelopmental disorder that permanently affects how an individual experiences and interacts with their surroundings. First identified by Leo Kanner in 1943, autism is characterised by an atypical pattern of behaviour evident from early childhood. Traditionally, autism is diagnosed by the presence of abnormality or impairment in three key areas; social and emotional development, language and communication and flexibility of thought (known as the ‘triad of impairments’ (Wing & Gould, 1979). Deficits in these three areas often has a significant effect on an individual’s quality of life which varies considerably between individuals and often across time, with significant implications for the development of an individual’s adaptive skills. The current conceptualisation of autism is that it is a

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**Footnote:** In line with current accepted practice, from here-on in, I will refer only to Autistic Spectrum Disorder (ASD) which will subsume all previous historical terminology.
spectrum disorder and is commonly termed Autistic Spectrum Disorder (ASD), meaning that individuals can present very differently due to differing levels of impairment in each of the ‘triad’ areas.

Abnormalities in social and emotional development associated with ASD typically include difficulty forming and maintaining relationships, inappropriate attempts to interact with others, difficulty recognising and responding appropriately to how other people are feeling and a preference not to share enjoyment with other people. Three subgroups of different types of social difficulties in individuals with ASD were identified by Wing and Gould (1979) ‘passive interaction’ where social interaction is neither initiated nor resisted if approached, ‘social aloofness’ where the individual is disinterested in interacting with others and ‘active but odd’ where social interaction with others is initiated but is very unusual and often inappropriate. Communication difficulties vary from a complete lack of verbal communication to unusual manifestations of language such as repeated phrases (echolalia), monotone voice pitch or lack of singular personal pronoun use. Inflexibility of thought in autism manifests as rigidity in behaviour and play, difficulty accepting and coping with change in routine and planning for future events.

A number of psychological models have been proposed to explain the pattern of presenting cognitive impairments presenting in individuals with ASD. Baron-Cohen, Leslie and Frith (1985) proposed that impaired ‘theory of mind’, the cognitive ability to understand the perspective of others’ and their mental states, provides an explanation for the social, communication and imagination difficulties associated with ASD (Happé & Ronald, 2008). Subsequent evidence supports an impairment in the development of
‘theory of mind’ in children with ASD (Brent et al., 2004), although whether such cognitive deficits are unique to individuals with ASD remains unclear (Happé & Ronald, 2008).

Specific impairments in areas of neuropsychological functioning are also associated with ASD, notably weak central coherence (Happé & Frith, 2006) and poor executive functioning (Ozonoff, 1995). Central coherence refers to the ability of typically developing individuals to understand the overall context of presented information without attending fully to the details of the information (i.e. “to give priority to the understanding of meaning” (Frith, 2003, p160). Individuals with ASD have been shown to tend to focus on details when processing information, resulting in weak central coherence and higher abilities in detail-focussed processing tasks (Happé & Frith, 2006). Executive functioning is the ability to purposefully regulate cognitive functions and behaviours to enable an individual to problem solve (Rabbitt, 2005). Individuals with ASD have generalised executive functioning impairments (Ozonoff, 1995; Geurts et al., 2004) which have been associated with the presentation of restricted and repetitive behaviours in ASD (Happé & Ronald, 2008).

ASD is diagnosed on the basis of behaviour using variety of screening measures and diagnostic instruments which assess these cognitive differences (Charman & Gotham, 2013). Diagnosis is traditionally acquired on the basis of the presence of impairment in each of Wing and Gould’s (1979) ‘triad’ areas (ICD-10, WHO, 1992; DSM-IV, APA, 1994), which must be evident from early childhood. Asperger’s syndrome is classified as a subtype of ASD (ICD-10, WHO, 1992; DSM-IV, APA, 1994) that is typically characterised by individuals with milder and/or fewer impairments in the triad areas.
and normal levels of intellectual ability (Ozonoff, Rogers & Pennington, 1991; Miller & Ozonoff, 2000).

However, the diagnostic framework for ASD has changed dramatically in the revised edition of the American Psychiatric Association’s Diagnostic and Statistical Manual (DSM), published in May 2013 (DSM-V, APA, 2013). This has altered the diagnostic framework of ASD from a ‘triad’ to a ‘dyad’ encompassing two domains of impairment in ‘social communication and interaction’ and ‘restricted, repetitive patterns of behaviour, interests or activities’ (DSM-V, APA, 2013). This is based on recent evidence that difficulties in the triad domains of social development and language may be caused by similar deficits in functioning (Geschwind, 2011; Lord & Jones, 2012). Sensory behaviours are also included in the diagnostic criteria for the first time in the second domain (DSM-V, APA, 2013). ‘Aspergers Syndrome’ has also been dropped with a more general ASD diagnosis applied to all (DSM-V, APA, 2013). The aim of the updated criteria in DSM-V (APA, 2013) is to make the ASD diagnosis simpler and more consistent for individuals, families and professionals as well as introducing a measure of the severity of an individual’s difficulties to inform decisions about the level of support they require (National Autistic Society website 2013). Although the main diagnostic framework in use in the UK is the World Health Organisation’s International Classification of Diseases (ICD), which contains more traditional ASD diagnostic criteria at present, the impact of the new DSM-V (APA, 2013) has been significant. Concerns about the impact of the new diagnostic criteria for ASD have been raised by individuals (e.g. Elder Robinson, 2012) and organisations (e.g. the National Autistic Society, 2010). A more indepth description of the nosological features of ASD can be found in the current DSM and ICD classification entries in Appendix 1.
Although widely estimated to be evident in around 1% of the population (Wing, 1996; Baird et al., 2006), the reported prevalence of ASD varies dramatically (Wing, 1996) with smaller studies tending to obtain higher rates (Fombonne, Quirke & Hagen, 2009). ASD may be much more common than this; for example Kim et al. (2011) observed an ASD prevalence rate of 2.64% in a South Korean population-based sample study and many of the affected children were previously undiagnosed. The prevalence of Asperger Syndrome is estimated to be 0.092% of the general population (Gillberg, Cederlund, Lamberg & Zeijlon, 2006).

Initially, ASD was viewed as an emotional response to indifferent parenting styles (Kanner, 1943; Harlow & McKinney, 1971) but contemporary research discredits this theory and points to a multi-aetiological explanation with a biological basis (Wing, 1981; Happé & Frith, 1996). Neurological abnormalities have been identified in studies comparing groups of individuals with ASD and controls, including structural differences in the frontal lobes (Carper & Courchesne, 2000) and cerebellum (Piven, Saliba, Bailey & Arndt, 1997). Atypical brain pathways (Rubenstein & Merzenich, 2003; Dhossche, 2004; Geschwind, 2011) and neuropsychological functioning have also been associated with ASD (Ozonoff, Strayer, McMahon & Filloux, 1994; Frith, 2003; Hare, Mellor & Azmi, 2007). There is much evidence for genetic factors underlying the aetiology of ASD (Rutter, 2000) with genetic causation determined for up to a fifth of the current ASD population (Geschwind, 2011) and susceptibility genes being identified on chromosome 15 (Dhossche, 2004) but a universal genetic marker remains to be identified and the neurobiology of ASD remains “diverse and non-specific” (Lord & Jones, 2012, p502). Indeed, many of the recently identified biological abnormalities
are also evident in other disorders (Geschwind, 2011), which indicates that the complex neurobiology of autism is far from being unravelled.

An emerging idea is that a single explanation at a biological level will not be found as ASD is actually an umbrella term for a number of independent clinical features which commonly co-occur rather than a coherent syndrome (Happé, Ronald & Plomin, 2006; Happé & Ronald, 2008; Williams & Bowler, 2014). The idea that it may be possible to fraction the triad of impairments associated with ASD into separable areas of deficit has the potential to shift aetiological understanding and allow the development of targeted clinical intervention for each dissociable triad area (Williams & Bowler, 2014). As part of this theory, it has been suggested, as previously mentioned, that poor executive functioning results in the repetitive and restricted behaviours of ASD and impaired theory of mind is the cause of the social and communication deficits of ASD (Happé & Ronald, 2008).

Kanner recognised that ASD is around four times more common in males than females, and this has been born out in subsequent research (Baird et al., 2006; Fombonne et al., 2009). Girls who have ASD tend to have higher levels of intellectual impairment (Lord, Schopler & Reviki, 1982; Volkmar, Szatmari & Sparrow, 1993) but this may be due to biases caused by poorer identification and diagnostic rates (Gould & Ashton-Smith, 2011; Mandy et al., 2012). Manifestation of ASD in girls seems to subtly differ from boys (Lord et al., 1982; Hartley & Sikora, 2009; Kopp & Gillberg, 2011; Mandy et al., 2012; Carpenter, 2012) which provides some evidence for differing phenotypes between the sexes. Others suggest that in biological terms, girls are more resistant to
ASD so require a greater number of inherited risk factors for ASD to manifest (Baron Cohen, 2002; Robinson, Lichtenstein, Anckarsäter, Happé & Ronald, 2013).

The reported prevalence of ASD is apparently rising (Gillberg & Wing, 1999; Baird et al., 2006; Fombonne et al., 2009), for as yet undetermined reasons (Insel, 2012), but due in part to some older individuals obtaining later diagnosis due to revised criteria (Gillberg & Wing, 1999) (e.g. prior to DSM-III (APA, 1980), ASD was not classified as a distinct clinical entity but as a subtype of ‘schizophrenic reaction, childhood type label’) and debate about whether the rising incidence of ASD due to increased detection or actual increased prevalence is ongoing (Lord, 2011; Insel, 2012).

There are many characteristics of ASD that are not included in current diagnostic criteria but which are common features of individuals with ASD. Associated features include sensory processing difficulties (Kern et al., 2006), motor abnormalities (Gowen & Hamilton, 2012) and a preference for restraint (Grandin, 1992). There are various co-morbidities associated with ASD, including psychosis (Gillberg & Steffenburg 1987; Billstedt et al., 2005), Attention Deficit Hyperactive Disorder (ADHD), Obsessive Compulsive Disorder (OCD) (Leyfer et al., 2006; Taurines et al., 2012) and intellectual disability (Gillberg & Billstedt, 2000). Other medical problems such as gastrointestinal problems (Schieve et al. 2012), eczema (Whiteley, Rodgers & Shattock, 1998), seizures (Gillberg & Steffenburg, 1987; Kohane et al., 2012), and motor problems (Gowen & Hamilton, 2013) occur at higher than expected rates in the ASD population. It can be difficult to diagnose co-morbid illnesses within ASD populations due to the intrinsic difficulties associated with the triad of impairments, for example when required to complete assessments reliant on intact verbal abilities, but these medical co-
Morbidities may be the cause of the significantly increased mortality rates associated with ASD (Bilder et al., 2013). Such common co-occurrences, particularly those involving motor and attention deficits, may be a result of underlying biological neuropathology (Gillberg & Billstedt, 2000) but a causal relationship remains to be established.

Although a life-long condition, the presentation of ASD changes over time (Rutter, 1970). In general, the long term outcomes for individuals with ASD remains mixed with some developing more complex presentations, particularly arising medical comorbidities (Gillberg & Billstedt, 2000) and increased levels of challenging behaviours (Billstedt et al., 2005), and others experiencing a decreased severity of symptoms associated with ASD as age increases (Périsse et al., 2010). There is evidence that individuals with ASD have more positive outcomes if they have milder cognitive impairments (Kobayashi, Murata & Yoshinaga, 1992; Périsse et al., 2010) and increased motor ability in childhood (Sutera et al., 2007).

1.2.4 Conceptualising Autistic Catatonia:
Catatonic-like symptoms such as repetitive and restricted behaviours, mutism and echolalia are also core features of ASD and many are included in the new dyadic model of ASD (Wing & Shah, 2006; Fink, Taylor & Ghaziuddin, 2006; Takota & Takata, 2007; Gowen & Hamilton, 2012; DSM-V, APA, 2013) and the presentation of one or two marked catatonic symptoms in individuals with ASD is common (Wing & Shah, 2006). Motor abnormalities in individuals with ASD are poorly understood, but it has been suggested to be related to impairments in processing and integrating sensory information which make it difficult to effectively plan motor action (Gowen &
Recent research indicates that some individuals with autism experience a change in presentation of altered motor movement, which in severe cases is characterised by gradual yet marked slowness of movement, increased passivity, freezing during motor movement and difficulty initiating actions (Wing & Shah, 2000; Hare & Malone, 2004; Wing, 2005). Such catatonic-like deterioration of key functional skills, termed ‘autistic catatonia’, has several consequences for quality of life and significant implications for the level of care individuals require with a higher proportion of those with autistic catatonia living in a residential placement (Wing & Shah, 2000).

Those individuals affected are usually adolescents with ASD who find it increasingly more difficult to maintain their existing independence (particularly personal care, expressive communication and engagement in activities) due to the onset of a range of unusual and chronic motor symptoms. Descriptive accounts depict individuals with hindered movement either in fluidity or quantity; remaining immobile for long periods of time, finding it difficult to start moving, performing movement at a very slow pace or getting ‘stuck’ part way through a gross motor action (Wing & Shah, 2000; Hare & Malone, 2004; Wing, 2005). Physical and verbal prompts are often reported to be required to complete motor actions successfully (Hare & Malone, 2004; Wing, 2005) and physical health is often a concern, with individuals eating and exercising significantly less.

There is variation in the presentation (Wing & Shah, 2000; Billstead et al., 2005) and severity (Dhossche, Shah & Wing, 2006; Neumarker, 2006) of autistic catatonia, with a core group of symptoms manifesting more frequently in individuals with autistic
catatonia, including increased slowness in completing movements and difficulty completing motor actions (Wing & Shah 2000). Autistic catatonia is often a chronic syndrome (Ohta, Kano & Nagai, 2006; Dhossche et al., 2006) but some individuals experience repeated shorter bursts of symptoms in possibly cyclical presentations (Realmuto & August, 1991; Ohta et al., 2006).

A commonly accepted diagnostic definition for autistic catatonia has not been established (Dhossche et al., 2006) and although there are numerous case descriptions of catatonia in the literature (e.g. Realmuto & August, 1991; Dhossche, 1998; Hare & Malone, 2004; Ghaziuddin, Quinlan & Ghaziuddin, 2005; Ohta et al., 2006; Schieveld, 2006; Takota & Takata, 2007; Kakooza-Mwesige et al., 2008), the number and severity of presenting symptoms reported varies and few individuals display all of the possible commonly associated symptoms (Wing & Shah, 2000). Some symptoms are commonly reported (e.g. reduced communication, slow motor movements, resistance to prompting, reduced eating) and others reported only once (e.g. visual hallucinations, diaphoresis, spontaneous crying) – see Table 1 in Appendix 3. Some authors suggest that an individual’s symptoms can vary during waking hours (Ohta et al., 2006). Currently, it is very unclear how to differentiate between autistic catatonia and other co-morbid conditions and some symptoms attributed to autistic catatonia may have another cause, potentially even ASD itself.

In their quantitative study of the prevalence of catatonic symptoms in 506 referrals to a specialist ASD clinic, Wing and Shah (2000) propose diagnostic guidelines for autistic catatonia, including a list of four ‘essential features’ (‘increased slowness affecting movement and verbal responses’, ‘difficulty in initiating and completing actions’, ‘increased reliance on physical or verbal prompts’ and ‘increased passivity and
apparent lack of motivation’) along with four frequently observed behavioural abnormalities (‘reversal of day and night’, ‘Parkinsonian features’, ‘excitement and agitation’ and ‘increase in repetitive, ritualistic behaviour’) (Wing & Shah, 2000, p357). This study represents the only large sample empirical analysis of catatonic symptoms in the population to date and provided firm evidence to support theories of a catatonic-like deterioration in a proportion of young people with ASD. However, it is important to note the potential biasing of their sample due to the tertiary nature of referrals to the ASD clinic and the lack of a suitable clinical measure available to researchers to assess catatonic symptoms in the population, which could have impacted the validity of the study findings. Despite these inherent limitations, Wing & Shah’s (2000) research has been key to conceptualising autistic catatonia and has provided a foundation for the current understanding of the condition. Subsequent researchers (Takota & Takata, 2007; Dhossche et al., 2009) have used Wing and Shah’s (2000) clinical definition as the basis of investigations into autistic catatonia, although some have noted its “idiosyncratic and broad” nature (Hare & Malone, 2004, p188) and there is little guidance regarding how long symptoms must be evident for diagnosis to be warranted. Individual researchers have subsequently adapted the criteria quite extensively by adding and removing specific diagnostic items, whilst others have used the DSM-IV (APA, 1994) definition of catatonia to diagnose the presence of catatonia in individuals with autism (Dhossche & Bouman, 1997; Zaw, Bates, Murali & Bentham, 1999; Ghaziuddin et al., 2005; Schieveld, 2006; Bozkurt & Mukaddes, 2010). Other researchers have proposed their own diagnostic criteria for autistic catatonia (Hare & Malone, 2004; Ohta et al., 2006; Fink, Taylor & Ghaziuddin, 2006). Although these studies provide a very useful starting point, a systematic
examination of the symptoms associated with catatonia in autistic populations is needed to enable a firm clinical definition to be delineated.

The prevalence of autistic catatonia is largely unknown, partly due to the lack of commonly accepted diagnostic criteria (Schieveld, 2006). Marked or moderate catatonic-like behaviour has been found to be a common feature in individuals with ASD (Wing & Shah, 2006) and many more individuals with ASD display catatonic symptoms in isolation (Wing & Shah, 2000). However, only a small proportion of individuals with ASD experience the catatonic-like deterioration associated with the concept of autistic catatonia (Wing & Shah, 2000). In their robust, large sample quantitative study, Wing & Shah’s (2000) found 6% (n=30) of their sample of 506 met their broad criteria for autistic catatonia, but a further 8 displayed milder catatonic-like symptoms which were judged to be below their defined threshold for diagnosis. All those who met Wing and Shah’s criteria for a diagnosis of autistic catatonia were above 15 years old; taking this into account increased the prevalence of autistic catatonia to 17% of referrals of age 15 years and over (Wing & Shah 2000). In their systematic population-based follow up study of ASD, Billstedt et al. (2005) found 8 of 73 adults with autistic disorder (11%) and 5 of 35 individuals with atypical autism (14%) had an existing clinical diagnosis of catatonia. Four additional individuals also met Wing and Shah’s (2000) criteria for autistic catatonia. Smaller studies have also picked up the presence of catatonia in individuals with ASD; Perisse et al. (2010) found 2 of 29 adults with ASD in an inpatient hospital population (7%) exhibited severe catatonia, and Nordin and Gillberg (1998) found a minimum of 3 of 46 individuals (7%) exhibited moderate to severe catatonia. Current estimates of the prevalence of autistic catatonia are considered to be underestimates (Dhossche, 2004; Wing, 2005), due to
milder symptoms of catatonia in ASD being overlooked (Stoppelbein, Greening & Kakooza, 2006). There is evidence that up to 22% of individuals with ASD experience some decline in functioning during adolescence (Gillberg & Steffenburg, 1987; Kobayashi et al., 1992; Nordin & Gillberg, 1998) so the prevalence of autistic catatonia may account for a larger proportion of this sub-group than currently estimated. Many now suggest that good clinical practice would dictate that all individuals with ASD who experience a decline in functioning are assessed for autistic catatonia (Ghaziuddin et al., 2005; Dhossche et al., 2006; Dhossche, Reti & Watchtel, 2009; Bozkurt & Mukaddes, 2010).

It is noteworthy that there are similarities in the identified brain abnormalities associated with catatonia and autistic catatonia (Takota & Takata, 2003; Fink et al., 2006; Dhossche & Rout, 2006), although this does not necessarily indicate they are a single neurobiological syndrome (Dhossche et al., 2006). The differences in age at presentation, manifestation of symptoms and prognosis between ASD and autistic catatonia does not rule out a common aetiology (Dhossche 2004). There is symptomatic overlap reported between ASD and catatonia, such as repetitive and restricted behaviours, but some common catatonic symptoms are unusual in ASD, for example auditory or visual hallucinations (Kakooza-Mwesige et al., 2008). Interestingly, a number of the individuals in Wing and Shah’s (2000) study developed OCD symptoms before the manifestation of autistic catatonia, which is an indicator of potential common neuropathology between these two conditions.

Opinion is divided as to whether autistic catatonia is a co-morbid condition of ASD or is part of ASD itself. Some researchers consider autistic catatonia to be a condition that develops in individuals with ASD in much the same way in individuals without ASD
(Gillberg, 1985; Kakooza-Mwesige et al., 2008; Dhossche et al., 2009; Wachtel, Griffin, Dhossche & Reti, 2010) so view autistic catatonia as a co-morbidity. Others view autistic catatonia as a presentation of co-morbid depression in individuals with ASD (Zaw et al., 1999; Takota & Takata, 2007). Ohta and colleagues (2006) postulate that although autistic catatonia may develop in some as a direct result of their ASD, it may then exist as an entirely separate co-morbid syndrome. Alternatively, other writers consider ASD and catatonia as more intrinsically linked (Realmuto & August, 1991), for example that ASD is an early expression of catatonia (Dhossche, 2004) or that the catatonic symptoms presenting in ASD are a late occurring expression of ASD itself (Hare & Malone, 2004). Whether there is a sub-group of individuals who are prone to developing autistic catatonia due to the abnormal neurocognitive, perceptual and sensory systems which cause their ASD (Hare & Malone, 2004) or whether some individuals with ASD develop catatonia independently, and indeed how to identify these populations, has not been determined.

