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The Quality of Everyday Eye Contact in Williams Syndrome: Insights from Cross-syndrome Comparisons

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Abstract

Past research shows that individuals with Williams syndrome (WS) have heightened and prolonged eye contact. Using parent report measures, we examined not only the presence of eye contact but also its qualitative features. Study 1 included individuals with WS (n=22, age 6.0–36.3). Study 2 included children with different neurodevelopmental (ND) conditions (WS, autism spectrum disorder, fragile X syndrome, attention-deficit hyperactivity disorder) and children with neurotypical development (NT) (n=262, age 4.0–17.11). Unusual eye contact features, including staring, were found in approximately half of the WS samples. However, other features such as brief glances were frequently found in WS and in all ND conditions, but not NT. Future research in ND conditions should focus on qualitative as well as quantitative features of eye contact.

Keywords: Williams syndrome; eye contact; neurodevelopmental condition; cross-syndrome comparison

2012; hyperarousal model, Hadjikhani et al., 2017), that autistic individuals have diminished eye contact. One problem is that the evidence for this view rests mainly on studies that report reduced frequency or presence of eye contact. However, there has been remarkable neglect in considering the nature of the quality of eye contact, which could possibly lead to a different understanding of eye contact in individuals with ND conditions. One reason for the past focus on quantity rather than quality is that much of the research knowledge on eye contact stems from a broader laboratory-based research tradition on eye gaze more generally, which tends to equate looking at the eyes of computerised facial stimuli with ‘eye contact’. While this paradigm affords a high level of experimental control, the passive viewing of socially-relevant stimuli is very different from how eye contact is experienced in everyday dyadic social interactions (see Kingstone, 2009). Research has shown that the realism of the stimuli used in social attention research (e.g. static versus dynamic images; isolated faces versus multiple faces in a social scene), impacts on eye contact (e.g. Hanley et al., 2013; Speer et al., 2007). Consequently, researchers have emphasised the importance of studying everyday situations to understand social attention in real-life interactive situations (e.g. Hanley et al., 2015; Kingstone, 2009; Risko et al., 2012). In the current investigation, we examine both the presence and quality of everyday eye contact of individuals with ND conditions, using the caregiver’s perspective of eye contact.

Eye contact behaviour in Williams syndrome

Williams syndrome (WS) is a genetic ND condition commonly associated with a heightened desire for social contact (termed “hyper sociability”; for a review of the WS social phenotype, see Thurman & Fisher, 2015). Indeed, WS is a really important ND condition to study various aspects of social behaviour because its genetic basis is well-defined (hemizygous deletion of ~25-28 genes on chromosome 7q11.23; Ewart et al., 1993), therefore research with this group has the potential to inform debate about gene-brain-behaviour links

73 and further our understanding of the “typical” social brain. Consequently, the WS social
74 profile has garnered a significant amount of research attention at the level of both brain and
75 behaviour. For example, evidence that WS is associated with structural and functional
76 atypicalities in key areas of the “social brain network” known to activate in response to eye
77 contact, such as the amygdala (Haas et al., 2009; Martens et al., 2009) and fusiform face area
78 (FFA; Golarai et al., 2010), has informed understanding of how different features of the WS
79 social phenotype may be subserved by neural substrates (for a review see Haas & Reiss,
80 2012). At the behavioural level there has been a great deal of interest in capturing various
81 aspects of social behaviour in WS, including eye gaze and eye contact behaviour. The
82 predominant evidence of gaze behaviour in WS comes from face scanning and eye-tracking
83 studies that have examined eye gaze behaviour towards images or movies on screen. These
84 studies show that the face, particularly the eye region, attracts and holds the attention of
85 individuals with WS for longer than is typical for children, adolescents and adults (Porter et
86 al., 2010; Riby & Hancock, 2008, 2009a, 2009b). This tendency for heightened, prolonged
87 looking to faces and eyes has been linked to a lack of habituation to faces (Järvinen et al.,
88 2012), to physiological reactivity and to attentional mechanisms related to arousal, suggesting
89 the possibility of hypo-arousal in this group (Doherty-Sneddon et al., 2009; Riby et al., 2012;
90 Skwerer et al., 2009, 2011).

91 Beyond laboratory studies using eye tracking and measuring gaze to computerized
92 images, a few other observational studies have also reported that young children with WS (<
93 5 years old) show intense and prolonged looking in real-world settings; during interactions in
94 clinics (Mervis et al., 2003) and with experimenters (Jones et al., 2000). Although studies
95 using a clinical measure, the Autism Diagnostic Observation Schedule (ADOS; Lord et al.,
96 2000), have reported up to 52% of children with WS had “definite abnormality” with eye
97 contact (Klein-Tasman et al., 2007, 2009), we know little about the nature of the unusual eye

98 contact, as the ADOS assessment does not capture quality features of eye contact. Given this
99 limited evidence of prolonged, intense eye contact in naturalistic settings, it is still not
100 established whether this quality of eye contact is common in individuals with WS, if it is a
101 feature distinctive to WS or frequently found in other ND conditions. Research that examines
102 eye contact behaviour in WS alongside other ND conditions will help to identify features of
103 eye contact that may be particularly distinctive to WS (syndrome-specific) or shared across
104 diagnostic groups (syndrome-general). See Asada and Itakura (2012) for further discussion.

105 **Eye contact behaviour across ND conditions**

106 While WS has been characterized by social interest associated with a heightened and
107 prolonged presence of eye contact, other ND conditions, particularly Autism, in contrast have
108 traditionally been associated with reduced presence of eye contact (Asada & Itakura, 2012;
109 Senju & Johnson, 2009). Reduced eye contact, in turn, has been connected to a lack of social
110 interest (Chevallier et al., 2012); an assumption that has been challenged by those with
111 subjective, lived experience of autism (Jaswal & Akhtar, 2019) who argue that reduced
112 quantity of eye contact does not necessarily equate with lack of interest. We propose that the
113 clarification of this issue has been hampered by a single dimensional approach to the
114 understanding of eye contact; that conflates presence and quality of eye contact.

