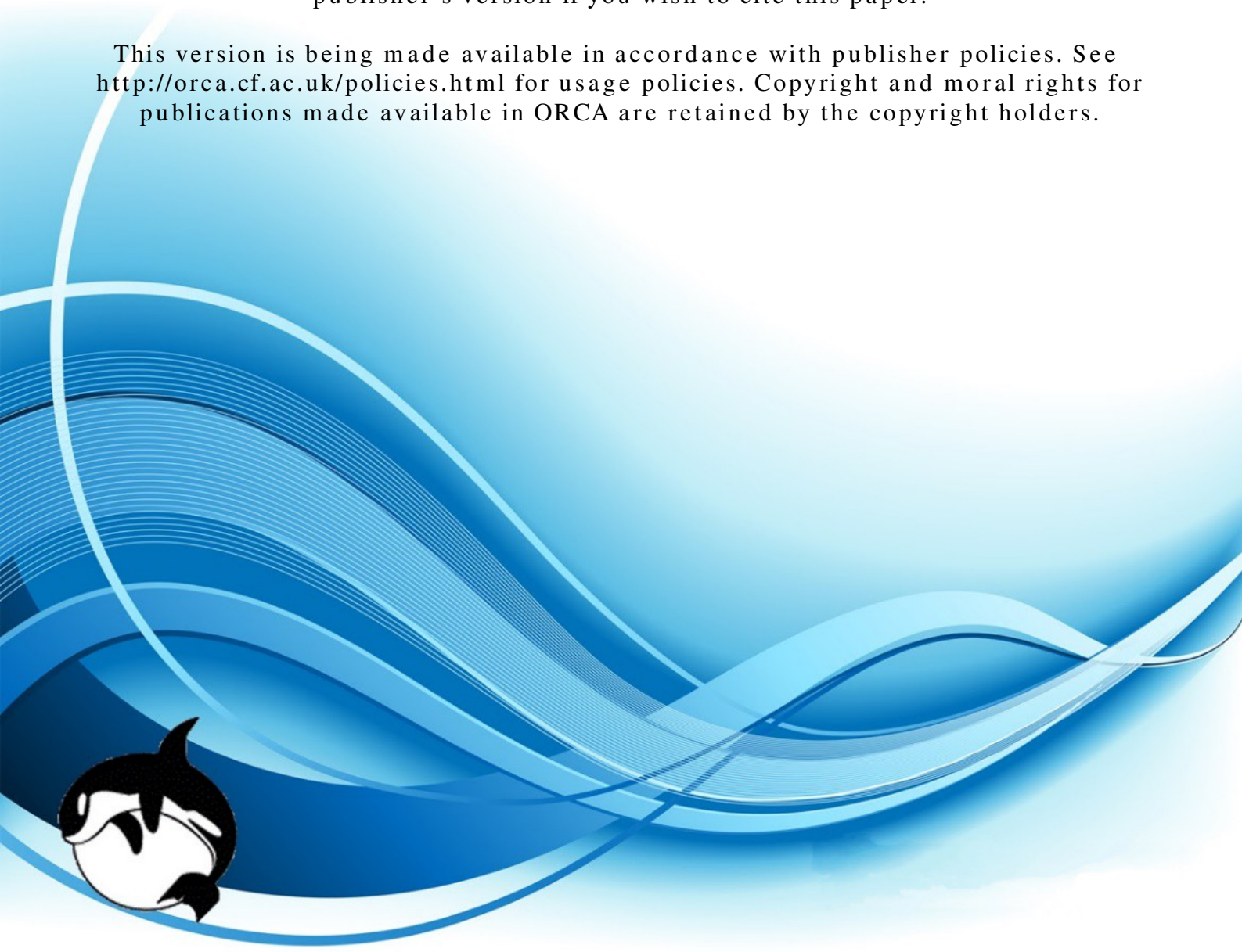

The nature and prevalence of catatonic symptoms in young people with autism

Breen J. & Hare D.J. (2017)

In press Journal of Intellectual Disability Research

DOI 10.1111/jir.12362

Abstract

Background A proportion of young people with autism are reported to show catatonic-like symptoms in adolescence. The aetiology and prevalence of such presentations is unknown but include a set of behaviours that can best be described as attenuated.

Method The current study empirically investigated the presence and nature of such attenuated behaviours in children and adolescents with autism using a newly developed 34-item third-party report measure, the Attenuated Behaviour Questionnaire. Caregivers or parents of young people with autism reported on the presentation of symptoms via the online completion of the Attenuated Behaviour Questionnaire and two established clinical measures of repetitive behaviour and depression.

