A cross-syndrome approach to the social phenotype of neurodevelopmental disorders:

Focusing on social vulnerability and social interaction style

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Highlights

- Heightened social vulnerability is evident across multiple neurodevelopmental disorders.
- The limitation of IQ to explain social vulnerability is shown by a cross-syndrome approach.
- Atypical social interaction styles vary within and across neurodevelopmental groups.
- Social interaction styles make a unique contribution to heightened social vulnerability.
- Social phenotypes are best understood as distributed across diagnostic boundaries.

Abstract

Background: Following Annette Karmiloff-Smith's approach to cognitive research, this study applied a cross-syndrome approach to the social phenotype, focusing on social vulnerability (SV) and the factors that contribute to it. **Aims:** To (i) identify syndrome-specific differences in SV across four neurodevelopmental disorder (NDD) groups, (ii) determine the contribution of intellectual disability (ID), age or gender to SV, and (iii) explore its relationship with social interaction style (SIS). **Methods and Procedures**: 262 parents of children: Autism (n = 29), Williams syndrome (n = 29), Attention Deficit Hyperactivity Disorder (n = 36), Fragile X syndrome (n = 18), and Neurotypical (n = 150) reported on their child's SV, quality of SIS and other factors (ID, age, gender). **Outcomes and Results:** Heightened SV was not syndrome-specific. Instead it was found equally across NDD groups (and not in the neurotypical group), and independently of ID, age and gender. Different atypical SISs were also distributed across NDD groups and each were significantly related to SV, independent of the factors above and beyond neurodevelopmental diagnosis. **Conclusions and Implications:** The findings emphasise that social phenotypes are best understood as distributed across diagnostic boundaries and offer opportunities to further test the role of varied atypical SISs in the development of heightened SV.

What this paper adds

In the first cross-syndrome comparison of social vulnerability (SV) profiles and social interaction styles (SISs), the current study emphasised that neurodevelopmental disorders (NDDs) of Autism, Williams syndrome, Attention Deficit Hyperactivity Disorder, and Fragile X syndrome are equally associated with atypical and heightened SV and that this cross-syndrome effect is not associated with intellectual disability (ID), age or gender. Furthermore, the study showed that SV is associated with the presence of distinctive, atypical patterns of SIS and that these are also found within and across these diagnostic groups. The results substantially extend previous evidence on cross-syndrome variability in both SV and SIS, highlighting the case for non-specificity in the social phenotype of different NDDs. The study also indicates the potential contribution of SIS as a factor in heightened SV beyond the effect of diagnostic group and other factors such as ID, age and gender. Crucially these initial findings strongly support a cross-syndrome approach to the study of SV in NDDs, and make a case for further consideration of the role of atypical SISs in our understanding of SV and the development of social phenotypes more generally.

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1. Introduction

Annette Karmiloff-Smith pioneered a cross-syndrome approach to the study of cognition in neurodevelopmental disorders (NDDs; Brown et al., 2003; Paterson et al., 2006; Scerif et al., 2004). Her cross-syndrome approach has advanced the understanding of a wide range of phenomena, including language development (Kelly et al., 2013; Lindgren et al., 2009), face and emotion recognition (Annaz et al., 2009; Dimitriou et al., 2015; Martínez-Castilla et al., 2015), attention (Cornish et al., 2012; Scerif et al., 2004), sleep (Ashworth et al., 2013, 2017; D'Souza et al., 2020), psychopathology (Rodgers et al., 2012; Royston et al., 2019; Woodcock et al., 2009), sensory processing (Hannant et al., 2018; Heald et al., 2020) and social/adaptive behaviour (Hamner et al., 2019; Sumner et al., 2016; Williams et al., 2013).

The goal of a cross-syndrome design is often to identify differences in abilities between disorders while capturing variability. For example, Karmiloff-Smith encouraged researchers to study cross-syndrome associations in order to understand cognitive mechanisms that drive development in specific disorders. However, in addition to helping identify these mechanisms, the focus on cross-syndrome associations in itself illuminates invariance in some areas of functioning in comparison to specific differences (see also Asada & Itakura, 2012; Farran & Karmiloff-Smith, 2012). The aim of the current study was to apply a cross-syndrome approach to the social domain in order to explore specificity and/or invariance in particular aspects of the social phenotype. The main focus of the study is social vulnerability (SV), defined as "*the disadvantages faced by an individual while he or she endeavours to survive as a productive member of society*" (Jawaid et al., 2012, p. 335) or an "*impaired ability to detect or avoid potentially harmful interpersonal interactions*" (Pinsker et al., 2006, p. 109).