115 Characterizing eye contact by a single dimension leads to a view that reduced eye contact is
116 poor eye contact and increased eye contact is good eye contact; an assumption that tends to
117 polarise the social phenotypes of ND groups into opposite profiles (see Asada & Itakura,
118 2012 for review of the Autism/WS distinction). By considering multiple qualitative features
119 of eye contact in everyday life contexts, across ND conditions, the current study attempts to
120 move away from examining eye contact through a quantitative, single dimensional lens.

121 Like studies of WS, much previous research on eye contact in autism has also tended
122 to focus on its presence or degree. Eye-tracking studies show that some autistic individuals

123 spend less time than is typical attending to face areas and eye areas on a screen (Sasson et al.,
124 2007; Shic et al., 2011). For reviews of the Autism eye tracking literature see Guillon et al.
125 (2014) and Papagiannopoulou et al. (2014). Both eye tracking studies (e.g. Hanley et al.,
126 2014, 2015) and face-to-face observational studies (e.g. Leekam & Ramsden, 2006) also find
127 differences in attentional orienting in autistic individuals compared to neurotypical (NT) and
128 intellectually disabled peers, and that reduced eye contact is very dependent on context (Jones
129 et al., 2017; Kasari et al., 1993). Furthermore, reduced presence of eye contact has been
130 associated with failure to automatically attend to the salience of social cues, rather than to
131 active avoidance of others in several eye tracking studies (Hanley et al., 2013; Klin et al.,
132 2002) and has been associated with over-arousal (Hadjikhani et al., 2017). First-hand insights
133 from autistic adults also describe reduced eye contact as a strategy for arousal reduction
134 (McGlensey, 2016; Trevisan et al., 2017) and report the use of qualitative strategies used
135 such as non-eye fixation, blurring focus and strategic fixation (Trevisan et al., 2017).
136 However, the perceived experience of unfocused eye gaze in these first-hand accounts has not
137 been measured from another person's perspective and the research reported here targets this,
138 by exploring parents' perspective of eye contact taken from their everyday experience.

139 While Autism and WS are two frequently studied ND conditions in the eye gaze and
140 eye contact literature, these are not the only ND conditions that are associated with social
141 difficulties related to eye contact. Like WS, fragile X syndrome (FXS) is a genetic condition
142 associated with mild to moderate intellectual disability (ID) and impacts upon social
143 functioning. The FXS social phenotype can be summarised as a mix of both social approach
144 (Cornish et al., 2008) and social withdrawal behaviours (Roberts et al., 2007, 2019),
145 alongside heightened social anxiety (Crawford et al., 2017). Studies to date consistently show
146 that FXS is associated with gaze avoidance (Hall et al., 2006, 2009, 2010; Hessel et al., 2006),
147 which increases when the interlocutor is unfamiliar (Hall & Venema, 2017), but which may

148 improve over the course of an interaction ('warm up effect'; Hall et al., 2009; Roberts et al.,
149 2007). People with FXS show a tendency for shorter gaze episodes towards another person
150 and for brief glances when the person is looking elsewhere rather than making direct eye
151 contact (Cohen et al., 1991; Hall et al., 2006, 2015; Klusek et al., 2020).

152 Although social difficulties are not part of the diagnostic criteria for attention-deficit
153 hyperactivity disorder (ADHD), there is a growing literature reporting socio-cognitive
154 difficulties, problematic peer relationships (for reviews see Gardner & Gerdes, 2015;
155 Soucisse et al., 2015) and high rates of social vulnerability (Ridley et al., 2020). Studies
156 reporting on aspects of gaze orienting and attention indicate impairments in attending to
157 socially relevant information (Airdrie et al., 2018; Marotta et al., 2014, 2017; Muszkat et al.,
158 2015), however everyday eye contact behaviors in this population have scarcely been
159 documented. One relevant study using the ADOS found that unusual eye contact was
160 reported statistically more frequently in a sample of autistic children compared to children
161 with ADHD (Grzadzinski et al., 2016). Nevertheless, 31% of the ADHD sample were
162 reported to have "abnormal" eye contact, yet the nature of the unusual eye contact was not
163 described.

164 **The current study**

165 In this study, we explored the quality of everyday eye contact in individuals with WS
166 in comparison with each of these ND groups using parent report. First, we studied the single
167 dimension of 'presence' (or degree of presence). Second, we included a specific measure of
168 different qualitative features that have been associated with different ND conditions. A two-
169 stage approach was adopted. First, given the gap in the literature on the quality of eye contact
170 in WS, particularly from a parent perspective, Study 1 used a set of standard interview
171 questions to explore the qualitative features that parents might observe in their son/daughter's
172 everyday eye contact. Although we expected a high presence of eye contact in WS, we also

173 expected, given the findings of Mervis et al. (2003) and Jones et al. (2000), that parents might
174 observe a quality of intense, prolonged eye contact (equated with staring in this study).
175 However, we did not know whether other qualitative features would be frequently seen or the
176 extent to which staring would be found across all WS individuals and across all ages.

177 In Study 2, we used a parent questionnaire method to examine further the eye contact
178 quality features used in Study 1 as well as other qualitative features, making cross-syndrome
179 comparisons across children with WS, Autism, FXS and ADHD. In addition, we included a
180 NT comparison group to examine whether particular qualitative aspects of eye contact were
181 specific to the presence of a ND condition. The research will contribute new evidence to an
182 ongoing debate about the similarities and differences in eye contact in ND conditions,
183 particularly between WS and Autism. The study will also add new findings to the literature
184 on eye contact behaviour in FXS, and in ADHD; a topic that has received limited attention.

185 **Study 1: Examining the nature of eye contact in Williams Syndrome**

186 The first study explored the presence and quality of eye contact used by individuals
187 with WS in their everyday life. A semi-structured set of interview items was used that
188 enabled parents to describe both the presence of eye contact and qualitative features, such as
189 brief glances, staring behavior and unfocused gaze. The individual's developmental level of
190 language and visuospatial ability was also recorded during the interview.

