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A systematic review of the evidence for hyporesponsivity in ASD

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Abstract:

Sensory modulation difficulties are common in children with Autism Spectrum Disorder (ASD).

Whilst both hyporesponsivity and hyperresponsivity have been established in ASD comparative to typically developing controls, it has been proposed that hyporesponsivity may distinguish the sensory profile of ASD from other neurodevelopmental conditions. This paper aimed to systematically evaluate evidence for a syndrome-specific profile of hyporesponsivity in individuals with ASD when compared to individuals from clinical comparison groups, evaluating 10 eligible papers. Support was cautiously identified for a syndrome-specific sensory profile of hyporesponsivity. Four factors that reduced variability in findings were: chronological age, type of comparison group, sensory measure, and quality of study. Whilst hyporesponsivity in ASD was identified, the use of poorly-defined comparison groups, over-representation of children with ASD and intellectual disabilities, and younger age ranges complicate generalisation of this body of work. Recommendations for further research in this field are offered.

Keywords: ASD, Sensory Processing, Hyporesponsivity.

Autism Spectrum Disorder (ASD) is a common neurodevelopmental condition recognised in approximately 1.1% of the UK population (Baird et al., 2006; Brugha et al., 2009, 2012), characterised by atypicality in two key domains: social communication and interaction; and restricted, repetitive and stereotyped patterns of behaviour, interests, and activities (APA, 2013). Whilst sensory modulation difficulties have been central to the clinical description of ASD since first-person accounts were reported by Asperger and Kanner (Asperger, as cited in Marco et al., 2011), only in 2013 did research recognising sensory modulation difficulties result in their inclusion within core diagnostic criteria for ASD in the Diagnostic and Statistical Manual of Mental Disorders (DSM-5) (APA, 2013). A robust literature establishing the presence of sensory features in ASD has emerged over the last decade, preparing the ground for exploration at a more detailed level.

Sensory modulation difficulties are common in ASD, and are reported in around 95% of those with the condition (Tomchek & Dunn, 2007). The term ‘sensory modulation’ refers to the central nervous system process by which sensory stimuli are received from multiple sense domains (taste, touch, etc.), and regulated into physiological or behavioural responses appropriate to the environment (Miller et al., 2001). Sensory modulation difficulties lie within an umbrella category of sensory processing disorders (Miller et al., 2007) (Figure 1), and consist of three main subtypes: sensory over-responsivity, sensory under-responsivity, and sensory seeking behaviour. Sensory over-responsivity is demonstrated by an exaggerated or extreme response to sensory stimuli that is experienced as aversive by the individual. Sensory under-responsivity is characterised as an absent or lesser than expected response to sensory stimuli.

[*Figure 1.* Subtypes of sensory modulation disorder within the overall umbrella of sensory processing disorders.]

Recognising the plethora of terms employed within the literature for these constructs, the current review will employ the terms ‘hyperresponsivity’ and ‘hyporesponsivity’ to denote over-arousal and under-arousal respectively.

Whilst not all individuals with ASD show behavioural signs of atypical sensory modulation (Baranek et al., 2005), difficulties are well established (Dawson & Watling, 2000; Rogers, Hepburn & Wehner, 2003; Tomchek & Dunn, 2007), and have been reported across the ASD spectrum, and across multiple sensory domains (Ben Sasson et al., 2009; O'Donnell et al., 2012). Sensory atypicalities are reported to occur in both social and non-social contexts (Baranek et al., 2006; Hilton et al., 2007; Liss et al., 2006), and are associated in ASD with anxiety (Ben-Sasson et al., 2008; Liss et al., 2006), caregiver stress (Ben-Sasson et al., 2013), and poorer educational outcome (Ben-Sasson et al., 2009).

Whilst common, difficulties in sensory modulation are not unique to ASD. Literature associates a number of other neurodevelopmental conditions with atypical sensory responsivity, including ADHD (Ahn et al., 2004; Broring et al., 2008; Cheung et al., 2009; Ghanizadeh, 2008, 2009), Fragile X syndrome (Baranek et al., 2008; Rogers, et al., 2003), Developmental Delay (Baranek et al., 2007), and Williams Syndrome (Blomberg et al. 2006; Dilts et al., 1990; Gallo et al., 2008). It has been proposed that elevated levels of hyporesponsivity, a sub-type of sensory responsivity, may be a more syndrome-specific feature of ASD than either hyperresponsivity, or overall sensory responsivity (Baker et al., 2008; Baranek et al., 2006, 2007; Ben-Sasson et al., 2009; Miller et al., 2005; Rogers & Ozonoff, 2005).

A meta-analysis of sensory modulation symptoms in ASD conducted by Ben Sasson and colleagues (2008) concluded that the greatest differences between ASD and comparison groups related to the heightened level of hyporesponsivity in ASD. Evidence for this responsivity style as a syndrome-specific characteristic of ASD is, however, substantially limited to studies employing typically developing (TD) controls (Azouz et al., 2014; Ben-Sasson et al., 2007; Kern et al, 2006; Kientz & Dunn, 1996; Reynolds et al., 2011, 2011b; Taylay-Ongan & Wood, 2000; Tomchek & Dunn, 2007). Whilst comparison with typically developing individuals offers convincing evidence of relative hyporesponsivity in ASD, such studies offer limited clinical utility. Clinicians are commonly faced with the challenge of differentiating children with ASD from those with other developmental

disorders, including conditions which share behavioural similarities in sensory features. Studies that employ clinical comparison groups rather than TD controls typically report smaller between-group differences in sensory responsivity, increasing the difficulty of diagnostic differentiation (Baranek et al., 2006; O'Brien et al., 2009). Indeed, a number of studies employing clinical comparison groups report no difference in sensory features compared to ASD (e.g. Sensory Modulation Disorder: Miller et al., 2001; Fragile X: Rogers et al., 2003). Such studies consider differences at the level of *overall* number of sensory atypicalities, rather than at a sub-construct level. Merging of hyperresponsivity and hyporesponsivity to provide an overall measurement of sensory features potentially mask syndrome-specific sensory responsivity profiles in which groups may differ. Studies which consider sensory responsivity at the sub-scale level are therefore indicated in order to comprehensively identify differences between clinical groups.

Aims Of This Review

This paper aims to systematically evaluate evidence for a syndrome-specific profile of hyporesponsivity in ASD, when compared to clinical comparison groups.

