Are language and social communication intact in children with congenital visual impairment?¹

Abbreviated title: Language and social communication in children with visual impairment

Valerie Tadić^{1,2}, Naomi Dale^{2,3}, Linda Pring¹

¹Goldsmiths, University of London, UK

²UCL/Institute of Child Health, UK

³Great Ormond Street Hospital NHS Trust, UK

¹ Word count: 7.557

ABSTRACT

Background: Development of children with congenital visual impairment (VI) has been associated with vulnerable socio-communicative outcomes that often bear striking similarities to those of sighted children with autism². Language has been proposed as a facilitative mechanism that mediates such outcomes in children with VI, although its contribution remains poorly understood. Methods: We examined language and socio-communicative profiles of 15 children with VI and normal-range verbal intelligence in the context of i) a structured language assessment, ii) a parental report of everyday communicative behaviours, and iii) in comparison to a group of typically developing sighted children of similar age and verbal ability. **Results:** Compared to their sighted peers, and relative to their good and potentially superior structural language skills, children with VI showed significantly poorer use of language for social purposes. The pragmatic language weaknesses in children with VI were observed within a broader socio-communicative profile, which in a substantial proportion of children with VI in this study is consistent with the pattern found in sighted children on the autism spectrum. Unique evidence that such characteristics are also prevalent amongst children with some limited levels of 'form' vision was offered. Conclusions: There is an ongoing socio-communicative and pragmatic language concern in children with good intellectual abilities and congenital VI at school age, despite having advanced linguistic skills. Very limited vision was not sufficient to ameliorate the effects of VI. Developmental potential in structural language may be an important candidate for future interventions with such children.

Keywords: Visual Impairment (VI), language, social communication, autism.

² Term 'autism' is used here to refer to all the individuals on the spectrum of the disorder, which is characterised by impairments in social interaction, communication, and repetitive behaviours and restricted interests (DSM-IV-TR, 2000).

INTRODUCTION

Congenital visual impairment (VI) has been associated with vulnerabilities in sociocommunicative and socio-cognitive development, including behavioural similarities with sighted children with autism (Pring, 2005). Difficulties have been reported in early social interaction and communicative competence (Preisler, 1991; Urwin, 1983); theory of mind (Green, Pring, & Swettenham, 2004; Peterson, Peterson, & Webb, 2000; Roch-Levecq, 2006), emotional expressiveness and recognition (Dyck, Farrugia, Shochet, & Holmes-Brown, 2004; Minter, Hobson, & Pring, 1991; Roch-Levecq, 2006); symbolic and functional play (M. Bishop, Hobson, & Lee, 2005; Lewis, Norgate, Collis, & Reynolds, 2000); behavioural mannerisms, rituals and stereotypes (Chess, 1971; Tröster, Brambring, & Beelmann, 1991) and autistic-like developmental regression in the preschool years (Cass, Sonksen, & McConachie, 1994; Dale & Sonksen, 2002).

The underlying reasons for such difficulties in children with VI are unknown. Similar to autism in the sighted, they have been attributed to disruptions in visually-driven processes (i.e., dyadic and triadic joint attention) and visual behaviours (i.e., eye-gaze monitoring, directing and following) in early childhood, which are seen as precursor milestones for subsequent social development (Hobson, 1993). However, empirical associations between the early concerns surrounding visual joint attention and later socio-communicative vulnerabilities in children with VI have not been established yet. This is because very little is known in general about the non-visual aspects of early social interaction and social attention that is not eye-gaze dependent, although there have been some recent empirical advances in understanding the mechanics of joint attention in young children with congenital VI (Tadić, Pring, & Dale, 2008).

Importantly, evidence suggests individual variation and greater success in social communication in some children with VI (Preisler, 1991; Urwin, 1978). It is possible that, while vision provides important means for social relating for children who are sighted, children with VI may be able to rely on alternative non-visual strategies. In this respect, language has been regarded as a particularly

important developmental domain (Landau & Gleitman, 1985; Pérez-Pereira & Conti-Ramsden, 1999), and some empirical evidence supports its role in social outcomes of children with VI. For instance, children with VI who passed a standard theory of mind task had significantly higher verbal IQs and verbal mental ages than did those who failed it (Green et al., 2004; Minter, Hobson, & Bishop, 1998). Similarly, Brown, Hobson and Lee (1997) found that children with VI who had higher verbal ability (VIQ>70) showed fewer autistic-like behaviours on an autism-screening observational schedule than did children with VI with lower verbal ability (VIQ<70).