191 **Participants**

192 Twenty-two individuals with WS and their families were recruited throughout the
193 North of England and Scotland following institutional ethical approval and study approval
194 from the Professional Advisory Panel of the Williams Syndrome Foundation. Informed
195 consent was given by all participants. The researcher (BA) conducting the interviews with
196 parents was trained in its use by SL. In all cases, it was the primary caregiver who completed
197 the interview with the researcher, either at home or in the University.

198 Individuals were sampled across a wide age range. At the time of the parent interview,
199 individuals with WS ranged between 6 years 0 months and 36 years 3 months of age (male n
200 = 10, female $n = 12$; $M_{\text{age}} = 196$ months, $SD = 98$ months). All individuals were attending
201 school, college or work placements; including five in mainstream school with support, 10 in
202 special educational provision and five in supported work or college (two had information
203 missing). All individuals had previously been diagnosed phenotypically by clinicians and
204 their diagnosis had been confirmed with positive fluorescent in situ hybridization (FISH)
205 testing.

206 Information on language delay, and current language and visuospatial ability was
207 collected from parents during the interview. As Table 1 shows, the group was
208 developmentally delayed. In terms of language delay, 78% of individuals (14/18, four
209 missing) were late to use 2-3 phrases and 84% (16/19, three missing) were late to understand
210 word meanings. In terms of current language ability, 21 participants (one missing) had
211 sentence-level expressive language and all but one participant had sentence-level receptive
212 language (simple or complex sentences). However, only two-thirds (14 individuals) used
213 expressive language at the highest level (complex age-appropriate grammatical constructions)
214 and only one third (seven individuals) understood language at this level. Visuospatial data
215 (two missing) showed that only three individuals (15%) had age-appropriate level of current
216 skill.

217 **Materials and procedure**

218 A research form of the Diagnostic Interview for Social and Communication Disorders
219 (DISCO; Leekam, 2020; Wing et al., 2002) was used. The DISCO is a semi-structured
220 clinical interview used with parents and carers. It is most commonly used for parents of
221 autistic individuals of any age, but is also suitable for use with individuals with other ND
222 conditions and includes items applicable for ADHD, WS and FXS. The current interview

223 followed the format of previous research that has used and published subsets of DISCO items
224 (e.g. Prior et al., 1998). The eye contact and language items used in Study 1 are included in
225 the published DSM-5 algorithm (Kent et al., 2013) and DISCO ICD-10 Childhood Autism
226 algorithm (Leekam et al., 2002), and the visuospatial skill item is a non-algorithm item in the
227 DISCO (Wing et al., 2002). Each of the four eye contact items and each of the language and
228 visuospatial items has a high level of inter-rater reliability ranging from $\kappa = .89$ to $\kappa = 1.00$
229 (Wing et al., 2002).

230 Information on language delay and current language and visuospatial ability was
231 collected using age-appropriate scales within the DISCO (see Table 1). Items from the
232 current language scales have been published (Honey et al., 2007). Age-equivalent
233 visuospatial skill was indicated by the ability to construct complex puzzles according to age
234 group. Language delay (use of phrases, comprehension of word meanings without visible
235 cue) was indicated by delay after 48 months old. Age-appropriate current sentence skills were
236 recorded when complex grammatical constructions and past, present and future tense were
237 present.

238 Information on the presence and quality of eye contact was collected using four eye
239 contact items and scored using the DISCO syntax rules that have previously been applied in
240 both interview (Kent et al., 2013) and questionnaire (Jones et al., 2020), research formats.
241 The first item related to the presence of eye contact. The interviewer asked the caregiver
242 whether it was easy to get eye contact with the individual. The item was scored as “eye
243 contact present” if the answer was “yes”, even if the eye contact given was described as
244 unusual in some way, and “no” if the parent reported little or no eye contact. The next three
245 questions related to quality of eye contact seen as usual behaviour on an everyday basis.
246 These were whether the individual (a) makes eye contact only in brief glances e.g. out of the
247 corner of eyes, but not for the purpose of gaining another’s attention, (b) whether the

248 individual has a blank, unfocussed gaze and (c) whether the individual stares too long and
249 hard, perhaps holding another person's face to make eye contact and/or looking closely into
250 another's eyes. Each item was sequentially assessed by the interviewer who established
251 whether this was a typical behaviour for the individual (used routinely with adults and age
252 peers) and whether it was marked (or frequent), occasional, or rarely/never seen. Following
253 DISCO syntax rules, each item was scored as having a markedly unusual quality if judged to
254 be "marked" (brief glances), "marked and frequent" (blank, unfocused gaze), and "marked
255 staring or otherwise inappropriate" (staring) in that individual, but not if the feature was
256 sometimes, rarely or never seen.

257 **Results and Discussion**

258 Case-by-case profiles of eye contact patterns are shown in Table 1, together with age
259 and language/visuospatial level. The cells that include the plus symbol (+) indicate
260 endorsement of a score for each individual (e.g. presence of eye contact or a marked quality
261 of eye contact), while the blank cells indicates non-endorsement. Results showed that 20
262 (91%) individuals (male $n = 9$, female $n = 11$) gave eye contact easily (even if
263 inappropriately), while two (9%), gave little or no eye contact. Subsequent analyses focused
264 on these 20 individuals, 13 of whom (65%; male $n = 7$, female $n = 6$), had a "marked"
265 unusual quality of eye contact, as indicated by at least one out of three unusual features –
266 brief glances, unfocused gaze, or staring. Brief glances at marked level were endorsed by
267 eight (40%), unfocused gaze by eight (40%), and staring was endorsed by 10 (50%; see Table
268 1). Six individuals had marked scores for all 3 features.

269 *INSERT TABLE 1 HERE*

270 Further exploration was made of the characteristics of the 13 individuals with marked
271 unusual quality of eye contact. More than half, nine of the 13 (69%), had early developmental
272 delay in understanding of word meanings (two had no delay, two had missing data), and of

273 these nine individuals, all but one (data missing) were also delayed in using 2-3 word
274 phrases. The gender distribution was also approximately equal for endorsement of each of the
275 three eye contact quality features.