Results Initial results indicate that the Attenuated Behaviour Questionnaire is a workable clinical measure in this population with a degree of discriminant validity with regard to catatonia. Attenuated behaviour indicative of catatonia were relatively common in young people with autism with up to 20.2% having an existing diagnosis of catatonia and evidence of a relationship between attenuated behaviours and measures of depression and repetitive and restricted behaviours.

Conclusion Catatonic symptoms are more prevalent in young people with autism than previously thought and the Attenuated Behaviour Questionnaire has potential as a clinical and research tool.

Keywords: autism; catatonia; attenuated behaviour

Introduction

Catatonia is characterised by abnormal hyper- or hypo-kinetic movements and behavioural alterations (Fink 1994; Bräunig, Krűger, Shuger, Höffler & Börner 2000) and is diagnosed on the presentation of positive and negative symptoms including mutism, catalepsy, facial grimaces, catalepsy, echolalia and akinesia in both International Classification of Diseases 10th Edition [ICD-10] (World Health Organization 1992) and Diagnostic and Statistical Manual of Mental Health Disorders 5th Edition [DSM-V] (American Psychiatric Association 2014). In severe cases, there is a sudden and dramatic loss of functional skills with subsequent difficulties with personal care, expressive communication and engagement in activities. Catatonia is rarely diagnosed in isolation and is usually co-morbid with other disorders including depression (Starkstein et al. 1996), encephalitis (Shill & Stacy 2000) and in developmental disorders including autism spectrum disorder [ASD] (Wing & Shah 2000) and Prader-Willi syndrome (Dhossche & Bouman 1997). Despite this, catatonia continues to be associated with psychosis by clinicians, despite being more prevalent in other clinical populations (Fink et al. 2010) and most people with psychosis are not catatonic (Heckers, Tandon & Bustillo, 2010) and there is a call for catatonia to be regarded as an independent syndrome or spectrum (Rosebush & Mazurek 2010). In order to accurately assess catatonia, several catatonia rating scales have been developed including the *Bush-Francis Catatonia Rating Scale* (Bush, Fink Petrides, Dowling & Francis 1996), the *Northoff Catatonia Scale* (Northoff, Wenke, Demisch Eckert, Gille & Pflug 1995) and the *Bräunig Catatonia Rating Scale* (Bräunig et al. 2000). Although these instruments have good inter-rater and test-retest reliability (Sienhart et al. 2011), the lack of a 'gold standard' definition of catatonia has restricted the development of catatonia rating scales and subsequent research (Dhossche & Rout. 2006; Sienhart, Rooseleer & De Fruyt 2011). Moreover, Carroll, Kirkhart, Ahuja, Soovere,, Lauterbach, Dhossche et al. (2008) propose that individual rating scales are required for the different populations of catatonic patients and there are problems with the rating scales, including inconsistent item definitions, a range of items (21-54), differing thresholds for diagnosis, omission of the affective symptoms (cf Abrams & Taylor 1976) and limited measurement of severity and frequency of symptoms. Moreover, none of these measures are validated for use with people with intellectual and developmental disorders and there are practical issues

including over-reliance on affective alterations that are difficult to accurately identify via third-party report and on alterations in speech or tone of voice and movement that be a feature of the developmental disorder itself (Carroll et al. 2008; Heckers, Tandon & Bustillo 2010)

In the case of ASD, there are reports of markedly abnormal motor movement, including extreme slowness in executing purposive movement, increased passivity, freezing during motor movement and difficulty initiating actions (Wing & Shah 2000; Hare & Malone 2004; Wing 2005) and such 'autistic catatonia' is usually onset in adolescence with a detrimental effect on quality of life (Wing & Shah 2006). Clinical accounts of autistic catatonia report hindered movements either in fluidity or quantity, remaining immobile for long periods of time, finding it difficult to start moving and performing movements very slowly or getting 'stuck' part way through a gross motor action (Wing & Shah 2000; Hare & Malone 2004), with physical and verbal prompts often being required (Hare & Malone 2004). Such apparent catatonia varies in presentation (Wing & Shah 2000; Billstead et al. 2005) and severity (Dhossche et al 2006) and is usually chronic (Ohta et al. 2006; Dhossche et al. 2006) but with some reports of cyclic (Realmuto & August 1991) and diurnal (Ohta et al. 2006) presentations. However, there is no commonly accepted diagnostic definition for 'autistic catatonia' and across the various case descriptions (Realmuto & August 1991; Dhossche 1998; Hare & Malone 2004; Ghaziuddin et al. 2005; Ohta et al. 2006; Schieveld 2006; Takota & Takata 2007; Kakooza-Mwesigeet al. 2008), the number and severity of presenting symptoms varies and few 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) [Appendix 1].