Whilst language, at least in the form of verbal IQ, appears to differentiate children with VI with differing socio-communicative competence and socio-cognitive outcomes, the contribution of the mechanisms that language provides for such children remains poorly understood. For children with VI, language-based measures are commonly used to assess general intelligence, making it more difficult to isolate the contribution of language irrespective of a child's general cognitive ability. The same issue arises from grouping children with VI with a wide range of intellectual abilities for research purposes. Thus, the better social outcomes of children with VI who have higher verbal intelligence may not be fully appreciated as a consequence of better language per se as much as a result of a higher intellectual level. It is, therefore, important to consider presentation of language in children with VI where cognition has been controlled for and a systematic comparison with a well controlled sighted sample is appropriate.

Language is a complex system, consisting of a rich network of functions and skills that provide building blocks for communicative and social interaction. While *structural* language skills, such as articulation of speech, use of grammar, vocabulary level and comprehension, may enable a person to converse fluently, it is the *pragmatic* language skills (i.e., use language socially and appropriately in a given context) that are required for successful socio-communicative functioning. Vision is implicated in language development in general, as visually-driven joint attention experiences in early childhood are seen as providing a framework within which language learning occurs (Tomasello & Farrar, 1986).

Despite some early delays and irregularities in the early vocabulary acquisition and production, syntactic knowledge, and the acquisition of semantic concepts (e.g., Andersen, Dunlea, & Kekelis, 1984; McConachie & Moore, 1994), the structural language skills in children with VI are believed to develop with relative ease (Landau & Gleitman, 1985; Mulford, 1988; Urwin, 1983). The early delays and differences generally seem to be overcome by school age, and despite following what seems like an alternative route of language development, children with VI ultimately seem to arrive at the same point as do sighted children (Landau & Gleitman, 1985; Mills, 1993; Reynell, 1978). Interestingly, the majority of evidence concerning language structure in children with VI concerns the early years whereas language ability at school age has been largely ignored.

The picture is less clear regarding the pragmatic language use of children with VI, which is suggested to have features similar to those of children with pragmatic language impairment (PLI) (Mills, 1993). These involve the extensive, and sometimes inappropriate, use of questions, an absence of communicative gestures, and the extensive use of imitative speech, repetitions and verbal routines (Mulford, 1983; Norgate, Collis, & Lewis, 1998; Preisler, 1991). It has been argued that such pragmatic language features in children with VI may have an important function in promoting their cognition and social interaction by providing an adaptive strategy by which to gather information, analyse speech, reduce memory load, and avoid isolation (see Pérez-Pereira & Conti-Ramsden, 1999). However, a concern has been raised regarding these language features in children with VI (e.g., stereotypic speech and echolalia), which are largely seen in children with autism and thus may contribute to the presentation of an autism-like syndrome in children VI (Brown et al., 1997; Fraiberg, 1977).

Based on the aforementioned studies, involving mostly preschool children with congenital VI, it generally appears that structural language is an area of relative strength for such children, whereas their pragmatic skills may be challenged. Irregular presentation of language skills in children with congenital VI has been illustrated recently in an uncontrolled study using a parental communication with a small sample of school-aged children (James & Stojanovik, 2007). However, the discrepancy between

structural and pragmatic language in children with VI, who do not have an additional diagnosis of autism or PLI, needs to be further substantiated by research.

Overall, little is known about presentation of structural and pragmatic language in children with congenital VI in middle childhood. Our understanding of the nature of autism-like socio-communicative difficulties in such children is still in its infancy, with learning difficulties being a major confounding factor. Empirical attempts to enhance this understanding remain a major challenge, given the rarity of children in question and the need to adapt assessment procedures to their available sensory channels. The aim of this study was to examine variation in language presentation and social communication in school-age children with congenital VI, while controlling for the children's general intelligence. To achieve this we i) focused on children with a significant congenital vision loss and intelligence in the normal range, ii) utilised an age and ability matched comparison group of typically developing sighted children, iii) used a standardised test designed specifically to assess language function in children in the context of a structured assessment, and iv) utilised a parental report of everyday language and communicative behaviours. We compared language and socio-communicative profiles of children with VI and sighted children with a view to examine the extent to which the two groups differ, and to gain better appreciation of specific strengths and weaknesses that may characterise the VI group.