276 To explore how each of the unusual eye contact quality features was affected by other
277 variables (current age, current language level, and visuospatial level), Mann-Whitney tests
278 were carried out with the 20 participants who were reported by parents as giving eye contact
279 easily. For each analysis, the group of individuals with ‘marked’ responses was compared with
280 the group without marked features (scoring ‘sometimes’ or ‘rarely/never’). Analyses were
281 repeated to examine brief glances, unfocused gaze, and staring features separately and
282 Bonferonni adjustment was applied to accommodate multiple comparisons ($.05/3, p = .02$). An
283 age difference was found (see Table 1), as the group with marked staring features was older,
284 having a mean age of 20 years 11 months ($M_{\text{age}} = 251.20$ months, $SD = 108.37, n = 10$) while
285 those without marked staring features had a mean age of only 12 years 6 months, ($M_{\text{age}} = 150.60$
286 months, $SD = 56.44, n = 10$), $U = 99.0, p < .010$. However, there were no age differences for
287 the other unusual quality features (unfocused gaze, $p = .92$; brief glances $p = 1.00$). No
288 differences were found in visuospatial ability, current expressive and receptive language for
289 those with marked unusual eye contact quality.

290 In summary, Study 1 used a set of parent interview questions for the first time, to
291 explore the qualitative features of everyday eye contact in individuals with WS. The results
292 showed positive presence of eye contact by 91%, together with an unusual quality of staring
293 in 50%. This pattern supports previous evidence from laboratory and clinic studies (Jones et
294 al., 2000; Mervis et al., 2003). However, in addition, new evidence was found. Results
295 showed that staring was more frequent among older ages. However, staring was not an
296 exclusive or predominant quality feature and parents endorsed features of unusual quality of
297 eye contact beyond staring, including brief glances and unfocused gaze. These were reported

298 by parents in 40% of individuals with least one of these features often co-occurring alongside
299 staring.

300

301 **Study 2: Comparing eye contact in WS, other neurodevelopmental conditions and**
302 **neurotypical development**

303 To gather a larger sample of reports, Study 2 asked the same questions as in Study 1
304 but used a questionnaire measure with parents of children with WS. In addition, we adopted a
305 cross-syndrome approach to examine potential syndrome-specific aspects of eye contact
306 behaviour in WS, Autism, FXS and ADHD, as well as NT development.

307 Given the research findings reviewed above and the results of Study 1, we predicted
308 1) a high presence of eye contact in WS compared with other ND groups 2) that unusual
309 qualitative features would be found in WS and also in the other ND groups, with staring
310 reported for children with WS (Klein-Tasman et al., 2007; Mervis et al., 2003), brief glances
311 and avoidance reported for FXS children (Klusek et al., 2020) and a blurred, blank or
312 unfocused gaze (Trevisan et al., 2017) and/or avoidant gaze (Senju & Johnson, 2009)
313 reported for autistic children. Given the results for the WS group in Study 1, we expected not
314 only staring but also other qualitative features to be reported. However, it was not known
315 whether other ND groups might have particular distinctive and predominating qualitative
316 features.

317 **Participants**

318 Parents/caregivers of children were recruited for this study as part of a larger
319 investigation of social interaction behaviours in children with and without ND conditions.
320 Survey responses were received for 276 caregivers/parents in total. Responses were included
321 for data analysis based on the child's primary diagnosis if the parent reported that their child:
322 (1) had a primary diagnosis of either WS, autism, FXS or ADHD, or had NT development

347 ethical opinion from the University ethics committee. Parents did not receive financial
348 remuneration.

349 **Materials**

350 Parents/caregivers reported on their child's eye contact behaviours as part of a larger
351 bespoke survey on social interactions throughout development², via online survey software
352 (www.onlinesurvey.ac.uk). In addition to the questions addressing the research aims, parents
353 provided demographic information concerning the child's date of birth, gender, diagnostic
354 status and ID-status. To gather information about language abilities we included the
355 following questions "does your child use language to communicate" (*none; single words;*
356 *simple phrases; full sentences*), and "does your child understand language" (*none; single*
357 *words; simple phrases; full sentences*).

358 The eye contact items corresponded exactly with interview items of Study 1 but the
359 method was distinct as the items were presented in a fixed response format more suitable for
360 a questionnaire. Items were presented as statements with options to select as follows: Item 1
361 "He/she makes eye contact (even if inappropriate, learned or occasional)" with a response
362 option "yes/no". The next set of items relating to quality of eye contact, unlike Study 1, were
363 not presented sequentially. Instead, they were presented as a forced choice format and
364 caregivers could select only one item in response to the following question: "Please tell us
365 more about the quality of eye contact. Which of the following applies most usually?" Six
366 response options were offered (shown in full in Table 3). In addition to the three items in
367 Study 1 (staring, unfocused gaze, brief glances), two other items were offered to capture a
368 wider range of qualitative features that might be seen in any of the children. These were (a)
369 "always appropriate and natural", and (b) "avoids eye contact". One of the six (indicating the

² The data reported in the current paper were not included in Ridley et al. (2020).

370 one that applies most usually) could be ticked. The next item, “If none of the above applies
371 you can give more information here if you wish (this is optional)” allowed parents to
372 elaborate on their child’s eye contact behaviour if it did not easily fit one of the pre-specified
373 categories

374 **Results and Discussion**

375 The first hypothesis, that there would be a high presence of eye contact in WS
376 compared with other ND groups, was not supported. Instead, results showed that the vast
377 majority of all children with a ND condition engaged in eye contact. Although as many as
378 93% (27/29) of parents of children with WS endorsed this item, similar to Study 1,
379 endorsement was also high for Autism: 86% (25/29), FXS: 72% (13/18) and ADHD: 86%
380 (31/36). A Chi-Square test of Independence showed no significant difference between the
381 four ND groups, $\chi^2(3) = 3.98, p = .264$. Nevertheless, the strong presence of eye contact in all
382 ND groups (96/112, 86%), was still lower than for the NT sample, virtually all of whom were
383 endorsed as showing eye contact (146/149, 98%, one missing response), $p < .001$ (Fisher’s
384 Exact Test).