The cognitive and social mechanisms that drive the development of SV are not yet understood. Jawaid et al.(2012) has proposed that a combination of intellectual disability (ID) and atypical social behaviours result in heightened SV. However, this proposal has not been

tested and research to date has been carried out only with one or two specific groups. First, with respect to the role of ID, evidence is sparse but preliminary studies suggest that this may not be a primary influence. For example, initial evidence with adults with Williams Syndrome who have heightened SV indicates that they do not differ across levels of ID (Lough & Fisher, 2016). Evidence also shows that those with Autism Spectrum Disorder (hereafter 'autism') who have heightened SV may not have IDs (Hofvander et al., 2009). Second, with respect to the role of atypical social interaction behaviour as a predictor of SV, to our knowledge there is no evidence available on this. Therefore, the current study explored and described for the first time the cross-syndrome variability of SV across five neurodevelopmental groups and the contribution made by ID and atypical social interaction style (SIS) as well as by other factors such as age and gender. Given the lack of previous evidence in this area, this exploratory method offers the potential to elicit factors relevant to understanding the development of heightened SV.

The current study focused on the relationship between SV and the variables above across a broad range of NDD groups (Autism, Williams Syndrome [WS], Attention Deficit Hyperactivity Disorder [ADHD], Fragile X Syndrome [FXS]) and neurotypical development [TD]. The motivation for including the four NDDs as well as TD was three-fold. First, all four NDDs are characterised by unusual social interactions in the literature. Social difficulties are definitive for autism (American Psychiatric Association, 2013) and an unusual over-approaching SIS is associated with WS (Doyle et al., 2004; Jarvinen et al., 2013; Järvinen-Pasley et al., 2010; Riby et al., 2014). FXS is associated with high social motivation alongside significant social anxiety and social communication difficulties (Cordeiro et al., 2011; Kau et al., 2004; Kaufmann et al., 2004; Roberts et al., 2007). Many children and youth with an ADHD diagnosis also show socio-cognitive impairments in areas of social problem-solving and perspective taking (Bora & Pantelis, 2016; Sibley et al., 2010) and experience interpersonal challenges, including an absence of mutual friends (Bagwell et al., 2001; Hoza et al., 2005), less stable and lower quality friendships (Normand et al., 2013) and high rates of peer rejection and victimisation (Holmberg & Hjern, 2008; Taylor et al., 2010). For an overview of peer difficulties in ADHD see Gardner & Gerdes (2015).

Secondly, we know there is significant within-disorder heterogeneity in all areas of cognition and behaviour in developmental disorders (Charman, 2015; Masi et al., 2017; Porter & Coltheart, 2005) as well as in TD. Research emphasises the overlapping characteristics between syndromes and a potential lack of discrete diagnostic boundaries at the behavioural level (Asada & Itakura, 2012; Bishop & Rutter, 2009; Dyck et al., 2011; Kaplan et al., 2006; Moreno-De-Luca et al., 2013; Zorlu et al., 2015). For example, studies often adopting a cross-syndrome approach have revealed the many shared social features between individuals with Autism and WS (Asada & Itakura, 2012; Hamner et al., 2019; Klein-Tasman et al., 2009; Vivanti et al., 2018). Consequently, the field has moved away from the notion of these two neurodevelopmental conditions as polar opposite of social functioning. Utilising a cross-syndrome approach to the study of SV and SIS in a much broader range of neurodevelopmental groups should help pinpoint where there are both group differences and shared features.

Finally, variability in social interaction abilities is also found in the TD population and therefore the inclusion of a TD group allows us to consider the behaviours that fall within the range of 'typical' variation including the extremes of individual differences. In a study design that examines both syndrome differences and cross-syndrome similarities, the issue of 'typicality', can only be considered by the inclusion of a TD group.

Both SV and SIS were operationalised using established methods. However, as these methods were adapted for the study, the measurement format of each construct was tested for the first time. SV was measured using a subset of items from the Social Vulnerability Questionnaire (Fisher et al., 2012), while SIS was measured using Wing and Gould's (1979) clinical classification system of SIS subtypes (see Scheeren et al., 2020 for recent description).