Some researchers have attempted to unpick the nosological features of autistic catatonia with limited success (e.g. Neumarker, 2006). Hare and Malone (2004) completed a comprehensive neuropsychological assessment of their case study ‘A’ and noted that his significant movement problems appeared to be a result of dysfunction in the initiation of motor movement rather than a difficulty in associated cognitive planning. Thus, it is suggested that autistic catatonia may be an extreme impairment in central coherence and so can be conceptualised as part of the phenotype of ASD. The debate about where the line is between ASD and catatonia in affected individuals is unlikely to be resolved until the diagnostic features of the condition are determined and further empirical investigative research completed (Schieveld, 2006).
Autistic catatonia is thought to often occur in adolescence (Wing & Shah, 2000; Ohta et al., 2006) with factors including stress and depression precipitating onset (Ghaziuddin et al., 2005; Shah & Wing, 2006; Stoppelbein et al., 2006). Onset is usually gradual with reports that some individuals experience more variation in symptom severity, for example those who are classified in the ‘passive’ social interaction subgroup and those with greater expressive language impairment seem to be at greater risk of developing autistic catatonia whilst those with higher IQ scores exhibit less marked catatonic symptoms (Wing & Shah, 2000). However, there are also case reports of individuals with expressive verbal communication skills developing autistic catatonia (Hare & Malone, 2004; Ghaziuddin et al., 2005; Bozkurt & Mukaddes, 2010) and Wing and Shah’s systematic study found almost half of those who met the criteria for autistic catatonia were in the Asperger’s diagnostic subgroup (n=14, 47%), although this was a non-significant variable in statistical analysis, and there was no correlation with level of intellectual disability. It is important to note that the number of catatonic symptoms presenting and the degree of symptom severity were not found to be correlated with language ability or social interaction subgroup (Wing & Shah, 2000). There are no studies which consider the effect on gender on the development of autistic catatonia, although Wing and Shah found a small but non-significant excess of males with autistic catatonia (Wing & Shah, 2000).

There is some evidence of early indicators of autistic catatonia that present in childhood; specifically hand flapping, holding and manipulating small objects and spinning on the spot (Thorndyke & Hare, 2008) and regression in early development (Dhossche & Rout, 2006). Similarly, dysfunction of the taste, tactile and olfactory sensory systems was also found to be associated with these potential ‘catatonic
markers’ in a small ASD population (Thorndyke & Hare, 2008) which may provide some tentative support to Hare and Malone’s (2004) proposal that autistic catatonia is part of the ASD phenotype. Additionally, individuals younger than 15 years old have been found to display some catatonic symptoms in childhood, namely brief episodes of freezing and difficulty crossing thresholds (Wing & Shah, 2000); which could potentially be additional prodromal symptoms of autistic catatonia.

1.2.5 Identifying and treating Autistic Catatonia:

Although accurate measurement of presenting symptoms is crucial for evidence-based intervention and research, there are limited options for clinical assessment tools that can be used within learning disability populations. Many of the existing measures remain largely un-validated and there are practical issues associated with importing catatonia rating scales to learning disability populations (Carroll et al., 2008; Heckers et al., 2010) including over-reliance on affective alterations which are intrinsically difficult to accurately identify via third party measures, altered speech or tone of voice (which may be abnormal or absent in individuals with learning disability) and non-typical movement in more general populations being evident pre-catatonia in individuals with ASD (e.g. repetitive stereotyped body movements).

Treatment options for autistic catatonia remain elusive (Ohta et al., 2006) and current research provides little guidance to clinicians (Dhossche et al., 2006). Autistic catatonia is a chronic condition and intervention is largely ineffective (Ohta et al., 2006). To date, no clinical trials have been completed and no evidence-based practice guidelines have been developed (Stoppelbein et al., 2006; Shah & Wing 2006). Some claim that autistic catatonia “responds to the same treatments that are effective in
other forms of catatonia” (Wachtel et al., 2010, p. 356), but this is not supported by empirical evidence. The evidence base consists mainly of single case reports of individual interventions (e.g. Dhossche, 1998; Hare & Malone, 2004; Ghaziuddin et al., 2005; Wachtel et al., 2010), and common treatment options include behavioural interventions (Hare & Malone, 2004; Wing, 2005; Shah & Wing, 2006), pharmacological prescription (Relmuto & August, 1991; Dhossche, 1998; Ohta et al., 2006) and ECT (Dhossche et al., 2006; Dhossche, Reti & Watchtel, 2009; Wachtel, Hermida & Dhossche, 2010). The impact of existing medications as a cause of catatonic symptoms must be ruled out before any intervention is attempted (Dhossche et al., 2006). Prompt identification and treatment are thought to be key to positive outcome (Dhossche et al., 2006).

Behavioural and environmental interventions have been described in detail in the literature (Hare & Malone 2004; Wing 2005; Dhossche et al., 2006) and include reducing stress (both resulting from external factors by implementing consistency and routine (Dhossche et al., 2006; Shah & Wing, 2006) and internal factors (e.g. biological or psychological) (Dhossche et al., 2006; Shah & Wing, 2006)), managing environmental factors such as minimising the presence of others during the presentation of marked catatonic symptoms (Hare & Malone, 2004), increasing caregiver’s knowledge and understanding of the condition (Dhossche et al., 2006) and implementing effective physical and verbal prompting (Hare & Malone, 2004; Dhossche et al., 2006). There is evidence that such approaches are effective in reducing (Hare & Malone, 2004) and reversing (Shah & Wing, 2006) autistic catatonia symptoms. These approaches are attractive due to the low risk associated with
implementing behavioural and environmental interventions for individuals and should be viewed as the basis of all treatment of autistic catatonia (Dhossche et al., 2006).

Pharmacological interventions mainly consist of the prescription of anti-psychotic medications (Realmuto & August, 1991; Ohta et al., 2006) or benzodiazepines such as Lorazepam (Dhossche, 1998; Dhossche et al., 2006; Ohta et al., 2006). There are reports of medication as a successful independent intervention for autistic catatonia (Realmuto & August, 1991; Takota & Takata, 2007; Bozkurt & Mukaddes, 2010) but questions remain about accurate diagnosis for these individuals, with suggestions that the medication may have relieved symptoms of depression rather than autistic catatonia (Stoppelbein et al., 2006). The commonly reported doses of pharmacological interventions prescribed to treat autistic catatonia are much higher than the maximum doses set by medicinal regulators in the UK. For example, the Medical Healthcare Regulations Agency recommend a safe maximum daily dose of 4 mg BD of Lorazepam per day for the short-term treatment (up to 4 weeks only) of severe and disabling anxiety (MHRA, 2007), whereas Dhossche et al. (2006) suggest daily doses of between 6 and 24 mg for a treatment phase lasting 6 to 12 months for autistic catatonia. Notably, there are serious side effects which are associated with Lorazepam and there have been 30 reports of fatal adverse drug reactions associated with Lorazepam since 1963 (MHRA, 2013).

ECT is stated to be an effective intervention for autistic catatonia (Dhossche et al., 2009; Wachtel et al., 2010) and it is often used to treat more severely affected individuals (Stoppelbein et al., 2006) after other treatment options have proved ineffective (Dhossche et al., 2006; Fink et al., 2006; Dhossche, 2009). However, the
function of ECT in the treatment of autistic catatonia remains unclear (Fink et al., 2006; Shah & Wing, 2006). Clinicians report that the required treatment courses of ECT are typically intensive and lengthy to maintain the treatment effects and prevent relapse (Kakooza-Mwesige et al., 2008; Dhossche, 2009; Wachtel et al., 2010). There are descriptive case reports describing individuals returning to their pre-autistic catatonia presentation after ECT treatment (Dhossche et al., 2006; Ghaziuddin et al., 2010; Weiss et al., 2012). Again, the recommended treatment courses for autistic catatonia using ECT are much higher than the maximum recommended by regulatory guidelines in the UK. The National Institute for Clinical Excellence (NICE) do not recommend that ECT is used to treat autistic catatonia and only permit use of ECT for short-term improvement of life-threatening symptoms in other patient populations once all other interventions have not been effective (NICE, 2009). Only courses of up to 12 sessions are permitted with any patient (NICE, 2009) whereas treatment algorithms suggested for autistic catatonia recommend continued ECT treatment courses of undetermined length (Dhossche et al., 2006; Wachtel et al., 2010).

As with the treatment of catatonia, the usual recommended treatment plan is for lower risk interventions to be attempted first, followed by higher risk options if there is no positive response identified (Dhossche et al., 2006). It is possible to classify individuals with autistic catatonia into mild, moderate and severe based on the impact of their symptoms on their ability to perform daily tasks independently (Wing & Shah, 2000) and it has been proposed to use these delineations to guide different treatment options in line with the medically standard expected risk to benefit ratio (Dhossche et al., 2006). However, without a valid and reliable clinical measure of autistic catatonia, it is very difficult to accurately determine the impact of any intervention. It is of note
that two of the leading specialists in autistic catatonia state that “they have never seen, in chronic catatonia-like deterioration in ASDs, the dramatic recoveries reported following the administration of lorazepam and/or ECT” (Shah & Wing, 2006, p251), and it seems reasonable to conclude that effective and safe treatments for autistic catatonia have yet to be established (Ohta et al., 2006).

1.3 Conclusion:

A catatonic presentation manifesting in adolescence is reported in around 8% of individuals with ASD, with core features of being very still for long periods of time, getting ‘stuck’ when trying to complete actions, difficulty stopping actions once they have been started, difficulty initiating actions, moving very slowly and taking a long time to finish actions or requiring prompts to complete actions. To date, little is known about the aetiology, phenomenology and course of such ‘autistic catatonia’, due to a lack of quantitative research in the literature base. Current knowledge consists mainly of practice-based qualitative literature, namely case examples and discussion papers, and no systematic literature reviews, randomised control trials or randomised sampling studies have been completed to date. Although the current literature base provides rich information and debate about the topic area, purposive sampling combined with a lack of empirical data has impeded the current understanding of autistic catatonia. Although there are numerous descriptions of individuals presenting with autistic catatonia in the research, the number and variation of symptoms varies widely with few individuals presenting with all of the reported symptoms of autistic catatonia. Different studies have also defined autistic catatonia very differently, which has resulted in inconsistencies in the research literature. The over-reliance on practice-based single case literature in the field of autistic catatonia
research has resulted in a weak evidence-base which must be corrected by empirical investigation into the prevalence of catatonic symptoms in the ASD population.

The lack of quantitative research and the ambiguity around a firm clinical definition has led to a longstanding lack of clarity for autistic catatonia, which has resulted in a reliance on individual clinician’s knowledge and experience for accurate assessment and diagnosis. It has also impacted on the development of targeted evidence-based treatments and impeded research.

This study aims to empirically investigate the phenomenon of ‘autistic catatonia’ in children and adolescents with ASD. Specifically, this study will:

1. Develop a valid and reliable clinical measure which can be used for assessment, research and to monitor the progression of the condition and the effectiveness of interventions.
2. Investigate how common catatonic symptoms are in young people with ASD.
3. Aim to develop better theoretical understanding of autistic catatonia.
CHAPTER 2: METHODOLOGY

2.0 Aim: This chapter describes the methodology of the study; specifically detailed accounts of clinical measures designed and used, research procedure and details about recruitment and participation.

2.1 Study design:
This study aims to investigate the prevalence of catatonic symptoms in children and adolescents with ASD in a cross sectional survey design. A 34-item Autistic Catatonia Questionnaire (ACQ) was developed from extant reports of autistic catatonia and comprises of six core features and an additional 28 items covering other aspects of repetitive, motoric and sensory functioning. Items on the ACQ are scored for presence, frequency and severity. A main caregiver or parent provided information about the presentation of a young person they care for via the online completion of the ACQ. Two additional measures were completed for validity reasons; the Repetitive Behaviour Questionnaire (Moss, Oliver, Arron, Burbidge & Berg, 2009) and the Carer Supplement to the Glasgow Depression Scale for people with Learning Disability (GDS-CS) (Cuthill, Espie & Cooper, 2003). Participants from the target population (i.e. parents and carers of individuals with ASD) were recruited via advertisement through relevant charities and support organisations in the United Kingdom.

Although this research is anonymous, general demographic and contextual information was collected in the study; specifically whether the participant is a parent or a carer (and if the latter, how long they have supported them), the age and gender
of the young person with ASD, whether the individual being reported on has an existing diagnosis of autistic catatonia and details of any other current diagnoses.

2.2 Ethics:

2.2.1 Ethical approval:

This study was ethically reviewed and approved by the University of Manchester Committee on the Ethics of Research on Human Beings (Research Committee 3) in November 2012 (ref: AJ/ethics/2410/03) for a period of 5 years. See Appendix 2 for a copy of the research study approval letter.

2.2.2 Ethical issues:

This research project was designed so that all collected data is anonymous. The researcher processed and analysed the data with no knowledge of each participant’s identity or personal details. Therefore, the design of this study ensures the total privacy of participants. However, it is important that some contextual and demographic information was collected, namely the age, sex and diagnosis of the individuals the participant is providing information about. This data is crucial for the validity of the study and does not provide enough detail about the individuals to enable their identification.

Participants were not put at risk of harm as a result of involvement in the study. Participation was voluntary and no incentives were offered for taking part. Although this research study involved the collection of information about vulnerable individuals, participants were adults in the general population who had official care responsibilities.
for vulnerable young people and so concern about the mental capacity of participants
to consent in taking part in the study is minimal.

Due to concerns that parents or carers may find reporting symptoms which someone
they care for experiences distressing or worrying, contact details of both the
researchers and the National Autistic Society helpline were clearly presented to
participants on completion of the online questionnaire. However, this was thought to
be unlikely given the nature of the questions and the relative commonality of reporting
abnormalities in movement for parents/carers of children with autism.

2.3 Description and administration of measures:

2.3.1 Assessment procedure:

Participation involved completing an anonymous online questionnaire using ‘Select
Survey’ software, a commonly used format for online questionnaires. A URL link to the
online study was included in the information advertised by organisations and sent on
by them to potential participants. Prior to the online questionnaire, clear and simple
information about the study was displayed to each participant which included a
statement that they are free to withdraw from participation in the study at any time
without giving a reason or consequence. To ensure that participants read this
information correctly, a number of questions were asked relating to information
provided and ensure each participant met the inclusion criteria of the study (see
Appendix 10). After completion of the ACQ, a debrief sheet was presented online to
each participant which provided information relating to requests for copies of the
research findings, contact details of the researcher and information about appropriate
help lines which can offer support in case participants experienced any emotional
distress or worry relating to taking part (see Appendix 11).

The study was completed at a time and place which is convenient to each participant.
The time that participants spent completing the online questionnaire varied from 5
minutes to 79 minutes, with the mean completion time for participation in this study
being 19.7 minutes.

2.3.2 Description of measures:
Three measures were completed by the participants and presented in the order as
detailed below:

2.3.2.1 Autism Catatonia Questionnaire (ACQ):
This 34 item, third party report clinical measure was designed as part of the current
research study and is described in detail below. The ACQ aims to assess the
prevalence and frequency of symptoms thought to be associated with autistic
catatonia in individuals with ASD. No training is required to complete the ACQ and
instructions for completion are included on the measure. The effectiveness of the ACQ
as a clinical measure will be assessed as part of this research.

The ACQ was developed by compiling commonly reported symptoms of autistic
catatonia in the literature into a screening tool similar to existing catatonia screening
tools (e.g. Bush-Francis Catatonia Rating Scale (Bush et al., 1996a), Northoff Catatonia
Rating Scale (Northoff et al., 1999)). A literature search was completed using
individual online healthcare databases which are relevant to the subject; ‘PsycInfo’ and
‘Web of Knowledge’. Each database was systematically searched using a number of key words identified as relevant; *autism, Autistic Spectrum Disorder, ASD, catatonia*, the wildcard *autis* and the exact phrase “*autistic catatonia*” along with appropriate AND operators (e.g. autism AND catatonia) including both the title and the abstract in the search. No US terminology or spelling differences were identified for alternative key words. Key words identified by existing literature was also studied for additional key words but no additional search terms were identified. No search limits were assigned to search terms nor any exclusion criteria to the literature search due to the existing poor knowledge base. Automated email alerts were set up on the databases so that the author would be notified about newly published literature. To ensure the literature search was not subjected to a publishing bias, efforts were made to locate relevant undergraduate theses and poster presentations. The references used by the journal articles were hand searched and additional literature identified. Author searches were completed for individuals who were found to have published numerous relevant journals; Dr. Dougal Julian Hare, Prof. Dirk Dhossche, Dr. Lorna Wing and Dr. Amitta Shah.

Once the literature search was complete, the number of reported symptoms connected to autistic catatonia in the research literature were tallied (see Table 1 in Appendix 3). Any symptom which was reported on only one occasion was excluded from the measure (e.g. finger tapping, diaphoresis). Any symptom which is a key feature of ASD and which did not indicate change to the individual’s previous presentation were also excluded (e.g. echolalia). As the ACQ is a third party measure, any symptom thought to be too vague or speculative (e.g. auditory hallucinations, anxiety, visual hallucinations) were also excluded from the measure. Any item which
exists on one of the other measures administered in this research study was also excluded. Altogether, eight reported symptoms were excluded from the measure (see Table 1 in Appendix 3). Existing measures of catatonia were studied and their structure replicated as appropriate. Using these guidelines, the ACQ consisted of 34 specific items which can be categorised into motor symptoms (n=15), affective alterations (n=5) and behavioural alterations (n=14), in a similar way to the Northoff Catatonia Scale (Northoff et al. 1999). Each symptom is clearly defined and many are accompanied with examples and descriptions, in line with concerns about varying definitions of symptoms across catatonia rating scales (Carrol et al. 2008). Table 2 below lists the items included in the ACQ, the order of presentation in the measure (‘ACQ question number’) the category assigned by the author (‘motor symptoms’, ‘affective alterations’ or ‘behavioural alterations’) and the associated examples and descriptors of symptoms provided to participants.

Table 2: Illustration of each symptom connected to autistic catatonia included in the ACQ measure, the associated question in the ACQ and the example or descriptor provided to participants:

<table>
<thead>
<tr>
<th>ACQ question number</th>
<th>Symptom:</th>
<th>Associated question in ACQ:</th>
<th>Example or descriptor given:</th>
<th>Supplementary questions re frequency &amp; severity of symptom?</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>'Freezing'/very still like a statue</td>
<td>Are there times when he/she is very still for long periods of time, almost like a statue?</td>
<td>Yes</td>
<td></td>
</tr>
<tr>
<td>2</td>
<td>Difficulty initiating actions/'stuckness’/akinesia</td>
<td>Does he/she seem to get ‘stuck’ when trying to do something?</td>
<td>(e.g. stopping mid-air halfway through reaching for something &amp; looking like they are trying to move but</td>
<td>Yes</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>---</td>
<td>---</td>
<td>---</td>
<td></td>
<td></td>
</tr>
<tr>
<td>3</td>
<td>Problems stopping actions once started</td>
<td>Does he/she seem to find it difficult to stop doing actions once they have started them?</td>
<td>(e.g. repeatedly putting on a coat &amp; taking it off again &amp; again for a long period of time)</td>
<td>Yes</td>
</tr>
<tr>
<td>4</td>
<td>Difficulty initiating movement</td>
<td>Does he/she seem to find it difficult to start moving?</td>
<td>(e.g. lying still and looking like he/she wants to get up or reach for something but can’t)</td>
<td>Yes</td>
</tr>
<tr>
<td>5</td>
<td>Slowness in movement</td>
<td>Does he/she move very slowly and takes a long time to finish actions?</td>
<td>(e.g. moving very slowly when doing things like picking up a cup to drink or eating dinner)</td>
<td>Yes</td>
</tr>
<tr>
<td>6</td>
<td>Requires prompts to complete actions</td>
<td>Are there times when he/she needs physical OR verbal prompts to complete actions?</td>
<td>(e.g. needing someone to tell them or touch their arm to enable them to lift a cup to their mouth to drink)</td>
<td>Yes</td>
</tr>
<tr>
<td>7</td>
<td>Waxy flexibility</td>
<td>Are there times when if you moved part of his/her body, they let you without taking much notice of what you are doing &amp; then stay in that position afterwards?</td>
<td>(e.g. would they offer no resistance to you curling their fingers into a fist &amp; then keep their hand curled up when you moved away)</td>
<td>No</td>
</tr>
<tr>
<td>8</td>
<td>Repetitive body movements</td>
<td>Does he/she like to move their body in repetitive ways?</td>
<td>(This includes any frequent body movement such as body rocking, twisting wrists, flicking fingers etc?)</td>
<td>No</td>
</tr>
<tr>
<td>9</td>
<td>Stiff posturing</td>
<td>Does he/she strike and hold stiff poses?</td>
<td></td>
<td>No</td>
</tr>
<tr>
<td>10</td>
<td>Noticeable resting tremor</td>
<td>When he/she is completely relaxed, does any part of their body tremble?</td>
<td></td>
<td>No</td>
</tr>
<tr>
<td>No.</td>
<td>Category</td>
<td>Question</td>
<td>Example</td>
<td>Answer</td>
</tr>
<tr>
<td>-----</td>
<td>-----------------------------------</td>
<td>--------------------------------------------------------------------------</td>
<td>-------------------------------------------------------------------------</td>
<td>--------</td>
</tr>
<tr>
<td></td>
<td><strong>Increased motor tics</strong></td>
<td>Does he/she experiences 'tics' (speech or movement)?</td>
<td>(e.g. suddenly &amp; repetitively move their body or saying a word/phrase in a way they seem unable to control?)</td>
<td>No</td>
</tr>
<tr>
<td>11</td>
<td>Waving or shaking extremities</td>
<td>Does he/she move their hands or feet in an odd way?</td>
<td>(e.g. twisting, waving or shaking)</td>
<td>No</td>
</tr>
<tr>
<td>12</td>
<td>Twisting or flicking hands in front of eyes</td>
<td>Does he/she twist or flick their hands in front of their eyes?</td>
<td></td>
<td>No</td>
</tr>
<tr>
<td>13</td>
<td>Moving in a jerky way</td>
<td>Does he/she move in a very jerky way?</td>
<td></td>
<td>No</td>
</tr>
<tr>
<td>14</td>
<td>Unusual gait/posture</td>
<td>Does he/she walk unusually?</td>
<td></td>
<td>No</td>
</tr>
</tbody>
</table>