Method

Systematic searches were conducted in August 2014, and updated in March 2016, using the following databases: PsycINFO, MEDLINE, Embase, and Scopus. Combination search terms included a diagnostic term (Autis*, Asperger*), a sensory term (Sensory, Reactiv*, Responsiv*), and a descriptor term (Threshold, Sensitiv*, Dysfunction, Pattern, Registration, Processing, Hypo*). Reference list searches and key author searches were conducted electronically, and two journals were hand searched (Journal of Autism and Developmental Disorders, American Journal of Occupational Therapy).

Eligibility Criteria

Eligibility criteria were set to include all primary studies in which individuals with a specific diagnosis of ASD were sampled as a distinct group, rather than as part of a wider developmental disorder sample (e.g. PDD-NOS). The ASD population was not restricted in any way (e.g. by age).

Criteria required that a clinical comparison group was used against which to measure the relative hyporesponsivity of the ASD group. Papers using any methodology were included in the initial literature search, providing that: the study provided a clinical (non-TD) comparison group; data on sensory processing were collected across multiple sensory modalities, rather than one specific modality (e.g. auditory); and that results related to sensory processing either clearly reference hyporesponsivity or one of the commonly employed similes for these terms, or provides data which allows the responsivity style to be calculated by the reader without further reference to the authors. The current search included all dates and languages. Unpublished and non-peer reviewed papers were included.

Development of Review Criteria

An evaluation tool was developed to assist in the analysis of studies: the Quality of Evidence Screening Tool (QuEST) (Table 1). Whilst the majority of existing tools for systematic review are developed for assessment of interventions, the current review sought to assess quality of evidence for the existence of a specific construct (hyporesponsivity). Development of the evaluation tool was informed by the frameworks of the Scottish Intercollegiate Guidance Network (SIGN) Guidelines for Cohort Studies (2012) and the Guidelines for Systematic Reviews and Meta-Analyses. Areas of assessment were based upon the checklist of Downs and Black (1998), and include additional topic-relevant criteria.

The evaluation tool addresses six key areas (Construct validity, External reliability, Internal reliability, External validity, Internal validity, and Statistical robustness), drawing upon 13 individual evaluation criteria. A rating scale providing comprehensive rating guidance for each criterion was developed to reduce rater-subjectivity in evaluation.

Methods of review resulting in allocation of an overall numerical score have attracted criticism (Colle et al., 2002; Juni et al., 2001; Lundh & Gotzsche, 2008). Consequently, the Cochrane Collaboration recommends against the use of such tools (Savovic, 2014). A fully qualitative rating scale was developed for the current review, following the recommendations of SIGN (2012). For the purposes

of the current review, it was decided to employ a four-category system for the evaluation of individual criteria: Excellent, Satisfactory, Poor, and Not Reported. Following Boulter (2013, unpublished thesis), the overall rating of papers was determined on the ability to provide quality evidence, and therefore this review makes use of the classification system employed: Decisive, Convincing, Fair, or Questionable, evidence.

Inter rater reliability.

A pilot version of the QuEST was developed prior to use in the full review to test the properties of the tool. Content and face validity were assessed by two reviewers with research experience in the field of sensory processing within ASD. Reliability was assessed by two independent raters scoring five studies (42% of studies in the review), finding moderate inter-rater agreement (Hallgren, 2012; Warrens, 2010). Alteration to the grid criteria was tested by a further independent rater, finding a good level of inter-rater reliability.

[Table 1 Quality of Evidence Screening Tool – Hyporesponsivity in ASD (QuEST)]

Results

62 papers were identified by electronic search, of which 10 met eligibility criteria and were selected for review using the QuEST. One paper did not directly provide results relating to hyporesponsivity (Cheung & Siu, 2009), and available data was hand calculated to provide a hyporesponsivity score for the study.

A summary of the key findings of identified studies and the associated overall quality ratings are provided as Table 2. Four factors were identified which reduced variability in findings: chronological age, type of comparison group, sensory measure, and quality of study.

[Table 2 Overview of reviewed studies]

Characteristics of the Evidence Base

The studies within the current review included a total of 524 individuals with ASD or Autistic Disorder. Studies represented both ASD and Autistic Disorder participants (5 ASD studies, 5 AD studies). Age range of participants varied from five months – 18 years (overall mean age = 5.07 years). Of studies that reported gender (8 of the 10), 82.91% of participants were male, consistent with the accepted prevalence of the condition (Fombonne et al., 2011). In all studies in which intellectual functioning was reported, the ASD or autism group demonstrated a level of intellectual function in the below average range. Nine of the 10 studies used a developmental delay (DD), learning disability (LD), or intellectual disability (ID) group as the clinical comparison group. The remaining study compared an ASD group to an ADHD group (Cheung & Siu, 2009). All studies included in the final review were peer-reviewed papers.

Quality of the Overall Evidence Base

Use of the QuEST tool suggested that four studies offered *decisive* evidence (Baranek et al., 2006, 2013; Kirby et al., 2015; O'Brien et al., 2009); three offered *convincing* evidence (Boyd et al., 2010; Brock et al., 2012; Watson et al., 2011); two provided *fair* evidence (Cheung & Siu, 2009; Joosten and Bundy, 2010); and one *questionable* evidence (Freuler et al., 2012).

Limitations to the generalisability of findings affected all studies reviewed. No study included in the current review included a population-based sample. Five higher quality studies which were produced by members of one research collaboration reported recruitment from a combination of clinical settings, a research database, and from schools (Baranek et al., 2006, 2013; Boyd et al., 2010; Kirby et al., 2015; Watson et al., 2011). Whilst not expressly indicated by any study, the commonality of co-authors and similarity of unusually broad recruitment method suggests that participants in these studies were drawn from the same pool, and potentially multiply measured (Baranek et al., 2006, 2013; Boyd et al., 2010; Kirby et al., 2015; Watson et al., 2011). Response/attrition rates were unreported in the majority of studies (8 of 10). Response rates alone were reported by Baranek et al. (2006) and Joosten & Bundy (2010). No study addressed potential bias within the sample.

Evidence

Seven of the 10 studies selected for this review report evidence to support a distinctive profile of hyporesponsivity in individuals with ASD when compared to a clinical comparison group. This proportion was not evenly distributed across the range of studies when considering the quality of evidence, as determined by the QuEST tool. Of studies which were determined as providing *decisive* or *convincing* evidence, 86% of studies found evidence for higher levels of hyporesponsivity in the ASD group (6 of 7). Of studies determined to have a *fair* or *questionable* ability to provide evidence, 33% (1 of 3) of studies found evidence for hyporesponsivity.