In their study of the prevalence of catatonic symptoms in 506 referrals to a specialist ASD clinic, Wing and Shah (2000) proposed a set of diagnostic guidelines for autistic catatonia, including 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*. Subsequent researchers have adapted these criteria by adding and

removing specific diagnostic items, whilst the DSM-IV definition of catatonia (APA, 1994) has been used by a number of researchers (Dhossche & Bouman 1997; 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 et al. 2006). A systematic investigation of such presentations is clearly required for research and clinical purposes, especially as prevalence is estimated at 6-17% (Wing & Shah 2006; Billstedt et al. 2005; Perisse et al. 2010; Nordin & Gillberg 1998) and catatonia is now specifically referenced in DSM-V (American Psychiatric Association 2013). Treatment options are limited (Ohta et al. 2006) and a recent systematic review of the treatment of 'autistic catatonia' (DeJong et al. 2014) indicated a paucity of evidence for all of the treatment modalities (behavioural, pharmacological and electro-convulsive therapy) reviewed.

Given the heterogeneous presentation and lack of consensus regarding aetiology and treatment with regard to 'autistic catatonia', it is proposed that the term *attenuated behaviour* is both accurate and more useful to researchers and clinicians and the current paper reports on the development of a novel measure of attenuated behaviour in young people with ASD.

Methods

Measures

The following measures were used in the study:

Attenuated Behaviour Questionnaire (ABQ) – a 34 item, third-party report measure was specifically designed for the present study to determine the prevalence and frequency of attenuated behaviours typically associated with 'autistic catatonia'. A literature search was completed using the on-line databases 'Psychinfo' and 'Web of Knowledge', which were systematically searched using key words *autism, Autistic Spectrum Disorder, ASD, catatonia,* the wildcard *autis** and the exact phrase *"autistic catatonia"*. To ensure the literature search was not subjected to a publishing bias, efforts were made to locate relevant undergraduate theses and poster presentations together with appropriate hand searching of journals. The numbers of each reported symptoms connected to autistic catatonia in the research literature were tallied

(Appendix 1). Any symptom reported only once (e.g. finger tapping, diaphoresis) was excluded from the measure as was any symptom which is a key feature of ASD and/or which did not indicate change to the individual's previous presentation were also excluded (e.g. echolalia). Any symptoms that were not amenable to accurate and reliable third-party report (e.g. auditory hallucinations, anxiety, visual hallucinations) were also excluded as any item that formed part of the other measures administered in the present study. A total of 88 reported symptoms were thus excluded.

Existing measures of catatonia were studied and their structure replicated as appropriate. Using this approach, the ABQ was developed consisting of 34 items, categorised as 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. 1995). Each symptom was defined with examples and behavioural descriptions to address concerns about inconsistent or vague symptom definitions in extant catatonia rating scales (Carrol et al. 2008). The items included in the ABQ, the order of presentation in the measure ('*ABQ 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 are shown in Appendix 2.

Items on the ABQ are rated on a five point scale designed to capture the presentation of a symptom over time, with symptoms that are progressively worsening over time being scored more highly:

0=-No, never

1= No, not at the moment but it used to happen

2=Yes but less than before

3=Yes, the same as before

4= Yes, more than before

The six most commonly reported autistic catatonia symptoms, representing difficulty with motor movement (ABQ item numbers 1-6 in Appendix 2) were termed 'core symptoms of autistic catatonia' as they are considered to be essential for diagnosis. Positive responses to these items (i.e. being rated 2, 3 or 4) triggered two supplementary questions measuring 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:

. *How <u>often</u> does the individual experience these periods of [symptom] <u>at the moment</u>?*

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*

1) *How <u>severely</u> does the individual experience these periods of [symptom] <u>at the moment</u>?*

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*

The pilot ABQ was examined by non-native English speakers prior to recruitment to ensure clarity and simplicity of the language used.

Repetitive Behaviour Questionnaire [RBQ] (Moss, Oliver, Arron, Burbidge & Berg. 2009) - a 19 item third party measure assessing prevalence and phenomenology of restricted and repetitive behaviours in people with intellectual disabilities with item is rated as occurring '*more than once a day'*, '*once a day*', '*once a week*', '*once a month*' or '*never*'. The items are scored into five sub-domains; '*Stereotyped behaviour'*, '*Compulsive behaviour'*, '*Restricted preferences'*, '*Repetitive speech'* and '*Insistence on sameness*' and a total score can be obtained from the sum of the sub-domains. The RBQ has good concurrent and construct validity when used with people with ASD aged 3-16.5 years (Honey et al. 2012). The RBQ was included in the study given the potential for phenomenological overlap between the various forms of repetitive and restricted behaviour as measured by the RBQ and features of autistic catatonia.