**Category: Affective Alterations**

<table>
<thead>
<tr>
<th>No.</th>
<th>Category</th>
<th>Question</th>
<th>Example</th>
<th>Answer</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Impulsive or excitable phases</td>
<td>Is he/she impulsive OR overexcitable?</td>
<td></td>
<td>No</td>
</tr>
<tr>
<td>15</td>
<td>Withdrawal from physical contact</td>
<td>Are there periods where he/she withdraws from contact with others?</td>
<td>(e.g. not want to be hugged or touched by anyone, shutting themselves in their room, sitting under a table alone etc)</td>
<td>No</td>
</tr>
<tr>
<td>16</td>
<td>Spontaneous crying, laughing or screaming</td>
<td>Does he/she scream, cry or laugh suddenly for no reason? If so, which?</td>
<td></td>
<td>No</td>
</tr>
<tr>
<td>17</td>
<td>Episodes of aggression</td>
<td>Is he/she agressive towards themselves or others at times? If so, which?</td>
<td></td>
<td>No</td>
</tr>
<tr>
<td>18</td>
<td>Reduced enjoyment in preferred activities</td>
<td>Has he/she lost enjoyment in their favourite activities?</td>
<td>(e.g. do they now get no enjoyment from activities they used to really like or now refuse to do them)</td>
<td>No</td>
</tr>
</tbody>
</table>

**Category: Behavioural Alterations**

<table>
<thead>
<tr>
<th>No.</th>
<th>Category</th>
<th>Question</th>
<th>Example</th>
<th>Answer</th>
</tr>
</thead>
<tbody>
<tr>
<td>19</td>
<td>Difficulty passing through doorways</td>
<td>Does he/she find it difficult to walk through</td>
<td></td>
<td>No</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>---</td>
<td>---</td>
<td>---</td>
<td></td>
<td></td>
</tr>
<tr>
<td>20</td>
<td>Difficulty crossing lines on the floor</td>
<td>Does he/she find it difficult to walk across lines on the floor or changes in flooring? (e.g. from a carpet to a wooden floor)</td>
<td>No</td>
<td></td>
</tr>
<tr>
<td>22</td>
<td>Requiring more encouragement to engage</td>
<td>Is he/she doing less than they used to? (e.g. is it harder than it used to be to encourage them to do activities?)</td>
<td>No</td>
<td></td>
</tr>
<tr>
<td>24</td>
<td>Reduced communication/muteness</td>
<td>Are there periods where he/she communicates with others less or not at all? (This includes all communication methods - it could be reduced speech or other communication such as signing, PECS etc)</td>
<td>No</td>
<td></td>
</tr>
<tr>
<td>25</td>
<td>Incontinence</td>
<td>Are there periods where he/she is incontinent OR refuses to use the toilet when they used to? (e.g. the person is not using skills that they have used in the past and are soiling themselves when they would use the toilet before)</td>
<td>No</td>
<td></td>
</tr>
<tr>
<td>26</td>
<td>Sleep problems</td>
<td>Does he/she have sleep problems? (e.g. finds it difficult to get to sleep at night, wants to sleep in the day but not at night, gets little sleep etc.)</td>
<td>No</td>
<td></td>
</tr>
<tr>
<td>27</td>
<td>Reduced eating</td>
<td>Are there periods where he/she refuses to eat OR eats less than they used to?</td>
<td>No</td>
<td></td>
</tr>
<tr>
<td>28</td>
<td>Eye rolling/ unusual eye movements</td>
<td>Does he/she move or roll their eyes unusually? (e.g. rolls their eyes around again &amp; again or looks from left to right again &amp; again)</td>
<td>No</td>
<td></td>
</tr>
<tr>
<td>29</td>
<td>Unusual facial expressions/’grimaces’</td>
<td>Does he/she pull unusual facial expressions or ‘grimaces’?</td>
<td>No</td>
<td></td>
</tr>
<tr>
<td>30</td>
<td>Ignoring instructions</td>
<td>Does he/she ignore instructions? (This must be for instructions that you know the individual understands)</td>
<td>No</td>
<td></td>
</tr>
<tr>
<td>31</td>
<td>Refusal to bathe or change clothes</td>
<td>Does he/she refuse to wash or change their clothes?</td>
<td>No</td>
<td></td>
</tr>
</tbody>
</table>
Occasional groans or unusual noises

Does he/she make groaning or other unusual noises regularly?

No

Staring into space/fixed gaze

Does he/she stare into space or fix their gaze onto certain things?

No

Unable to lift head

Does he/she seem unable to lift their head?

(e.g. does their head look like it is too heavy for them to lift)

No

Statistical analysis will be completed to determine if there is evidence for a clinical cut-off ACQ score for autistic catatonia, as in existing measures like the Northoff Catatonia Scale (Northoff et al. 1999). Items on the ACQ are rated on a Likert scale as in existing measures of catatonia (Modified Rogers Scale (McKenna et al. 1991); Bush-Francis Catatonia Rating Scale (Bush et al. 1996); Northoff Catatonia Scale (Northoff et al. 1999); Braunig Catatonia Rating Scale (Braunig et al. 2000)). For each item, participants are asked to rate whether the symptom has been displayed by the individual they care for, either currently or in the past. Whether the symptom has presented consistently is also measured as participants are asked about whether it is more, less or the same amount of a problem as previously. Thus the ACQ provides a measure of each symptom, both currently and in the past. The attributes for each variable on the ACQ are:

1. No – never
2. No – not at the moment but it used to happen
3. Yes but less than before
4. Yes but the same as before
5. Yes – more than before
Six of the most commonly reported symptoms of autistic catatonia represent difficulty with motor movement (see ACQ item numbers 1-6 in Table 2), are termed the ‘core symptoms of autistic catatonia’ in this study and are thought to indicate essential elements for diagnosis. Planned statistical analysis will determine if this could be used as a clinical screening measure, as present in other measures such as the Northoff Catatonia Scale (Northoff et al. 1999). Positive responses to these items (i.e. attributes 3, 4 or 5) trigger two supplementary questions which measure the current frequency (i.e. the usual amount of time the symptom is present during waking hours) and severity (i.e. the effect of the symptom on the individual’s ability to perform tasks or activities) of the symptom. This ensures that the ACQ is an effective and sensitive clinical measure of autistic catatonia. The supplementary questions presented for positive responses related to these six ‘core’ symptoms of autistic catatonia are:

1) **How often does the individual experience these periods of [symptom] at the moment?**

Choose from the following options:

- All or almost all of the time they are awake
- Most of the time they are awake
- Some of the time they are awake
- Rarely when they are awake

2) **How severely does the individual experience these periods of [symptom] at the moment?**

Choose from the following options:
• Very severely – they seem unable to focus on or do anything else at these times
• Quite severely – it is difficult for them to focus on or do anything else at these times
• Moderately – there seems to be an effect on their ability to focus on or do things
• Slightly – this seems to have little or no effect on their life

2.3.2.2 Repetitive Behaviour Questionnaire (RBQ):

The RBQ (Moss et al., 2009) is a 19 item third party measure which assesses the prevalence and phenemology of restricted and repetitive behaviours displayed by an individual and is suitable for use within learning disability populations (Moss et al., 2009). Each item is rated as occurring ‘more than once a day’, ‘once a day’, ‘once a week’, ‘once a month’ or ‘never’ and scored on a four point Likert scale. The items are scored into five subdomains; ‘Stereotyped behaviour’, ‘Compulsive behaviour’, ‘Restricted preferences’, ‘Repetitive speech’ and ‘Insistence on sameness’. A total score for each participant can also be obtained by summing the scores of the subdomains. See Appendix 4 for a copy of the RBQ measure.

2.3.2.3 Carer Supplement to the Glasgow Depression Scale for people with Learning Disability (GDS-CS):

The GDS-CS (Cuthill, Espie & Cooper, 2003) is a 16 item measure which assesses current presentation of co-morbid depression in individuals with learning disabilities, as rated by a third party. Each item is rated as ‘never/no’, ‘sometimes/a little’ or ‘always/a lot’. The items are scored in 0-2 format and a cut off point of 13 to indicate
a clinical level of depression is recommended by the authors (Cuthill, Espie & Cooper, 2003). See Appendix 5 for a copy of the GDS-CS measure.

2.4 Validity and reliability of measures:

2.4.1 Validity:
Questionnaire validity is the extent to which an instrument measures what it intends to measure (Anastasi, 1982).

2.4.1.1 Validity of the ACQ:
The ACQ has good face validity due to its objectivity; scoring is based on third party reports of behaviours and all reported symptoms of autistic catatonia which ask raters to infer internal states were excluded from the measure. Additional support for the face validity of the ACQ comes from the similarity of the questions in the measure to the theoretical understanding of autistic catatonia; due to how the measure was designed, the symptoms included in the ACQ are the most commonly reported symptoms associated with autistic catatonia. Content validity is also good for the ACQ due to the adopted empirical approach of including all items in the measure which are reported twice or more in the research literature. Additionally, the ACQ is comparable to non-ASD clinical measures of catatonia, which indicates validity. The ACQ was also examined by non-native English speakers prior to recruitment to ensure clarity and simplicity of the language used. No participants requested help completing the ACQ, which indicates that the measure is clear, readable and easy to use. In summary, we can be confident that the ACQ provides strong face validity of autistic catatonia.
Quantitative assessments of the validity of the ACQ will be detailed in the results section.

2.4.1.2 Validity of the RBQ and GDS-CS:

The RBQ is a valid measure of repetitive and restricted behaviours, showing concurrent and content validity in a sample of 103 individuals aged 10 to 28 years old with intellectual disability when compared to existing measures (Moss et al. 2009). Good levels of concurrent and construct validity have also been shown in an ASD population of 180 individuals aged three to 16.5 years (Honey et al. 2012).

The GDS-CS has good face validity and correlates strongly with existing established clinical measures of depression in a sample of 76 carers ($r=0.88$) (Cuthill, Espie & Cooper 2003). Content validity, discriminant validity and criterion validity have all been shown to be high in this sample (Cuthill, Espie & Cooper 2003).

2.4.2 Reliability:

Reliability is the consistency of a measure; reliability can be proved if a clinical instrument is dependable, reproducible and consistent.

2.4.2.1 Reliability of the ACQ:

The reliability of the ACQ is not assessed quantitatively in this study.

2.4.2.2 Reliability of the RBQ and GDS-CS:

In their sample of 103 individuals with intellectual disability, Moss and colleagues (2009) demonstrated the RBQ has high internal consistency (Cronbach’s $\alpha>0.80$),
Spearman coefficients measuring inter-rater (0.46 - 0.80) and test-retest (0.61 - 0.93) reliability when measuring repetitive and restricted behaviours in this population. Good levels of the internal consistency of the RBQ (Cronbach’s α>0.88) have also been shown in an ASD population of 180 individuals aged three to 16.5 years (Honey et al. 2012).

There is evidence that GDS-CS is a reliable measure of depression in individuals with learning disability; Cuthill and colleagues (2003) administered the GDS-CS to 76 carers and found high internal consistency (Cronbach’s α>0.88), Spearman coefficients measuring test-retest (r=0.98) and inter-rater reliability (r=0.98) (Cuthill, Espie & Cooper 2003).

2.5 Recruitment:

2.5.1 Recruitment of participants to the study:

Recruitment for the study was targeted at organisations which supported young people with ASD in England and their families. Specialist care providers, parent support groups and charities were identified and contacted via letter or email informing them of the aims of the research study, why it is important that the research is completed and what participating in the study will entail. Contact details were included so that potential participants were able to contact the researcher and ask any questions about the study. See Appendix 6 for a copy of the initial contact letter. If no contact was made by the organisation within a month, a follow up phone call was made to discuss the research project further (as stated in the initial contact letter) to ascertain whether the care provider was interested in being involved in the study and
answer any questions that they may have. If this was the case, the applicant offered to arrange a meeting to discuss the project further and/or send some more information about the study (i.e. the participant information approved by the ethics committee).

Consent was required from organisations on an opt-in basis and written consent was obtained from each before information about the research was passed on to individuals associated with the organisation (see Appendix 7). Once this was received, information about the study (specifically the Participant Information Sheet v1.0 (see Appendix 8) and a flyer about the research project (see Appendix 9) were distributed by email and post by the care providers. This information was disseminated by the organisation so the researchers had no way of knowing who had been invited to participate in the study unless the participant themselves contacts the applicant with questions about the study and volunteered information (the applicant did not ask for any personal information).

Information about the research project was also posted on the National Autistic Society website in the ‘Requests for Research Participants’ page (http://www.autism.org.uk/about-autism/research/research-projects-be-a-participant.aspx).

2.5.2 Study inclusion and exclusion criteria:
Participants were recruited based on the following inclusion criteria: Parent or long-term carer (>2 years) of a young person aged 12-25 years with an existing diagnosis of ASD. The inclusion criteria was developed as individuals who develop autistic catatonia have an existing diagnosis of autistic spectrum disorder. As research
indicates that the condition develops during adolescence, it is important to assess the presentation of autistic catatonia in those aged 12-25 years old. Some items refer to comparison to the individual’s previous presentation of symptoms so it was important to only include participants who have known the individual they are reporting about for a reasonable length of time. Therefore, participants were recruited who are a medium or long term carer of an individual with autism spectrum disorder aged 12-25 years and who can report on their current and past presentation.

There were no exclusion criteria in this study.

2.5.3 Sample size

Initially the number of participants reflect an opportunity sample. A minimum of 80 participants was required to test the internal validity of the ACQ. Recruitment for the study exceeded this minimum.

2.6 Data storage and confidentiality:

All data involved in this study is anonymous and was inputted online by participants, directly onto a secure website hosted by the University of Manchester.

All collected data is kept confidential via restricted access. The data will be stored on a password protected drive at the University of Manchester for 5 years. No third party will be informed of any of the data gathered in the study, except for the overall published results.
CHAPTER 3: RESULTS

3.0 Aim: This chapter will describe the data analyses conducted to examine the delineation and the distribution of catatonic symptoms, the assessment of the psychometric properties of the ACQ (i.e. validity) and the relationship between the ACQ and the two additional measures included in the study (RBQ and GDS-CS).

3.1 Statistical analysis:

Data analysis was completed using Statistical Package for the Social Sciences (SPSS) version 20.0. Prior to conducting the analyses, preliminary data checks were completed including that all participants met the inclusion criteria for the study (see Appendix 10) and that assumptions of data normality and independence allowed parametric analysis of data. There was no missing data due to the use of a computerised programme to conduct the survey, however 12 participants dropped out of the study after completing at least up to item six on the ACQ and so their data was excluded beyond this point.

Statistical analysis focussed on answering the research questions stated in Section 1.3. Parametric tests were used as part of the statistical analysis due to the normal distribution of data. Where appropriate checks of skewness and kurtosis were completed on non-normally distributed data (see Appendix 16).
In general, results sections would not include qualitative discussion of the findings, however due to the inclusion of a study aim which intended to design a clinical measure, discussion will be used to support and structure analysis and findings.

3.2 Demographic information:

Eighty-seven participants completed the full questionnaire and an additional 12 participants completed at least the first part of the questionnaire (i.e. at least the demographic information and the items relating to the six core symptoms of autistic catatonia).

Table 3: Summary of demographic information of the sample:

<table>
<thead>
<tr>
<th>Demographic Information</th>
<th>Total sample (n=99)</th>
<th>Full participation (n=87)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age (years)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Range</td>
<td>12 – 25</td>
<td>12 – 25</td>
</tr>
<tr>
<td>Mean</td>
<td>15.7</td>
<td>15.6</td>
</tr>
<tr>
<td>Gender</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Male (%)</td>
<td>79 (79.8%)</td>
<td>67 (77.0%)</td>
</tr>
<tr>
<td>Female (%)</td>
<td>20 (20.2%)</td>
<td>20 (23.0%)</td>
</tr>
<tr>
<td>Relationship</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Parent (%)</td>
<td>91 (91.9%)</td>
<td>81 (93.1%)</td>
</tr>
<tr>
<td>Carer (%)</td>
<td>8 (8.1%)</td>
<td>6 (6.9%)</td>
</tr>
<tr>
<td>Existing Autistic Catatonia</td>
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<td>Diagnosis</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Yes (%)</td>
<td>20 (20.2%)</td>
<td>18 (20.7%)</td>
</tr>
<tr>
<td>No (%)</td>
<td>79 (79.8%)</td>
<td>69 (79.3%)</td>
</tr>
</tbody>
</table>
The distribution of the sample in terms of age can be seen in Graph 1 below. The data are not asymmetrically distributed by age; there is a positive skew towards participants who are younger in the sample. This could be a result of the recruitment procedure for the study as more specialist education providers were identified and targeted than adult care providers.

The gender ratio of the sample reflects the 4:1 ratio commonly identified in ASD populations (Baird et al., 2006; Fombonne et al., 2009), and so indicates that the sample is representative of the study population in terms of gender.

The majority of the participants who took part in the study were parents (91.9%) and so can be assumed to have good knowledge of the presentation of the symptoms exhibited by the person they are reporting about over time. The inclusion criteria specified that carers must have supported the individual for at least two years but all of the recruited carers exceeded this minimum; the six carers who participated fully in the study had cared for the individual they were reporting about for between three
and 22 years (mean = 7.6 years) and the eight carers who participated in the first part of the study had cared for the individual they were reporting about for between three and 22 years (mean = 8.1 years). This information indicates that participants recruited to the study can be expected to have good knowledge of the presentation of symptoms in the young person they are reporting about, both currently and in the past.

All participants indicated that they were reporting symptoms presenting in a young person with an existing diagnosis of ASD. A proportion of participants reported additional diagnoses, with 41 of the total sample (41.4%) having additional diagnostic labels in addition to ASD. Thirteen participants had multiple diagnostic labels (13.1%), resulting in a total of 77 different co-morbid diagnoses (excluding autistic catatonia). See Table 4 (Appendix 12) for details of co-morbid diagnoses reported in the sample.

As shown in Table 4 (Appendix 12), 20 individuals had an existing diagnosis of autistic catatonia. Nine of these individuals had additional diagnostic labels (47.4%), which are displayed in Table 5 (Appendix 12). It is not possible to complete statistical analysis for co-morbid conditions other than autistic catatonia due to small sample size in each additional diagnosis sub-group. 14 boys (18.7%) and six girls (25.0%) in the sample had an existing diagnosis of autistic catatonia. There were more girls than boys in the sample with an existing diagnosis of autistic catatonia (see Graph 2).
3.3 Prevalence of catatonic symptoms in the sample of young people with ASD

Catatonic symptoms present frequently in the study sample. All participants displayed at least four of the broad range of symptoms reported to be connected to autistic catatonia and covered in the 34 items on the ACQ measure. Eighty-four participants (85%) had displayed at least one of the core symptoms associated with autistic catatonia (i.e. ACQ items 1-6) either currently or in the past. Graph 3 shows the number of six core symptoms of autistic catatonia currently displayed by participants in the sample (mean=2.38, sd=1.85).
An independent samples t-test revealed that individuals with an existing diagnosis of autistic catatonia displayed a significantly higher number of the six core symptoms of autistic catatonia (mean=3.10, sd=1.86) compared to those without a diagnosis of autistic catatonia (mean=2.20, sd=1.81); t(97)=1.97, p=0.05.

Graph 4 displays the frequencies that each core symptom was reported to be currently presenting in the sample. This shows that all six of the core symptoms associated with autistic catatonia were commonly reported in the sample, with difficulty initiating movement being the least reported symptom (n=18) and physical and/or verbal prompts required being the most frequently reported (n=60).
3.4 The ACQ as a clinical measure:

There are two options for categorising the data set; by existing autistic catatonia diagnosis or by the presentation of symptoms connected to autistic catatonia. Analysis using existing autistic catatonia diagnosis provides comparison of scores on the ACQ measure with existing clinical measures of autistic catatonia. However, due to the design of the study as a cross-sectional online survey design, not all participants in the populations sample have been clinically assessed for autistic catatonia and under-diagnosis is assumed in the general population (Wing & Shah, 2000). Therefore, using existing diagnosis as data categorisation means that the between group differences may be greater than they appear. An empirical approach to between-group analysis using a clinical cut-off would provide a more unbiased and robust measure of autistic catatonia in the sample.

As the six core symptoms of autistic catatonia are presumed to indicate essential elements for diagnosis, the generated hypothesis for a clinical cut-off point focussed on the number of currently presenting core symptoms. The distribution of the data in the sample (Graph 3) lack a stark categorical division but indicate a potential bimodal distribution which could suggest that autistic catatonia may become a syndrome when individuals display three or more of the six core symptoms of autistic catatonia. Additionally, individuals in the sample with an existing diagnosis of autistic catatonia were shown to display significantly higher numbers of the six core symptoms of autistic catatonia, which indicates that the data supports the ACQ as a valid indicator of autistic catatonia. The lower threshold was assigned as a working parameter for a
diagnosis of autistic catatonia in this study (i.e. three or more of the six core symptoms of autistic catatonia) due to the accepted notion that these are key to the presentation of autistic catatonia and will be explored statistically in this chapter. Both options for categorisation of the data set into diagnostic groups provide useful and distinct analysis of the data and so between group statistical analyses were completed and reported for both options in this study.

3.4.1 Core symptom scoring options for the ACQ measure:

All items on the ACQ involve a five point scoring system (no never, No – not at the moment but it used to happen, Yes but less than before, Yes but the same as before, Yes – more than before). Scoring the ACQ using a 0-4 Likert scale scoring system involves a weighted scoring method of 0-1-2-3-4 and so captures the presentation of the symptom over time (as worsening symptoms score more highly).