385 The second hypothesis was that unusual qualitative features would be found in WS
386 and in other ND groups. This hypothesis was examined in several ways. Table 3 presents the
387 distribution of responses (i.e. children with endorsement of “yes” to Item 1 reporting
388 presence of eye contact). First, taking the responses for “eye contact always natural and
389 appropriate” (Column 3 of Table 3), this was the most highly endorsed option for 87% of the
390 parents of NT children and significantly higher than endorsement for the ND sample as a
391 whole (31%; $p < .001$ Fisher’s Exact Test) or for the WS group alone (44%; $p < .001$ Fisher’s
392 Exact Test). This evidence supports the prediction that even when children with a ND
393 condition do give eye contact, the quality of their eye contact is not predominantly natural or
394 appropriate. Nevertheless, the WS group did show a significantly higher frequency of

395 appropriate eye contact compared to the Autism group (12%; $p = .01$), but no difference
396 compared to FXS (15.4%; $p = .09$) or ADHD groups (42%; $p = 1$).³

397 *Insert Table 3 here*

398 Second, initial examination of the pattern of unusual qualitative features revealed that
399 the option “avoids eye contact” was rarely selected for any of the ND groups. This was
400 surprising, given descriptions of avoidance in the Autism and FXS literature (e.g. Hall et al.,
401 2006; Senju & Johnson, 2009), but it demonstrates parents’ interpretation of their child’s eye
402 contact quality when selecting from different behavioural options.

403 Subsequent analysis therefore focused on the three unusual quality descriptors from
404 Study 1 (staring, brief glances, and blank, unfocused gaze). Results showed that the majority
405 of parents in the ND sample selected one of these features as the most usual qualities of their
406 child’s eye contact (ranging from 48% to 77% of each group and 54% of the total ND
407 sample) in comparison to only 8% of the NT group. A Fisher’s Exact Test confirmed higher
408 endorsement any of these three (see Table 3) in the ND groups taken together (54%)
409 compared to the NT group ($p < .001$).

410 Given the result of Study 1, we did not predict specificity or dominance in one
411 qualitative feature (e.g. staring) for the WS group. However, it was not known whether other
412 ND groups might have specific qualitative features that are distinctive or dominating. To
413 analyse this, a series of 2 x 2 Fishers Exact Chi-square analyses were carried out, using only
414 the samples endorsed with brief glances, unfocused gaze or staring (totals from columns 4-6
415 of Table 3 (i.e., WS $n = 13$; Autism $n = 14$; FXS $n = 10$; ADHD $n = 15$). The categories
416 “unfocused, blank gaze” and “staring” were collapsed together (due to small expected

³ Tested in a series of 2 x 2 Fishers Exact Chi-square analyses, with WS compared with each ND group for responses to the “appropriate” option versus the remaining response options.

417 frequencies) and compared with “brief glances”. This confirmed a different distribution of
418 response: brief glances were more frequently selected for Autism (78.6%, $p = .05$), FXS
419 (90%, $p = .03$) and ADHD (86.7%, $p = .02$) groups compared to the WS group (5/13, 38.5%),
420 while the presence of staring behaviour (with unfocused gaze) was more frequently endorsed
421 in the WS group (7/13, 61.5%) This finding supports previous descriptions of persistent and
422 prolonged eye contact in young children (Klein-Tasman et al., 2007; Mervis et al., 2003),
423 showing these behaviours are also found in older children and adolescents. In summary,
424 although dominance of one specific qualitative feature was neither predicted nor found, the
425 results indicate that when given a forced choice format, a small but significant proportion of
426 parents of children with WS tend to preferentially select “staring/unfocused gaze” in favour
427 of “brief glances”, while the majority of parents of all other ND groups select “brief glances”.

428 Only a very small minority of parents selected the option “none of the above apply”
429 (5.4% of the full sample; NT $n = 5$, ND $n = 8$), indicating that the options provided were
430 mostly consistent with the range of parent experiences. All of these parents also answered “if
431 none of the above apply please leave further information here (this is optional)”. The majority
432 of the free-text responses (NT $n = 4$, ND $n = 5$) reported that the child might show more than
433 one type of eye contact behaviour according to situational or person context.

434 Follow-up analyses examined the relationship between eye contact behaviour, first for
435 presence and then for quality (“blank, unfocused gaze” collapsed with “staring” as above)
436 and the demographic variables: age, gender, ID-status (yes/no) and language-status
437 (with/without full sentences) analysed using Chi-square tests. Small samples limited the
438 opportunities for finding significant associations with other demographic variables
439 throughout. No significant associations were found between type of unusual eye contact and
440 language ability (expressive or receptive), ID, gender or age, and it was not meaningful to test
441 the comparison between staring and age found in Study 1 because of the sample sizes.

442

General Discussion

443 Eye contact strengthens the communication process during human social interaction
444 and shapes our judgements about others (Conty et al., 2016; MacDonald, 2009). For this
445 reason, it is important to understand how eye contact manifests in everyday life for those with
446 WS and with other ND conditions. The results of Study 1 and 2 show that parents of
447 individuals with WS, nearly all of whom described their child as making eye contact, also
448 described their child's eye contact as unusual rather than natural and appropriate. Our
449 findings support previous evidence showing prolonged and intense looking in individuals
450 with WS and Study 1 also found first evidence of an association between staring and
451 increased age. However, importantly, staring was not the only type of unusual feature as
452 many parents also reported the use of brief glances and blank, unfocused gaze.

453 The cross-syndrome comparison with other ND groups in Study 2 revealed surprising
454 insights. First, the research literature for Autism and FXS, often describes individuals as
455 having reduced or avoidant eye contact. But parents of these children, who must be looking at
456 their children's eyes on an everyday basis, tend not to describe a lack of eye contact. Like the
457 parents of children with WS, most parents in the Autism, FXS and ADHD groups reported
458 that their child does make eye contact; however, when given different options to indicate the
459 quality of that eye contact, they indicate an unusual quality to it. The most frequently
460 endorsed feature for parents of all three groups was brief glances, whereas this was not the
461 case for the parents of the WS group who more frequently than the other groups, selected
462 staring or unfocused gaze in this forced choice question format. However, staring/unfocused
463 gaze was not unique to WS and many parents also endorsed brief glances in their children
464 with WS.