Carer Supplement to the Glasgow Depression Scale for people with Learning Disability [GDS-CS] (Cuthill, Espie & Cooper 2003) – a 16 item third-party measure which assessing current presentation of co-morbid depression in people with intellectual disabilities. 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 for clinical depression. The GDS-CS correlates with established measures of depression (r=0.88) and has high content validity, discriminant validity and criterion validity (Cuthill et al. 2003). The GDS-CS was included given the potential for phenomenological overlap between autistic catatonia and the psychomotor retardation component of depression, particularly as the latter is likely to particularly evident to parents and carers.

In addition to descriptive statistics relating to ABQ-derived scores in the present sample, further analyses were performed to determine:

- The relationship of ABQ-derived scores to the reported presence/absence of extant diagnoses of autistic catatonia
- A clinical cut-off for identifying cases of autistic catatonia via a ROC analysis of a derived total ABQ score.
- The association of ABQ-derived scores with demographic variables and with GDS-CE and RBQ scores

Participants

Informants were recruited via specialist care providers, parent support groups and charities and were included in the study if they were parent or long-term carer (>2 years) of a young person aged 12-25 years with an existing diagnosis of ASD. A minimum of 80 participants was required to test the internal validity of the ABQ. Participants completed measures anonymously via an online questionnaire (*Select Survey*) by URL link to a secure website hosted by the University of Manchester. Mean completion time was 19.7 minutes (range 5-79 minutes). No participants requested help completing the ABQ, which was taken as indicating that it was clear, readable and easy to use. The study was reviewed and approved by the University of Manchester Committee on the Ethics of Research on Human Beings.

Results

Data were analysed with *Statistical Package for the Social Sciences* (SPSS) version 20.0. Prior to conducting the analyses, the data was examined to ensure that all participants met inclusion criteria and that parametric data analysis was permissible. The majority of participants the informants were parents (91.9%). There were no missing data but 12 informants dropped out of the study after completing at least up to item six on the ABQ. A total of N=87 informants completed the full questionnaire and an additional N=12 informants completed the demographics information and the six core ABQ.

Table 1 here

41 respondents (41.4%) reported additional diagnostic labels in addition to ASD resulting in a total of 77 different co-morbid diagnoses (excluding catatonia). N=20 reported an existing diagnosis of catatonia, of whom N=9 had additional diagnostic labels. Further analysis was not feasible due to the small sample size, but there were significantly more females than males with diagnoses of catatonia (t (99) =-1.53, p=0.006). N=84 (85%) reported at least one core symptom either currently or in the past (Figure 1) with a bimodal distribution of 0 and 3 symptoms.

Figure 1 here

Independent samples t-tests indicated that subjects with extant diagnoses of catatonia displayed significantly more core symptoms (mean=3.10, SD=1.86) compared to those without a diagnosis (mean=2.20, SD=1.81) (t (97) =1.97, p=0.05; d=0.49). All six core symptoms were commonly reported, with difficulty initiating movement (ABQ item 2) being least reported (n=18) and physical and/or verbal prompts required (ABQ item 6) most reported (n=60).

Further preliminary analysis of the core symptoms involved computing four summary variables (ABQ-CAB, ABQ-CS, ABQ-CF & ABQ-TOT) as described in Table 2:

Table 2 here

The ABQ-CAB scores were bi-modally distributed (0 and 9) with a mean of 7.48 (SD=5.70; range 0-24) and N=15 subjects had an ABQ-CAB score = 0 (15.2%) i.e. no reported core symptoms to date. There was no significant difference in ABQ-CAB scores between those with (mean=9.35, SD=6.00) and without (mean=7.01, SD=5.57) an existing diagnosis of autistic catatonia [t(97)=1.65, p=0.10] whereas those currently displaying three or more core symptoms had a significantly higher ABQ-CAB scores (mean=12.04; SD=4.24) than those displaying less than three (mean=3.2; SD=2.86)(t(97)=-12.23, p<0.01; d=2.44). The Mean ABQ-CS score was 5.23 (SD=4.81; range of 0-23). No ABQ-CS score was recorded for N=22 participants. There was no significant difference in mean ABQ-CS score between those with (mean=6.95, SD=4.70) and those without (mean=4.80, SD=4.77) an existing diagnosis of catatonia [t (97) =1.81, p=0.07]. Subjects who currently displayed three or more core symptoms had significantly higher ABQ-CS scores (mean=8.94; SD=4.09) than those displaying less than three (mean=1.75; SD=2.01) (t (97) =-11.20, p<0.01; d= 2.23).

The mean ABQ-CF score was 4.73 (SD=3.87; range 0-15) with no ABQ-CF score being recorded for N=21 participants. Subjects with an existing diagnosis of catatonia had a significantly higher mean ABQ-CF score (mean=6.45, SD=3.90) than those without (mean=4.34, SD=3.72) (t(97)=2.24, p<0.05; d=0.55) and those displaying three or more core symptoms had a significantly higher mean ABQ-CF score (mean=7.85; SD=2.78) than those displaying less than three (mean=1.86; SD=1.93) (t(97)=-12.51, p<0.01; d= 2.50).