Age.

The majority of studies (7 out of 10) considered children in early childhood (5 months – 5.4 years; overall mean age = 4.27 years). Three studies focused upon participants in the mid-childhood age range (overall mean age = 8.32 years) (Joosten & Bundy, 2010; Kirby et al., 2015; O'Brien et al., 2009). Of the seven studies that found evidence of hyporesponsivity, six sampled an early childhood age range (range of mean ages = 40.5 – 64.8 months). Only Kirby et al. (2015) used a mid-childhood sample and found evidence for hyporesponsivity (mean age = 85.68 months). Of the three studies that found no evidence for hyporesponsivity, two used a mid-childhood sample (Joosten & Bundy, 2010; O'Brien et al., 2009). The third study which did not find evidence of hyporesponsivity (Cheung & Siu, 2009) used a younger sample when considering mean age only, but had a wider spread of age than the other studies in the younger child group (range= 32.4-139.2 months). No study eligible for this review included participants above 18 years of age. The mean age in months of participants in studies reporting hyporesponsivity was $M=55.18$, $SD=18.15$, compared to $M=88.67$, $SD=48.24$ for studies not reporting hyporesponsivity. Accounting for the wide age range in the Cheung and Siu study (2009), all studies using an exclusively younger childhood sample reported hyporesponsivity in children with ASD.

Intellectual functioning.

8 of the 10 studies in the current review provided an indication of the level of intellectual functioning (IF) of the ASD/autism group (excepting Cheung & Siu, 2009; and Freuler et al., 2012). In all cases where level of intellectual ability was reported, the ASD/autism group were in the below average range. Two papers reported an association of mental age (MA) with hyporesponsivity, such that higher MA predicted lower hyporesponsivity (Baranek et al., 2006, 2013), however IQ was unrelated to hyporesponsivity (Baranek et al., 2006). Three papers found that MA was unrelated to level of hyporesponsivity (Joosten & Bundy, 2010; Kirby et al., 2015; Watson et al., 2011). Three papers measured level of IF, but did not indicate whether this affected findings related to hyporesponsivity. Evidence *for* an association between mental age and hyporesponsivity and for *no* association between MA and hyporesponsivity was evenly distributed in the higher quality categories.

Sensory responsivity measures.

Whilst five studies employed a single measure of sensory responsivity, five studies used a battery of measures, deriving measures of hyporesponsivity from individual items across multiple measures (Baranek et al., 2013; Boyd et al., 2010; Brock et al., 2012; Freuler et al., 2012; Watson et al., 2011). Parent/carer questionnaire format assessment was most common. The Sensory Experiences Questionnaire (Baranek et al., 2006) was used in seven of the studies reviewed. The Sensory Profile (Kientz & Dunn, 1997) was used in six studies, with the truncated version, the Short Sensory Profile (McIntosh, Miller, Shyu, & Dunn, 1999), used in one study. The Sensory Processing Assessment (play-based observation) was used as an additional measure in five of the studies that used parental report. Use of the Sensory Experiences Questionnaire (SEQ), or SEQ plus a further measure (Sensory Processing Assessment, Sensory Profile, Short Sensory Profile) was positively associated with the finding of hyporesponsivity when compared to a DD comparison group. 6 of the 7 studies using the SEQ or SEQ/SP/SPA combination battery found evidence for hyporesponsivity. No study which used the Sensory Profile or Short Sensory Profile alone as its measure of sensory responsivity found evidence for hyporesponsivity (Cheung & Siu, 2009; Joosten & Bundy, 2010; O'Brien et al., 2009).

All studies rated ‘*decisive*’ in their ability to provide quality evidence employed the SEQ as a sole or contributory measure. Three quarters (3 of 4) of studies rated ‘*convincing*’ used the SEQ as a contributory measure. One third (1 of 3) of studies rated ‘*fair*’ or ‘*questionable*’ used the SEQ.

Sensory modality.

Of the 10 papers reviewed, only three provided results relating to sensory modality (Baranek et al., 2013; Cheung & Siu, 2009; O’Brien et al., 2009), of which two did not link modality scores with sensory responsivity (Baranek et al., 2013; Cheung & Siu, 2009). The one study that reported an association of sensory modality to sensory responsivity style reported the ASD group to differ from the comparison group by low auditory hypersensitivity and visual seeking (O’Brien et al., 2009).

Effect sizes.

It was possible to calculate effect sizes for hyporesponsivity in five of the papers reviewed (Baranek et al. 2006, 2013; Boyd et al., 2010; Kirby et al., 2015; Watson et al., 2011). Effect sizes were converted to d using the means and standard deviations of group hyporesponsivity scores. Sizes varied from $d=.02$ (Baranek et al., 2013) to $d=.91$ (Baranek et al., 2013). The weighed mean of effect sizes of hyporesponsivity in the current review was $d=.57$

Discussion

This review sought to systematically evaluate evidence for a syndrome-specific profile of hyporesponsivity in individuals with ASD when compared to individuals from clinical comparison groups. Evidence was found for hyporesponsivity in seven of the 10 studies reviewed. Four factors which reduced variability in findings were: chronological age, type of comparison group, sensory measure, and quality of study. It was not possible to draw conclusions about hyporesponsivity in older childhood or adult samples, in relation to non-DD comparison groups, or associations between hyporesponsivity and level of intellectual functioning, due to limitations in the literature. Limitations of the general field will be outlined.