In summary, the present study aimed to address the following research questions: First, are there syndrome-specific differences between NDD groups in SV? Second, can SV be explained by other factors such as ID, age, or gender? Third, is there a cross-syndrome *association* between SV and SIS and if so, does SIS itself make a unique contribution to SV, independent of other factors above?

2. Method

2.1. Participants

276 parents or guardians were recruited for the study. Data from 14 participants were removed prior to analysis due to parents reporting that their child did not meet the inclusion criteria because they (i) presented with a variety of difficulties changeable over time or had a diagnosis beyond the focus of the study (n = 5), (ii) fell outside of the age range (n = 1), or (iii) were recruited to the TD group but parents reported an intellectual disability or presence on the special educational needs register (n = 8). The final sample included parents or guardians of 262 4- to 17-year-old children (M_{Age} = 112 months, SD = 42.43) living in the UK (93% Mothers), of which 118 were parents of children with a diagnosis of a NDD and 150 were parents of TD children. The children were categorised into 4 NDDs: Autism (n = 29), WS (n = 29), ADHD (n = 36), FXS (n = 18; see Table 1). Parents were recruited through a university research participation database for local families, social media, and via UK charity networks (e.g. Williams Syndrome Foundation, ADHD Foundation, and Fragile X Society). The study complied with ethics (as per BPS requirements) and GDPR legislation (as per University requirements) and received favourable ethical opinion from the local ethics committee. Parents opted-in to the study and were not reimbursed for their time.

Age was normally distributed for each of the neurodevelopmental groups but not for the TD group. Preliminary analysis using Kruskall-Wallis analysis across all 5 groups (M_{Age}: TD = 107 months, Autism = 127 months, WS = 100 months, ADHD = 126 months, FXS = 120 months) found a main effect of chronological age ($H(4) = 16.25$, $p = .003$) however this difference was confined to a difference between the TD and NDD subgroups (specifically Autism and ADHD groups). Follow up tests using the Bonferroni correction revealed no significant differences in age between the four NDD subgroups.

The NDD subgroups differed in parent-reported ID status as seen in Table 1, $\chi^2(df = 3) = 50.98$, p < .001. With respect to language, parents reported that the majority of participants in all groups had expressive language and receptive language at the level of full sentences, Table 1. However, the number of children with and without full sentences differed between

the four NDD subgroups for receptive language χ^2(df = 3) = 10.41, p = .015) and expressive language χ^2(df = 3) = 24.31, $p < .001$).

2.2. Materials

Parents completed a bespoke online questionnaire about their child's social functioning and social interactions, via Online Survey software (www.onlinesurvey.ac.uk). Of the items included in the online questionnaire, only the quality of SIS and SV items are reported here[1]. A set of demographic questions were asked at the end (e.g. parents provided their child's date of birth, gender, Special Educational Needs status (SEN) and gave information about diagnosis, schooling and presence of ID, some of which are provided below in terms of describing the sample [also see Table 1]).

[TABLE 1]

2.2.1. Measure of level of SV

SV was measured using nine items from the Social Vulnerability Questionnaire (SVQ). The SVQ is a 30-item parent-report measure of vulnerability, which taps *Emotional Bullying, Risk Awareness, Social Protection, Perceived Vulnerability, Parental Independence* and *Credulity* (Fisher et al., 2012). The SVQ was validated on 144 parents of individuals with intellectual and developmental disabilities and has previously been used to examine SV in autism, WS and Downs syndrome (Fisher et al., 2012, 2013; Lough & Fisher, 2016). As the SVQ focuses on many broader issues of vulnerability than the current study aimed to explore (e.g. physical threat) we selected nine items specifically social in nature (SVQ items 1, 3, 4, 12, 13, 14, 16,

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[1] This research formed part of a larger study exploring social interactions in children with and without developmental disabilities

19, 25). Parents rated statements on a Likert scale of 0 ("not true/never") to 3 ("very true/always"). Potential SV total scores (SV-Total) ranged from 0-27, with a higher SV-Total score indicative of greater SV. Cronbach's alpha for the nine SV items on the current total sample was 0.87 (per NDD subgroup: TD: $\alpha = 0.69$, Autism: $\alpha = 0.70$, WS: $\alpha = 0.64$, ADHD: $\alpha = 0.7$, FXS: $\alpha = 0.81$).