The mean ABQ-TOT score was 54.28 (SD=21.23; range 5-110) with no significant differences in mean ABQ-TOT scores between those with (mean=59.33, SD=19.77) and without (mean=52.96, SD=21.54) existing diagnoses of catatonia [t(85)=1.14, p=0.259], but those currently displaying three or more core symptoms had a significantly higher ABQ-TOT score (mean=61.04; SD=21.07) than those displaying less than three (mean=44.33; SD=22.02) (t(82)=-3.53, p<0.01; d= 0.77).

A clinical cut-off score for identifying cases of autistic catatonia was derived from a Receiver Operating Curve (ROC) analysis using the ABQ-CAB scores (Appendix 3). The area under curve was computed as 62.4% and participants with a diagnosis of catatonia group was identified by the ABQ-CAB score at significantly higher than chance (p<0.05), Analysis of the co-ordinates of the ROC curve (sensitivity and

specificity values) indicated that a conservative clinical cut-off point for catatonia was an ABQ-CAB score greater than 8.

All ABQ items can be assigned to one of three sub-domains, motor symptoms (n=15), affective alterations (n=5) or behavioural alterations (n=14). Sub-domain total scores were derived from the scores (0-4) for the items in each sub-domain. In the current sample (N=87), mean *ABQ Motor Score* [ABQMS] was 20.61 (SD=11.25; range 0-55), mean *ABQ Affective Alterations Score* [ABQAAS] was 11.30 (SD=4.42; range 0-20) and mean *ABQ Behavioural Alterations Score* [ABQBAS] was 22.37 (SD=8.78; range 3-47) and all were positively correlated with ABQ-CABS.

As autistic catatonia appears to onset in adolescence, the older individuals in the sample are more likely to have catatonia but independent samples t-tests revealed 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 catatonia [t (96) = -0.647, p=0.519] and there were no significant differences between those currently displayed three or more core behaviours (mean=16.32, SD=4.57) and those who displayed less than three core behaviours (mean=15.10; SD=3.29) for age; t (96) =-1.53, p=0.130. Pearson Chi-Square analysis of catatonia diagnoses and gender indicated that these were independent of each other [X^2 (1, n=99) =0.45, p=0.501] and independent samples t-tests indicated no significant difference in ABQ-CAB 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]. Similarly, there no gender differences were found for the ABQ-CS or ABQ-CF scores.

The mean GDS-CS score for the study sample was 10.56 (SD=5.98) and the scores range from 0-20. Between- group analyses showed no statistically significant difference in mean GDS-CS score 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) but subjects currently displaying three or more core symptoms had a significantly higher GDS-CS score (mean=12.74; SD=5.07) than those who displayed less than three core symptoms (mean=8.43; SD=6.09); t(85)=-3.586, p=0.01; d= 0.77). Regression analysis of the correlation between these variables indicates a significant linear association with *ABQ-CAB* accounting for approximately 15% of the variation in *GDS-CS Score (*r^2=0.15, p<0.001).

The mean RBQ score for the sample was 32.82 (SD=16.73; range1-68) and independent samples t-test indicates that subjects with an existing diagnosis of autistic catatonia have had significantly higher RBQ total scores (mean=40.56, SD=15.73) than those without an existing diagnosis (mean=30.80, SD=16.49) ($t(85)=2.256$, $p<0.05$; d= 0.60) and those currently presenting with three or more core symptoms also had significantly higher RBQ total scores (mean=39.58, SD=13.74) than those presenting with less than three core symptoms (mean=26.20, SD=16.86) ($t(85)=-4.05$, $p<0.01$; d=0.87) Regression analysis of the correlation between these variables indicates a significant linear association between ABQ-CABS and *RBQ Total Score* with ABQ-CABS, accounting for approximately 12% of the variation in *RBQ Total Score* ($r^2=0.12$, $p<0.001$).

The between-group analyses were repeated for the five RBQ sub-domain scores (*Stereotyped behaviour*, *Compulsive behaviour*, *Restricted preferences*, *Insistence on sameness* and *Repetitive speech*), which identified that subjects currently presenting with three or more core symptoms had higher *Compulsive behaviour* sub-domain scores (mean=11.37, SD=6.28), *Restricted preferences* sub-domain scores (mean=7.23, SD=3.64), *Insistence on sameness* sub-domain scores (mean=5.95, SD=2.39) and *Repetitive speech* sub-domain scores (mean=6.79, SD=3.94). Similarly, subjects with an existing diagnosis of autistic catatonia have significantly higher *Repetitive speech* sub-domain scores (mean=7.39, SD=4.18) than those without (mean=5.12, SD=4.14) ($t(85)=2.07$, $p<0.05$; d=0.54). There were significant positive correlation between ABQ-CACS and sub-domain scores for *Compulsive behaviour* (r=0.342, n=87, p=0.001); *Restricted preferences* (r=0.358, n=87, p=0.001), *Insistence on sameness* sub-domain scores (r=0.323, n=87, p=0.002) and *Repetitive speech* (r=0.336, n=87, p=0.001).