METHOD

Participants

15 children with congenital VI and 26 sighted children took part in the study. The children with VI were recruited through the Developmental Vision Clinic at the UCL/ Institute of Child Health and Great Ormond Street Hospital, London, UK where they had been referred to for developmental and functional vision assessments in their early years. The sighted children were recruited through primary schools in the UK.

The children with VI all had a degree of vision loss which was present from birth and was of peripheral, rather than cerebral origin. The inclusion criteria, based on the origin and the site of the VI, was adopted from the taxonomy by Sonksen and Dale (2002) (i.e., 'potentially simple' congenital disorders of peripheral visual system). The group consisted of children with varying levels of VI, including children whose VI was profound - PVI (light perception or worse - no form vision; n=6) and those children whose VI was severe - SVI (severely degraded form vision - able to perceive a non-light reflecting spinning ball sized 12.5cm from a distance of 30 cm, or better; visual acuity in the better eye worse than 6/30, n=9). None of the children with VI had a known additional paediatric diagnosis involving the central nervous system or a severe hearing impairment.

No formal vision assessment was carried out at the time of the current research. The information on children's vision levels was obtained from the latest preschool assessment of functional vision of each child before their participation in this study (i.e., archived clinical records containing each child's history of comprehensive formal functional vision assessments by the paediatrician across the preschool years). The greatest development to the visual system occurs across the early years of life and the visual level is usually stable by the early school years (Sonksen, Petrie, & Drew, 1991).

The two groups were well matched in terms of their VIQ ($t_{(39)}$ = -.105; p=.917), age ($t_{(39)}$ = -.502; p=.618) or gender ratio ($\chi^{2}_{(1)}$ = .702; p=.754) (Table 1).

(Insert Table 1 here)

Materials

The Verbal scale from the Wechsler Intelligence Scales for Children-III (WISC-III) (Wechsler, 1992) was used for developmental matching. The VIQ for each child was derived from five verbal subtests that did not require presentation of visual stimuli (Information, Similarities, Vocabulary, Comprehension and Digit Span).

The Clinical Evaluation of Language Fundamentals-3 (CELF-3) (Semel, Wiig, & Secord, 2000) was used to assess language function. Only 2 core and 2 supplementary subtests were suitable for use with children with VI, as they did not require visual stimuli. Subsequently, the Receptive and Expressive Language composites were calculated as a pro-rata of two subtests for each language domain (which potentially reduces their reliability). Receptive language subtests were Word Classes (i.e., semantic word grouping) and Listening to Paragraphs (i.e., verbal recall, comprehension and interpretation). Expressive language subtests were Recalling Sentences (i.e., short-term memory test) and Word Associations (i.e., word fluency test).

The Children's Communication Checklist-2 (CCC-2) (D. V. M. Bishop, 2003), which is a parental questionnaire, was used to evaluate communicative skills that are not easy to assess in a context of a traditional structured assessment. The CCC-2 assesses language *structure*, *pragmatic* language and *social* behaviours that are usually impaired in cases of autism spectrum disorder (ASD). The General Communication Composite (GCC) is used to identify children likely to have clinically significant communication problems. The Social Interaction Deviance Composite (SIDC) can help identify children in whom pragmatic language skills and social interaction skills are disproportionately impaired relative to their structural language. Although not diagnostic, the CCC-2 can be seen as useful in screening for a potential communication disorder (e.g., ASD and specific language impairment/SLI). All of the items on the CCC-2 were considered appropriate for use with children with VI except item 14 (i.e., 'does not look at the person s/he is talking to'). In over 50% of the cases, the parents of children with VI omitted this item, which was subsequently removed from analyses for both groups.

The Social Communication Questionnaire (SCQ) (Lifetime Autoscore) (Rutter, Bailey, & Lord, 2003) was used to screen for socio-communicative behaviours associated with ASD (scores of ≥15 are considered to be of potential clinical significance). The SCQ can be broken down into three behavioural domains of the autism diagnosis: Reciprocal Social Interaction, Communication and the Restricted, Repetitive and Stereotyped Patterns of Behaviour.

Procedure

The children took part in the language tasks while their parents completed the questionnaires. All of the questionnaires were completed by the parents, except in the case of two children with VI where the questionnaires were completed by a teacher who knew the children well.

The study, including the recruitment with informed parental consent, conduct towards the participants and study procedure, was carried out following the research protocol approved by the NHS research ethical committee for the UCL/Institute of Child Health and the ethical committee for the Goldsmiths, University of London (UK).