2.2.2. Measure of SIS

SIS was measured using Wing & Gould's (1979) original clinical classification system of SIS subtypes ('typical', 'aloof', 'passive' and 'active-but-odd (hereafter 'active-but-unusual'). This classification system shows good internal consistency when used in a parent questionnaire format (Castelloe & Dawson, 1993; Roeyers, 1997; Scheeren et al., 2012). The classification also shows good external validity (Borden & Ollendick, 1994; Waterhouse et al., 1996). Extended versions of the classification system have been developed and these have been validated in behavioural observation studies using a single checklist judgement by naïve observers (Roeyers, 1997) and in parent interview studies with judgements by interviewers blind to diagnosis (Leekam et al., 2002; Wing et al., 2002). The current study followed the extended interview classification developed by Wing (2006) described in Kent (2014) and used within the validation study of Leekam et al., (2002). For the current study it was adapted into a parent questionnaire context (see Table 2). Parents/guardians were asked to select one description that best described their child's social interactions. From the original classification, five subtypes were selected, four of which were, 'typical', 'aloof', 'passive' and 'active-but-unusual. The fifth, "shy but social contact is appropriate for mental age with well-known people, including age peers" was also selected because together with the 'typical' description it formed part of the original measure of 'appropriate' social styles previously validated (Leekam et al., 2002). Several subtype descriptions were combined to form the 'aloof' subtype. Subtypes that specifically referred to (a) WS and (b) FXS, and also the subtypes; "selective mutism" and "over- formal, stilted, rigid, over-polite and calmly outspoken" were excluded.

[Table 2]

2.3. Analytic Approach

Shapiro-Wilk test of normality indicated that the data for SV-Total was not normally distributed for the sample as whole, or for the TD and Autism groups (when normality tests were run per diagnostic group). Therefore, nonparametric tests were used but results from parametric tests were reported if they did not differ. To examine RQ1, group differences in SV were tested using Kruskal-Wallis H test / one-way ANOVA. To examine effects of ID status, age and gender (RQ2) Spearman's correlations and Mann Whitney tests/ t tests were used. Finally, to examine associations between SIS and SV and whether SIS was uniquely related to level of SV (SV-Total score), a multiple regression analysis was conducted with SIS subtypes as the predictor variables, and SV-Total as the dependent variable (RQ3), while statistically controlling for age, gender, ND status and ID status. For all tests, an alpha value of .05 was set, unless multiple comparisons required Bonferroni adjustment. In addition, analyses were re-run to equalise the size of the TD comparison group.

3. Results

3.1. SV and NDD

Mean SV-Total scores for each diagnostic group are shown in Table 1. To examine differences across groups (TD, Autism, WS, ADHD and FXS), SV-Total scores were analysed using a one-way ANOVA. A significant group difference was found, $F(4,257) = 90.81$, $p < 0.001$, $\eta_p^2 = 0.59$. Scheffe post hoc comparisons revealed that SV-Total score for the TD group was significantly lower than all four NDD subgroups (all p's < .001; see Table 1). Pairwise comparisons showed no significant difference in SV-Total score between each of the NDD subgroups (all p's > .05). Atypically heightened SV was a feature of NDD diagnosis and distinctive from TD, but it was not syndrome-specific; instead scores were equivalently elevated across all the four NDD subgroups. Given the unequal size of the TD group, the ANOVA was re-run using the first 35 respondents who were recruited into the TD group. Results showed a significant group difference in SV-Total score $F(4,142) = 38.38$, $p <$ .001, $\eta_p^2 = 0.52$. As above, mean SV-Total score for the TD group was significantly lower

than all four ND groups, with no significant difference between the ND groups. The result from the full sample analysis was therefore maintained.