Analysis focussed on the six core symptoms associated with autistic catatonia as it is assumed that these commonly reported motor symptoms are key to conceptualising autistic catatonia. A number of preliminary variables were computed from the data to conduct the statistical analysis and are described below:

1. ‘Core Autistic Catatonia Score’: This computed variable represents a measure of the presence of each of the six core symptoms associated with autistic catatonia in each individual. The total score for these six items are summed to obtain each participant’s Core Autistic Catatonia Score.

2. ‘Core Severity Score’: This computed variable represents a measure of the severity of the currently presenting core symptoms associated with autistic
catatonia in each individual. As stated in Section 2.3.2.1, each positive response to the six core symptoms of autistic catatonia triggers a supplementary question which measures the current severity (i.e. the effect of the symptom on the individual’s ability to perform tasks or activities) of the symptom. The total score for these six supplementary questions are summed to obtain each individual’s Core Severity Score.

3. ‘Core Frequency Score’: This computed variable represents a measure of the frequency of the currently presenting core symptoms associated with autistic catatonia in each individual. As stated in Section 2.3.2.1, each positive response to the six core symptoms of autistic catatonia triggers a supplementary question which measures the current frequency (i.e. the usual amount of time the symptom is present during waking hours) of the symptom. The total score for these six supplementary questions are summed to obtain each individual’s Core Frequency Score.

*Core Autistic Catatonia Score, Core Severity Score and Core Frequency Score* are computed using a weighted scoring method of 0-1-2-3-4 for the total sample (n=99). The maximum possible score for each computed variable is 24.

### 3.4.1.1 Core Autistic Catatonia Score:

The distribution of the *Core Autistic Catatonia Score* data can be seen in Graph 5. The mean *Core Autistic Catatonia Score* for the sample was 7.48 (sd=5.70) with a score range of 0-24. Fifteen participants had a *Core Autistic Catatonia Score* of 0 (15.2%).
An independent samples t-test found no significant difference in **Core Autistic Catatonia Score** between those with (mean=9.35, sd=6.00) and those without (mean=7.01, sd=5.57) an existing diagnosis of autistic catatonia; t(97)=1.65, p=0.10). As could be expected, individuals who currently displayed three or more core symptoms of autistic catatonia had a significantly higher **Core Autistic Catatonia Score** (mean=12.04; sd=4.24) than those who displayed less than three core symptoms of autistic catatonia (mean=3.2; sd=2.86); t(97)=-12.23, p<0.01.

### 3.4.1.2 Core Severity Score:

The distribution of the **Core Severity Score** (i.e. how severely the presenting core symptoms were rated) in the sample can be seen in Graph 6. The mean **Core Severity Score** for the sample was 5.23 (sd=4.81) with a score range of 0-23. Twenty-two participants had a **Core Severity Score** of 0 (22.2%).
No significant difference was found in the Core Severity Score between those with (mean=6.95, sd=4.70) and those without (mean=4.80, sd=4.77) an existing diagnosis of autistic catatonia; t(97)=1.81, p=0.07). Individuals who currently displayed three or more core symptoms of autistic catatonia had a significantly higher Core Severity Score (mean=8.94; sd=4.09) than those who displayed less than three core symptoms of autistic catatonia (mean=1.75; sd=2.01); t(97)=-11.20, p<0.01.

3.4.1.3 Core Frequency Score:

The distribution of the Core Frequency Score (i.e. how frequently the presenting core symptoms were rated) in the sample can be seen in Graph 7. The mean Core Frequency Score for the sample was 4.73 (sd=3.87) with a score range of 0-15. Twenty-one participants had a Core Frequency Score of 0 (21.2%).
Individuals with an existing diagnosis of autistic catatonia were found to have significantly higher Core Frequency Scores (mean=6.45, sd=3.90) than those without an existing diagnosis of autistic catatonia (mean=4.34, sd=3.72); t(97)=2.24, p<0.05. Therefore, the core symptoms associated with autistic catatonia were rated to occur for a higher proportion of waking hours for those participants with an existing diagnosis of autistic catatonia.

Individuals who currently displayed three or more core symptoms of autistic catatonia had a significantly higher Core Frequency Score (mean=7.85; sd=2.78) than those who displayed less than three core symptoms of autistic catatonia (mean=1.86; sd=1.93); t(97)=-12.51, p<0.01.
3.4.2 ACQ scoring comprising all items in the ACQ:

3.4.2.1 Total ACQ Score:

Statistical analysis including all 34 items on the ACQ measure was completed to assess if a ‘Total ACQ Score’ would be an appropriate scoring method for the ACQ. Total ACQ Score is computed by summing the total score for the 34 items using a 0-4 Likert scoring method, providing a maximum score of 136 for each participant (n=87). The distribution of the Total ACQ Score in the sample can be seen in Graph 8. The mean Total ACQ Scores for the sample was 54.28 (sd=21.23) with a score range of 5-110.

Graph 8: Histogram displaying Total ACQ scores (n=87)

Independent samples t-tests revealed that there were no statistically significant difference between those with (mean=59.33, sd=19.77) and those without (mean=52.96, sd=21.54) an existing diagnosis of autistic catatonia for Total ACQ Score; t(85)=1.14, p=0.259.
However, individuals who currently displayed three or more core symptoms of autistic catatonia had a significantly higher *Total ACQ Score* (mean=61.04; sd=21.07) than those who displayed less than three core symptoms of autistic catatonia (mean=44.33; sd=22.02); t(82)=-3.53, p<0.01.

### 3.4.2.2 ACQ Supplementary Score:

An ‘ACQ Supplementary Score’ comprising of the 28 items on the ACQ which are not related to core symptoms of autistic catatonia was also computed in a similar way, providing a maximum score of 112 for each participant (n=87). The distribution of the ACQ Supplementary Scores in the sample can be seen in Graph 9. The mean ACQ Supplementary Score for the sample was 46.56 (sd=17.53) with a score range of 5-87.

![Graph 9: Histogram displaying ACQ Supplementary scores (n=87)](image)

There were no statistically significant difference between those with (mean=50.33, sd=17.42) and those without (mean=45.58, sd=17.58) an existing diagnosis of autistic catatonia for *ACQ Supplementary Score*; t(85)=1.03, p=0.308. There were no statistically significant difference between individuals who currently displayed three or
more core symptoms of autistic catatonia (mean=49.42, sd=17.39) and those who
displayed less than three core symptoms of autistic catatonia (mean=43.77; sd=17.41)
for ACQ Supplementary Score; t(85)=-1.51, p=0.134.

3.4.3 Scoring recommendation for the ACQ measure:
The recommended primary scoring strategy for the ACQ as a diagnostic screening tool
is to focus on the six core symptoms associated with autistic catatonia as these are
viewed as essential elements for diagnosis, namely the computed variable Core
Autistic Catatonia Score. An analysis of the sensitivity of this scoring strategy was
completed using a Receiver Operating Curve (ROC) analysis to determine the ability of
Core Autistic Catatonia Score to identify individuals in the sample who met diagnostic
criteria for autistic catatonia. Appendix 13 shows the ROC curve assessing the ability of
Core Autistic Catatonia Score to identify individuals with an existing diagnosis of
autistic catatonia and indicates that this parameter is suitable for use as a scoring
strategy for the ACQ with the area under curve computed as 62.4% and the autistic
catatonia group being identified by Core Autistic Catatonia Score at significantly higher
than chance probability rates (p<0.05). For the proposed clinical cut-off for autistic
catatonia, the sensitivity value is shown to be 0.914 and the specificity value 0.179 for
predicting existing autistic catatonia diagnosis (Appendix 13). This suggests that using
Core Autistic Catatonia Score provides a sensitive scoring strategy for the ACQ measure
as this variable is able to identify individuals who are presenting with and have a
current diagnosis of autistic catatonia. This will be discussed further in Chapter 4.
Appendix 13 indicates that for an ACQ score of 6.5, the sensitivity is 0.882 and 1-
specificity is 0.687, which changes only slightly for an ACQ score of 7.50, which has a
sensitivity of 0.824 and 1-specificity of 0.597, suggesting an alternative clinical cut-off
score of 7 which provides an appropriate balance of false negatives and false positives could be proposed.

An alternative clinical cut-off for autistic catatonia could be proposed using the Core Autistic Catatonia Score variable. Analysis of the co-ordinates of the ROC curve (i.e. sensitivity and specificity values) in Appendix 13 indicates that a clinical cut off point for autistic catatonia could be a Core Autistic Catatonia Score of greater than 7 or 8. The validity of both proposed clinical cut offs require further investigation which is beyond the scope of the current study. As the presentation of the six core symptoms of autistic catatonia are generally accepted to be indicators of autistic catatonia and the ACQ measure is not presently an accepted clinical measure of autistic catatonia, the presentation of three or more of these has been chosen as the proposed clinical cut-off in the current study.

3.4.4 Checking for redundant items on the ACQ measure:

In line with other clinical measures of catatonia and as already stated in Section 2.3.2.1, items on the ACQ can be assigned to three subdomains; motor symptoms (n=15), affective alterations (n=5) and behavioural alterations (n=14). Subdomain scores can be computed by summing the weighted scoring method (0-1-2-3-4) for items assigned to each subdomain (n=87). It is important to ensure that all items on the measure are clinically relevant; a statistical examination of the correlation between individual items in the subdomain and the overall subdomain score indicates if individual items are contributing to the measure or can be dropped.

3.4.4.1 Motor Symptom subdomain:
An ‘ACQ Motor Score’ can be computed by summing the total score for the 15 motor items using a 0-4 Likert scoring method, providing a maximum score of 60 for each participant (n=87). The distribution of the ACQ Motor Score in the sample can be seen in Graph 10. The mean ACQ Motor Score for the sample is 20.61 (sd=11.25) with a score range of 0-55.

Graph 10: Histogram displaying ACQ Motor scores (n=87)

ACQ Motor Score correlates with Core Autistic Catatonia Score and the other two subdomain scores (p<0.01), as shown in Table 6 below, which indicates that this subdomain is contributing to the measure and additional motor items not related to the six core symptoms associated with autistic catatonia should not be dropped from the ACQ.

Table 6: Pearson’s parametric correlations between ACQ computed variables:
Pearson’s parametric correlations between the *ACQ Motor Score* and the individual items included in the motor subdomain can be seen in Table 7 below.

**Table 7: Pearson’s parametric correlations between *ACQ Motor Score* and the individual items included in the motor subdomain**

<table>
<thead>
<tr>
<th></th>
<th>VS</th>
<th>S</th>
<th>VSM</th>
<th>IM</th>
<th>VSM</th>
<th>PR</th>
<th>WF</th>
<th>MVC</th>
<th>SP</th>
<th>TSM</th>
<th>OMHF</th>
<th>TFHE</th>
<th>JM</th>
<th>WU</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Core_AC_Score</strong></td>
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<td>.831**</td>
<td>.368**</td>
<td>.513**</td>
<td>.559**</td>
<td>.729**</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td><strong>ACQ_MotorScore</strong></td>
<td>.831**</td>
<td>1</td>
<td>.540**</td>
<td>.598**</td>
<td>.808**</td>
<td>.889**</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td><strong>ACQ_AffectiveScore</strong></td>
<td>.368**</td>
<td>.540**</td>
<td>1</td>
<td>.720**</td>
<td>.840**</td>
<td>.792**</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td><strong>ACQ_BehaviourScore</strong></td>
<td>.513**</td>
<td>.598**</td>
<td>.720**</td>
<td>1</td>
<td>.900**</td>
<td>.880**</td>
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<tr>
<td><strong>SupplimentaryACQScore</strong></td>
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<td></td>
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</tr>
</tbody>
</table>

**. Correlation is significant at the 0.01 level (2-tailed).**

* Correlation is significant at the 0.05 level (2-tailed).
**Correlation is significant at the 0.01 level (2-tailed).**  
*Correlation is significant at the 0.05 level (2-tailed).

<table>
<thead>
<tr>
<th></th>
<th>ACQ Behavioural Score p&lt;0.01</th>
<th>ACQ Motor Score p&lt;0.01</th>
</tr>
</thead>
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<td>RBM</td>
<td>.72** .27 .24 .37** .36** .28** .34** .16 1 .28** .26 .59** .68** .51** .34** .25</td>
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</tr>
<tr>
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</tr>
<tr>
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</tr>
<tr>
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<td>.61** .18 .13 .30** .28** .21 .23 .03 .59** .22 .31** 1 .53** .37** .24 .29**</td>
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<tr>
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<td></td>
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<tr>
<td>TFHE</td>
<td>.63** .34** .23 .16 .46** .34** .21 .23 .52** .39** .15 .37** .46** 1 .28** .11</td>
<td></td>
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<td></td>
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<tr>
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<td>.42** .06 .14 .18 .08 .08 .11 .18 .25 .25 .06 .29** .32** .11 .42** 1</td>
<td></td>
</tr>
</tbody>
</table>

**. Correlation is significant at the 0.01 level (2-tailed).  
*. Correlation is significant at the 0.05 level (2-tailed).

WU=walk unusually, JM=jerky movements, TFHE=twist/flick hands in front of eyes, OMHF=odd movements hands/feet, TSM=tics speech/movement, RT=resting tremor, SP=striking/holding stiff poses, RBM=repetitive body movements, WF=waxy flexibility, PR=prompts required, VSM=very slow movements, PIM=problems initiating movements, SA=stopping actions, S=stuck, VS=very still

All items are highly correlated with the ACQ Behavioural Score (p<0.01). Generally, correlation is high between items in the motor subdomain. There is no one item which is perfectly correlated with another in the subdomain which indicated that there are no duplicate items which can be collapsed. Table 7 indicates that ‘Waxy flexibility’ (item number 7) and ‘Noticeable resting tremor’ (item number 10) are poorly correlated with other motor items and so these are dropped from the ACQ measure.

### 3.4.4.2 Affective Alterations subdomain:

An ‘ACQ Affective Alterations Score’ can be computed by summing the total score for the five affective alteration items using a 0-4 Likert scoring method, providing a maximum score of 20 for each participant (n=87). The distribution of the ACQ Affective Alterations Score in the sample can be seen in Graph 11.
The mean ACQ Affective Alterations Score for the sample was 11.30 (sd=4.42) with a score range of 0-20. ACQ Affective Score correlates with Core Autistic Catatonia Score and the other two subdomain scores (p<0.01) (see Table 6 above), which indicates that this subdomain is contributing to the measure and items relating to affective alterations should not be dropped from the ACQ.

Pearson’s bivariate correlations between the ACQ Affective Alterations Score and the individual items included in the Affective Alterations subdomain can be seen in Table 8 below.

Table 8: Pearson’s bivariate correlations between the ACQ Affective Alterations Score and the individual items included in the affective alterations subdomain:
All items are highly correlated with the ACQ Affective Alterations Score (p<0.01). Generally, correlation is high between items in the affective alterations subdomain. Table 8 indicates that there is evidence that there is higher levels of correlation between internalising affective alteration items (e.g. withdrawal from others and lost enjoyment in favourite activities) and externalising affective alteration items (e.g. spontaneous screaming, laughing or crying and impulsive or overexcitable) so the five items in this subdomain will be collapsed into two items on the ACQ measuring emotional affective state; internalising and externalising (with appropriate examples).

3.4.4.3 Behavioural Alterations subdomain:

An ‘ACQ Behavioural Alterations Score’ can be computed by summing the total score for the 14 affective alteration items using a 0-4 Likert scoring method, providing a maximum score of 56 for each participant (n=87). The distribution of the ACQ Behavioural Alterations Scores in the sample can be seen in Graph 12. The mean ACQ
**Behavioural Alterations Score** for the sample was 22.37 (sd=8.78) with a score range of 3-47.

**ACQ Behaviour Score** correlates with **Core Autistic Catatonia Score** and the other two subdomain scores (p<0.01) (see Table 6 above), which indicates that this subdomain is contributing to the measure and items relating to behavioural alterations should not be dropped from the ACQ.

Pearson’s bivariate correlations between the **ACQ Behavioural Alterations Score** and the individual items included in the behavioural alterations subdomain can be seen in Table 9 below.

**Table 9: Pearson’s bivariate correlations between the ACQ Behavioural Alterations Score and the individual items included in the behavioural alterations subdomain**
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**. Correlation is significant at the 0.01 level (2-tailed).
*. Correlation is significant at the 0.05 level (2-tailed).

ILH=inability to lift head, FG=fixed gaze, UN=unusual noises, RWCC=refusal to wash/change clothes, II=ignore instructions, UEM=unusual eye movements, RE=reduced eating, SP=sleep problems, I=incontinence, REC=reduced expressive communication, LEFA=lost enjoyment in favourite activities, DWALF=difficulty walking across lines on the floor, DWTD=difficulties walking through doorways.

There is no one item which is perfectly correlated with another in the subdomain which indicated that there are no duplicate items which can be collapsed. Table 9 indicates that all items are highly correlated with the ACQ Behavioural Alterations Score (p<0.01) except for ‘inability to lift head’ (item number 34), which can be dropped from the ACQ measure. Unlike the other subdomains, lots of items in the behavioural alterations subdomain are not highly correlated with each other.

### 3.4.4.4 Amended ACQ measure:

Statistical analysis has reduced the number of items on the ACQ from 34 to 28 items (12 motor symptoms, two affective alterations and 13 behavioural alterations). It is
not possible to re-run statistical analysis using this modified 28 item ACQ due to the collapsing of the five items in the *Affective Alterations subdomain* into two items. Although further statistical analysis could be completed to determine approximate scores for each participant, for example by calculating mean secondary scores for internalising and externalising items, this is beyond the scope of the current study.

### 3.5 Additional information which adds to the theoretical understanding of autistic catatonia:

#### 3.5.1 Age:

As autistic catatonia is thought to onset in adolescence, it could be assumed that older individuals in the sample are more likely to have autistic catatonia. Independent samples t-test reveals that there is no significant different in age between those with (mean=16.20, sd=4.420) and those without (mean=15.55, sd=3.89) an existing diagnosis of autistic catatonia; t(96)=-0.647, p=0.519. There were no statistically significant difference between individuals who currently displayed three or more core symptoms of autistic catatonia (mean=16.32, sd=4.57) and those who displayed less than three core symptoms of autistic catatonia (mean=15.10; sd=3.29) for age; t(96)=-1.53, p=0.130. Therefore there is no evidence that older individuals in the sample were more likely have an existing diagnosis of autistic catatonia or to present with symptoms connected to autistic catatonia. However, it is important to note the non-typical distribution of age within the study sample.

#### 3.5.2 Gender:
As already stated, there were significantly more girls than boys with an existing diagnosis of autistic catatonia (see Graph 2); p<0.05. Pearson Chi-Square analysis of independence of existing autistic catatonia and gender (see Appendix 14) indicate that these variables are independent of each other and so there is insufficient evidence to conclude that gender is associated with receiving a diagnosis of autistic catatonia; $\chi^2(1, n=99)=0.45$, p=0.501. Independent samples t-tests reveal that there is no significant difference in Core Autistic Catatonia Score between males (mean=8.89, sd=5.15) and females (mean=8.64, sd=5.26) in the sample; t(82)=0.195, p=0.846. Similar gender differences were also not found in Core Severity Score (p=0.683) and Core Frequency Score (p=0.822).

3.5.3 Assessment of the six core symptoms as key to an existing diagnosis of autistic catatonia:

As the six core symptoms are believed to be key to identifying autistic catatonia, the current presentation of these symptoms in the individuals in the sample with an existing diagnosis of autistic catatonia was examined. No clear pattern of these symptoms which could be used as defined diagnostic criteria for autistic catatonia could be identified in the current study. Graph 13 shows the frequency of each of the six core symptoms reported to be currently presenting in the individuals with an existing diagnosis of autistic catatonia in the sample (n=20).

Graph 13: Histogram displaying the frequency of current core symptom presentation in individuals with an existing diagnosis of autistic catatonia (n=20)
3.5.4 Potential under-diagnosis of autistic catatonia:

Statistical investigation can be completed to determine if there is evidence of potential under-diagnosis of autistic catatonia in the sample using the clinical cut-off point suggested in this study by comparing individuals with an existing diagnosis of autistic catatonia and those who meet the clinical cut-off for current presentation of core catatonia symptoms.

Fourty-two individuals (48.3%) displayed three or more of the core symptoms of autistic catatonia in the current study sample. To investigate if there is possible evidence that autistic catatonia has been under-diagnosed, crosstabulations and Pearson’s Chi-Squared test of independence ($\chi^2$) were completed for existing diagnosis of autistic catatonia and whether individuals displayed more or less than three core symptoms of autistic catatonia (see Table 10 in Appendix 15). This variable was chosen due to the proposal that there is evidence that this represents a clinical cut-off point for autistic catatonia. No association at the chosen threshold was found between individuals displaying three or more core symptoms of autistic catatonia and an
existing diagnosis of autistic catatonia; $\chi^2(1, n=99)=2.74, p=0.098$. Therefore there is no evidence that individuals in the sample who meet the proposed clinical cut-off for autistic catatonia have an existing diagnosis of the condition.

Twenty-three individuals (26.4%) displayed four or more core symptoms of autistic catatonia in the study sample. Crosstabulations and Pearson’s Chi-Squared test of independence ($\chi^2$) analysis was repeated with a new computed variable where individuals were recoded into whether they displayed more or less than four of the six core symptoms associated with autistic catatonia (see Table 11 in Appendix 15). A significant association at the chosen threshold was found between individuals displaying four or more core symptoms of autistic catatonia and an existing diagnosis of autistic catatonia; $\chi^2(1, n=99)=6.53, p<0.05$. Thus there is evidence that an existing diagnosis of autistic catatonia is associated with presentation of four or more core symptoms of autistic catatonia.