465 This study contributed to the literature by moving beyond the conventional
466 measurement of eye contact as being either present or absent, in varying degree. By

467 separating the measurement of “presence” from an additional measurement of “quality”, we
468 found different results from studies that have used a single measure of presence of eye
469 contact as an indicator that eye contact is good versus poor. In contrast, our results suggest
470 that nearly all individuals with WS (study 1), and nearly all children whether WS, Autism,
471 FXS or ADHD (study 2), do make eye contact even if in an unusual manner. The type of this
472 unusual quality also seems to be consistently identified by parents as taking the form of brief
473 glances, unfocused gaze or staring, as evidenced by the fact the option “none apply” was
474 rarely endorsed in Study 2. In Study 2 we also found that the option of “avoids eye contact”
475 was rarely endorsed by parents in preference to these other three items. However, it is not
476 clear why they made this preference. Possibly, the choice of one of six forced choice options
477 constrained them and resulted in few cases of “avoids eye contact”. Further research is
478 needed to test out why parents did not choose “avoids” in preference to other items and to
479 evaluate whether this is because it is not a feature of eye contact according to caregiver
480 perspective, or whether it is because other types of contact behaviour are merely more
481 common.

482 We learn from the cross-syndrome comparison design of Study 2 that unusual eye
483 contact is found across multiple ND conditions, rather than specific conditions being
484 associated with specific patterns of eye contact. It is unclear the extent to which this is due to
485 direct yet variable effects of the ND condition on eye contact, or whether these behaviours
486 are differently acquired through factors which may vary but show commonalities across ND
487 conditions, along with external and internal environment. To disentangle this further, the next
488 stage of research enquiry may benefit from moving towards a more transdiagnostic design. In
489 a recent review on the transdiagnostic model for understanding neurodevelopment, Astle et
490 al. (2021) outline a spectrum of study designs that can offer transdiagnostic insights, which
491 vary in the emphasis placed on diagnostic status. Based upon this classification, studies like

492 ours that test for syndrome-specific associations offer value in elucidating where aspects of
493 cognition and behaviour cross over different ND conditions, or are distinctive. However, this
494 traditional, categorical approach is problematic as it rests on the assumption that ND
495 conditions are homogenous and have clear-cut boundaries; an assumption that does not match
496 up with the clinical reality. Consequently, researchers have argued for the need to
497 reconceptualise neurodevelopment and embrace more transdiagnostic features of design
498 throughout the research process (Astle et al., 2021; Casey et al., 2014; Sonuga-Barke &
499 Thapar, 2021). In the case of research on eye contact, there would be value in following a
500 model similar to that used in research areas of cognition and learning (e.g. Bryant et al., 2020;
501 Mareva et al., 2019), by recruiting a large heterogeneous sample of individuals with ND
502 conditions known to impact on social attention and social interaction, and stratifying on the
503 basis of particular eye contact styles (see the “diagnostic-blind” approach in Astle et al.,
504 2021).

505 An important consideration for studies such as ours that do compare groups according
506 to diagnostic label, is that children and adults who receive a diagnosis of any ND condition
507 may also receive other associated diagnoses (Cleaton & Kirby, 2018). Autism frequently co-
508 occurs with other conditions and as atypical eye contact is a diagnostic feature of Autism, this
509 might explain unusual eye contact differences in other conditions as well. As information on
510 co-occurring Autism diagnoses had been collected at the time of recruitment, we were able to
511 carry out further analysis of those with associated diagnoses (WS $n = 2$, FXS $n = 9$, ADHD n
512 $= 9$). The pattern of results for presence of eye contact and for unusual quality of eye contact
513 remained unchanged; therefore, significant effects of an associated autism diagnosis were not
514 evident in this study, but given the small sample sizes, future research designs should test
515 more fully for the effect of co-occurring diagnoses on eye contact presence and quality (see
516 model of study designs outlined in Astle et al., 2021).

517 Limitations

518 There are several important limitations to this study. While the results from parent
519 reports in these studies appear striking, it should also be remembered that there are problems
520 using subjective methods of this kind. Parents were aware that this was an interview or
521 questionnaire studying social interactions in those with ND conditions and responses could be
522 attributed to a response bias. Therefore, a recommendation for future research would be for
523 the inclusion of different measures that combine insights from direct observations and
524 experiments, along with multi-informant reports of everyday eye contact. Teacher insights
525 would make a valuable addition given teachers are interacting with children on a regular
526 basis, but within a different setting compared to parents.

527 Another limitation was that the measure adapted from Study 1 for use in Study 2, did
528 not use exactly the same format. Parents were given a forced choice which did not include
529 options for reporting overlapping types of eye contact quality, as measured in Study 1. This
530 means we cannot make exact comparisons between the measures. Nevertheless, despite
531 differences in the presentation format, the measurement of common behaviour indicators of
532 quality of eye contact (staring, unfocused gaze, brief glances) in each of the two studies
533 contributes new evidence to this sparse literature on the quality of eye contact within WS and
534 across other ND groups. Further testing and replication is still a priority however. Although
535 we might be encouraged by the endorsement rates for Study 2 across the options linked to
536 Study 1, with few choosing the option “none of these apply”, still further validation of the
537 Study 2 method is needed. For example, we recommend further testing of internal,
538 convergent and discriminant validity as has been carried out for other questionnaires using
539 DISCO items (e.g. Jones et al., 2020).

540 The most serious limitation of the study was that the lack of associations with ID, age
541 and gender, were likely due to a lack of power due to small samples distributed across the ND

542 groups. Although the sample size for the WS group in both studies was the same as the
543 sample size for other studies (Klein-Tasman et al., 2007, 2009), there were limitations in
544 making group-wise comparison for each ND condition and in drawing conclusions on the
545 effects of ID, age, gender and language level. As this was compounded by the constraint on
546 caregivers to select only one of six options to describe their child's eye contact, further
547 replication is needed by comparing larger participant groups and testing different research
548 designs.