This review found consensus within the literature of the definition of hyporesponsivity, although a number of different terms were used to label this construct, including ‘under-arousal’, ‘low arousal’, ‘hypo-arousal’, ‘hyposensitivity’, ‘hyporesponsivity’, and ‘high threshold for sensory responsivity’. As identified by Bishop (2014) in her study of the disparity of terms referring to specific language impairments, lack of clarity in diagnostic labels impedes progress in research, identification of difficulties, and access to services. While sensory hyporesponsivity is not, of itself, a disorder, the consequences of inconsistency of construct labelling have similar implications. Nonetheless, whilst the use of multiple related terms hampered the identification of relevant literature, the different terms were used consistently to refer to the same construct. Imprecision was found in all studies in the current review around the distinction between individual *experience* and behavioural *response* to sensory stimuli. This imprecision was mirrored in the wider literature. Hyporesponsivity was often defined as ‘[a] lack of response, or insufficient intensity of response to sensory stimuli’ (Baranek et al., 2006). This definition fails to isolate whether hyporesponsivity is causally an internal (sensory) or an external (behavioural) phenomena: is the child who fails to *recognise* stimuli hyporesponsive, or is the child who *registers* stimuli, but *fails to respond* to this hyporesponsive¹? Measures of sensory responsivity based on external behaviour identify all individuals who are behaviourally hyporesponsive, but amalgamate those whose hyporesponsivity is associated with neurological under-arousal with individuals whose behavioural hyporesponsivity is a psychological response to over-arousal.

Literature offers theoretical support for the concept of subgroups within behavioural hyporesponsivity. Schoen and colleagues found physiological low arousal and low neurological reactivity in an ASD group when compared to TD and SMD controls, however physiological and neurological low arousal did not correlate with hyporesponsivity as measured by the Short Sensory Profile (2009). Similarly, Azouz and colleagues (2014) identified neurological evidence of prolonged inter-peak short-latency somatosensory-evoked potentials (neurological hyporesponsivity), in the

¹ There is, of course, the third possibility that an individual may show neurological signs of registration, in the absence of *conscious recognition* of this registration, and thus no behavioural response.

context of no behaviourally-apparent sensory abnormalities (behavioural hyporesponsivity). These studies suggest that neurological and physiological hypo-arousal are not well associated with behavioural hyporesponsivity, and that neurological hypo-arousal may not be the sole causal factor predicting behavioural hyporesponsivity. This offers support for the idea of hypo subgroups with different causal factors: those who are neurologically under aroused and who attempt to regulate this to by sensory-seeking (potential low-registering/non-responders), and those who are not neurologically under aroused, but who fail to respond to sensory stimuli (potential registering/non-responders)². The potential of subgroups with different causal motivation for hyporesponsivity calls for more precise distinction and definition of this construct within the field. The assumption of association between neurological and behavioural responsivity is a significant limitation of the field.

Whilst there is, at present, no reliable decision-making method of associating different types of behavioural responsivity with specific treatment, the potential confounding of hyperresponsivity with hyporesponsivity adds an additional challenge to the effective clinical application of these findings. The absence of a clear causal link between neurological responsivity and behavioural responsivity raises a number of challenges with regards to the application of findings to clinical interventions, and thus the translational value of these studies is not at the moment clear.

Much of the limitation introduced by exclusive use of parent report measures is attributable to the relative expense, difficulty, and ensuing reduction in participant numbers of collecting physiological data when compared to using parent-report or observational methods. These latter methods are therefore highly represented in the field. This review found that findings of hyporesponsivity were moderated by measure used, such that use of the SEQ parent report measure associated with the finding of relative hyporesponsivity in the ASD group. It should be noted that the SEQ is the only measure for which there is published psychometric data supporting its construct validity in isolating

² There must be an alternative causal explanation for the hyporesponsivity of registering/non-responders other than hypo-arousal. It is possible that the registering/non-responders are motivated by *hyper*responsivity, rather than hyporesponsivity. Grandin writes, 'My hearing is like having a hearing aid with the volume control stuck on "super loud." It is like an open microphone that picks up everything. I have two choices: turn the mike on and get deluged with sound, or shut it off. Mother reported that sometimes I acted like I was deaf' (in Melillo, 2015).

hyporesponsivity and hyperresponsivity, rather than at the level of overall sensory abnormalities (Ausderau et al., 2014). In the wider literature however, a significant proportion of studies comparing ASD to TD controls have utilised measures other than the SEQ, and found evidence of hyporesponsivity in the ASD group. The Infant/Toddler Sensory Profile (Ben-Sasson et al., 2007); Sensory Profile (Dunn et al., 2002; Reynolds et al., 2011, 2012); Short Sensory Profile (Tomchek & Dunn, 2007); and Sensory Sensitivity Questionnaire – Revised (Talay-Ongen & Wood, 2000) have all been used with success to identify hyporesponsivity in ASD compared to TD groups. It is possible that, whilst the SP/SSP successfully discriminates hyporesponsivity in individuals with ASD when compared to a TD group, that it is a less sensitive measure when discriminating hyporesponsivity in ASD individuals from a DD group. Whilst the current review found an association between use of the SEQ and evidence for hyporesponsivity, it is important to note the parallel association between higher quality of study and use of the SEQ. The current review cannot therefore definitively indicate whether the SEQ has proven superiority in discriminating hyporesponsivity between ASD and DD groups. As noted, there was considerable overlap in authorship in the higher quality studies of the current review, in which use of the Sensory Experiences Questionnaire (SEQ) featured within the battery of measures employed by all but one higher quality study. This measure was developed by Baranek (2006), who was a lead or senior author within seven of the ten papers reviewed, and who co-authored two papers addressing the psychometric validation of the same measure. The conclusions drawn by the current review are therefore weighted towards the findings of this one research group, and would suggest that replication of these findings by other groups utilising additional measures is warranted.

Evidence for hyporesponsivity by sensory modality was largely absent from the literature base, due to the absence of reporting of individual modality scores and associations with responsivity style. It is therefore not possible to draw conclusions about the specificity of sensory responsivity to particular modalities in the ASD group. This absence represents a notable gap in the current field, given the plethora of single-modality research which shows differences in sensory responsivity between individuals with ASD and other groups. Studies which report sensory responsivity, and also the

breakdown of these findings by single sensory modality are underrepresented. Of the limited findings reported, differences found in low auditory hyperresponsivity and high auditory hyporesponsivity are consistent with the wider literature base, which suggests that the largest difference between ASD groups and controls relates to auditory differences (Klintwall et al., 2011; Tomchek & Dunn, 2007). Between 93 – 100% of participants with ASD are reported to show auditory processing problems (Greenspan and Wieder, 1997; Lane et al., 2010), with both hyperresponsivity and hyporesponsivity to auditory stimuli recorded (Baranek et al., 1997; Baranek, 1999; Osterling and Dawson, 1994; Matsushima & Kato, 2013). Whilst the hyporesponsivity identified within the current review may represent a general profile of sensory modulation within the ASD group, it is possible that responsivity styles vary by sensory modality. Findings of research which has failed to find hyporesponsivity may be affected by between-modality differences in hypo and hyperresponsivity, thus calling for studies which report sensory responsivity style by individual modality.