It is interesting to consider the possibility of under-diagnosis of autistic catatonia in the current study sample. It may be possible that under-diagnosis of autistic catatonia is present here if the proposed clinical cut-off point of an individual currently displaying three or more of the core symptoms associated with autistic catatonia is proven to be accepted as diagnostic criteria. Data analyses suggests that autistic catatonia is more likely to be diagnosed in the current sample when four or more core symptoms of autistic catatonia are present, although it may be that autistic catatonia is clinically evident when three core symptoms of autistic catatonia are present. This will be further discussed in Chapter 4, along with evidence relating to data comparing the ACQ to established clinical measures in this population presented in section 3.6.
It is important to note that although these chi squared analyses do not reach the chosen threshold for a clinical cut off point, significance is demonstrated to a less stringent threshold. Furthermore, the ROC analysis of *Core Autistic Catatonia Scores* (Appendix 13) indicates that the presentation of these core symptoms is connected to a diagnosis of autistic catatonia. Thus the debate around under-diagnosis of autistic catatonia requires further investigation, which is beyond the scope of the current study.

3.6 *Investigation into a relationship between the ACQ and the other administered measures:*

3.6.1 *Carer Supplement to the Glasgow Depression Scale for people with Learning Disability (GDS-CS):*

The GDS-CS is scored using a 0-2 Likert scoring method, providing a maximum score of 32 for each participant. The distribution of the GDS-CS scores in the study sample can be seen in Graph 14. Further analysis of skewness and kurtosis of this data was completed to investigate the non-typical distribution further; the GDS-CS data is moderately negatively skewed and an acceptable platykurtic kurtosis is evident (see Table 12 in Appendix 16). The mean GDS-CS score for the study sample is 10.56 (sd=5.98) and the scores range from 0-20.
Between group analyses were completed for the measure of co-morbid depression in the study. There were no statistically significant difference between those with (mean=13.33, sd=5.02) and those without (mean=12.77, sd=4.17) an existing diagnosis of autistic catatonia for GDS-CS scores; t(85)=-0.491, p=0.625).

However, individuals who currently displayed three or more core symptoms of autistic catatonia had a significantly higher GDS-CS Score (mean=12.74; sd=5.07) than those who displayed less than three core symptoms of autistic catatonia (mean=8.43; sd=6.09); t(85)=-3.586, p=0.01.

The significant difference in GDS-CS scores between groups may be due to an association between the presentation of symptoms associated with depression and core symptoms connected to autistic catatonia; i.e. Core Autistic Catatonia Score and GDS-CS score may be correlated. A simple linear regression analysis was conducted to determine if Core Autistic Catatonia Score can be predicted from GDS-CS score. Assumptions required for regression analysis were statistically checked and verified,
including homoskedasticity and the presence of a linear association between variables (see Appendix 17). Regression analysis of the correlation between these variables indicates a significant linear association between ACQ Core Score and GDS-CS Score, which indicates that Core Autistic Catatonia Score describes around 15% of the variation in GDS-CS Score; $r^2=0.15$, $p<0.001$ (see SPSS output in Appendix 18).

3.6.2 Repetitive Behaviour Questionnaire (RBQ): The RBQ is scored using a 0-4 Likert scoring method, providing a maximum score of 76 for each participant (n=87). The distribution of Total RBQ scores in the sample can be seen in Graph 15. Further analysis of skewness and kurtosis of this data was completed to investigate the non-typical distribution further; the RBQ data is moderately negatively skewed and an acceptable platykurtic kurtosis is evident (see Table 13 in Appendix 16). The mean RBQ score for the sample is 32.82 (sd=16.73) and the scores range from 1-68.

Independent samples t-test indicates that individuals with an existing diagnosis of autistic catatonia have statistically higher RBQ total scores (mean=40.56, sd=15.73)
than those without an existing diagnosis of autistic catatonia (mean=30.80, sd=16.49); t(85)=2.256, p<0.05. Participants who currently presented with three or more core AC symptoms also had significantly higher RBQ total scores (mean=39.58, sd=13.74) than participants presenting with less than three core AC symptoms (mean=26.20, sd=16.86); t(85)=-4.05, p<0.01.

The significant difference in RBQ scores between groups may be due to an association between the presentation of repetitive and restricted behaviours and core symptoms connected to autistic catatonia; i.e. Core Autistic Catatonia Score and RBQ score may be correlated. A simple linear regression analysis was conducted to determine if Core Autistic Catatonia Score can be predicted from RBQ Total score. Assumptions required for regression analysis were statistically checked and verified, including homoskedasticity and the presence of a linear association between variables (see Appendix 19). Regression analysis of the correlation between these variables indicates a significant linear association between Core Autistic Catatonia Score and RBQ Total Score, and indicates that Core Autistic Catatonia Score describes around 12% of the variation in RBQ Total Score; \( r^2=0.12, p<0.001 \) (see SPSS output in Appendix 20).

3.6.2.1 Repetitive Behaviour Questionnaire subdomains:

Five subdomain scores can also be computed from the RBQ data; Stereotyped behaviour, Compulsive behaviour, Restricted preferences, Insistence on sameness and Repetitive speech. Similar between-group analyses can also be completed for these subdomain scores. Pearson’s bivariate correlations between the ACQ Core Score and individual subdomain scores can be seen in Appendix 21.
3.6.2.1.1 Stereotyped behaviour subdomain:

There were no statistically significant difference between those with (mean=8.61, sd=3.47) and those without (mean=6.57, sd=4.41) an existing diagnosis of autistic catatonia for Stereotyped behaviour subdomain scores; t(85)=1.824, p=0.072. There were no statistically significant difference between individuals who currently presented with three or more core AC symptoms (mean=7.16, sd=4.13) and those presenting with less than three core AC symptoms (mean=6.82, sd=4.49) for Stereotyped behaviour subdomain scores; t(85)=-0.372, p=0.711. Appendix 21 demonstrates a significant correlation was found between Stereotyped behaviour subdomain scores and Core Autistic Catatonia Score; r=0.369, n=87, p=0.000.

3.6.2.1.2 Compulsive behaviour subdomain:

There were no statistically significant difference between those with (mean=11.28, sd=7.37) and those without (mean=8.70, sd=6.44) an existing diagnosis of autistic catatonia for Compulsive behaviour subdomain scores; t(85)=1.471, p=0.145. However, individuals who currently presented with three or more core AC symptoms had significantly higher Compulsive behaviour subdomain scores (mean=11.37, sd=6.28) than those presenting with less than three core AC symptoms (mean=7.14, sd=6.45); t(85)=-3.10, p<0.05. Appendix 21 demonstrates a significant correlation was found between Compulsive behaviour subdomain scores and Core Autistic Catatonia Score; r=0.342, n=87, p=0.001.

3.6.2.1.3 Restricted preferences subdomain:

There were no statistically significant difference between those with (mean=7.28, sd=3.86) and those without (mean=5.71, sd=3.76) an existing diagnosis of autistic
catatonia for Restricted preferences subdomain scores; t(85)=1.567, p=0.121. However, individuals who currently presented with three or more core AC symptoms had significantly higher Restricted preferences subdomain scores (mean=7.23, sd=3.64) than those presenting with less than three core AC symptoms (mean=4.86, sd=3.65); t(85)=-3.03, p<0.05. Appendix 21 demonstrates a significant correlation was found between Restricted preferences subdomain scores and Core Autistic Catatonia Score; r=0.358, n=87, p=0.001.

3.6.2.1.4 Insistence on sameness subdomain:
There were no statistically significant difference between those with (mean=6.00, sd=2.54) and those without (mean=4.71, sd=2.66) an existing diagnosis of autistic catatonia for Insistence on sameness subdomain scores; t(85)=1.85, p=0.068. Individuals who currently presented with three or more core AC symptoms were found to have significantly higher Insistence on sameness subdomain scores (mean=5.95, sd=2.39) than those presenting with less than three core AC symptoms (mean=4.02, sd=2.61); t(85)=-3.60, p=0.001. Appendix 21 demonstrates a significant correlation was found between Insistence on sameness subdomain scores and Core Autistic Catatonia Score; r=0.323, n=87, p=0.002.

3.6.2.1.5 Repetitive speech domain:
Independent samples t-test reveal that participants with an existing diagnosis of autistic catatonia have significantly higher Repetitive speech subdomain scores (mean=7.39, sd=4.18) than those without an existing diagnosis of autistic catatonia (mean=5.12, sd=4.14); t(85)=2.07, p<0.05. Individuals who currently presented with three or more core AC symptoms were also found to have significantly higher
Repetitive speech subdomain scores (mean=6.79, sd=3.94) than those presenting with less than three core AC symptoms (mean=4.41, sd=4.20); t(85)=-2.73, p<0.01. Appendix 21 demonstrates a significant correlation was found between Repetitive speech subdomain scores and Core Autistic Catatonia Score; r=0.336, n=87, p=0.001.
CHAPTER 4: DISCUSSION

4.0 Aim: This chapter will interpret and further discuss the statistical findings of the research study that were presented in Chapter 3. The findings will be discussed in relation to the aims of the research and implications for clinical practice and recommendations for future research will be outlined, along with the inherent limitations of the current study.

4.1 Overview of the study:

Autistic catatonia is an under-researched and poorly understood neurological condition, and little is known about the presentation and variation of symptoms. Although there are numerous descriptions of autistic catatonia in the literature, the number and extent of reported symptoms varies widely with few individuals presenting with the full range of core symptoms.

The primary aims of the study were to focus on the phenomenology of autistic catatonia via a systematic examination of the prevalence and presentation of symptoms reported to be associated with autistic catatonia in young people aged 12-25 years old with Autistic Spectrum Disorder (ASD). The Autistic Catatonia Questionnaire (ACQ), a third party observational measure, was developed from extant reports of symptoms as an aim of this study for use as a clinical assessment and research tool. Preliminary investigations of the internal properties of the ACQ were completed, along with correlation with two established clinical measures assessing...
depression (GDS-CS - Cuthill, Espie & Cooper, 2003) and repetitive and restricted behaviour (RBQ - Moss et al., 2009).

4.2 Summary of the results:

Statistical analyses of the data indicate that catatonic symptoms are commonly presented in this population of young people with ASD. At least one of the six core symptoms of autistic catatonia, comprising motor items regarded as essential elements for diagnosis, were reported to have been displayed by 85% (n=84) of the sample. No clear pattern of symptoms associated with autistic catatonia were found in the current study. Participants reported on a broad range of symptoms connected to autistic catatonia in the present study and all reported the presentation of at least four of these symptoms.

The current results indicated that the ACQ is a workable clinical measure for use with this population with a degree of discriminant validity. On the basis of the initial statistical analysis, the ACQ was reduced from 34 to 28 items. Items can be assigned to three subdomains (Motor symptoms, Affective alterations and Behavioural alterations) in a similar format to the existing catatonic measures used in other clinical populations. There are a number of scoring options and Autistic Catatonia Score is recommended as the primary scoring strategy for the ACQ.

There was evidence from the present study that the number of core symptoms currently presenting in each individual provides key information about the presentation of autistic catatonia. The current results also support the suggestion of
an autistic catatonia continuum and there is support for the notion that the items relating to the six core symptoms of autistic catatonia (ACQ item numbers 1-6) could be used as a working clinical screening tool for autistic catatonia, with a proposed diagnostic cut off of three or more of these core symptoms. On the basis of the current findings, there may be an under recognition of autistic catatonia with 20 participants having an existing diagnosis of autistic catatonia but 42 displaying three or more core symptoms of autistic catatonia (the proposed clinical cut off suggested in this study) and 23 displaying four or more core symptoms of autistic catatonia.

There is also evidence of a relationship between the presentation of autistic catatonia and measures of depression and repetitive and restricted behaviours, although a causal relationship was not determined.

4.3 The ACQ as a clinical measure:

The results indicate that the ACQ is a valid clinical measure of symptoms indicative to autistic catatonia. There is evidence of discriminant validity of the ACQ measure against existing measures of autistic catatonia (i.e. existing diagnosis) and when using a clinical cut-off point of three or more of the six core symptoms of autistic catatonia. The computed variables and subdomain scores are highly inter-correlated to the 0.01 probability level, as can be seen in Table 6. The distribution of computed variables for scoring appear to be normally distributed, particularly Core Autistic Catatonia Score (see Graph 5), Total ACQ Score (see Graph 8) and Supplementary ACQ Score (see Graph 9). The distribution of the three subdomain scores also appear close to normal, as
shown in Graphs 10, 11 and 12 further contributing to the good psychometric properties of the ACQ.

The validity of the ACQ was examined using Pearson’s bivariate correlations with items compared to each other and to the associated total subdomain score (Tables 6-8). Items with poor correlation coefficients were dropped from the measure, reducing the number of items on the ACQ by six to a total of 28, comprising six core symptom items and 22 supplementary items. No two items were perfectly correlated suggesting that there are no duplicate or redundant items present on the ACQ.

The ACQ allows a comparison of the change in presentation of symptoms over time due to the scoring metric for each question asking participants to rate the symptom compared to its past presentation. Assuming the face and ecological validity of the ACQ to be reasonable on the basis that the items relate to observable behaviours and characteristics, the ACQ facilitates analysis of changes to the presentation of symptoms over time and so would provide information about the course of autistic catatonia and the effectiveness of clinical intervention. However, the ACQ would not indicate when a symptoms presentation changes, only that it is more, less or the same as before.

The ACQ permits analysis on a between-group basis to examine existing diagnosis of autistic catatonia or using a clinical cut-off point to determine the presence of autistic catatonia. A clinical cut-off point for the presence of autistic catatonia has been provisionally set at three or more currently presenting core symptoms, based on the
distribution of the number of core symptoms of autistic catatonia in the study sample (n=99).

4.3.1 Scoring the ACQ:

The recommended primary scoring strategy for the ACQ is to focus on the six core symptoms associated with autistic catatonia and there is support from the present study for using a 0-4 Likert scale scoring for core autistic catatonia symptoms, including the frequency and severity of the presenting symptoms, provides a valid clinical screening tool for autistic catatonia. There is evidence that the computed variable *Core Autistic Catatonia Score* provides a scoring strategy which has the ability to sensitively identify individuals presenting with autistic catatonia. Further empirical investigation needs to be completed into which computed variables provide the most useful information, but initial analysis indicates that the *Core Autistic Catatonia Score* and the number of core items currently present may be of clinical utility.

4.3.1.1 Core symptom scoring:

The computed variables appertaining to the six core symptoms associated with autistic catatonia are *Core Autistic Catatonia Score, Core Severity Score* and *Core Frequency Score*. A significant difference in *Core Autistic Catatonia Score* was only found when the data was divided into groups on the basis of the proposed clinical cut-off point for autistic catatonia rather than by existing diagnosis. Similarly, individuals currently displaying three or more core symptoms of autistic catatonia had significantly higher *Core Severity Scores* but there was no significant difference found when using existing autistic catatonia diagnosis as a between-group variable. Significant between-group
differences in Core Frequency Score were found for both the catatonia diagnosis and no catatonia diagnosis groups.

Core Autistic Catatonia Score was normally distributed in the sample with a slight skew towards the lower end of the scale (Graph 5). Normal distributions were not found for either Core Severity Score (Graph 6) or Core Frequency Score (Graph 7). Inter-rater reliability of items rating the frequency and severity of symptoms is also required. Further investigation is required to determine if these supplementary questions concerning the frequency and severity of key motor symptoms connected to autistic catatonia can provide useful clinical information about the presentation of the condition.

The core symptom items were highly inter-correlated, and also highly correlated with both ACQ Motor Score and Core Autistic Catatonia Score (Table 6 in Appendix 15), whilst the Core Autistic Catatonia Score was also highly correlated with Total ACQ Score, ACQ Supplementary Score and the three subdomain scores (Table 6), possibly indicating the centrality of the previously identified core items.

The assessment of the internal validity of the core items on the ACQ thus indicated that these six items represent a useful screening tool for the symptoms of autistic catatonia. ROC analysis also indicated that Core Autistic Catatonia Score is a sensitive measure of autistic catatonia. Therefore, the core items on the ACQ measure are recommended as a working clinical screening measure for autistic catatonia. It is noteworthy that these six core autistic catatonia items are essentially similar to the criteria in Wing & Shah’s (2000) proposed diagnostic guidelines for autistic catatonia.
4.3.1.2 Scoring options comprising the total ACQ measure:

The current analysis provides support for including the supplementary items contained in the ACQ as highly significant correlations were observed between scores obtained from supplementary items and the computed variables connected to autistic catatonia presentation. Additionally, the distributions of the computed variables that contain information about all items on the ACQ appear normal. Therefore, it is proposed that the supplementary 22 items should remain as part of the broader ACQ measure, but further investigation will need to be completed to determine whether any of these items should be dropped from the ACQ.

Two variables were computed that included the supplementary items on the ACQ measure, namely the Total ACQ Score and ACQ Supplementary Score. A significant difference in Total ACQ Score was only found when the data was divided into groups on the basis of the proposed clinical cut-off point for autistic catatonia rather than by existing diagnosis. There were no statistically significant difference when using both between-group categorisation options for ACQ Supplementary Scores. These computed variables may be less useful than others computed from core symptom scores due to the wide ranging symptoms included in the ACQ measure and so Total ACQ Score and ACQ Supplementary Score may actually provide less information about the presentation of specific catatonic symptoms, or indeed autistic catatonia, in the sample. However, there is evidence that core item scores highly correlate with Total ACQ Score and ACQ Supplementary Score (Table 6), which indicates that autistic catatonia is probably associated with the computed variables containing supplementary items, although this could be a result of multicollinearity. Graph 9
indicates that the distribution of ACQ Supplementary Scores in the sample is normal which suggests that it is a meaningful computed variable.

The items on the ACQ can also be split into three subdomains; Motor Symptoms (n=15), Affective Alterations (n=5) and Behavioural Alterations (n=14). Each of these subdomain scores are strongly inter-correlated, with Pearson’s correlation values of 0.54 for Motor Symptom Score and Affective Alterations Score, 0.598 for Behavioural Alterations Score and Motor Symptom Score and 0.72 for Affective Alterations Score and Motor Symptom Score (n=87, p=0.001). The subdomain scores are also correlate with Core Autistic Catatonia Score (Motor Symptom Score r=0.83, n=87, p=0.001, Affective Alterations Score r=0.37, n=87, p=0.001, Behavioural Alterations Score r=0.51, n=87, p=0.001), which indicates that these subdomains represent meaningful groupings of symptoms. Note that items in the Affective Alterations subdomain have been altered after statistical analysis with the number of items reduced, which will affect the strength of correlations. Although it was beyond the scope of the current study to re-run statistical analysis for the amended 28 item ACQ measure, it can be assumed that this would further increase the correlations between variables and increase the evidence for the usefulness of including the supplementary items in the ACQ measure.

4.4 Additions to the theoretical understanding of autistic catatonia:

The current data indicate that catatonic-like symptoms are common in young people with ASD, which is in line with predictions from the putative models of autistic catatonia (Wing & Shah, 2006; Fink, Taylor & Ghaziuddin, 2006; Takota & Takata, 2007;
The data also supports the notion that previously reported variation in the presentation of autistic catatonia (Wing & Shah, 2000; Billstead et al., 2005; Dhossche, Shah & Wing, 2006; Neumarker, 2006) might in part be due to the absence of an empirically derived criteria. Moreover, the number of the six core symptoms presenting in individuals with an existing diagnosis of autistic catatonia varied with between one and six core symptoms (mean=3.1), which would support Wing & Shah’s (2000) clinical observations that the presentation of symptoms varies and few individuals display all of the possible commonly associated symptoms. In the present study, only two individuals with an existing diagnosis of autistic catatonia (10.0%) displayed all six core symptoms of autistic catatonia in the current study.

The number of core symptoms currently displayed by an individual appears to be a good indicator of autistic catatonia as statistical investigation identified that individuals with an existing diagnosis of autistic catatonia in the sample currently displayed significantly higher number of these core symptoms. A clear median split is evident in the number of core symptoms currently presented by participants (Graph 3), which indicates that catatonic symptoms present commonly in the sample and point towards autistic catatonia as spectrum condition. This generated a hypothesis that autistic catatonia may become a clinical syndrome when individuals display more than three core symptoms of autistic catatonia.

However, there is evidence from the present study that individuals in the study sample without an existing diagnosis of autistic catatonia also present with significant symptomatology; for example four individuals without an existing diagnosis of autistic catatonia were reported to display all six core symptoms (4.0%). On this basis, if
under-diagnosis of autistic catatonia is present in this sample, between group differences may be greater than they currently appear. The hypothesed clinical cut off point for the ACQ potentially permits further empirical investigation into theories of under-diagnosis of autistic catatonia. Despite evidence that autistic catatonia may become a clinical syndrome when three or more core symptoms of autistic catatonia are present, evidence from this sample suggests that individuals with a diagnosis of autistic catatonia typically display four or more of these core symptoms, which may suggest under diagnosis of autistic catatonia in the sample (see Tables 10 and 11 in Appendix 15).

The prevalence of autistic catatonia in the current study varied depending on the criteria used for diagnosis. Information about 20 individuals with an existing diagnosis of autistic catatonia (20.2%) is included in the data and 18 of these fully participated in the study (20.7%). Forty-two individuals in the sample (48.3%) displayed three or more of the six core symptoms of autistic catatonia (i.e. above the proposed clinical cut-off point) and 23 individuals (26.4%) displayed four or more core symptoms. Therefore, the potential prevalence of autistic catatonia in the current study sample may exceed the estimates in the literature (7% in Nordin & Gillberg, 1998; 6% in Wing & Shah, 2000; 11%-14% in Billstedt et al., 2005; 7% in Perisse et al., 2010) and support suggestions of under-diagnosis of autistic catatonia in young people with ASD (Dhossche, 2004; Wing, 2005; Schieveld, 2006). It could be argued that this was a result of bias in the sample during recruitment, as participants who cared for individuals presenting with autistic catatonia were more likely to participate in the study. However, autistic catatonia was not specified in the participation information used for recruitment and the research title indicated that the study was investigating
how common movement problems were in young people with ASD. Additionally, the majority of the sample did not have an existing diagnosis of autistic catatonia (79.8%).