RESULTS

Structured language assessment

First, we examined whether the CELF-3 would discriminate between the two groups in terms of their language ability (see Table 2 for group means and standard deviations). Despite being comparable on verbal IQ (individual WISC-III subtests, p values>.214), the VI group showed significantly better performance on the CELF-3 than the sighted group (Total Language: $t_{(39)} = 2.674$, p = .011). This language strength did not seem to be confined to a specific language sub-domain as the VI group achieved higher performance in terms of both their receptive and expressive language ($t_{(39)} = 2.528$, p = .016; $t_{(39)} = 2.352$, p = .024).

The between-group difference in CELF-3-related language competence appeared to vary as a function of a specific language skill, as the only individual subtest discriminating the children with VI as significantly better than the sighted children was Recalling Sentences ($t_{(39)}$ = 2.956; p=.005). There were trends towards a significant difference on Word Classes ($t_{(39)}$ =1.742; p=.089) and Listening to Paragraphs ($t_{(38.9)}$ =1.702; p=.097), while the group difference on Word Associations was not significant ($t_{(39)}$ =1.256; p=.217).

(Insert Table 2 about here)

The performance of the sighted group on the CELF-3 Total Language was significantly discrepant from their VIQ ($t_{(25)} = 4.231$; $p \le .001$), whereas the scores of the VI group seemed comparable across the two composites ($t_{(14)} = -1.262$; p = .228). However, the overall performance on the two tests (CELF-3/Total Language and WISC- III / VIQ) was significantly correlated in both children with VI (r = .559; p = .03) and sighted children (r = .715; $p \le .001$) signifying that the skills required for the two tests may not necessarily be independent.

Parental reports

Table 3 shows the group means and standard deviations on the CCC-2 and the SCQ. A Profile Analysis was used to compare the communicative profiles of the two groups on the CCC-2. Consequently, when averaged across the groups, the children's performance was found to differ across different CCC-2 subtests (i.e., significant test of Flatness - Pillai's Trace criterion: $F_{(9, 29)} = 5.323$; $p \le$.001). Additionally, there was a significant between-group difference when the children's scores were averaged across different CCC-2 scales (i.e., significant test of Levels - $F_{(1, 37)} = 26.6$; $p \le$.001). These tests are qualified by a significant test of Parallelism, indicating distinguishable profiles between the two groups across individual CCC-2 scales (Pillai's Trace criterion: $F_{(9, 29)} = 7.266$; $p \le$.001).

(Insert Table 3 about here)

Figure 1 graphically illustrates divergent CCC-2 profiles in the two groups. Here, the pattern of parental rating for sighted children's behaviours appears relatively consistent across different scales, averaging at around the mean scaled score of 10, which is in line with the CCC-2 developmental norms. In contrast, the profile of the VI group appears uneven. Despite this, the mean scores of children with VI across the CCC-2 scales are largely within the normal range limits (i.e., scaled score≥6), except for the Non-verbal and Social scales, where the mean scores of the VI group fall below normal limits (i.e., scaled score<6).

(Insert Figure 1 about here)

Following from the results of the Profile Analysis, we examined the between-group difference on individual CCC-2 scales. With regards to the scales tapping *structural language*, the children with VI were found to be comparable to sighted children on Speech ($t_{(37)}$ =- 0.401; p=.691) and Syntax (equal variances not assumed – $t_{(16.99)}$ = -1.250; p=.228), but poorer than their sighted peers on Semantics ($t_{(37)}$ =- 2.717; p=.01) and Coherence ($t_{(37)}$ =- 2.404; p=.021). Children with VI also obtained significantly poorer ratings than sighted children on all four *pragmatic* scales (Inappropriate Initiation: $t_{(37)}$ = - 3.838; p≤.001; Stereotyped language: $t_{(37)}$ = - 3.18; p≤.003; Use of Context: $t_{(37)}$ = - 5.105; p≤.001; and Non-

verbal: $t_{(37)}$ = - 7.49; $p \le .001$). With respect to the two CCC-2 scales tapping *social interaction*, the VI group obtained significantly lower ratings than the sighted group on both scales (Social: $t_{(37)}$ = - 5.17; $p \le .001$; and Interests: $t_{(37)}$ = -3.15; p = .003).