This review suggests that evidence of hyporesponsivity in ASD is more readily identified in children up to the age of around five years. Hyporesponsivity was reported by every study that used an exclusively younger childhood sample. Whilst a meta-analysis of sensory modulation symptoms in ASD (Ben-Sasson et al., 2009) found higher effect sizes for under-responsivity (hyporesponsivity) to be reported in the 6-9 year old age group (compared to 0-3, 3-6, and >9 years groups), the majority of studies in this meta-analysis employed TD controls. It is possible that trajectories of hyporesponsivity may differ in DD groups compared to TD groups, thus explaining difference in findings between the meta-analysis and those studies included in the current review. Children with ASD may show elevated levels of hyporesponsivity compared to children with DD at a young age (≤ 5.4 years), but show the greatest difference in levels of hyporesponsivity when compared to TD children in mid-childhood. Only one study in the current review explored profiles of hyporesponsivity in children aged between 12-15 years. This study had too small a sample size to allow for discriminant analysis of the age group (Joosten & Bundy, 2010), and thus a definitive commentary on hyporesponsivity within middle childhood age cannot be made. Studies considering older children were limited either by small sample size, or wide age range. It appears that hyporesponsivity may have an association

with age in children with ASD, but that the age at which hyporesponsivity peaks and declines has not been definitively proven. Higher quality studies using exclusively mid and older childhood samples are indicated. No study eligible for this review included individuals in the adult age range. This review is therefore able only to comment upon evidence of hyporesponsivity in children in ASD.

Studies with developmental delay control groups were over-represented in the current review, limiting comparison of hyporesponsivity in ASD with other clinical groups. The selection of developmental delay groups is clinically justifiable, given that individuals with intellectual impairments often experience sensory hyper/hypo-responsiveness (Padankatti, 2005), and are well-represented in the developmental disability clinics from which many studies recruit participants. Developmental delay groups, however, frequently include wide variation in individual profiles. This complicates interpretation of results with respect to specific clinical groups, and provides findings which may not be replicable. The literature could be considerably enriched by use of more focused comparison groups, such as clearly defined neurodevelopmental disorder groups. Whilst lack of clarity around the measure of hyporesponsivity rendered the study of Schoen and colleagues (2009) ineligible for the current review, this study compared an ASD/Autism group without intellectual disability ($FSIQ > 70$) with a Sensory Modulation Disorder group. Sensory Modulation Disorder is characterised by the appropriate gradation of one's response to everyday sensory experiences (Miller et al., 2007a), and therefore comparison of an SMD group to an ASD group is of great relevance. Findings of similarities and difference between ASD and SMD highlight which elements of sensory responsivity may be part of a shared behavioural phenotype between the conditions, and which are specific to diagnostic group. Future studies that use highly-specific clinical comparison groups would add more to the literature than further TD or DD studies.

The evidence for an association between hyporesponsivity and level of intellectual functioning is equivocal. This uncertainty is mirrored in the wider literature, which, whilst finding predominantly no association between level of intellectual functioning and hyporesponsivity in ASD (Ben-Sasson et al., 2007; Klintwall et al., 2011; Wiggins et al., 2009), has produced potential evidence of a

relationship between the two variables (Patten et al., 2013). The current review identified associations between intellectual functioning and hyporesponsivity extended only to individuals with *below average* cognitive function. This limitation is mirrored in the wider literature, which disproportionately considers children with ASD with lower levels of intellectual functioning. Research has not established whether hyporesponsivity is present in individuals with ASD and average ability when compared to a comparison group, and future efforts should be expended in this direction. No difference was identified in the finding of hyporesponsivity between papers using an Autistic Disorder sample compared to those using an Autism Spectrum Disorder sample. This finding fits within a mixed literature, which has found both evidence for, and evidence counter to, an association of sensory responsivity with severity of ASD traits. Three studies which considered responsivity subtype within analyses found no association between level of ASD traits and level of sensory responsivity (Azouz et al., 2014; Lane et al., 2014; Uljarevic et al., 2016). Three studies which did not consider responsivity subtype found no evidence of an association (Hilton et al., 2010; Kern et al., 2007; Zachor & Ben-Itzhak, 2013).

The recruitment of a representative sample of individuals with ASD continues to present a challenge to researchers (Hulley et al., 2001; Woods et al., 2000). Studies were limited by their inability to offer a true population sample, drawing frequently upon clinic-attending individuals. Studies which potentially utilised a larger shared resource in the interests of increasing generalisability raise questions about potential non-independence of results (Cooper, 2009). Challenges of recruitment and attrition remain regularly unreported (Dowling & Weiner, 1997), as was the case in the majority of studies considered in the current review, and no study provided a comprehensive consideration of non-response bias. Future research considering sensory processing styles in ASD would be strengthened by use of population based rather than convenience samples, which are frequently gained from developmental disability clinic samples.

Directions for Future Research

This review recognises a number of limitations of the current literature, and recommendations for future research are identified. Progress in the field is hindered by inconsistency in definition and specificity of hyporesponsivity in ASD. Future studies might consider subgroups of hyporesponsivity, based on association and dissociation between neurological, physiological responsiveness, and behavioural responsiveness. Specifically, studies which consider the potential different causal mechanisms of hyporesponsivity will enrich understanding of this response. Whilst research has identified dissociations in somatosensory-behavioural and auditory-behavioural relationships, future research will most effectively consider all sensory modalities. A more established term or terms to refer to hyporesponsivity would support future work in the area. Future studies which go beyond use of parental report and behavioural observation will be necessary in identifying causal differences within presentations of hyporesponsivity. Limitations in the field of the focus on children with lower levels of intellectual ability mean that it is not possible to generalise findings, nor is it possible to comprehensively explain the possible association between level of intellectual functioning and sensory sensitivity. Given that approximately half of individuals with ASD have a level of intellectual functioning in the average range (Joseph, 2011); this focus represents a gap in the literature. Future studies which use children of average intellect will round out the literature in this respect. The finding of this review with respect to hyporesponsivity and age suggests that future studies should consider the use of samples from older childhood groups. The attempted replication of findings of hyporesponsivity in middle and older children's age groups (5-11, 12-18) is indicated, which may clarify potential trajectories of hyporesponsivity with respect to age. Finally, this review suggests that studies which use developmental delay comparison groups are over-represented in the field, and may now add little to our understanding of the nature of hyporesponsivity in ASD. More focused and clinically relevant comparison groups may now be used to address defined questions about the profile of sensory responsiveness in ASD.