There is no evidence for autistic catatonia remission in the study. Additionally, there is no evidence to indicate that older participants were more likely to present with autistic catatonia which could be assumed as age is linked to onset (Wing & Shah, 2000; Ohta et al., 2006). However, as previously stated, the distribution of the ages in the sample is non-typical which could affect the validity of this statistical investigation.

4.5 Summary of the assessment of a relationship to the RBQ and the GDS-CS:

Statistically significant relationships were found between scores on the ACQ and established clinical measures of depression (GDS-CS - Cuthill, Espie & Cooper, 2003) and repetitive and restricted behaviour (RBQ - Moss et al., 2009) appropriate for the study population.

Individuals in the sample who met the generated clinical cut-off criteria for autistic catatonia (i.e. who displayed more than three of the core symptoms of autistic catatonia) were found to score significantly higher on the GDS-CS measure of depression. Investigations into correlations between these variables indicates a significant linear association (see Appendix 20), which suggests that Core Autistic Catatonia Score and GDS-CS Score are intrinsically linked by a predictive correlation. It is unclear why individuals with ASD who are displaying catatonic symptoms are also scoring highly on a clinical measure of depression; it may be that these conditions are co-occurring or that the presentation of one of these conditions is being picked up by
the other measure. A number of the items on the two measures are duplicated or overlap; specifically reduced eating (ACQ item 27, GDS-CS item 11), reduced communication or muteness (ACQ item 24, GDS-CS item 5), increased aggression (ACQ item 18, GDS-CS item 2), avoidance of contact with others (ACQ item 16, GDS-CS item 3), decreased personal hygiene and/or concern about appearance (ACQ item 31, GDS-CS item 4), crying (ACQ item 17, GDS-CS item 6), reduced engagement in preferred activities (ACQ item 21, GDS-CS item 8), requiring more encouragement (ACQ item 22, GDS-CS item 10) and sleep problems (ACQ item 26, GDS-CS item 12). As both measures are third-party ratings of observable behaviours, it may be that individuals are presenting with either autistic catatonia or depression, but both clinical measures are interpreting this symptom as an indication of the presentation of that condition without being sensitive enough to determine aetiology; for example reduced eating could be due to inability to execute motor action in the body or reduced appetite as a result of depression, but both causes would present identically and be scored on a third party measure of autistic catatonia and third party measure of depression. Further investigation is required to fully understand the evident relationship between the ACQ and the GDS-CS.

Statistical analysis also indicated a link between scores on a clinical measure of repetitive and restricted behaviours and the ACQ. Individuals in the sample who met both criteria for autistic catatonia (i.e. existing diagnosis and proposed clinical cut-off) displayed significantly increased RBQ Scores. A significant linear association between these variables was also found (see Appendix 20), which suggests that Core Autistic Catatonia Score and RBQ Total Score are intrinsically linked by a predictive correlation. Further statistical investigation of RBQ subdomain scores indicates that individuals
with an existing diagnosis of autistic catatonia had significantly higher repetitive speech subdomain scores (p<0.05) and non-significant between-group differences were found for the other four subdomain scores. Examination based on the clinical cut-off point for autistic catatonia suggests between-group differences in the compulsive behaviour subdomain (p<0.05), restricted behaviour subdomain (p<0.05), insistence on sameness subdomain (p<0.01) and repetitive speech subdomain (p<0.01). Significant correlations were also found between all five individual subdomain score and Core Autistic Catatonia Score (p<0.01). Again, some duplication of items is present on both clinical measures; specifically body stereotopy (ACQ item 8, RBQ item 2) and hand stereotopy (ACQ item 13, RBQ item 3). However, the minimal duplication between these clinical measures indicates an alternative hypothesis to explain this correlation.

The association demonstrated between the presentation of catatonic symptoms in individuals with ASD via the ACQ and repetitive and restricted behaviours via the RBQ may prove to be important to conceptualising and understanding autistic catatonia. If these variables prove to be predictive in nature as suggested by regression analysis, this could be key to unravelling the phenomenology of autistic catatonia. Additionally, this association has the potential to allow the identification of a sub-group of individuals with ASD who are more likely to develop autistic catatonia via risk markers, which has previously been elusive. Alternatively, it is possible that both measures are associated with each other due to a third factor which is independently associated with both variables, potentially the autistic spectrum itself.
Movement abnormalities are clearly a feature of ASD and it may be that autistic catatonia is part of the ASD phenotype rather than being a co-morbid condition. The current study indicates that autistic catatonia is a spectrum condition and it is possible that this reflects the ASD spectrum; the ACQ and the RBQ may be both measuring the repetitive and restricted behaviours, activities and interests subdomain in the DSM-V dyadic ASD criteria (APA, 2013). Further investigation into the association between the ACQ and the RBQ is required to resolve this, along with an assessment of whether other core features of ASD such as social and communication deficits correlate with the presentation of catatonic features in young people with ASD.

4.6 Limitations of the current study:

The current study has inherent associated limitations, due to both the design and execution of the research. Study design limitations include that the relevance of literature encompassed in the literature base was judged solely on the inclusion of key words in the title or abstract of published articles. Therefore, it is possible that some relevant articles have not been included in the literature review. However, this is unlikely due to standard publishing practice of including key words prominently in the abstract and/or title. The research design relies on the retrospective reporting of current symptoms with previous presentation, and so accuracy in terms of true frequency and severity comparison may be an issue resulting in reporting bias. However, the main aims of the study are concerned with current presentation of symptoms so this is a minor issue. Additionally the online nature of the study survey results in no direct contact between the researchers and the participants. Although this allows anonymity for participants, it prevents a check of individual participants
against the inclusion criteria for the study. The insertion of pre-participation questions (Appendix 10) which included confirmation about inclusion criteria removes the potential for participant error before completion of the online questionnaire. Although the data distribution was generally acceptable, the distribution of participants age in the sample was non-typical, with some age groups being very under-represented in the study, which could affect the validity of data analysis relating to age. The reliability of the ACQ was not assessed as part of this study, which impacts on the case for the clinical usefulness of the measure and will need to be examined in a future study. There was also no assessment of test-retest validity of the ACQ, although this can be assumed due to the third party observational nature of the measure.

Recruitment to the study was completed over a number of months and targeted a wide range of organisations and support groups, yet this small-scale study involved a relatively small sample size of 99 participants, with only 20 individuals with an existing diagnosis of autistic catatonia. This can partly be attributed to a lack of awareness about autistic catatonia, poor diagnostic rates and a lack of organisations providing support and information about the condition. However, the sample size and small number of individuals with an existing diagnosis of autistic catatonia resulted in unbalanced groupings for between group analyses which will have impacted on the validity of the results. Alpha corrections for statistical tests were also not completed throughout the analyses. A factor analysis was not completed to analyse the results, although this would have been appropriate, as the data analysis was influenced by the methods used in the development of the Manchester Attachment Scale – Third Party Observational Measure (Penketh et al., 2014). Caution is recommended in the
interpretation of the findings of the current exploratory study as a result of these statistical limitations.

There are also limitations relating to the execution of the study. This research was undertaken by a neophyte researcher as part of a postgraduate research degree without the support of an extensive research team. Cost implications due to an inability to access funding to support this study has further impacted on this research; specifically accessing two journal article of interest that were unavailable in the University of Manchester Library (Barnhill J (2012): 'Catatonia and Autism Spectrum Disorders', Journal of Neuropsychiatry and Clinical Neurosciences, 24(2):4); Dijkxhoorn Y (2012): ‘Autism and Catatonia’ IASSIDD Conference Systems, 2012 IASSID World Congress) nor information received from the author when requested via email. Another article identified as relevant was excluded due to it being published in Dutch and no funds were available for a translation service (Sienaert P (2012): 'The ethical duty to treat children and adolescents with autism and catatonia', Tijdschrift voor Psychiatrie, 54:6). Although interesting, these articles were judged to be non-essential so access was not pursued and so they were excluded from the literature review. Additionally, inability to access financial support for the study also impacted recruitment for the study as it limited the ability of the researcher to effectively access a wide range of potential participants by accepting requests to visit services and pass on information or ask questions about the study. Participation in the research relied on computer literacy and access to the internet, so this also biases the cohort somewhat, although this is unlikely to impact on the suitability of the population sample.
4.7 Implications of the current study on future research:

This research project has multi-factorial implications for future research in the autistic catatonia field and provides important insight into the condition. By collecting an empirically valid data set reporting on the presentation of symptoms connected to autistic catatonia, the current study represents the first systematic attempt to assess the prevalence and presentation of observable catatonic symptoms in young people with ASD. Therefore, this study has provided some clarity to the presentation and variation of catatonic symptoms in a sample of young people with ASD which can be used by future researchers to investigate autistic catatonia.

The empirical assessment of the presentation of symptoms connected to autistic catatonia in the current study represent unique investigative enquiry into the descriptive accounts of autistic catatonia in the literature and provides empirical evidence to support theory. This will allow future researchers to clearly define autistic catatonia, empirically examine the accurate prevalence rate of the condition, inform epidemiological studies, complete investigations into the course of the condition and inform evidence-based treatment intervention. This study also provides information which will enable researchers to investigate potential risk markers of autistic catatonia enabling earlier identification and treatment, which have been shown to be crucial for positive outcomes (Carrol et al., 2008; Heckers et al., 2010). Investigations into the impact of age on the development of autistic catatonia needs to be completed using a more appropriately distributed data set as adolescence is commonly linked to onset (Wing & Shah, 2000; Ohta et al., 2006). Further investigation needs to be completed into potential gender differences as the current study sample contained significantly
more females with an existing diagnosis of autistic catatonia whereas there has been other indications of male dominance (Wing & Shah, 2000), to establish whether autistic catatonia represents a difference in manifestation or phenotype of ASD in terms of gender.

Participants in this study were not asked to report on level of severity of participant learning disability, expressive language impairment or ASD social interaction subgroup classification, which have all been identified as potentially increasing risk of autistic catatonia development (Wing & Shah, 2000) and so need further empirical investigation and comparison with ACQ Scores. There was also no assessment of onset of symptoms in the current study, including the speed of onset of catatonic symptoms and precipitating factors such as stress, which have been highlighted as potentially important (Shah & Wing, 2000; Ghaziuddin et al., 2005; Shah & Wing, 2006, Stoppelbein et al., 2006).

As already mentioned, it is possible that the ACQ is actually measuring the continuum of ASD and that high ACQ scores reflect individuals with extreme manifestations of ASD. As the new diagnostic criteria for ASD reflects a dyadic model comprising a restricted and repetitive behaviours, interests and activities domain and a social communication domain (DSM-V, APA, 2013), this would explain the demonstrated association between ACQ scores and RBQ scores. Further investigation is required into whether ACQ scores correlate positively with measures of social and communication deficits; if this is found to be the case, it would indicate that the ACQ may be measuring the autistic spectrum and that autistic catatonia is linked to an extreme manifestation of ASD as suggested by Hare and Malone (2004). Investigations into
executive functioning deficits linked to theories of ASD as a fractionable triad (Happé, Ronald & Plomin, 2006; Happé & Ronald, 2008; Williams & Bowler, 2014) would also indicate whether autistic catatonia may be part of the ASD phenotype.

The ACQ has the capability to assess the course of autistic catatonia and to empirically measure the effectiveness of treatment interventions over time via the attributes contained on the measure. Therefore, future research can fully explore the benefits of different treatment options and develop evidence-based best practice guidelines for clinicians. This is particularly important due to high-risk interventions available to treat autistic catatonia, including ECT and extensive pharmacological prescription. Prognosis of the condition can also be investigated by using the ACQ to assess the course of autistic catatonia in longitudinal research studies.

The ACQ is a useful clinical tool to support investigation into autistic catatonia and can be used to further investigate the symptoms associated with autistic catatonia to enable a clear clinical definition to be developed. Focus on the six core symptoms of autistic catatonia is recommended for empirical enquiry, including information relating to the frequency and severity of these symptoms. Focussed lines of enquiry into supplementary items, which have been shown to contain useful information via statistical analysis, is also recommended to investigate whether these provide additional information about the presentation of autistic catatonia, either individually or via computed variables. Individual supplementary items may provide information about vulnerability to autistic catatonia or information relating to the presentation of autistic catatonia which should be included in diagnostic criteria. The internal properties of the ACQ need to be further assessed using factor analysis to further
explore the validity of the subdomains and whether further redundant items could be dropped from the measure, along with investigations into reliability (including test-retest and inter-rater reliability). It is recommended that wherever possible all participants in future studies are clinically assessed for autistic catatonia to ensure valid between-group statistical comparisons.

4.8 Implications of the current study on future clinical practice

The current research study also has numerous potential benefits for future clinical practice. It is hoped that the current empirical investigation of autistic catatonia will raise the profile of the condition and result in higher rates of clinical recognition, as this would have a direct impact on patients. Future research enabled by the current study could also have significant clinical impact, for example via the development of evidence-based treatment algorithms for autistic catatonia and best practice guidelines to inform clinicians.

The six core items of the ACQ is recommended as a clinical screening tool for autistic catatonia, which can be used by clinicians to quickly assess individuals for the potential presence of the condition. This short clinical assessment tool could be used in clinical practice to screen for the presence of autistic catatonia and enable prompt identification of the condition to allow patients speedy access to clinical intervention, which has been demonstrated as important for positive outcome (Dhossche et al., 2006, Dhossche, 2009). It could be recommended that the ACQ clinical screening tool could be routinely administered as part of clinical practice to adolescents with ASD to assess for autistic catatonia onset. This study also has the potential to enable clinicians
to monitor the effectiveness of treatment interventions in individuals with autistic catatonia using the ACQ.

In line with the approach in DSM-V (APA, 2013), the current research may also have clinical utility in enabling ASD to be measured (rather than simply diagnosed) if autistic catatonia is demonstrated to be part of the phenotype of ASD. This would enable clinicians to obtain more specific information about the presentation of ASD in each individual and provide more tailored intervention by informing decisions about appropriate level of care support required.

4.9 Conclusions

The current study represented a unique quantitative assessment of the prevalence and presentation of catatonic symptoms in young people with ASD and provided information about autistic catatonia which adds considerably to current understanding about the condition. The focus in the current study was on the phenomenology of autistic catatonia but the results could by future researchers to explore the nosology, epidemiology, aetiology and course of the condition.

A clinical measure for autistic catatonia was developed as part of this study, and preliminary investigations into the validity and internal properties of the Autistic Catatonia Questionnaire (ACQ) are promising. The systematic method used to design this third-party clinical measure ensured that the ACQ includes all observable catatonic symptoms connected to autistic catatonia and the development of the ACQ should obviate the need to ‘import’ other assessment tools for use with this population. The
ACQ is quick to administer and score, requiring minimal instruction and no training to do so, which increases its clinical and research usefulness. This research study suggests that the ACQ could be used as an effective tool for the assessment of autistic catatonia, research purposes and to monitor the course of the condition and the effectiveness of treatment interventions.

The onset of autistic catatonia can have devastating consequences for quality of life and significant implications for the level of care that individuals require. Prompt identification has been linked to positive outcomes yet autistic catatonia appears to be poorly identified and awareness of the condition is low. It is crucial that future researchers further investigate autistic catatonia empirically and that diagnostic criteria are finalised to allow the progression of theoretical understanding, enable the development of safe and effective evidence-based treatments and raise the profile of autistic catatonia in mainstream ASD research.
REFERENCES:


Carpenter L (2012): “Mummy, I’m not a naughty girl”. Sunday Times, 7/4. Available at: http://www.thetimes.co.uk/tto/magazine/article3369688.ece


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Mahendra B (1981): “Where have all the catatonics gone?”, Psychological Medicine, 11:669-671


National Autistic Society Website (2010): “Proposed changes to autism and Aspergers Syndrome diagnostic criteria” [online]. Available at:


APPENDICIES:

Appendix 1 - Current DSM and ICD classification entries for ASD


1. Persistent deficits in social communication and social interaction:
   - Deficits in social-emotional reciprocity
   - Deficits in non-verbal communicative behaviours used for social interaction
   - Deficits in developing, maintaining and understanding relationships

2. Restricted, repetitive patterns of behaviour, interests or activities:
   - Stereotypes or repetitive motor movements
   - Insistence on sameness
   - Highly restricted, fixated interests that are abnormal in intensity or focus
   - Hyper- or hypo-reactivity to sensory input or unusual interest in sensory aspects of the environment.


(a) Qualitative impairment in reciprocal social interaction, three from the following five areas:
   - failure to use eye gaze, body posture, facial expression and gesture to regulate interaction adequately;
a failure to develop (in a manner appropriate to mental age, and despite ample opportunity) peer relationships that involve a mutual sharing of interests, activities and emotions;

rarely seeking and using other people for comfort and affection at times of stress or distress and/or offering comfort and affection to others when they are showing distress or unhappiness;

a lack of shared enjoyment in terms of vicarious pleasures in other people's happiness and/or a spontaneous seeking to share their own enjoyment through joint involvement with others;

a lack of socio-emotional reciprocity, as shown by an impaired or deviant response to communicative behaviours;

(b) Qualitative impairments in communication, two from the following five areas

- a delay in, or total lack of, spoken language that is not accompanied by an attempt to compensate through the use of gesture or mime as alternative modes of communication;
- a relative failure to initiate or sustain conversational interchange (at whatever level of language skills is present) in which there is a reciprocal to and fro responsiveness to the communication of the other person;
- stereotyped and repetitive use of language and/or idiosyncratic use of words or phrases;
- abnormalities of pitch, stress, rate, rhythm and intonation of speech;
- a lack of varied spontaneous make-believe play, or when young, social imitative play.

(c) Restricted repetitive and stereotyped patterns of behaviour, interests and activities, two from the following six areas

- an encompassing preoccupation with stereotyped and restricted patterns of interest;
- specific attachments to unusual objects;
- apparently compulsive adherence to specific, non-functional routines and rituals;
- stereotyped and repetitive motor mannerisms that involve either hand/finger
- flapping or twisting or complex whole body movements;
- preoccupation with part-objects or non-functional elements of play materials (such as odour, the feel of their surface, or the noise/vibration that they generate);
- distress over changes in small, non-functional details of their environment.

(d) Developmental abnormalities must be present in the first three years for the diagnosis to be made

(e) Clinical picture is not attributable to other varieties of pervasive developmental disorder, specific developmental disorders of receptive language with secondary socio-emotional problems; reactive attachment disorder or disinhibited attachment disorder, mental retardation with some associated emotional/behavioural disorder, schizophrenia of unusually early onset; and Rett syndrome.
Appendix 2 - University of Manchester Committee on the Ethics of Research on Human Beings approval letter

Miss. Breen  
School of Psychological Sciences  
20th November 2012

Dear Miss. Breen

Research Ethics Committee 3  

Breen, Hare: An investigation into how common 'catatonic' symptoms are in children and young people with ASD (ref 12260)  

I write to confirm that the Chair is now satisfied that you have addressed the concerns of the Ethics Committee of the 24th of October 2012 and has therefore given the above research project a favourable ethical opinion.  

This approval is effective for a period of five years and if the project continues beyond that period it must be submitted for review. It is the Committee's practice to warn investigators that they should not depart from the agreed protocol without seeking the approval of the Committee, as any significant deviation could invalidate the insurance arrangements and constitute research misconduct. We also ask that any information sheet should carry a University logo or other indication of where it came from, and that, in accordance with University policy, any data carrying personal identifiers must be encrypted when not held on a university computer or kept as a hard copy in a location which is accessible only to those involved with the research. Finally, I would be grateful if you could complete and return the attached form at the end of the project or by October 2013.

We hope the research goes well.  