With a view to statistically examine this apparent discrepancy between structural and pragmatic language skills in children with VI, the scaled scores on the structural and pragmatic scales respectively were summed in order to derive a Structural and Pragmatic Index for each child. Subsequently, a 2x2 mixed ANOVA revealed a significant main effect of Language Index ($F_{(1, 37)}$ = 32.471, p≤.001) and Group ($F_{(1, 37)}$ =789.94, p≤.001), qualified by a significant Language Index x Group Interaction ($F_{(1, 37)}$ =34.261, p≤.001). As their relatively flat profile suggested previously, there was no difference in structural and pragmatic language skills in sighted children ($t_{(24)}$ = -.125; p=.901). However, the structural language skills of the VI group were significantly better than their pragmatic language ($t_{(13)}$ =7.716; p≤.001).

Sighted children obtained significantly higher General Communication Composite (GCC) scores on average, signifying their higher communicative competence, than in the VI group ($t_{(37)}$ = - 5.105; p≤.001). A between-group comparison on the SIDC was not considered useful as this composite provides qualitative information about the pattern of impairment on an individual child's level and cannot be interpreted without the GCC. Instead, the SIDC scores of each child have been examined qualitatively in relation to their GCC's.

Three reference lines plotted in Figure 2 indicate the clinical cut-offs used to subgroup children with specific communication difficulties (D. V. M. Bishop, 2003). The top-right section of the scatter-plot (i.e., GCC>55 and SIDC≤15) marks the distribution of individual GCC/SIDC profiles that are considered to be in a typically developing range. The bottom-left section of the scatter-plot (i.e., GCC<55 and SIDC<0), marks a region of profiles where both composite scores are considered to be below normal range, and such profiles are typical of a broader autism spectrum. Finally, the bottom-right section of the scatter-plot highlights the profiles of those children whose GCCs are within normal range (GCC>55), but

whose SIDC is considered to be deviant (SIDC<-15), and such profiles are frequently seen in Asperger Syndrome (AS).

(Insert Figure 2 about here)

Figure 2 shows that the CCC-2 socio-communicative profiles of all the children in the sighted group were within a typical range. It is also important to highlight that the profiles of five children with VI were also distributed within this section. However, four children with VI showed CCC-2 profiles that are typical of ASD and five that are associated with AS. Overall, 64% of the children with VI in this study (9/14, with the data of one child missing) showed socio-communicative characteristics that are consistent with a broader autism phenotype.

Parental ratings of the children's socio-communicative behaviours on the SCQ were in line with this pattern, as children in the VI group obtained significantly higher scores, than the sighted, on the SCQ total score ($t_{(38)}=7.727$; $p\leq.001$), as well as on the individual SCQ domains (Reciprocal Social Interaction: equal variances not assumed - $t_{(16.9)}=5.306$; $p\leq.001$; Communication Domain: $t_{(38)}=4.835$; $p\leq.001$; and Restricted, Repetitive and Stereotyped Patterns of Behaviour Domain: $t_{(38)}=4.941$; $p\leq.001$).

Five children in the VI group (34%) obtained SCQ scores of potential clinical significance. Four of these children were also within a domain for clinical concern of a potential communicative disorder on the CCC-2 (Figure 2). Additionally, a number of other children in the VI group achieved overall SCQ scores that were just below the clinical cut-off of 15. Children obtaining such scores are frequently considered worthy of further clinical evaluation where there has been a raised concern of a potential ASD (Rutter et al., 2003). Thus, it is also worth noting that the profiles of three of these children fell within the section of the CCC-2 that marks the profiles associated with AS.

Consideration of individual differences

The prevalence of autistic-like characteristics in the present sample of children with VI could not be explained by differing levels of VI or the individual differences in verbal intelligence and language competence. Although the confounding effect of VI severity could not be examined statistically due to small numbers, the clinically elevated scores on the two socio-communicative measures were not confined to the group of children whose VI is of greater severity, and were also seen in children with SVI. Additionally, no significant correlations were found between the children's overall performance on the WISC-III and CELF-3, and socio-communicative behaviour ratings on the SCQ and the CCC-2 (p values>.05). Data examination at an individual child's level did not shed further light on why some children with VI showed atypical profiles while others did not. None of the clinical or language characteristics (e.g., age, gender, visual level, diagnosis, VIQ) of those children with VI with better CCC-2 and SCQ outcomes seemed to distinguish them from the children whose profiles were atypical. Interestingly, the two children with VI with some of the lowest SCQ scores in the VI group were still in line with the 10% of children who showed the highest prevalence of undesirable SCQ behaviours within the sighted group. Similarly, the four children with VI, whose CCC-2 profiles were considered to be in the typical domain, achieved scaled scores that were below the sighted group mean on Social, Non-Verbal and Context scales.