Strengths and Limitations of the Review

This review controlled for bias in methodological analysis through use of three independent systematic raters, and reduced publication bias by author correspondence. No exclusion was made on the basis of language of publication, thus preserving access to the literature. This review is, nonetheless, subject to several limitations. Papers published in the field of neurology address the relationship between individuals with ASD and sensory processing, and it is highly likely that some of these studies would find neurophysiological evidence of atypical responsivity profiles (e.g. Azouz et al., 2014; Donkers et al., 2015; Schaaf et al., 2015). Whilst these studies did not meet eligibility criteria for the current review, and thus neurophysiological evidence was excluded, a full review of the field would undoubtedly yield findings of interest. In addition, studies which focused upon only one sensory modality were excluded from review. Whilst retaining focus to the current review, future inclusion and synthesis of single-modality studies may offer further evidence for a syndrome-specific profile of hyporesponsivity in ASD. The small sample size of studies included in the current review (n=10) potentially introduces further limitation in addressing the research question. Finally, it must be noted that the measure used to screen evidence was purpose-developed for the current review. Whilst this offers a high level of specificity to the measure, the psychometric properties of the tool have not been evaluated beyond the current study.

Conclusions

In summary, the current review finds cautious support for a profile of relative hyporesponsivity in children with ASD. Four factors that reduced variability in findings were: chronological age, type of comparison group, sensory measure, and quality of study. Hyporesponsivity was highest in children with ASD aged ≤ 5.4 years, in comparison to a DD group, using the Sensory Experiences Questionnaire, and in higher quality studies. It was not possible to establish hyporesponsivity in adults, in children of average ability with ASD, or in comparison to non-DD groups due to limitations in the field. Higher quality studies provided conflicting evidence for a relationship between level of intellectual functioning and hyporesponsivity. Studies in the current review addressed solely

individuals with lower than average level of intellectual functioning, and thus findings cannot be generalised to individuals with average ability and ASD. Future contributions to the literature base could usefully consider measuring levels of hyporesponsivity by sensory modality in individuals with average ability, across an older childhood sample (≥ 5.4 years). Potential comparison groups which utilise a more focused DD group, or a neurodevelopmental disability group, would enrich the field beyond the current plethora of TD or mixed DD samples.

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Figure 1. Subtypes of sensory modulation disorder within the overall umbrella of sensory processing

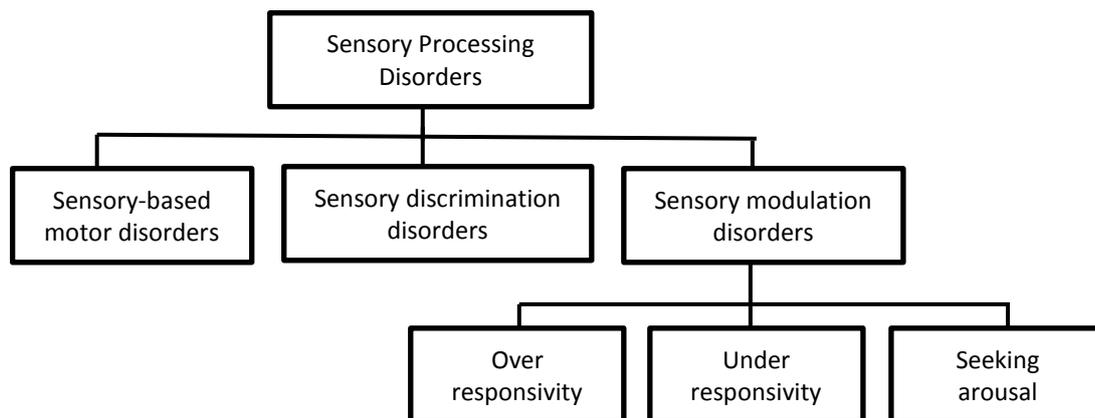


Table 1
Quality of Evidence Screening Tool

Criterion	Quality assessment	Rating criteria	Area summary
Construct validity			
1. The construct of sensory under-responsiveness/hyporesponsivity is defined * Alternative suitable terms may include the following: hyporeactivity, hyposensitivity, underreactivity, low registration, high threshold.	Excellent	The term 'hyporesponsivity' (or an alternate suitable term*) is comprehensively operationalised, leaving the reader clear as to the exact definition of the term as employed within the study. This definition may be provided either in the introduction or discussion sections.	
	Satisfactory	The term 'hyporesponsivity' (or an alternate suitable term*) is introduced, and a definition offered. This definition will differ from an 'Excellent' rating in that the exact definition is not thoroughly clarified, however an acceptable understanding of the construct as employed in the paper is gained.	
	Poor	'Hyporesponsivity' (or an alternate suitable term*) is introduced, although both context and definition is minimal, leaving a non-expert reader uncertain as to the exact meaning of the term as employed within the paper.	
	Not reported	'Hyporesponsivity' (or an alternate suitable term*) is not introduced within the background to the study.	
2. The sensory measures used are appropriate to capture the construct of hyporesponsivity: *Examples of measures which would meet 'Excellent' criteria are (but are not limited to): SEQ (Sensory Experiences Questionnaire) SSP (Short Sensory Profile) SP (Sensory Profile)	Excellent	Sensory measures are fully appropriate to capture hyporesponsivity, referring directly to this construct (but possibly using the terms hyposensitivity, low registration, hyporeactivity)*	
	Satisfactory	Sensory measures are appropriate to capture hyporesponsivity. Whilst the measure was not developed for the purpose of measuring hyporesponsivity, the study makes the method by which this is measured clear.	
	Poor	It is not made clear whether the measure is capable of capturing this construct.	
	Not reported	There is no discussion of the suitability of the measures used to capture this construct.	
Criterion			
Quality assessment			
Rating criteria			
Area summary			
3. The validity of the sensory measures within the relevant population(s) have been addressed	Excellent	The validity of the sensory measures within the relevant population(s) have been fully addressed with information of studies testing validity provided and full validity statistics reported.	
	Satisfactory	The validity of the sensory measures within the relevant population(s) has been fully addressed with information of studies testing validity provided. The reader is able to access validity information using this.	