3.2. SV and age, ID and gender

SV-Total score was not related to Age for the sample as a whole (TD, Autism, ADHD, WS, FXS combined; $r_s(259) = .08$, $p = .21$), or for the NDD group taken together ($r_s(259) = .10$, $p = .29$). Follow-up comparisons for each sub-group also showed no significant SV correlation with Age for the Autism, WS, or ADHD groups (all p's > .05) and although there was a significant, positive relationship between SV-Total score and Age for the FXS group ($r_s(17) = .54$, $p = .03$) and a significant negative relationship for the TD group ($r_s(149) = -.19$, $p = .02$, the significance level did not survive when Bonferroni adjustment for multiple comparison was applied ($.05/6 = p.01$)

For the analysis of ID, only the NDD subgroups were included as none of the TD group had ID. For the NDD group as a whole, there was no significant difference in SV-Total score associated with the presence of ID ($M = 16.67$, $SD = 4.66$) / absence of ID ($M = 15.96$, $SD = 5.24$), $t(104) = 0.74$, $p = .46$, $d = 0.14$. For Gender, taking the sample as a whole, there was a significant difference in SV-Total score due to Gender, with higher SV reported for males ($M = 11.70$, $SD = 7.12$) than females ($M = 8.62$, $SD = 5.70$); $t(248.93) = 3.87$, $p < .001$, $d = 0.48$. This difference was not significant when all NDD subgroups were analysed together ($t(64.44) = 1.03$, $p = .31$, $d = 0.21$), but was significant for the Autism group independently, as males ($M = 18.52$, $SD = 5.13$) scored higher than females ($M = 14.38$, $SD = 3.54$; $t(18.53) = 2.47$, $p = .02$, $d = 0.94$), although this effect did not survive Bonferroni adjustment ($.05/4 = p.01$). Note that there was a substantial imbalance in gender in the autism group and other NDD groups (see Table 1 for a breakdown of gender per group). There was no significant gender difference for each of the remaining four developmental groups (including TD) (all p's > .05). The aforementioned results remained unchanged when the analysis was applied with the reduced TD sample.

3.3. SV and SIS

To examine the relation between SV and different types of SIS, several analyses were conducted. First, the relation between SV-Total score and SIS was explored for the whole sample independently of NDD subgroup status. An initial ANOVA, with SIS subtype as the independent variable (5 categories as shown in Table 2) and SV-Total score as the dependent variable showed a significant effect of SIS subtype on SV-Total score, $F(5, 256) = 36.02$, $p < .001$, $\eta_p^2 = 0.41$. Post hoc comparisons also revealed that children with SIS subtypes 'appropriate (1)' ($M = 7.03$, $SD = 5.06$) and 'shy (2)' ($M = 9.03$, $SD = 6.00$) had significantly lower SV-Total scores than children with the atypical SIS subtypes 'active-but-unusual (3)' ($M = 16.69$, $SD = 4.60$), 'passive (4)' ($M = 16.34$, $SD = 5.94$) and 'aloof (5)' ($M = 16.67$, $SD = 4.80$). However, SV-Total scores did not differ between 'appropriate' and 'shy' SIS subtypes ($p > .05$). Neither were differences found between each of the three atypical SIS subtypes (all p's > .05). This result remained even when subtype 5 'aloof' (n = 6) was collapsed with subtype 4 'passive' (n = 29) into a social withdrawal subtype ($M = 16.40$, $SD = 5.70$), and compared with subtype 3, 'active-but-unusual' (n = 51; $M = 16.69$, $SD = 4.60$) due to unequal samples (*t* test; $p = .8$, $d = 0.06$). The results reported above were maintained when analysed using the reduced TD sample.

Next, to examine whether each of the atypical SIS subtypes uniquely predicted SV independent of other factors, including diagnostic status, a regression analysis was conducted. The SV data were entered for the whole sample, including TD data (original sample) in order to increase variability. An initial model was run with only SIS subtypes as predictor and SV-Total as the dependent variable. The model generated (adjusted $R^2 = .40$) was a significant predictor of overall SV-Total score, $F(4,257) = 44.58$, $p < .001$, with each of the four interaction subtypes entered (excluding "appropriate") making a significant contribution to the model (all p's < .05). In order to probe the unique contribution of SIS, a sequential, multiple-regression strategy was conducted where Age, Gender, ID status (presence/absence of ID) and NDD status (the presence of a NDD compared to TD) were entered in Model 1; Model 2 added SIS subtypes. Of the variables entered in Model 1, only NDD status was a significant predictor of SV-Total ($p < .001$). Age ($p = .39$), Gender ($p = .68$) and ID status ($p = .43$) did not significantly contribute therefore, the regression was rerun

with only NDD status statistically controlled for. The regression revealed that at Model 1, NDD status entered alone contributed significantly to the model ($p < .001$) and accounted for 57.3% of the variation in SV-Total. Introducing the SIS subtypes explained an additional 5.3% of variation in SV-Total and this change in R^2 was significant ($p < .001$). With ND status statistically controlled for, the SIS subtypes of *active-but-unusual, passive* and *aloof* significantly contributed to the model (all p's <.001). The SIS subtype *shy* was no longer a significant predictor of SV-Total ($p = .2$).