Overall, it was found that in the present sample, higher numbers of the core symptoms and severity but not frequency thereof being reported when participants had extant diagnoses of autistic catatonia. On the basis of a visual inspection of the distribution of the distribution of the number of reported core symptoms, the presence of any three core symptoms was taken as a putative clinical cut-off. An alternative clinical cut-off of a score of 8 on the ABQ-CAB sub-total was also derived using a ROC analysis. In addition, there was an association between the presence of more than three core symptoms and higher scores on the GADS-CS and RBQ, with these two scales accounting for 12 % and 15% of the variance in the ABQ-CAB scores.

Discussion

The ABQ appears to have potential as a valid and practical clinical measure with a degree of discriminant validity with the six core attenuated behaviours (ABQ items 1-6) being used as a clinical screening tool for catatonia in ASD with a proposed diagnostic cut-off of three or more core attenuated behaviours. It can be noted that these six core attenuated behaviours are essentially the same as Wing and Shah's (2000) proposed diagnostic criteria. In particular, the current results support the notion of catatonia as a continuum in ASD and that catatonia may be under-identified as N=20 participants had an existing diagnosis of catatonia whereas N=42 displayed three or more core attenuated behaviours. There is also evidence of a relationship between attenuated behaviours and measures of depression and repetitive and restricted behaviours, although a causal relationship was not determined.

The ABQ permits comparison of the change in presentation of attenuated behaviour over time due to the scoring metric for each question asking participants to rate each presenting behaviour compared to its past presentation. Assuming the face and ecological validity of the ABQ to be reasonable on the basis that the items relate to directly observable behaviours, the ABQ facilitates analysis of changes in the presentation of symptoms over time and could therefore would provide information about the course of catatonia and the effectiveness of any intervention. However, the ABQ would not indicate when a specific behaviour changes, only that it is more, less or the same as before.

The data from this initial study indicate that attenuated behaviours associated with catatonia are common in young people with ASD in line with predictions from the putative models of catatonia in ASD (Wing & Shah 2006; Fink et al. 2006; Takota & Takata 2007; Gowen & Hamilton 2013). The data also supports the notion that previously reported variation in the presentation of catatonia in people with ASD (Wing & Shah 2000; Billstead et al. 2005; Dhossche et al. 2006; Neumarker 2006) might in part be due to the absence of empirically derived criteria. Moreover, the number of core behaviours present in individuals with an existing diagnosis of catatonia varied with between one and six (mean=3.1), supporting Wing and Shah's (2000) clinical observations of heterogeneity in presentation. In the present study, only two individuals

with an existing diagnosis of catatonia displayed all six core behaviours. However, the *number* of core behaviours currently displayed appears to be a good indicator of catatonia as individuals with an existing diagnosis currently presented with significantly more core behaviours. However, four individuals without an existing diagnosis of catatonia presented with all six core behaviours.

The estimated prevalence of catatonia in the current study varied depending on the cut-off applied. Twenty (20.2%) subjects had an existing diagnosis of catatonia whereas 42 (48.3%) displayed three or more core behaviours (i.e. above the proposed clinical cut-off point). Therefore, the current data indicate that possible prevalence of catatonia in the current study sample may exceed previous estimates of 6-14% and support suggestions of an under-diagnosis of catatonia in young people with ASD. It should be noted that catatonia was not specified in the information used for recruitment and the research title indicated that the study was investigating movement problems in young people with ASD. Additionally, the majority of the sample did *not* have an existing diagnosis of catatonia (79.8%).

Statistically significant relationships were found between scores on the ABQ and measures of depression and repetitive and restricted behaviour (appropriate for the study population). In the case of the former, these conditions may be co-occurring or this may be due to the apparent overlap in the items on each measure, specifically reduced eating (ABQ item 27, GDS-CS item 11), reduced communication or muteness (ABQ item 24, GDS-CS item 5), increased aggression (ABQ item 18, GDS-CS item 2), avoidance of contact with others (ABQ item 16, GDS-CS item 3), decreased personal hygiene and/or concern about appearance (ABQ item 31, GDS-CS item 4), crying (ABQ item 17, GDS-CS item 6), reduced engagement in preferred activities (ABQ item 21, GDS-CS item 8), requiring more encouragement (ABQ item 22, GDS-CS item 10) and sleep problems (ABQ item 26, GDS-CS item 12). As both measures are third-party ratings of observable behaviours, it may be that a young person with ASD is presenting with either catatonia or depression, but this is being attributed to either or both conditions as extant measures lack the sensitivity 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 third-party measures of catatonia and depression. Further investigation is therefore required to elucidate the apparent relationship between ABQ and GDS-CS scores. The