549 **Future directions and implications**

550 The relationship between older age and staring behaviour in Study 1 is an intriguing
551 finding. One explanation is that staring behaviour emerges throughout development in WS.
552 Another interpretation is that the reporting of marked staring in adults relates more to a
553 change in the perception of this behaviour. From the perspective of the interlocutor, an adult
554 showing staring behaviour may be more striking and deemed less socially acceptable
555 compared to a child staring. However, it is important to note this association with age was not
556 found in the child-only sample of Study 2; therefore, future research should help to
557 corroborate differences and similarities across age and ND groups.

558 Future cross-syndrome comparisons will also benefit from a fine-grained analysis of
559 the differential qualitative aspects of unusual eye contact in relation to social interaction and
560 communication. Klein-Tasman et al. (2007, 2009) noted findings of "abnormal eye contact"
561 in young children with WS as measured within the ADOS domain of reciprocal social
562 interaction. Common difficulties were also found in the ADOS domains of declarative
563 pointing, showing and giving objects, reciprocal social interactions and social
564 communication, and cognition. However, as the qualitative nature of unusual eye contact
565 (e.g. specific type of qualitative features) is not recorded by the ADOS, follow up research
566 using the ADOS, DISCO or other assessment measures could help to clarify the relation

567 between particular qualitative types of eye contact and other social interaction,
568 communication and social cognition difficulties. The prediction would be that unusual
569 qualitative features have particular implications for other aspects of social interaction and for
570 social cognition as the flow of interaction is affected.

571 Our findings may also prove useful in future transdiagnostic research, with respect to
572 (1) separating out the cognitive processes involved in attention and arousal, (2) elucidating
573 the neural circuitry associated with eye contact, and (3) the psychosocial factors associated
574 with qualities of eye contact. In terms of the cognitive processes, it may be possible to test
575 whether unfocused gaze is related to slow allocation of automatic attention (Kuhn et al.,
576 2010), whether staring is related to attentional shifting and hypo arousal (Riby et al., 2011),
577 and whether brief glances are linked to gaze aversion strategies during information
578 processing (Doherty-Sneddon et al., 2012). In the case of neural processes, a more
579 transdiagnostic analysis would be particularly informative for revealing the neural processes
580 associated with qualities of eye contact in people with genetic and non-genetic ND
581 conditions. Not only is there a dearth of research documenting how the brain circuitry
582 responds to eye contact in people with ND conditions, to our knowledge, no research has
583 examined how qualitative features of eye contact are subserved by neural substrates. Indeed,
584 the characteristic use of qualitative features of eye contact early in life may itself have a role
585 in neural development, indicating bi-directional biology-behaviour relations, rather than a
586 simple underpinning of neural processes driving eye contact quality. The results also address
587 psychosocial influences on eye contact and how different qualitative features may serve as
588 adaptive functions to increase or avoid social contact when eye contact is experienced as
589 overly stimulating, distracting in some way, or not as socially rewarding. With respect to
590 brief glances for example, for some people who find it aversive to look in the eyes of others
591 (hyperarousal), brief glances may serve to reduce the uncomfortable sensation, as indicated

592 by evidence of increased activation of the subcortical system when focusing on the eye region
593 (Hadjikhani et al., 2017) and first-hand insights from autistic people (McGlensey, 2016;
594 Trevisan et al., 2017). However, brief glances may also indicate an opportunity for
595 information processing during gaze aversion (Doherty-Sneddon et al., 2012). Collecting
596 further parental data on the quality of eye contact used by their child in varying contexts (e.g.
597 interaction partners, social situations) would add valuable insights into the psychosocial
598 factors that may influence eye contact behaviour.

599 The findings also point to the direction for future research priorities in the areas of
600 FXS and ADHD. Our findings regarding brief glances support previous research with
601 children with FXS. However, the previous research has largely referred to brief glances made
602 while the individual looks elsewhere rather than as part of making eye contact; therefore,
603 further fine-grained observational research is needed to examine the extent to which the well
604 documented finding of brief glances in FXS (e.g. Hall et al., 2015) provides a communication
605 strategy for eye contact, at least as far as parents are concerned. At the same time, the results
606 open a new direction of research in ADHD; a ND condition in which eye contact profiles
607 have previously been neglected. The fact that only 42% of this group showed eye contact that
608 is always appropriate and natural, and similarities in the pattern of unusual eye contact
609 quality to that seen in other ND conditions, should be investigated in relation to their known
610 challenges establishing and maintaining friendships (Normand et al., 2011, 2013) and broader
611 socio cognitive skills (Bora & Pantelis, 2016; Sibley et al., 2010; Uekermann et al., 2010).
612 Further research is also needed with this group to understand eye contact patterns in those
613 with co-occurring ADHD and autism.

614 From a clinical and societal perspective, the findings emphasise that eye contact given
615 by people with ND conditions may look different from the NT preference of direct, steady
616 gaze, but that the observable qualities may vary across individuals with the same diagnosis.

617 Difference from a NT pattern of eye contact should not be interpreted as a call for
618 intervention, given these behaviours likely serve an adaptive role. One important
619 consideration however, is the potential impact that different eye contact behaviours may have
620 on the wider social interaction, in terms of impression formation and potential stigma
621 (Morrison et al., 2020; Sasson et al., 2017). Unusual qualities of eye contact may
622 miscommunicate information about the intentions and attitudes of people with ND
623 conditions. For example, brief glances may infer that the person is disinterested in the
624 interaction. Equally, being on the receiving side of prolonged eye contact may be an
625 uncomfortable experience. Prolonged staring at a time of greater social independence during
626 adolescence and young adulthood is particularly important, given the vulnerability issues that
627 have been emphasised in people with ND conditions (Fisher et al., 2013; Jawaid et al., 2012;
628 Ridley et al., 2020).

629 To conclude, it is known that measurement differences lead to particular
630 interpretations of eye contact (Jongerius et al., 2020). We argue that the previous single
631 dimension interpretation, based on measurement of the degree or strength of eye contact, has
632 led to the oversimplified assumption that reduced eye contact equates to poor eye contact,
633 while eye contact that is not reduced equates to good eye contact. This in turn has led to an
634 interpretation that polarises different ND groups, such as WS and Autism, and makes the
635 incorrect assumption about underlying social motivational and cognitive factors. Given our
636 findings on similarities across ND conditions, we think it is time to focus on describing eye
637 contact profiles more in terms of different qualitative styles, and less in terms of a single
638 dimension (i.e. degree of presence/absence). This new perspective would have implications
639 for research on psychological and neural mechanisms related to eye contact, as it indicates
640 that quality of eye contact subtypes may be studied independently of traditional diagnostic
641 groupings and divisions.