4. Discussion

Inspired by Annette Karmiloff-Smith's approach to cognitive research, this study applied a cross-syndrome approach to the social phenotype; focusing on SV and its relation to SIS, ID and other factors.

4.1. Are there syndrome specific differences in SV?

One of the main findings was that heightened SV was found across multiple NDDs. Parents/guardians in the Autism, WS, ADHD and FXS groups all endorsed higher levels of SV compared to parents of neurotypical children. While autism and WS are already known to be two particularly socially vulnerable populations (Fisher et al., 2012, 2013; Griffiths et al., 2019; Jawaid et al., 2012; Lough et al., 2015; Lough & Fisher, 2016; Riby et al., 2017; Sofronoff et al., 2011) this is the first evidence of heightened SV in ADHD and FXS groups. This finding suggests that heightened SV may be a clinical phenomenon that is a shared feature of NDDs, even those distinguished by specific genetic and biological aetiologies.

To date we know little about the developmental mechanisms of SV and further research beyond this study will be needed in order to isolate and test out these mechanisms. Our starting point was to address Jawaid et al's. (2012) proposal that a combination of ID and atypical social behaviours contribute to heightened SV and we used parent questionnaires to explore the concurrent contribution of these and other factors (age, gender) to SV.

4.2. Can SV be explained by ID, age, or gender?

First, we found, like previous studies, that ID did not fully explain SV (Hofvander et al., 2009; Lough & Fisher, 2016; Wilson et al., 1996). Comparable, heightened SV was also found in the NDD groups not characterised by ID (ADHD and Autism, of whom 72% and 79% did not have an ID as reported by parents, respectively).

Age also did not explain the presence of heightened SV scores either for the whole sample or for the NDD groups, except for small correlations with FXS and TD which did not survive adjustment for multiple testing. Although these findings for IQ and age might be surprising, the participants were young. Therefore, further research should investigate whether the lack of age and ID would replicate in older groups of individuals. A major limitation is also that our measure of ID was limited by parent-report ID (yes/no response) and formal standardised measurement of ID would be needed in order to probe this more accurately. Finally, gender also did not significantly contribute to SV although there were indicative findings of higher scores in males in the autistic group only. However, inequality in gender grouping size constrained the analysis. Further research is needed with matched gender samples to clarify these effects.

The results for ID, age and gender support other studies using the SVQ (e.g. Lough & Fisher, 2016) and extend findings of heightened SV for the first time to ADHD and FXS, as well as consolidating the finding of reduced SV within a large sample of neurotypical children. As the study used only a small subset of items from the longer SVQ this may indicate the effectiveness of this format for this purpose. However, while internal consistency was good, the lack of psychometric testing on this abbreviated version was a limitation, potentially restricting its capacity to capture differences and further research comparing the question sets is needed.

Nevertheless, these findings emphasise that SV may potentially be an issue that transcends diagnostic boundaries, irrespective of ID, and this reinforces the view that "*individuals with intellectual and developmental disabilities can be vulnerable in multiple, potentially*

unrelated ways, and it is important for researchers and clinicians to try to capture these distinct patterns of vulnerability" (Fisher et al., 2018, p. 8).

4.3. Understanding SV through the lens of SIS

To explore the role of SIS, we used the classification of quality of social interaction based on Wing and Gould's original typology (1979), drawing on Wing's (2002, 2006) clinical classification and applying this within a parent questionnaire. Five SIS subtypes were analysed; three atypical (aloof, passive and active-but-unusual) together with two appropriate styles (typical and shy). Although Scheeren et al., (2012) found an active-but-unusual style in children with autism who were also reported as having ADHD features and/or disruptive or social-emotional behaviours, this is the first study to apply Wing & Gould's classification system to those with diagnoses beyond autism and pervasive developmental disorder. The results showed that atypical SIS was strongly associated with SV and this association was not specific to any particular type of atypical SIS subtype (aloof, passive, active). Furthermore, like the results for SV we also found no syndrome-specific effects; atypical SIS was found across all the NDD groups.