Yours sincerely

Adrian Jarvis  
Ethics Committee 3 Secretary
Appendix 3 – Illustration of the symptoms reported to be connected to autistic catatonia in the literature (Table 1)

<table>
<thead>
<tr>
<th>Symptom</th>
<th>Number of times reported in literature:</th>
<th>Included in the measure?</th>
</tr>
</thead>
<tbody>
<tr>
<td>‘Freezing’/very still like a statue</td>
<td>6</td>
<td>Yes</td>
</tr>
<tr>
<td>Difficulty initiating actions/‘stuckness’/akinesia</td>
<td>6</td>
<td>Yes</td>
</tr>
<tr>
<td>Problems stopping actions once started</td>
<td>3</td>
<td>Yes</td>
</tr>
<tr>
<td>Difficulty initiating movement</td>
<td>5</td>
<td>Yes</td>
</tr>
<tr>
<td>Slowness in movement</td>
<td>5</td>
<td>Yes</td>
</tr>
<tr>
<td>Requires prompts to complete actions</td>
<td>4</td>
<td>Yes</td>
</tr>
<tr>
<td>Waxy flexibility</td>
<td>3</td>
<td>Yes</td>
</tr>
<tr>
<td>Repetitive body movements</td>
<td>4</td>
<td>Yes</td>
</tr>
<tr>
<td>Stiff posturing</td>
<td>9</td>
<td>Yes</td>
</tr>
<tr>
<td>Noticeable resting tremor</td>
<td>2</td>
<td>Yes</td>
</tr>
<tr>
<td>Increased motor tics</td>
<td>3</td>
<td>Yes</td>
</tr>
<tr>
<td>Waving or shaking extremities</td>
<td>2</td>
<td>Yes</td>
</tr>
<tr>
<td>Twisting or flicking hands in front of eyes</td>
<td>2</td>
<td>Yes</td>
</tr>
<tr>
<td>Moving in a jerky way</td>
<td>2</td>
<td>Yes</td>
</tr>
<tr>
<td>Impulsive/excitable phases</td>
<td>4</td>
<td>Yes</td>
</tr>
<tr>
<td>Withdrawal from physical contact</td>
<td>2</td>
<td>Yes</td>
</tr>
<tr>
<td>Spontaneous crying, laughing or screaming</td>
<td>4</td>
<td>Yes</td>
</tr>
<tr>
<td>Episodes of aggression</td>
<td>6</td>
<td>Yes</td>
</tr>
<tr>
<td>Difficulty passing through doorways</td>
<td>2</td>
<td>Yes</td>
</tr>
<tr>
<td>Difficulty crossing lines on the floor</td>
<td>2</td>
<td>Yes</td>
</tr>
<tr>
<td>Reduced enjoyment in preferred activities</td>
<td>4</td>
<td>Yes</td>
</tr>
<tr>
<td>Condition</td>
<td>Frequency</td>
<td>Present</td>
</tr>
<tr>
<td>-----------------------------------------------</td>
<td>-----------</td>
<td>---------</td>
</tr>
<tr>
<td>Requiring more encouragement to engage</td>
<td>4</td>
<td>Yes</td>
</tr>
<tr>
<td>Unusual gait/posture</td>
<td>5</td>
<td>Yes</td>
</tr>
<tr>
<td>Reduced communication/muteness</td>
<td>10</td>
<td>Yes</td>
</tr>
<tr>
<td>Incontinence</td>
<td>4</td>
<td>Yes</td>
</tr>
<tr>
<td>Sleep problems</td>
<td>4</td>
<td>Yes</td>
</tr>
<tr>
<td>Reduced eating</td>
<td>6</td>
<td>Yes</td>
</tr>
<tr>
<td>Eye rolling/ unusual eye movements</td>
<td>5</td>
<td>Yes</td>
</tr>
<tr>
<td>Unusual facial expressions/‘grimaces’</td>
<td>5</td>
<td>Yes</td>
</tr>
<tr>
<td>Ignoring instructions</td>
<td>2</td>
<td>Yes</td>
</tr>
<tr>
<td>Refusal to bathe or change clothes</td>
<td>2</td>
<td>Yes</td>
</tr>
<tr>
<td>Occasional groans or unusual noises</td>
<td>3</td>
<td>Yes</td>
</tr>
<tr>
<td>Staring into space/fixed gaze</td>
<td>5</td>
<td>Yes</td>
</tr>
<tr>
<td>Unable to lift head</td>
<td>2</td>
<td>Yes</td>
</tr>
<tr>
<td>Echolalia</td>
<td>2</td>
<td>No – common feature of ASD</td>
</tr>
<tr>
<td>Finger tapping</td>
<td>1</td>
<td>No – reported once</td>
</tr>
<tr>
<td>Diaphoresis</td>
<td>1</td>
<td>No – reported once</td>
</tr>
<tr>
<td>Withdrawal</td>
<td>1</td>
<td>No – reported once</td>
</tr>
<tr>
<td>Auditory hallucinations</td>
<td>1</td>
<td>No – reported once/speculative</td>
</tr>
<tr>
<td>Auditory hypersensitivity</td>
<td>1</td>
<td>No – reported once/speculative</td>
</tr>
<tr>
<td>Depressed</td>
<td>5</td>
<td>No - too vague or speculative</td>
</tr>
<tr>
<td>Anxious</td>
<td>2</td>
<td>No - too vague or speculative</td>
</tr>
</tbody>
</table>


These next 19 questions will examine different behaviours. Each behaviour comes with a brief definition to give a greater understanding for the behaviours and symptoms we are looking for.

Please circle the appropriate number in the scale to indicate frequency of these behaviours. If a behaviour does not apply for any specific reason please circle 0.

<table>
<thead>
<tr>
<th>Behaviour</th>
<th>Scale</th>
<th>Frequency</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Never</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td><strong>Once a month</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td><strong>Once a week</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td><strong>Once a day</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td><strong>More than once a day</strong></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

1. **Object stereotypy**: repetitive, seemingly purposeless movement of objects in an unusual way. 
   *E.g. twirling or twiddling objects, twisting or shaking objects, banging or slapping objects.*
   | 0 | 1 | 2 | 3 | 4 |

2. **Body stereotypy**: repetitive, seemingly purposeless movement of the whole body (other than hands) in an unusual way. *E.g., body rocking or swaying, or spinning, bouncing, head shaking, body posturing. (Does not include self-injurious behaviour)*
   | 0 | 1 | 2 | 3 | 4 |

3. **Hand stereotypy**: repetitive, seemingly purposeless movement of hands in an unusual way.
   *E.g. finger twiddling, hand flapping, wigging or flicking fingers, hand posturing. (Does not include self-injurious behaviour).*
   | 0 | 1 | 2 | 3 | 4 |

4. **Cleaning**: Excessive cleaning, washing or polishing of objects or parts of the body. *E.g. polishing windows and surfaces excessively, washes hands and face excessively.*
<p>| 0 | 1 | 2 | 3 | 4 |</p>
<table>
<thead>
<tr>
<th></th>
<th></th>
<th></th>
<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>5. <strong>Tidying up:</strong> Tidying away any objects that have been left out. This may occur in situations when it is inappropriate to put the objects away. Objects may be put away into inappropriate places. <em>E.g. putting cutlery left out for dinner in the bin, removes all objects from surfaces.</em></td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>3</td>
</tr>
<tr>
<td>6. <strong>Hoarding:</strong> Collecting, storing or hiding objects to excess, including rubbish, bits of paper, and pieces of string or any other unusual items</td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>3</td>
</tr>
<tr>
<td>7. <strong>Organising objects:</strong> Organising objects into categories according to various characteristics such as colour, size or function. <em>E.g. ordering magazines according to size, ordering toy cars according to colour, ordering books according to topic.</em></td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>3</td>
</tr>
<tr>
<td>8. <strong>Attachment to particular people:</strong> Continually asking to see, speak or contact a particular ‘favourite’ person. <em>E.g. continually asks to see or speak to particular friend, carer, babysitter or school teacher.</em></td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>3</td>
</tr>
<tr>
<td>9. <strong>Repetitive questions:</strong> Asking specific questions over and over again. <em>E.g. always asking people what their favourite colour is, asking who is taking them to school the next day over and over.</em></td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>3</td>
</tr>
<tr>
<td>10. <strong>Attachment to objects:</strong> Strong preferences for a particular objects to be present at all times e.g. carrying a particular piece of string everywhere, taking particular red toy car everywhere, attachment to soft toy or particular blanket.</td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>3</td>
</tr>
<tr>
<td>11. <strong>Repetitive phrases/signing:</strong> Repeating particular sounds, phrases or signs that are unrelated to the situation over and over. <em>E.g. repeatedly signing the word ‘telephone’.</em></td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>3</td>
</tr>
</tbody>
</table>
12. **Rituals**: Carrying out a sequence of unusual or bizarre actions before, during or after a task. The sequence will always be carried out when performing this task and will always occur in the same way. *E.g.* turning round three times before sitting down, turning lights on and off twice before leaving the room, tapping door frame twice before passing through it.

13. **Restricted conversation**: Repeatedly talks about specific, unusual topics in great detail. *E.g.*, conversation restricted to trains, buses, dinosaurs, particular films, country, or sport.

14. **Echolalia**: Repetition of speech that has either just been heard or has been heard more than a minute earlier. *E.g.* Mum: 'Jack don't do that', Jack: 'Jack don't do that'.
Appendix 5 – The Carer Supplement to the Glasgow Depression Scale for people with Learning Disability (GDS-CS)

APPENDIX 2

Carer Supplement to the Glasgow Depression Scale for people with a Learning Disability (GDS-CS)

What is the name of the person you look after? ________________________________
(referred to as X in the following questions)
What is your relationship to X? ________________________________
The following questions ask about how you think X has been in the last week. There is no right or wrong answer. Please circle the answer you feel best describes X in the last week.

In the last week . . .

<table>
<thead>
<tr>
<th></th>
<th>Never/</th>
<th>Sometimes/</th>
<th>Always/</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Has X appeared depressed?</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
<td>2</td>
<td>Has X been more physically or verbally aggressive than usual?</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
<td>3</td>
<td>Has X avoided company or social contact?</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
<td>4</td>
<td>Has X looked after his/her appearance?</td>
<td>2</td>
<td>0</td>
</tr>
<tr>
<td>5</td>
<td>Has X spoken or communicated as much as he/she used to?</td>
<td>2</td>
<td>0</td>
</tr>
<tr>
<td>6</td>
<td>Has X cried?</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
<td>7</td>
<td>Has X complained of headaches or other aches and pains?</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
<td>8</td>
<td>Has X still taken part in activities which used to interest him/her?</td>
<td>2</td>
<td>0</td>
</tr>
<tr>
<td>9</td>
<td>Has X appeared restless or irritable?</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
<td>10</td>
<td>Has X appeared lethargic or sluggish?</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
<td>11</td>
<td>Has X eaten too little/too much?</td>
<td>0</td>
<td>1</td>
</tr>
</tbody>
</table>

If no problem, score 0. (A positive answer to either question means it should be scored. Please tick which response is relevant, beside the question.)

12 Has X found it hard to get a good night’s sleep? Please also tick which one of the following options is relevant.
   Has X had difficulty falling asleep when going to bed at night? ☐
   Has X been waking in the middle of the night and finding it hard to get back to sleep again? ☐
   Has X been waking very early in the morning and finding it hard to get back to sleep? ☐

13 Has X been sleeping during the day? | 0           | 1        | 2        |
14 Has X said that he/she does not want to go on living? | 0           | 1        | 2        |
15 Has X asked you for reassurance? | 0           | 1        | 2        |
16 Have you noticed any change in X recently? Please explain what changes you have noticed, in either mood or behaviour. | 0           | 1        | 2        |

Thank you for answering these questions.
Appendix 6 – Initial contact letter about the study sent to specialist care providers, parent support groups and charities targeted for recruitment

Dear Sir/Madam,

I am a postgraduate student at the University of Manchester who is completing a research project investigating autistic catatonia, supervised by Dr Dougal Julian Hare (Senior Lecturer in Clinical Psychology). We are attempting to investigate the prevalence of catatonic behaviours in children and young people with Autistic Spectrum Disorder.

It is estimated that 6-8% of people with autism develop autistic catatonia, often during adolescence, but it is thought that it is often missed by clinicians, except in the most severe cases. Autistic catatonia is a very under-researched area and little is known about the presentation and variation of symptoms. My research aims to find out how common movement difficulties are in young people with a diagnosis of autism spectrum disorder (ASD).

The development of autistic catatonia has a serious impact on an individual’s quality of life and the level of care they require. Commonly individuals experience increased slowness in movement, difficulty initiating gross motor actions and reduced expressive communication (along with a range of other possible symptoms), which results in a marked loss of skills in many areas (particularly independence skills) and an increased reliance on others.

Although there are numerous descriptions of the effects of autistic catatonia in the literature, the number and extent of symptoms are reported to vary widely with few individuals presenting with all of the possible symptoms. Different studies have also defined autistic catatonia very differently, which results in inconsistencies in research literature.
This study will aim to more accurately define autistic catatonia and increase understanding about the symptoms that are part of the condition. This is important as a lot of ambiguity and speculation exists about which problems the individual is experiencing are part of autistic catatonia and which are attributable to other things. An assessment tool would provide clinicians with a measure that could be used to easily and accurately diagnose autistic catatonia, as well as enabling more accurate research to be completed in the future.

At the moment, I am in the process of a University Ethics Procedure so data collection is envisaged to start within the next few months. Participation in this study will involve someone who knows an individual aged 12-25 and with a diagnosis of autism spectrum disorder well (i.e. a staff member or parent) completing an online questionnaire reporting on symptoms connected to autistic catatonia. This should take approximately 30-40 minutes, and can be completed online at a time that is convenient for the participant. All data will be anonymous and processed and stored with the highest confidentiality. Please note that the presence of ‘autistic catatonia’ is not needed for participation in the study, only a diagnosis of autism spectrum disorder.

I would be very interested in including your service in my research if possible. If you have any questions at all or would like to arrange a meeting to discuss the project, please do not hesitate to contact either myself or Dr Hare by email (jennifer.breen@postgrad.manchester.ac.uk or dougal.hare@manchester.ac.uk) or telephone (0161 306 0400). If I do not hear from you, I will contact you again in a few weeks to discuss this project further with you.

Kind regards,

Jennifer Breen

Postgraduate Student, University of Manchester
Appendix 7 – Consent form for Care Providers (v1.0)

Consent form for Care Providers – v1.0

Title: An investigation into how common ‘catatonic’ symptoms are in children and young people with ASD.

Researcher: Jenny Breen (jennifer.breen@postgrad.manchester.ac.uk)

Please initial all boxes and sign below:

I confirm that I have read the participant information (v1.0) for the above study and have had the opportunity to answer questions. All my questions have been answered satisfactorily.

I understand that our service’s participation in this study is voluntary and can be withdrawn at any time without consequence or having to give a reason.

I agree for my service to take part in the above study

Signed: ..........................  Print name: ..........................  Date: ..........................

(Manager of service)

Signed: ..........................  Print name: ..........................  Date: ..........................

(Researcher)
Appendix 8 - Participant Information Sheet (v1.0)

Participant Information Sheet – v1.0

**Title:** An investigation into how common ‘catatonic’ symptoms are in children and young people with ASD.

**Researcher:** Jenny Breen (jennifer.breen@postgrad.manchester.ac.uk)

**Summary of the study:**

This research project is trying to find out more about how common ‘catatonic’ symptoms are in young people with autism spectrum disorder (ASD). There is evidence that a small number of young people with autism spectrum disorder (ASD) develop ‘Autistic Catatonia’ when they are teenagers. Usually, individuals with autistic catatonia find it difficult to move and communicate with others. Little is known about the symptoms and behaviours associated with autistic catatonia.

This research project will investigate how common symptoms thought to be part of ‘autistic catatonia’ are in children and young people with autism spectrum disorder (ASD). At the moment, it is not known how many young people with autism spectrum disorder (ASD) experience movement difficulties (or ‘catatonic symptoms’ as they are often called).

Some people may experience this but it is not picked up by doctors. It is hoped that this study will help us understand more about catatonic symptoms in young people with autism spectrum disorder (ASD) so that more effective care and treatments can be developed.

Not everyone involved in this study needs to have movement difficulties or ‘autistic catatonia’ but they do need to have a diagnosis of autism spectrum disorder (ASD).
hy am I being asked to take part in this study?

You are being invited to take part in this research because you care for a young person with autism spectrum disorder (ASD). This research is looking to gather information about symptoms and behaviours present for young people with autism spectrum disorder (ASD) aged 12 to 25 years old.

What will I have to do if I take part?

Taking part involves completing an online questionnaire answering questions about whether the person you care about displays certain ‘catatonic’ symptoms. This is to find out how common movement difficulties are in young people with autism spectrum disorder (ASD).

You will also be asked to complete two short well-known questionnaires (the Glasgow Scale and the Repetitive Behaviour Questionnaire). The Repetitive Behaviour Questionnaire measures how often a person chooses to do restricted or repetitive activities such as rocking or lining up objects. The Glasgow Scale screens for depression in people with learning disabilities and/or autism spectrum disorder (ASD) as this could be a reason why people do less (rather than because they have movement problems).

Participation in this study should take between 30 and 40 minutes. This is the only thing you will be asked to do as part of this study.

How do I take part?

The questionnaire is completed online so you can take part wherever and whenever it is convenient for you. The link to the questionnaire is: https://apps.mhs.manchester.ac.uk/surveys//TakeSurvey.aspx?SurveyID=m4LH8855

Will I have to give any personally identifiable information in this study?

No. Taking part in this study is anonymous. We do not need any personally identifiable information about you or the young person with autism spectrum disorder (ASD) you are answering questions about. We only ask for some basic demographic information (such as the age of the person you care for, how long you have known them for etc) to check that all the information collected is about young people with autism spectrum
disorder (ASD) and the questions have been answered by someone who knows them well. We will not ask for names, addresses, telephone numbers etc.

**Do I have to take part in the study?**

No. Whether you take part is totally up to you. Whether you take part or not will not affect any care or treatment that you or the person you care for receive. Nobody will know if you took part or not as the study is entirely anonymous.

**Can I withdraw from the study if I decide I no longer want to take part?**

You can exit the questionnaire at any time while you are completing it online and the data that you have input will not be used as part of the study. However, as the questionnaire is anonymous we are not able to delete information after you have finished and submitted the questionnaire as we will not be able to identify which responses are yours.

**Is taking part in the study confidential?**

Yes. You do not need to tell anyone that you have taken part if you do not want to. We will not know who has taken part either as all the information collected is anonymous. We store all information securely on password protected computers and in locked offices. Information security is very important to us and we will take all steps possible to keep the data secure.

**What are the benefits and risks associated with taking part in the study?**

There are no risks associated with taking part and you will not benefit directly from being involved. However, it is hoped that this study will improve understanding of autistic catatonia and improve treatment for people with ASD who develop autistic catatonia in the future.

**Who is running the study?**

This study is being completed as part of a MPhil postgraduate degree at the University of Manchester. Dr Dougal Hare is supervising the researcher, Jenny Breen, to complete the study. The study has been ethically reviewed by the University of
Manchester Ethics Research Committee. Appropriate insurance connected with this study is provided by the University of Manchester.

**What will happen to the results of the study?**

General overall findings of the study will be published in articles in journals. This is to share information with others and add to knowledge and understanding of autistic catatonia. No information about specific individuals will be published; only summaries of the overall findings.

**Is there anyone I can talk to about the study?**

If you have any questions about the study or would like to find out more, you can contact the researcher, Jenny Breen ([jennifer.breen@postgrad.manchester.ac.uk](mailto:jennifer.breen@postgrad.manchester.ac.uk)) or her academic supervisor Dr Dougal Hare ([dougal.hare@manchester.ac.uk](mailto:dougal.hare@manchester.ac.uk)). We are more than happy to talk to you about the study and answer any questions you may have.

*Thank you for taking the time to think about being involved in this study.*
Appendix 9 – Flyer/Poster advertising the study

A research project investigating how common ‘catatonic’ symptoms are in children and young people with Autism Spectrum Disorders (ASD).

Are you the Parent or Carer of a young person aged 12-25 who has a diagnosis of autism spectrum disorder (ASD)?

(This includes those who have been diagnosed with any type of autism or aspergers syndrome)

If so, we would like to invite you to be involved in a research project running at the University of Manchester.

Researchers at the University of Manchester want to find out how common movement problems are in young people with autism spectrum disorder (ASD).

What would happen if I took part in the study?

If you decide to take part in this study, you will complete an online questionnaire answering questions about whether the person you care for displays certain behaviours or symptoms. This will take about 30 minutes to complete. More indepth information about the study is attached to this letter/email or can be requested by contacting the researcher (see how below). If after reading this information, you would like to take part in the study, the link to the questionnaire can be found at: https://apps.mhs.manchester.ac.uk/surveys//TakeSurvey.aspx?SurveyID=m4LH8855

If you have any questions or would like some more information about this study, please contact Jenny on jennifer.breen@postgrad.manchester.ac.uk

Thank you for taking the time to read about this study.
Appendix 10 – Pre-participation questions presented to participants at the beginning of the online questionnaire
Participant Debrief Sheet

Thank you for taking the time to participate in this research study – we really appreciate it.

We hope that this study will help us understand more about catatonic symptoms in young people with autism spectrum disorder (ASD) so that more effective care and treatments can be developed.

If you would like some information about the findings of the study, please email jennifer.breen@postgrad.manchester.ac.uk. A summary of the overall findings will be sent to you once the data is analysed.

If you have any questions about the study or would like to discuss it, please contact the researcher, Jenny Breen (jennifer.breen@postgrad.manchester.ac.uk) or her academic supervisor Dr Dougal Hare (dougal.hare@manchester.ac.uk). We are more than happy to talk to you about the study and answer any questions you may have.

If taking part in this study has made you upset or worried about the person you care for there are a number of organisations that can help you or give advice.

The National Autistic Society can be contacted via their telephone helpline on 0808 800 4104. Lines are open 10am – 4pm on weekdays and are free from landlines and most mobiles. This helpline provides impartial, confidential information, advice and support for people with autism spectrum disorders and their families and carers.

Further information can also be found on their website: www.autism.org.uk

Other useful resources are:

The Autistic Society - www.autisticsociety.org

Focuses on Autistic Spectrum Disorders and aims to provide a supportive and friendly community for parents and families of children and adults with ASD.