DISCUSSION

The present study highlights three important findings. Firstly, despite being comparable on age and verbal intelligence, children with VI performed significantly better than developmentally matched sighted children on a standardised test of language function. Secondly, and in contrast to the first finding, the children with VI showed a significantly poorer range of skills than their sighted peers in terms of their communicative functioning in an everyday context (based on parental reports), with a particular weakness in use of language for pragmatic and social purposes. Thirdly, judging by the parental reports of children's everyday socio-communicative behaviours, a substantial proportion of children with VI showed a level of behavioural difficulties that is consistent with a broader autism phenotype in sighted children.

As discussed previously, language has been generally seen as an area of strength in the development of children with VI. However, what is especially remarkable about the present findings is that they differentiated children with VI as better than their sighted peers. This is particularly significant in light of the early language irregularities and delays in language development of children with VI reported by previous research (Andersen et al., 1984; McConachie & Moore, 1994), and bear important implications for educational language-based interventions. Crucially however, how can we explain the language strength that children with VI have demonstrated in this study?

In line with Pérez-Pereira and Conti-Ramsden (1999), the language domain may be more salient to children with VI than sighted children and is therefore likely to serve a different function in the two groups. Importantly, the CELF-3 was shown to be a successful tool in separating this language function from general intelligence, allowing us to illuminate the strength of children with VI that may be specific to their dominant domain. Being a test of language ability, a child's performance on the CELF-3 is also likely to be related to their verbal IQ as assessed by the WISC-III (Semel et al., 2000), a pattern which is also supported by the significant correlations between the two tests in the present research. However, unlike the majority of verbal WISC-III subtests, which essentially measure crystallised

intelligence and 'fluid' reasoning, the CELF-3 is less reasoning based, largely tapping linguistic elements such as content and structure, which can be evaluated independently and out of context. This potential of the CELF-3 to isolate language-specific strengths and difficulties in children has also been demonstrated in research with other clinical groups (e.g., children with specific language impairment/SLI or autism) (Lloyd, Paintin, & Botting, 2006).

However, the CELF-3 assessment in the current study was based only on four out of the six possible subtests (two of which were supplementary), as the excluded subtests involved visual stimuli. The language profiles provided by the CELF-3 in this study are therefore incomplete, and the composite language scores may be less reliable. This reduced assessment may explain why there was a significant discrepancy between the CELF-3/Total Language and the VIQs in the sighted group, who did not perform on the CELF-3 as it would be expected based on their verbal IQ. Since both groups were administered the same auditory-dependent subtests, it is not known why the sighted group performed less successfully than the VI group. It is possible that the composite auditory subtests were especially favourable to children with VI, an advantage which was particularly obvious on Recalling Sentences.

Even though the CELF-3 subtests generally all have a strong memory component, the Recalling Sentences subtest is particularly verbal short-term memory (STM) based. It is possible that, given adequate intelligence, verbal STM may play a particularly important role for children with VI, because of their reliance on auditory and verbal information. The VI group's superior performance on Recalling Sentences may reflect such advantage, which traditionally has been demonstrated using the Digit Span WISC subtest (Hull & Mason, 1995; Smits & Mommers, 1976). The Digit Span superiority of children with VI has not been replicated here. It is likely that Recalling Sentences places slightly different demands on the child than does the Digit Span, in that the words to be recalled need to be recognised within a language context (i.e., syntax and semantics). For this reason, Recalling Sentences may better capture a language-specific STM advantage than would a traditional Digit Span test, although this requires further investigation in the future.

Importantly, STM advantage may not only be obvious at the level of STM tests, but may also feed into all the other skills required for successful language function.³ Hence, for a child with ample vocabulary, grammar and semantics, good verbal STM may especially boost an overall language outcome. This may explain why the overall CELF-3 performance of the VI group was more successful than for the sighted children, even though at the level of an individual subtest, only Recalling Sentences had the power to differentiate the two groups. Interestingly, STM strength may also provide an explanation for certain characteristics of the language of children with VI that are considered to be a disadvantage, such as modelled and imitative speech, although this needs to be further substantiated with research.