association between repetitive and restricted behaviours and attenuated behaviour as measured by the ABQ appears to be more robust and not necessarily artefactual. There is some duplication of items across both measures, specifically body stereotypy (ABQ item 8, RBQ item 2) and hand stereotypy (ABQ item 13, RBQ item 3). Overall, the effect sizes for the significant results were found to be moderate to high.

The current study has a several limitations with respect to both design and execution. It is possible that relevant material was omitted from the initial literature review, which was based on key words in the title or abstract of published articles. The research design relied on retrospective reporting of current symptoms with previous presentation, which may have compromised the accuracy of the data on frequency and severity. However, the main focus of the study was on the current presentation of observable attenuated behaviours and this may have off-set any such bias. The online nature of the study survey resulted in no direct contact between researchers and participants, which whilst guaranteeing anonymity for participants also precluded checking against inclusion criteria and therefore the insertion of pre-participation questions that included confirmation about inclusion criteria was intended to minimise the possibility of error prior to completion of the online questionnaire. With regard to the determination of clinical cut-offs, the composite nature of the ABQ meant that both categorical and nominal cut-offs were necessary, the latter being computed via the ROC curve analysis, with the majority of the reported analyses being undertaken using the former cut-off, which should be regarded at this stage as provisional. Further work is required to identify which cut-off score is the more valid. Finally, the test-retest and inter-rater reliability of the ABQ were not assessed as part of the present study and should be incorporated in future research.

Future research could examine the association between ABQ scores and the degree of global intellectual disability, expressive language impairment and severity of ASD , all of which have been identified as potentially risk markers for catatonia development (Wing & Shah, 2000) as well as investigation of speed of onset of attenuated behaviour symptoms and precipitating factors such as stress, which have been *a priori* identified as of prognostic importance (Shah & Wing 2000; Ghaziuddin et al. 2005; Shah & Wing 2006).

The ABQ has the capability to assess the course of catatonia and to empirically measure the effectiveness of treatment interventions over time to support the development of evidence-based treatments (DeJong et al 2014) and practice guidelines. It could also have value as a routine screening tool for early detection of catatonia in adolescents with ASD

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Appendix 1 –Symptoms of autistic catatonia

Symptom	Frequency in case reports	Included in ABQ
'Freezing'/very still like a statue	6	Yes
Difficulty initiating actions/'stuckness'/akinesia	6	Yes
Problems stopping actions once started	3	Yes
Difficulty initiating movement	5	Yes
Slowness in movement	5	Yes
Requires prompts to complete actions	4	Yes
Waxy flexibility	3	Yes
Repetitive body movements	4	Yes
Stiff posturing	9	Yes
Noticeable resting tremor	2	Yes
Increased motor tics	3	Yes
Waving or shaking extremities	2	Yes
Twisting or flicking hands in front of eyes	2	Yes
Moving in a jerky way	2	Yes
Impulsive/excitable phases	4	Yes
Withdrawal from physical contact	2	Yes
Spontaneous crying, laughing or screaming	4	Yes
Episodes of aggression	6	Yes
Difficulty passing through doorways	2	Yes
Difficulty crossing lines on the floor	2	Yes
Reduced enjoyment in preferred activities	4	Yes
Requiring more encouragement to engage	4	Yes
Unusual gait/posture	5	Yes
Reduced communication/muteness	10	Yes

Incontinence	4	Yes
Sleep problems	4	Yes
Reduced eating	6	Yes
Eye rolling/ unusual eye movements	5	Yes
Unusual facial expressions/'grimaces'	5	Yes
Ignoring instructions	2	Yes
Refusal to bathe or change clothes	2	Yes
Occasional groans or unusual noises	3	Yes
Staring into space/fixed gaze	5	Yes
Unable to lift head	2	Yes
Echolalia	2	No – common feature of ASD
Finger tapping	1	No – reported once
Diaphoresis	1	No – reported once
Withdrawal	1	No – reported once
Auditory hallucinations	1	No – reported once/speculative
Auditory hypersensitivity	1	No – reported once/ speculative
Depressed	5	No - too vague or speculative
Anxious	2	No - too vague or speculative

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Appendix 2: Attenuated Behaviour Questionnaire