UK Autism Awareness - www.autism-awareness.org.uk

A meeting point for the vast network of people concerned with autism.
### Appendix 12 – Additional information relating to the diagnostic labels reported in the study (Table 4 and 5)

Table 4: Frequency of additional diagnostic labels reported in the study

<table>
<thead>
<tr>
<th>ADDITIONAL DIAGNOSTIC LABEL</th>
<th>n</th>
</tr>
</thead>
<tbody>
<tr>
<td>Autistic Catatonia</td>
<td>19</td>
</tr>
<tr>
<td>Dyspraxia</td>
<td>12</td>
</tr>
<tr>
<td>Epilepsy</td>
<td>8</td>
</tr>
<tr>
<td>Attention Deficit Hyperactive Disorder</td>
<td>7</td>
</tr>
<tr>
<td>Anxiety</td>
<td>7</td>
</tr>
<tr>
<td>Dislexia</td>
<td>6</td>
</tr>
<tr>
<td>Obsessive Compulsive Disorder</td>
<td>6</td>
</tr>
<tr>
<td>Intellectual Disability</td>
<td>5</td>
</tr>
<tr>
<td>Depression/mood disorder</td>
<td>4</td>
</tr>
<tr>
<td>Oppositional Defiance Disorder</td>
<td>3</td>
</tr>
<tr>
<td>Pervasive Demand Avoidance</td>
<td>3</td>
</tr>
<tr>
<td>Sensory Integration Disorder</td>
<td>3</td>
</tr>
<tr>
<td>Tics/Tourettes</td>
<td>2</td>
</tr>
<tr>
<td>Post Traumatic Stress Disorder</td>
<td>2</td>
</tr>
<tr>
<td>Psychosis</td>
<td>2</td>
</tr>
<tr>
<td>Mitochondrial Disease</td>
<td>1</td>
</tr>
<tr>
<td>Ehlers Danlos syndrome</td>
<td>1</td>
</tr>
<tr>
<td>Cerebral Palsy</td>
<td>1</td>
</tr>
<tr>
<td>Hyperacusis</td>
<td>2</td>
</tr>
<tr>
<td>Underlined Bone Disorder</td>
<td>1</td>
</tr>
<tr>
<td>Marfans Syndrome</td>
<td>1</td>
</tr>
</tbody>
</table>
### Table 5: Frequency of additional diagnostic labels reported by participants who had an existing diagnosis of autistic catatonia

<table>
<thead>
<tr>
<th>Diagnostic label</th>
<th>n</th>
</tr>
</thead>
<tbody>
<tr>
<td>Dyslexia</td>
<td>1</td>
</tr>
<tr>
<td>Epilepsy</td>
<td>1</td>
</tr>
<tr>
<td>Dyspraxia</td>
<td>1</td>
</tr>
<tr>
<td>Intellectual Disability</td>
<td>1</td>
</tr>
<tr>
<td>Psychosis</td>
<td>1</td>
</tr>
<tr>
<td>Oppositional Defiance Disorder</td>
<td>1</td>
</tr>
<tr>
<td>Mitochondrial Disease</td>
<td>1</td>
</tr>
<tr>
<td>ADHD, Dyspraxia, Obsessive Compulsive Disorder, Anxiety, Sleep Disorder</td>
<td>1</td>
</tr>
<tr>
<td>Anxiety, Obsessive Compulsive Disorder, Tics</td>
<td>1</td>
</tr>
</tbody>
</table>
Appendix 13 - SPSS output - Receiver Operating Curve (ROC) analysis using existing diagnosis of autistic catatonia as the state variable

**Case Processing Summary**

<table>
<thead>
<tr>
<th>AC_diagnosis</th>
<th>Valid N (listwise)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Positive</td>
<td>20</td>
</tr>
<tr>
<td>Negative</td>
<td>79</td>
</tr>
</tbody>
</table>

Larger values of the test result variable(s) indicate stronger evidence for a positive actual state.
a. The positive actual state is Yes.

Area Under the Curve

<table>
<thead>
<tr>
<th>Test Result Variable(s): Core AC Score</th>
<th>Area</th>
<th>Std. Error</th>
<th>Asymptotic Sig.</th>
<th>Asymptotic 95% Confidence Interval</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>.624</td>
<td>.072</td>
<td>.047</td>
<td>.483</td>
</tr>
</tbody>
</table>

The test result variable(s): Core_AC_Score has at least one tie between the positive actual state group and the negative actual state group. Statistics may be biased.
a. Under the nonparametric assumption
b. Null hypothesis: true area = 0.5
### Coordinates of the Curve

**Test Result Variable(s): Core_AC_Score**

<table>
<thead>
<tr>
<th>Positive if Greater Than or Equal To</th>
<th>Sensitivity</th>
<th>1 - Specificity</th>
</tr>
</thead>
<tbody>
<tr>
<td>-1.00</td>
<td>1.000</td>
<td>1.000</td>
</tr>
<tr>
<td>.50</td>
<td>.850</td>
<td>.848</td>
</tr>
<tr>
<td>1.50</td>
<td>.850</td>
<td>.797</td>
</tr>
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<td>2.50</td>
<td>.850</td>
<td>.747</td>
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<td>.800</td>
<td>.696</td>
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<td>4.50</td>
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<td>.620</td>
</tr>
<tr>
<td>5.50</td>
<td>.750</td>
<td>.582</td>
</tr>
<tr>
<td>6.50</td>
<td>.750</td>
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</tr>
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<td>7.50</td>
<td>.700</td>
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<td>.380</td>
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<td>10.50</td>
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<td>.127</td>
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<td>.150</td>
<td>.114</td>
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<td>23.50</td>
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<td>.013</td>
</tr>
<tr>
<td>25.00</td>
<td>.000</td>
<td>.000</td>
</tr>
</tbody>
</table>

The test result variable(s): Core_AC_Score has at least one tie between the positive actual state group and the negative actual state group.

a. The smallest cutoff value is the minimum observed test value minus 1, and the largest cutoff value is the maximum observed test value plus 1. All the other cutoff values are the averages of two consecutive ordered observed test values.
Appendix 14 – SPSS output - Pearson Chi-Square analysis of independence of existing autistic catatonia and gender

Case Processing Summary

<table>
<thead>
<tr>
<th>Cases</th>
<th>Valid</th>
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<th></th>
<th></th>
<th>Missing</th>
<th></th>
<th></th>
<th></th>
<th></th>
<th>Total</th>
<th></th>
<th></th>
</tr>
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<tbody>
<tr>
<td></td>
<td>N</td>
<td>Percent</td>
<td>N</td>
<td>Percent</td>
<td>N</td>
<td>Percent</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>AC_diagnosis *</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Gender</td>
<td>99</td>
<td>100.0%</td>
<td>0</td>
<td>0.0%</td>
<td>99</td>
<td>100.0%</td>
<td></td>
<td></td>
<td></td>
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<td></td>
<td></td>
</tr>
</tbody>
</table>

AC_diagnosis * Gender Crosstabulation

<table>
<thead>
<tr>
<th></th>
<th>Gender</th>
<th>Total</th>
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<th></th>
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<tbody>
<tr>
<td></td>
<td>Male</td>
<td>Female</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>No AC_diagnosis</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
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<tr>
<td>Count</td>
<td>61</td>
<td>18</td>
<td>79</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Expected Count</td>
<td>59.8</td>
<td>19.2</td>
<td>79.0</td>
<td></td>
<td></td>
</tr>
<tr>
<td>% within AC_diagnosis</td>
<td>77.2%</td>
<td>22.8%</td>
<td>100.0%</td>
<td></td>
<td></td>
</tr>
<tr>
<td>% within Gender</td>
<td>81.3%</td>
<td>75.0%</td>
<td>79.8%</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Yes AC_diagnosis</td>
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<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Count</td>
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<td>6</td>
<td>20</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Expected Count</td>
<td>15.2</td>
<td>4.8</td>
<td>20.0</td>
<td></td>
<td></td>
</tr>
<tr>
<td>% within AC_diagnosis</td>
<td>70.0%</td>
<td>30.0%</td>
<td>100.0%</td>
<td></td>
<td></td>
</tr>
<tr>
<td>% within Gender</td>
<td>18.7%</td>
<td>25.0%</td>
<td>20.2%</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Total</td>
<td>75</td>
<td>24</td>
<td>99</td>
<td></td>
<td></td>
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<tr>
<td>Expected Count</td>
<td>75.0</td>
<td>24.0</td>
<td>99.0</td>
<td></td>
<td></td>
</tr>
<tr>
<td>% within AC_diagnosis</td>
<td>75.8%</td>
<td>24.2%</td>
<td>100.0%</td>
<td></td>
<td></td>
</tr>
<tr>
<td>% within Gender</td>
<td>100.0%</td>
<td>100.0%</td>
<td>100.0%</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>
## Chi-Square Tests

<table>
<thead>
<tr>
<th>Test</th>
<th>Value</th>
<th>df</th>
<th>Asymp. Sig.</th>
<th>Exact Sig. (2-sided)</th>
<th>Exact Sig. (1-sided)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Pearson Chi-Square</td>
<td>.452a</td>
<td>1</td>
<td>.501</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Continuity Correctionb</td>
<td>.145</td>
<td>1</td>
<td>.704</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Likelihood Ratio</td>
<td>.437</td>
<td>1</td>
<td>.509</td>
<td>.562</td>
<td>.343</td>
</tr>
<tr>
<td>Fisher's Exact Test</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Linear-by-Linear Association</td>
<td>.448</td>
<td>1</td>
<td>.503</td>
<td></td>
<td></td>
</tr>
<tr>
<td>N of Valid Cases</td>
<td>99</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

a. 1 cells (25.0%) have expected count less than 5. The minimum expected count is 4.85.
b. Computed only for a 2x2 table
Appendix 15 – SPSS output – Investigation into potential under-diagnosis of autistic catatonia in the sample (Table 10 and 11)

Table 10: Crosstabulations and Pearson’s Chi-Squared test of independence ($\chi^2$) were completed for existing diagnosis of autistic catatonia and whether individuals displayed more or less than 3 core symptoms of autistic catatonia ('ACcorecurrent' variable; 1 = <3, 2 = >=3)

<table>
<thead>
<tr>
<th>AC_diagnosis * ACcorecurrent</th>
<th>Cases</th>
<th></th>
<th></th>
<th></th>
<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>N</td>
<td>Percent</td>
<td>N</td>
<td>Percent</td>
<td>N</td>
<td>Percent</td>
</tr>
<tr>
<td>AC_diagnosis *</td>
<td>99</td>
<td>100.0%</td>
<td>0</td>
<td>0.0%</td>
<td>99</td>
<td>100.0%</td>
</tr>
</tbody>
</table>

AC_diagnosis * ACcorecurrent Crosstabulation

<table>
<thead>
<tr>
<th>AC_diagnosis</th>
<th>ACcorecurrent</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>1.00</td>
<td>2.00</td>
</tr>
<tr>
<td>No</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Count</td>
<td>44</td>
<td>35</td>
</tr>
<tr>
<td>Expected Count</td>
<td>40.7</td>
<td>38.3</td>
</tr>
<tr>
<td>% within AC_diagnosis</td>
<td>55.7%</td>
<td>44.3%</td>
</tr>
<tr>
<td>% within ACcorecurrent</td>
<td>86.3%</td>
<td>72.9%</td>
</tr>
<tr>
<td>Yes</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Count</td>
<td>7</td>
<td>13</td>
</tr>
<tr>
<td>Expected Count</td>
<td>10.3</td>
<td>9.7</td>
</tr>
<tr>
<td>% within AC_diagnosis</td>
<td>35.0%</td>
<td>65.0%</td>
</tr>
<tr>
<td>% within ACcorecurrent</td>
<td>13.7%</td>
<td>27.1%</td>
</tr>
<tr>
<td>Total</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Count</td>
<td>51</td>
<td>48</td>
</tr>
<tr>
<td>Expected Count</td>
<td>51.0</td>
<td>48.0</td>
</tr>
<tr>
<td>% within AC_diagnosis</td>
<td>51.5%</td>
<td>48.5%</td>
</tr>
<tr>
<td>% within ACcorecurrent</td>
<td>100.0%</td>
<td>100.0%</td>
</tr>
</tbody>
</table>
### Table 11: Crosstabulations and Pearson’s Chi-Squared test of independence ($\chi^2$) were completed for existing diagnosis of autistic catatonia and whether individuals displayed more or less than 4 core symptoms of autistic catatonia (‘ACcorecurrent2’ variable; 1 = <4, 2 = >=4)

#### Case Processing Summary

<table>
<thead>
<tr>
<th>Cases</th>
<th>Valid</th>
<th>Missing</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>N</td>
<td>Percent</td>
<td>N</td>
</tr>
<tr>
<td>AC_diagnosis</td>
<td>99</td>
<td>100.0%</td>
<td>0</td>
</tr>
<tr>
<td>ACcorecurrent2</td>
<td>99</td>
<td>100.0%</td>
<td>0</td>
</tr>
<tr>
<td>AC_diagnosis * ACcorecurrent2 Crosstabulation</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>----------------------------------------------</td>
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<td></td>
<td></td>
</tr>
<tr>
<td>ACcorecurrent2</td>
<td>Total</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>1.00</td>
<td>2.00</td>
<td></td>
</tr>
<tr>
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<td>62</td>
<td>17</td>
<td>79</td>
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<tr>
<td>Expected Count</td>
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<td>79.0</td>
</tr>
<tr>
<td>% within AC_diagnosis</td>
<td>78.5%</td>
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<td>100.0%</td>
</tr>
<tr>
<td>% within ACcorecurrent2</td>
<td>86.1%</td>
<td>63.0%</td>
<td>79.8%</td>
</tr>
<tr>
<td>Count</td>
<td>10</td>
<td>10</td>
<td>20</td>
</tr>
<tr>
<td>Expected Count</td>
<td>14.5</td>
<td>5.5</td>
<td>20.0</td>
</tr>
<tr>
<td>% within AC_diagnosis</td>
<td>50.0%</td>
<td>50.0%</td>
<td>100.0%</td>
</tr>
<tr>
<td>% within ACcorecurrent2</td>
<td>13.9%</td>
<td>37.0%</td>
<td>20.2%</td>
</tr>
<tr>
<td>Count</td>
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<td>Expected Count</td>
<td>72.0</td>
<td>27.0</td>
<td>99.0</td>
</tr>
<tr>
<td>% within AC_diagnosis</td>
<td>72.7%</td>
<td>27.3%</td>
<td>100.0%</td>
</tr>
<tr>
<td>% within ACcorecurrent2</td>
<td>100.0%</td>
<td>100.0%</td>
<td>100.0%</td>
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</tbody>
</table>

Chi-Square Tests

<table>
<thead>
<tr>
<th></th>
<th>Value</th>
<th>df</th>
<th>Asymp. Sig. (2-sided)</th>
<th>Exact Sig. (2-sided)</th>
<th>Exact Sig. (1-sided)</th>
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</thead>
<tbody>
<tr>
<td>Pearson Chi-Square</td>
<td>6.527a</td>
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<td>.011</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Continuity Correctionb</td>
<td>5.170</td>
<td>1</td>
<td>.023</td>
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<td></td>
</tr>
<tr>
<td>Likelihood Ratio</td>
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<td>.014</td>
<td>.022</td>
<td>.014</td>
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<tr>
<td>Fisher's Exact Test</td>
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<td></td>
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<td></td>
<td></td>
</tr>
<tr>
<td>Linear-by-Linear</td>
<td>6.461</td>
<td>1</td>
<td>.011</td>
<td></td>
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</tr>
<tr>
<td>Association</td>
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<td></td>
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<td></td>
<td></td>
</tr>
<tr>
<td>N of Valid Cases</td>
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<td></td>
<td></td>
<td></td>
<td></td>
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</tbody>
</table>

a. 0 cells (0.0%) have expected count less than 5. The minimum expected count is 5.45.
b. Computed only for a 2x2 table
Table 12: Analysis of skewness and kurtosis of GDS-CS Total Scores:

<table>
<thead>
<tr>
<th>Statistics</th>
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</tr>
</thead>
<tbody>
<tr>
<td>GDSLD_Score</td>
<td></td>
</tr>
<tr>
<td>Valid</td>
<td>87</td>
</tr>
<tr>
<td>N</td>
<td>87</td>
</tr>
<tr>
<td>Missing</td>
<td>12</td>
</tr>
<tr>
<td>Skewness</td>
<td>-.247</td>
</tr>
<tr>
<td>Std. Error of Skewness</td>
<td>.258</td>
</tr>
<tr>
<td>Kurtosis</td>
<td>-1.068</td>
</tr>
<tr>
<td>Std. Error of Kurtosis</td>
<td>.511</td>
</tr>
</tbody>
</table>

Table 13: Analysis of skewness and kurtosis of RBQ Total Scores:

<table>
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</tr>
</thead>
<tbody>
<tr>
<td>RBQtotal</td>
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</tr>
<tr>
<td>Valid</td>
<td>87</td>
</tr>
<tr>
<td>N</td>
<td>87</td>
</tr>
<tr>
<td>Missing</td>
<td>12</td>
</tr>
<tr>
<td>Skewness</td>
<td>-.097</td>
</tr>
<tr>
<td>Std. Error of Skewness</td>
<td>.258</td>
</tr>
<tr>
<td>Kurtosis</td>
<td>-.908</td>
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<tr>
<td>Std. Error of Kurtosis</td>
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</tr>
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</table>
Appendix 17 – SPSS output - Statistical checks of assumptions required for regression analysis between Core ACQ score and GDS-CS score
Appendix 18 - SPSS output - Linear regression analysis between Core ACQ score and GDS-CS score

Variables Entered/Removed\(^a\)

<table>
<thead>
<tr>
<th>Model</th>
<th>Variables Entered</th>
<th>Variables Removed</th>
<th>Method</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>GDSLD_Score(^b)</td>
<td>.</td>
<td>Enter</td>
</tr>
</tbody>
</table>

\(^a\) Dependent Variable: No_CoreSymptoms_Currently
\(^b\) All requested variables entered.

Model Summary

<table>
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<tr>
<th>Model</th>
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<th>R Square</th>
<th>Adjusted R Square</th>
<th>Std. Error of the Estimate</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>.389(^a)</td>
<td>.151</td>
<td>.141</td>
<td>1.694</td>
</tr>
</tbody>
</table>

\(^a\) Predictors: (Constant), GDSLD_Score

ANOVA\(^a\)

<table>
<thead>
<tr>
<th>Model</th>
<th>Sum of Squares</th>
<th>df</th>
<th>Mean Square</th>
<th>F</th>
<th>Sig.</th>
</tr>
</thead>
<tbody>
<tr>
<td>Regression</td>
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<td>43.433</td>
<td>15.132</td>
<td>.000(^b)</td>
</tr>
<tr>
<td>1</td>
<td>243.969</td>
<td>85</td>
<td>2.870</td>
<td></td>
<td></td>
</tr>
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<td>287.402</td>
<td>86</td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

\(^a\) Dependent Variable: No_CoreSymptoms_Currently
\(^b\) Predictors: (Constant), GDSLD_Score
### Coefficients

<table>
<thead>
<tr>
<th>Model</th>
<th>Unstandardized Coefficients</th>
<th>Standardized Coefficients</th>
<th>t</th>
<th>Sig.</th>
<th>95.0% Confidence Interval for B</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>B</td>
<td>Std. Error</td>
<td>Beta</td>
<td></td>
<td>Lower Bound</td>
</tr>
<tr>
<td>1</td>
<td>(Constant)</td>
<td>1.182</td>
<td>.370</td>
<td>3.192</td>
<td>.002</td>
</tr>
<tr>
<td></td>
<td>GDSLD_Score</td>
<td>.119</td>
<td>.031</td>
<td>3.890</td>
<td>.000</td>
</tr>
</tbody>
</table>

a. Dependent Variable: No_CoreSymptoms_Currently
Appendix 19 – SPSS output - Statistical checks of assumptions required for regression analysis between ACQ score and RBQ Total score

Histogram checking the assumption of normally distributed residuals
Dependent Variable: Core_AC_Score

Normal P-P Plot of Regression Standardized Residual
Dependent Variable: Core_AC_Score

Scatterplot
Dependent Variable: Core_AC_Score

Regression Standardized Residual vs. Regression Standardized Predicted Value
Appendix 2 – SPSS output - linear regression analysis between Core ACQ score and RBQ Total Score

Variables Entered/Removeda

<table>
<thead>
<tr>
<th>Model</th>
<th>Variables Entered</th>
<th>Variables Removed</th>
<th>Method</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>RBQtotalb</td>
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</tr>
</tbody>
</table>

a. Dependent Variable: Core_AC_Score
b. All requested variables entered.

Model Summaryb

<table>
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<tr>
<th>Model</th>
<th>R</th>
<th>R Square</th>
<th>Adjusted R Square</th>
<th>Std. Error of the Estimate</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>.342a</td>
<td>.117</td>
<td>.105</td>
<td>4.939</td>
</tr>
</tbody>
</table>

a. Predictors: (Constant), RBQtotal
b. Dependent Variable: Core_AC_Score

ANOVAb

<table>
<thead>
<tr>
<th>Model</th>
<th>Sum of Squares</th>
<th>df</th>
<th>Mean Square</th>
<th>F</th>
<th>Sig.</th>
</tr>
</thead>
<tbody>
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<td>236.395</td>
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<tr>
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<td>Total</td>
<td>2016.987</td>
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</tbody>
</table>

a. Dependent Variable: Core_AC_Score
b. Predictors: (Constant), RBQtotal

Coefficientsa

<table>
<thead>
<tr>
<th>Model</th>
<th>Unstandardized Coefficients</th>
<th>Standardized Coefficients</th>
<th>t</th>
<th>Sig.</th>
<th>95.0% Confidence Interval for B</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>B</td>
<td>Std. Error</td>
<td>Beta</td>
<td></td>
<td>Lower Bound</td>
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<tr>
<td>1</td>
<td>(Constant)</td>
<td>5.07</td>
<td>3</td>
<td>.11</td>
<td>.036</td>
</tr>
<tr>
<td></td>
<td>RBQtotal</td>
<td>1.381</td>
<td>.038</td>
<td>.342</td>
<td>3.67</td>
</tr>
</tbody>
</table>

a. Dependent Variable: Core_AC_Score
**Appendix 2 – Correlation of Repetitive Behaviour Questionnaire subdomains and Core Autistic Catatonia Score**

<table>
<thead>
<tr>
<th></th>
<th>Core_AC_Score</th>
<th>RBQ_Stereotypsed</th>
<th>RBQ_Comulsive</th>
<th>RBQ_RestrictedPreferences</th>
<th>RBQ_InistenceSameeness</th>
<th>RBQ_RepetitiveSpeech</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Correlation</strong></td>
<td></td>
<td></td>
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<td></td>
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**. Correlation is significant at the 0.01 level (2-tailed).