		Poor	The validity of the sensory measures within the relevant population(s) has been claimed with no supporting information provided.
		Not reported	Information on validity is not provided.
4.	The sensory measure is capable of measuring sensory processing within individual modalities, and this is reported (i.e. tactile, auditory, etc.)	Excellent	It is evident which modalities are affected by hyporesponsivity or other forms of sensory processing abnormality. Report is provided for each modality, even where no statistical significance was found.
		Satisfactory	It is evident precisely which modalities are affected by hyporesponsivity or other forms of sensory processing abnormality, and a reference to these is offered without provision of full data per modality (e.g. reference is made to significant hyporesponsivity is auditory and taste, without discussion of findings in other modalities)
		Poor	It is unclear which modalities were found to be affected, OR a report referring to overall profiles of responsivity was provided, leaving the reader unable to determine sole modality performance.
		Not reported	No information provided.
5.	The procedure used to isolate the construct of hyporesponsivity using each measure is clear	Excellent	Information provided clarifies the method of isolating the construct, and reports clearly. It would be possible for the reader to recalculate the level of hyporesponsivity, if provided with the raw data.
		Satisfactory	Information provided largely clarifies the method of isolating the construct, but omits the finer detail of this procedure.
		Poor	Information provided is insufficient to clearly understand how the construct was isolated.
		Not reported	Procedural information is not provided.

Overall Construct validity:

Excellent
Satisfactory
Poor
Not reported

Criterion	Quality assessment	Rating criteria	Area summary
External and internal reliability			
6.	The reliability of the sensory measures within the relevant population(s) have been addressed	Excellent	The reliability of the sensory measures within the relevant population(s) have been fully addressed with information of studies testing reliability provided and full reliability statistics reported (both external and internal).
		Satisfactory	The reliability of the sensory measures within the relevant population(s) has been fully addressed with information of studies testing reliability provided. The reader is able to access reliability information using this, however full details of both external and internal reliability may be missing.
		Poor	The reliability of the sensory measures within the relevant population(s)

			has been claimed with no supporting information provided.
		Not reported	Information on reliability is not provided.
External validity (generalisability)			
7.	Eligibility (inclusion/exclusion) criteria and demographic information about the target (ASD) group are comprehensively stated	Excellent	Eligibility criteria are comprehensively stated, including <u>key areas of age, general level of ability, gender, and comorbidities</u> . Demographic information is provided for age, gender, general level of ability, comorbidity, and ASD type. Standard deviations are provided where appropriate
	Cont'd: Eligibility (inclusion/exclusion) criteria and demographic information about the target (ASD) group are comprehensively stated	Satisfactory	Eligibility criteria are clearly stated, but lack one key area (<u>key areas of age, ability level, gender, and comorbidities</u>). Demographic information is provided for age, gender, and general level of ability, although this may be group means only.
		Poor	Eligibility criteria are stated, but lack at least two key areas. Scant demographic information is provided which would not allow for a rating of 'Satisfactory'.
		Not reported	Eligibility criteria are not reported Demographic information is not reported.
8.	Diagnostic assessment of the ASD group is appropriate	Excellent	The diagnostic assessment tool used was a standardised measure, and was the same assessment measure used for all participants
		Satisfactory	A mixture of different standardised measures was used for participants.
		Poor	Non-standardised measures were used in the diagnosis of participants.
		Not reported	Diagnostic information is not reported.
9.	Capacity for generalisability	Excellent	A population based sample is offered, and differences controlled for.
	This item is half weighted. Whilst the importance of generalisability is significant, of itself low generalisability would not threaten the conclusions drawn by the study.	Satisfactory	A population based sample is offered, but differences are unacknowledged or not controlled for.
		Poor	A selectively recruited sample is used (clinic based)
		Not reported	Representation information is not reported.
Overall External validity:			Excellent
			Satisfactory
			Poor
			Not reported
Internal validity (Bias)			Excellent
			Satisfactory
			Poor
			Not reported

10.	The matching process for the comparison group to the ASD group is appropriate, or differences have been controlled for	Excellent	Groups are matched on both CA and MA, OR CA and a general level of ability; OR It is clear that potential confounds related to age and ability level have been appropriately discussed and controlled for, potentially by post-hoc analyses.
		Satisfactory	Groups are matched on <u>one</u> aspect of age or general level of ability; OR Potential confounds related to age and ability levels are acknowledged, with some attempt to control for this. Post-hoc analyses are incomplete or lacking, although this is acknowledged and discussed.
		Poor	Groups are unmatched on any key area (age, cognitive functioning); OR While groups are matched on one aspect of age or ability level, there is no evidence of attempts to control for confounds related to age or ability level, and no evidence of post-hoc analyses of these variables or discussion of the same.
		Not reported	The matching process is unreported.
Internal validity (confounding)			
11	Adequate adjustments or analyses for potential confounds was carried out	Excellent	Confounds are thoroughly discussed; appropriate management is applied.
		Satisfactory	Confounds are managed, but may be less robust than in an 'Excellent' rating.
		Poor	Confounds are not discussed; no evidence of management/evidence of poor management.
		Not reported	No report of confounds.
Overall Internal validity (bias)			Excellent
			Satisfactory
			Poor
			Not reported
Overall Internal validity (confounds)			Excellent
			Satisfactory
			Poor
			Not reported
12	Statistical matters		
The study is sufficiently powered	Excellent	Power analysis is clearly reported, and is fully appropriate.	
	Satisfactory	There is no score of 'Satisfactory for this item'.	
	Poor	Reporting of power calculations is unreported or shows the design to be insufficiently powered.	

		Not reported	Power calculations are unreported.
13.	Reporting of statistical results evidences the strength of findings claimed	Excellent	Where appropriate, and for all analyses related to hyporesponsivity : P-values are reported Confidence intervals are reported Standard deviations are reported Effect sizes are reported
		Satisfactory	The above statistical results are offered, but for main or significant results only OR P-values, effect sizes and one of CI or SD are consistently reported.
		Poor	Statistical results are offered in a confusing or patchy manner, leaving the reader unsure as to aspects of findings related to hyporesponsivity .
		Not reported	Statistical results are not reported.