To further explore the possibility of a unique contribution made by SIS, the regression analysis revealed that each of the atypical SISs (aloof, passive and active) made a significant contribution to SV even when neurodevelopmental status was accounted for. In contrast the shy style did not significantly contribute. However, the magnitude of contribution made by SIS to SV was smaller than that of NDD status. Furthermore, of the child characteristics examined (ID, age, gender), only the presence /absence of a neurodevelopmental diagnosis was a significant predictor in the model, indicating a qualitative difference between typical and atypical development (Autism, WS, ADHD or FXS) regardless of a range of other factors.

In summary, these findings show that atypical SIS uniquely contributes to SV. Importantly not one, but each of these SISs separately (aloof, passive, active-but-unusual) were significant predictors of SV. Each of these SISs are very different from each other, yet each may still be relevant to the characterisation of any NDDs. Given that between half to three

quarters of the children in all NDD groups studied here were classified with one of these particular social interaction profiles (see Table 2), it is possible that these profiles are not part of the normal distribution across the population and that difficulties with social interaction may serve as a consistent flag or indicator when clinical neurodevelopmental diagnosis is considered.

4.4. Considerations and future research

The current study has a number of limitations that require consideration. In terms of measurement, the two main measures had been adapted from pre-existing measures and used for the first time in this research. While the adapted measure of SV showed good internal validity, an assessment of external validity is needed. Similarly, although the SIS checklist judgement method used for the first time with parents appeared to be effective, it requires validation against clinical judgement and in comparison with the well-established Wing Subtypes Questionnaire (e.g. Castelloe & Dawson, 1993). A considerable measurement concern is also that both measures collected parent information, raising the possibility of informant bias and follow up studies using cross-informant analysis and other forms of testing are needed. In terms of design, correlational studies of this kind are insufficient to provide insight into the directionality of the relationships between variables. An experimental design would help to disentangle the concepts of SV, for example, by separating particular types of individual style (e.g. aloof, passive, active-but-unusual) from particular types of behaviours by others (e.g. taunting, exploiting a child (e.g. for favours) or rejecting a child's social approach). An experimental design would also be necessary in order to test the effect of interventions, both to support children and reduce stigma and victimisation by other people.

It has been argued that the SIS of children with autism may be a predictor of intervention success, with several studies giving insights on how to tailor interventions to support different children with different SIS (Begeer et al., 2015; Beglinger & Smith, 2005). Also, studies with autistic children show that an active-but-unusual style is seen more commonly in older children with higher IQ and that these children have less severe autism symptoms especially across time (Scheeren et al., 2020), while those classified as 'aloof' make fewer

improvements after intensive intervention (Beglinger & Smith, 2005). Our findings across NDD groups suggest the need for further exploration of this evidence, given that half of the WS group also show the active-but-unusual SIS accompanied by ID. For this group, an approaching social style may lead to greater social learning opportunities while their ID may limit their ability to take up the opportunity to learn. In the case of higher IQ in children with autism and ADHD, their higher IQ may be a protective factor to enable them to learn and adapt to complex social challenges and in turn possibly change their social interactions (Scheeren et al., 2020).Yet even for this group, at some point the demands of complex social environments may exceed adaptive capacity.

In the current study, presence of ID was controlled in the analysis as we examined the contribution of SIS on SV; however future studies should further examine the role played by cognitive ability, both general IQ and specific cognitive skills (e.g. executive functioning, theory of mind) in a cross-syndrome approach extending existing work on SIS and autism intervention (Begeer et al., 2015; Beglinger & Smith, 2005). Like these studies, future work should be directed towards developing interventions that are sensitive to SIS. However new work should also focus on SV to provide a cross-syndrome understanding of how SIS and adaptive cognitive skills can help the individual to buffer particular kinds of challenges that they face in the social environment. Such focus on SV ensures that interventions can also work to assess and intervene on disadvantages experienced by the individual that can be identified in their social environment, including the contribution of other individuals, the social group and organisations.