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Table 1

Data for each Individual with Williams Syndrome (WS) for Diagnostic Interview for Social and Communication Disorders (DISCO) Items Assessing Eye Contact, and Language and Visuospatial Skill Level

Age in months	M/F	Language Delay		Age-appropriate level of current skill			Eye contact present	Unusual quality of eye contact (marked or frequent)		
		Late to use 2-3 phrases	Late to understand word meanings	Expressive language Level 1-9 ^a	Receptive language Level 1-7 ^b	Visuospatial skill Level 1-12 ^c		Brief glances	Blank unfocused gaze	Staring
72	F	Yes	Yes	8	5	9	+	+	+	+
89	M	Yes	Yes	8	5	5				
100	F	No	No	9	6	9	+			
101	M	-	-	9	3	12	+			
106	M	Yes	Yes	9	7	10	+	+		
115	F	Yes	Yes	7	6	10	+			
124	F	Yes	Yes	8	7	10	+			
153	M	Yes	Yes	9	7	9	+		+	
159	M	Yes	Yes	9	7	9	+			
161	F	Yes	Yes	9	5	12	+			
172	F	Yes	Yes	9	4	10	+	+	+	+
193	M	No	-	9	4	10	+	+	+	+
193	M	-	Yes	8	5	8	+	+	+	+
205	F	Yes	Yes	8	4	-				
206	M	Yes	Yes	9	4	12	+		+	+
210	F	No	Yes	9	7	6	+			
258	F	Yes	Yes	8	5	8	+			
277	M	Yes	Yes	9	5	6	+	+		
286	F	-	No	9	7	8	+	+	+	+
301	M	Yes	Yes	9	7	3	+			+
396	F	No	No	9	6	8	+			+
435	F	-	-	-	6	-	+	+	+	+

Note. Dash sign (–) = parent data was not available. Cells with plus sign (+) indicate endorsement of either (a) presence of eye contact and (b) unusual quality of eye contact at a marked level. M = male; F = female.

^a Language expression: 0-2 = No speech or babbles, 3-4 = Says names for things only, 5 = says phrases of 2 words only, 6 = Says longer phrases, 7 = Uses spontaneous sentences, present tense only, 8 = Uses sentences/phrases including ‘but’ and ‘because’, 9 = Uses past, present and future tenses and complex grammatical constructions.

^b Language comprehension: 0-1 = No response or responds to name only, 2 = Understands simple words from phrases in context (learned from gestural cues, e.g. time for bed), 3 = Knows the meaning of some words and can responds e.g. ‘give me your cup’, 4 = Follows instructions involving 2 new objects “Put the doll on the chair”, 5 = Can reliably follow instruction to fetch 2 or more objects from outside of the room, 6 = understands a sequence of commands, 7 = Understands instructions involving decisions (conditionals) “see if my phone is in my bedroom and if not look for it in the bathroom”.

^c Visuospatial skill: 0 = does not hold objects in hands, 1 = holds objects in hands, 2 = examines objects, 3 = handles objects, 4 = rolls toys on floor, 5= builds tower of 2-5 bricks, 6 = builds tower of 6 bricks, 7 = arranges objects in size order, 8 = completes puzzle 6 pieces, 9 = completes puzzle 10 pieces, 10 = completes puzzle 20-30 pieces, 11 = completes puzzle 50 pieces, 12 = completes puzzle 150 pieces.

Table 2*Demographic Characteristics of the Sample (% Reported) Split by Diagnostic Group*

Demographic variables	Autism (<i>n</i> = 29)	WS (<i>n</i> = 29)	ADHD (<i>n</i> = 36)	FXS (<i>n</i> = 18)	NT (<i>n</i> = 150)
Males/females/prefer not to say	72/28/0	59/41/0	78/19/3	94/6/0	48/51/1
Age (months)					
<i>M</i> (<i>SD</i>)	127 (28.4)	100 (36.3) ^a	127 (38.8) ^b	118 (36.9)	107 (45.8) ^c
Range	59-187	48-204	54-179	54-197	48-215
Presence of an intellectual disability	21	90	28	89	0
Expressive language					
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
Receptive 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

Note. WS = Williams syndrome; ADHD = attention-deficit hyperactivity disorder; FXS = fragile X syndrome; NT = neurotypical.

^aMissing data (*n* = 1). ^bMissing data (*n* = 1). ^cMissing data (*n* = 1).

Table 3*Quality of Eye Contact Behaviour Endorsed by Parents in Each Group*

Group	<i>n</i> ^a	Quality of eye contact applied most usually					
		Eye contact always appropriate and natural	Brief glances	Blank, unfocused gaze	Staring	Avoids eye contact	None of these apply
WS	27	12 (44.4)	5 (18.5)	1 (3.7)	7 (25.9)	1 (3.7)	1 (3.7)
Autism	25	3 (12)	11 (44)	2 (8)	1 (4)	3 (12)	5 (17.2)
FXS	13	2 (15.4)	9 (69.2)	0 (0)	1 (7.7)	0 (0)	1 (7.7)
ADHD	31	13 (41.9)	13 (41.9)	1 (3.2)	1 (3.2)	2 (6.5)	1 (3.2)
NT	143 ^b	124 (86.7)	11 (7.7)	1 (0.7)	0 (0)	2 (1.4)	5 (3.5)
Total ND	96	30 (31.3)	38 (39.6)	4 (4.2)	10 (10.4)	6 (6.3)	8 (8.3)

Note. Percentages are presented in parentheses. WS = Williams syndrome; FXS = fragile X syndrome; ADHD = attention-deficit hyperactivity disorder; NT = neurotypical; ND = neurodevelopmental.

^aParents who reported “yes” to Q1 about the presence of eye contact. ^bOf the 146 TD parents who reported yes to Q1, 3 data points were missing.