Overall statistical matters

- Excellent**
- Satisfactory**
- Poor**
- Not reported**

Key: ASD= Autism Spectrum Disorder; CA= Chronological Age; MA= Mental Age; ES= Effect Size; CI= Confidence Interval; SD= Standard Deviation

Table 2
Overview of reviewed studies

Author(s) Date published Title	Main aim	Participants (ASD), and sampling method	Comparison group	Sensory measures used	Main findings related to this review	QuEST-HA rating and most significant threats to conclusions
Baranek et al. (2006) Sensory Experiences Questionnaire: discriminating sensory features in young children with autism, developmental delays, and typical development.	To explore whether the SEQ could satisfactorily discriminate sensory features in children with ASD.	ASD=56 5-80 months Mixed sample: clinical, community, research database and school	PDD=24 DD/MR=33 Other DD=35 TD=110	SEQ	Evidence of greater hyporesponsivity compared to DD group	Decisive evidence Potential non- independence of results.
Baranek et al. (2013) Hyporesponsiveness to social and non-social sensory stimuli in children with autism, children with developmental delays, and typically developing children.	To consider the relationship of age to hyporesponsiveness.	ASD=63 11-105 months Mixed sample: clinical, community, research database and school	DD=47 TD=68	Sensory Processing Assessment (Play based observation) SEQ	Evidence for greater hyporesponsiveness from 6-60 months compared to DD and TD. No hyporesponsiveness after 60 months.	Decisive evidence Potential non- independence of results.
Kirby et al. (2015) Caregiver strain and sensory features in children with autism spectrum disorder and other developmental disabilities.	Exploration of caregiver strain comparing ASD and DD groups	ASD=71 2-12 years Mean 85.7 months Mixed clinical, community, research and school	DD=36 2-12 years Mean 88.8 months	SEQ	Hyporesponsivity was related to caregiver strain in the ASD but not the DD group.	Decisive evidence Potential non- independence of results.
Author(s) Date published Title	Main aim	Participants (ASD), and sampling method	Comparison group	Sensory measures used	Main findings related to this review	QuEST-HA rating and most significant threats to conclusions
O'Brien et al. (2009) Discriminating children with autism from children with	To discriminate ASD from LD using the Short Sensory Profile	ASD=34 Mean age 9.8 years (4.5)	LD=22 Mean age 9.3 years (5.0) TD=33	Short Sensory Profile	No overall profile of hyporesponsivity found when compared to the LD group.	Convincing evidence LD control group

learning difficulties with an adaptation of the Short Sensory Profile.		Specialist school sample	Mean age 9.7 years (5.3)			poorly defined.
Boyd et al. (2010) Sensory features and repetitive behaviors in children with autism and developmental delays.	Examination of the association between sensory processing and RRBs.	ASD=67 Mean age 51.69 months (17.07) Mixed sample: clinical, community, research database and school	DD=42 Mean age 49.45 months (24.19)	SEQ Sensory Profile Sensory Processing Assessment for Young Children Tactile defensiveness Discrimination Test	Evidence of greater hyporesponsivity than DD.	Convincing evidence Potential non-independence of results. DD control group poorly defined.
Brock et al. (2012) Temperament and sensory features of children with autism.	To explore differences in temperament, and the relationship between sensory responsiveness and temperament, between ASD and DD groups	ASD=54 36-84 months Sampling method unclear	DD=33	As for Boyd et al. (2010)	Evidence of greater hyporesponsivity than DD.	Convincing evidence Sampling method unclear. DD control group poorly defined.
Author(s) Date published Title	Main aim	Participants (ASD), and sampling method	Comparison group	Sensory measures used	Main findings related to this review	QuEST-HA rating and most significant threats to conclusions
Watson et al. (2011) Differential association between sensory response patterns and language, social, and communication measures in children with autism or other developmental disabilities.	To examine patterns of sensory responsiveness as factors accounting for variability in social-communicative symptoms in ASD or DD.	ASD=72 Mean age 52.3 months Mixed sample: clinical, community, research database and school	DD=44 Mean age 48.1 months	As for Boyd et al. (2010)	Evidence of greater hyporesponsivity than DD.	Convincing evidence DD control group poorly defined. Statistical management of unequal sample size unclear.
Cheung & Siu (2009) A comparison of patterns of sensory processing inn	To compare patterns of sensory processing between ASD and ADHD	ASD=72 2.7-11.6 years Clinical sample	ADHD=114 4.8-12 years TD=1840 3-10 years	Sensory Profile	No difference found between ASD and ADHD group in hyporesponsivity or hyperresponsivity.	Fair evidence Characterisation of the ASD sample

children with and without developmental disabilities.

limited to children accessing a psychiatric unit.

Statistical reporting is absent. Description is not provided of management of the unequal sample sizes used, raising questions as to the statistical findings claimed.

Author(s) Date published Title	Main aim	Participants (ASD), and sampling method	Comparison group	Sensory measures used	Main findings related to this review	QuEST-HA rating and most significant threats to conclusions
Joosten et al. (2010) Sensory processing and stereotypical and repetitive behaviour in children with autism and intellectual disability.	To explore sensory processing and RRBs in children with ASD/ID and ID	ASD/ID= 29 5-18 years Mean age 9.7 years Specialist school sample	ID= 23 5-18 years Mean age 9.5 years	Sensory Profile	No hyporesponsivity found when compared to ID alone.	Fair evidence Generalisability is low. Confounds have not been addressed. Relatively small sample size. ID control group poorly defined.
Freuler et al. (2012) Precursors and trajectories of sensory features: Qualitative analysis of infant home videos.	To explore precursors and trajectories of extreme sensory patterns in children with ASD	ASD=6 Video: 0-2 years Research database sample	DD=6 Video: 0-2 years	SEQ Sensory Profile Sensory Processing Assessment for Young Children Tactile defensiveness and Discrimination Test	Children with ASD found to exhibit hyporesponsivity when compared to DD group, and for this construct to remain stable to preschool age	Questionable evidence Whilst ecological validity was excellent, generalisability is low. Internal validity is poor, both in failure to acknowledge or address bias and confounds. Small sample size Reliability of measures is not

addressed.
Statistical reporting
is absent.
DD control group
poorly defined.

Key: ASD= Autism Spectrum Disorder; DD= Developmental Delay; MR= Mental Retardation; PDD= Pervasive Developmental Disorder; PDD-NOS= Pervasive Developmental Disorder-Not Otherwise Specified; LD= Learning Disability; ID= Intellectual Disability; TD= Typically Developed; SEQ= Sensory Experiences Questionnaire.