In summary, the current study is the first to compare SV profiles and the role of SIS, in TD children and children with a range of NDDs. While exploratory in nature, this study provides preliminary evidence that SISs may play an important role in SV and opens up potential avenues for future research to delineate the nature of the association more comprehensively. We know that "equivalent behavioural outcomes stem from different underlying processes" (Karmiloff-Smith, 1997, p. 513), therefore studies adopting a cross-syndrome approach are key in understanding whether pathways to SV are the same or different across neurodevelopmental groups.

Declaration of Competing Interest

None.

Table 1. Demographic characteristics of the sample (% reported) and SV-Total, split by diagnostic group

	Autism (N = 29)	WS (N = 29)	ADHD (N = 36)	FXS (N = 18)	TD (N = 150)
Males/females/prefer not to say	72/28/0	59/41/0	78/19/3	94/6/0	48/51/1
Age					
Mean (SD) (months)	127 (28.4)	100 (36.3)	126 (35.5)	120 (43.7)	107 (45.8)
Range (months)	59-187	48-204	54-197	52-178	48-215
Presence of a physical disability	21	21	3	17	1
Presence of a hearing impairment	0	10	6	0	0
Presence of a visual impairment	10	37	14	11	6
Presence of an intellectual disability	21	90	28	89	0
Stage of education					
Preschool	3	10	3	11	7
Primary	62	77	53	45	63
Secondary	35	10	44	45	27
Post-16 education	0	0	0	0	3
Educational provision					
Mainstream school	66	50	86	22	97
Special Educational school	17	43	11	67	0
Home-schooling	10	3	3	0	3
Other[1]	7	3	0	11	0
Special educational needs register					
Yes	69	69	50	67	0
No	17	3	33	6	98
I don't know	14	24	17	28	2
Statement of SEN/EHCP					
Yes	52	90	33	89	-
No	45	7	61	6	-
I don't know	3	3	6	6	-
Use of language to communicate					
None	3	7	0	11	1
Single words	3	7	0	17	0
Simple phrases	7	24	6	33	0

Full sentences	86	62	94	39	99
Understanding of language					
None	0	0	0	6	0
Single words	0	7	0	0	0
Simple phrases	17	28	6	28	0
Full sentences	83	66	94	67	100
SV-Total					
Mean (SD)	17.83 (5.05)	17.07 (4.29)	14.83 (4.61)	16.78 (5.61)	6.08 (4.01)

[1]Four parents reported 'other'. This included children not currently in educational provision (due to pupil/school Choice) and pupils with a mix of provision.

Table 2. Percentage of children within each Quality of Social Interaction Style subtype across diagnostic groups (%)

Quality of Social Interaction (%)	Interaction label	Group					
		Autism (n=29)	WS (n=28)	ADHD (n=36)	FXS (n=17)	TD (n=150)	NDD[3] (n=112)
1. Social contacts with children and adults are appropriate for level of ability. Looks up with interest and smiles when approached. Responds to the ideas and interests of people of similar mental/developmental age and contributes to the interaction	Appropriate	6.9	31	38.9	0	74.0	22.3
2. Shy but social contact is appropriate for mental age with well-known people, including age peers. Might refuse to talk to adults but interacts with other children	Shy	17.2	0	8.3	23.5	17.3	10.7
3. Makes social approaches actively but these are usually inappropriate / the behaviour is not modified according to the needs, interests and responses of the person approached	Active-but-unusual	48.8	51.7	36.1	17.6	4.0	40.2
4. Generally does not initiate but responds to social contact if others make approaches. May join in passively and shows pleasure in passive role and may try to copy but with little understanding	Passive	24.1	13.8	13.9	41.2	4.0	20.5
5. Does not interact; aloof and indifferent (though may interact to obtain physical needs, including physical contact needs, rough and tumble play, cuddle)	Aloof	3.4	0	2.8	17.6	0.7	4.5
Unusual / inappropriate for mental age[1]		75.9	65.5	52.8	72.2	8.7	65.2
Typical / appropriate for mental age[2]		24.1	31.0	47.2	22.2	91.3	33.0

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[1] Descriptions 3, 4 and 5 collapsed to form one category
[2] Descriptions 1 and 2 collapsed to form one category
[3] NDD = Autism, Williams syndrome, ADHD and FXS collapsed to form one category

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