ABQ question number:	Symptom:	Associated question in ABQ:	Example or descriptor given:	Supplementary questions re frequency & severity
Category: Motor Symptoms				
1	'Freezing'/very still like a statue	Are there times when s/he is very still for long periods of time, almost like a statue?		Yes
2	Difficulty initiating actions/'stuckness'/akinesia	Does s/he seem to get 'stuck' when trying to do something?	Stopping mid-air half way through reaching for something & looking like they are trying to move but cannot OR beginning to pick up a cup to drink but lifting it only half way and then putting it down again	Yes
3	Problems stopping actions once started	Does s/he seem to find it difficult to stop doing actions once they have started them?	Repeatedly putting a coat on & taking it off again & again for a long period of time	Yes
4	Difficulty initiating movement	Does s/he seem to find it difficult to start moving?	Lying still and looking like S/he wants to get up or reach for something but cannot	Yes
5	Slowness in	Does s/he move very	Moving very slowly	Yes

	movement	slowly and takes a long time to finish actions?	when doing things like picking up a cup to drink or eating dinner	
6	Requires prompts to complete actions	Are there times when s/he needs physical and/or verbal prompts to complete actions?	Needing someone to tell them or touch their arm to enable them to lift a cup to their mouth to drink	Yes
7	Waxy flexibility	Are there times when if you moved part of their body, they let you without taking much notice of what you are doing & then stay in that position afterwards?	Offer no resistance to you curling their fingers into a fist & then keep their hand curled up when you moved away	No
8	Repetitive body movements	Does s/he like to move their body in repetitive ways?	Any frequent body movement such as body rocking, twisting wrists, flicking fingers etc	No
9	Stiff posturing	Does s/he strike and hold stiff poses?		No
10	Noticeable resting tremor	When s/he is completely relaxed, does any part of their body tremble		No
11	Increased motor tics	Does s/he experience 'tics' (speech or movement)?	Suddenly & repetitively move their body or saying a word/phrase in a way they seem unable to control	No

12	Waving or shaking extremities	Does s/he move their hands or feet in an odd way?	Twisting, waving or shaking	No
13	Twisting or flicking hands in front of eyes	Does s/he twist or flick their hands in front of their eyes?		No
14	Moving in a jerky way	Does s/he move in a very jerky way?		No
23	Unusual gait/posture	Does s/he walk unusually?		No
Category: Affective Alterations				
15	Impulsive or excitable phases	Is s/he impulsive OR over-excitable?		No
16	Withdrawal from physical contact	Are there periods where s/he withdraws from contact with others?	Does not want to be hugged or touched by anyone, shutting themselves in their room, sitting under a table alone etc	No
17	Spontaneous crying, laughing or screaming	Does s/he scream, cry or laugh suddenly for no reason? (If so, which?)		No
18	Episodes of aggression	Is s/he aggressive towards themselves or others at times? (If so, which?)		No
21	Reduced enjoyment in preferred activities	Has s/he lost enjoyment in their favourite activities?	Currently gets no enjoyment from activities they used to enjoy or refuses to do	No

			them	
Category: Behavioural Alterations				
19	Difficulty passing through doorways	Does s/he find it difficult to walk through doorways?		No
20	Difficulty crossing lines on the floor	Does s/he find it difficult to walk across lines on the floor or changes in flooring?	Such as from a carpet to a wooden floor	No
22	Requiring more encouragement to engage	Is S/he doing less than they used to?	It is now harder than it used to be to encourage them to do activities	No
24	Reduced communication/ muteness	Are there periods where s/he communicates with others less or not at all?	This includes all communication methods including reduced speech or in other communication such as PECS etc	No
25	Incontinence	Are there periods where s/he is incontinent *or* refuses to use the toilet when they used to?	Not using skills that they have used in the past and are soiling themselves when they would have previously used the toilet.	No
26	Sleep problems	Does s/he have sleep problems?	Finding it difficult to get to sleep at night, wants to sleep in the day but not at night, getting little sleep etc	No
27	Reduced eating	Are there periods		No

		where s/he refuses to eat *or* eats less than they used to?		
28	Eye rolling/ unusual eye movements	Does s/he move or roll their eyes unusually?	Repeatedly rolling their eyes or repeatedly looking from left to right	No
29	Unusual facial expressions/'grimaces'	Does s/ he pull unusual facial expressions or grimaces?		No
30	Ignoring instructions	Does s/he ignore instructions?	This refers to instructions that they understands	No
31	Refusal to bathe or change clothes	Does s/he refuse to wash or change their clothes?		No
32	Occasional groans or unusual noises	Does S/he regularly make groaning or other unusual noises?		No
33	Staring into space/fixed gaze	Does s/he stare into space or fix their gaze onto certain things?		No
34	Unable to lift head	Does s/he seem unable to lift their head?	Their head looks like it is too heavy for them to lift up	No

Appendix 3: ROC curve analysis