Furthermore, it is likely that the observed language competence in children with VI is dependent on the context within which it is assessed, given that their parental ratings of specific structural language skills (CCC-2 Semantics and Coherence) revealed a potentially contrasting picture. It is possible that the children with VI benefited from the context of a traditional one-to-one assessment, which is structured and therefore rigid and scripted. This context, in addition to the provision of clear instructions by the assessor, may provide scaffolding for achieving successful performance in a child with VI and may better capture the strength that is not necessarily apparent in an everyday conversation with such a child. Conversely, an everyday context (within which language is generally used) is largely spontaneous and inherently social. Thus, parental reports may be more likely to reveal a VI-related disadvantage in Coherence and Semantics than would be evident in the structured context. However, more rudimentary language elements, such as Speech and Syntax, may be less susceptible to contextual influences, explaining why the two groups did not differ on these two components.

In contrast to robust structural language skills, parental reports captured a particularly striking weakness of children with VI in their use of language for socio-communicative purposes. Concerns

³ In relation to this, it is important to note that the Recalling Sentences subtest has been demonstrated in previous research as a highly sensitive measure for discriminating between children with and without language impairment, including those with SLI (Botting & Conti-Ramsden, 2003; Conti-Ramsden, Botting, & Faragher, 2001).

about poor pragmatic skills in children with congenital VI have been raised previously in research studies looking primarily at young preschool children (Fraiberg, 1977; Preisler, 1991; Urwin, 1978), and research attempts to address this issue systematically with school-age children have been methodologically limited (James & Stojanovik, 2007). The current research demonstrates that pragmatic language concerns in children with VI are present at school age and in those children who are linguistically advanced, highlighting the non-verbal aspects of pragmatics as a particular challenge.

It is possible that the parental ratings on the CCC-2 were negatively biased towards the children with VI, as this questionnaire has not been developed with such children in mind and is therefore less sensitive to their strengths. On the other hand, such strong disadvantage of children with VI may reflect the possibility that pragmatic language skills are particularly vision-driven. The non-verbal aspects of pragmatics in particular, such as use of facial expressions and gestures, may be the most potent communicative tools in maintaining a conversational partner's focus of attention. Similarly, the ability to initiate conversations appropriately, understand irony and sarcasm, and adjust conversational topics based on others' levels of interest may be easier to achieve through monitoring of the conversational partner's facial expressions and bodily gestures. Interestingly, the visual nature of pragmatics may be the reason for why pragmatic language may not benefit from scaffolding in the same way as structural language does. This is because pragmatic language is possibly more dependent on successful development of joint attention in early childhood than are structural language skills, which seem to develop without much difficulty in children with VI. On the other hand, early joint attention is a recognised area of developmental vulnerability in such children, and poor communicative pragmatics of verbally able children with VI at school age may be a consequence of this vulnerability. This is certainly in line with the developmental patterns observed in autism, as even high-functioning children with autism, who show better language outcomes, show poor use of socio-pragmatic language (Dennis, Lazenby, & Lockyer, 2001; Klin, 2000).

Following from this, it may not be surprising that the VI group obtained notably impoverished ratings on the CCC-2 scales targeting autism-related social difficulties, as well as on the SCQ. Their uneven CCC-2 profiles, marked by a disproportional weakness in social and pragmatic skills relative to the presentation of structural language skills, are reminiscent of the presentation of autism in sighted children, and a substantial proportion of the children with VI reached or exceeded the clinical cut-off for autism concern on both the CCC-2 and the SCQ.

These findings support the previous research that raised a concern about the alarming prevalence of autism-like features in children with congenital VI of different ages (Brown et al., 1997; Cass et al., 1994; Dale & Sonksen, 2002; Hobson, Lee, & Brown, 1999). Hobson and collaborators, for instance, reported autism-like clinical features across a cognitively heterogeneous population of children with congenital and total sight loss of preschool and primary school age, although it was significantly more prominent in those children who had learning difficulties. The present research provides an important additional insight in that autism-like socio-communicative vulnerabilities have been observed in an intellectually homogeneous group of children VI with advanced language skills, some of whom have above average verbal IQs. Interestingly, sighted children with high-functioning autism and good language outcome may provide a useful comparison group in further research, to help illuminate some of the subtleties that underlie the autistic-like presentation in verbally proficient children with congenital VI. Furthermore, while these researchers investigated autistic-like characteristics exclusively in children with total sight loss, the present research provides unique evidence that such characteristics are also prevalent amongst children with some limited levels of 'form', although severely degraded, vision. It appears that children with a congenital vision loss, whether profound or severe, are at risk of autistic-like socio-communicative difficulties, as presentation of autistic-like features in the current sample of children with VI was not confined to children with profound VI.

The reasons for why such children are at risk of adverse socio-communicative outcomes are not known. One explanation is that significant vision loss in early childhood may impose seriously limited

opportunity to engage in that very special form of relatedness, affective sharing and perspective taking that the context of joint attention provides to the child and their interactional partner, creating a developmental vulnerability with possible long-term consequences (Hobson, 1993). This vulnerability may also account for why even those children with VI whose socio-communicative profiles were within normal range limits did not reach the levels of socio-communicative competence that is typical of the majority of sighted children. Another explanation is that visual impairment may interact with an inherent neurological susceptibility and an adverse environmental climate at a critical stage of development (Cass et al., 1994; Sonksen & Dale, 2002). Individual variation within the VI sample provides suggestive evidence for a multi-factorial aetiology. Despite their good language and verbal intelligence, some children with VI presented with autistic-like behaviours more strongly than others, and to the extent that may warrant further clinical evaluation. The individual characteristics of those children whose socio-communicative profiles scores are in the normal range did not provide any insights with respect to the potential factors that may contribute to their seemingly better socio-communicative outcomes and further research may be necessary to clarify the underlying reasons for this variation.

Finally, utilising more structured measures and direct clinical assessments may be a useful addition to the parental questionnaires in future studies of this kind. However, the existing clinical measures that target autism-related socio-communicative problems are not developed for children with VI and are likely to be less sensitive to their developmental strengths, emphasising the need for VI-specific measures to be developed. With this in mind, parents are a valuable source of knowledge about their children and are likely to provide a window into their children's characteristics that may not be easy to evaluate otherwise. Therefore, parental reports provided a valuable insight into the everyday socio-communicative competencies and vulnerabilities of children with VI in the present study. Further research is now required to establish the developmental cause, genesis and maintaining factors of these vulnerabilities. Greater insight into these aspects may provide the platform for potential

preventative and habilitative interventions that can assist socio-communicative and pragmatic development of children with VI.

Acknowledgements: The authors would like to thank the children and their families who have kindly agreed to take part in this research, which was supported by a 1+3 Economic and Social Research Council studentship awarded to the first author (PTA-031-2004-0021).

Key points:

- Children with congenital VI have been associated with poor socio-communicative outcomes, including resemblances to sighted children with autism, although learning difficulties have been identified as a confounding factor.
- The current study demonstrates that, even with intact verbal intelligence and superior structural language skills, children with congenital VI are at risk of socio-communicative difficulties, with an ongoing concern at primary school age.
- Pragmatic language skills of such children seem to be disproportionately impaired compared to their strengths in structural language.
- Future interventions may particularly benefit from such children's developmental potential in structural language.

Matching criteria	VI	Sighted	
	N=15	N=26	
VIQ/WISC-III			
Mean (SD)	105.9 (10.7)	106.3 (11.1)	
Range	84-128	80-130	
Age			
Mean in months (SD)	103.1 (23.0)	106.5 (20.3)	
Range in years	6:06-12:11	6:02-11:11	
Gender			
Female	60%	54%	

Table 1: Matching characteristics of the sample

Measure	VI	Sighted	p level
Mean (SD)			
CELF - 3			
Receptive Language Composite	104.5 (10.3)	96.7 (9.1)	*
Word Classes	11.4 (2.4)	10.04 (2.4)	n. s.
Listening to Paragraphs	9.9 (1.5)	8.8 (2.6)	n. s.
Expressive Language Composite	113.8 (15.6)	102.5 (14.4)	*
Recalling Sentences	12.6 (2.7)	9.8 (3.1)	**
Word Associations	11.9 (3.5)	10.7 (2.6)	n. s.
Total Language Composite	109.6 (12.9)	99.3 (11.3)	**

Table 2: CELF-3 - Group means and standard deviations (SD)

n. s. – not significant; * significant at $p \le .05$; ** significant at $p \le .01$

Measure	VI	Sighted	p level
Mean (SD)			
CCC – 2	N missing = 1	N missing = 1	
Structure domain			
Speech	9.9 (3.5)	10.3 (2.6)	n. s.
Syntax	9.6 (3.6)	10.9 (1.9)	n. s.
Semantics	8.6 (3.1)	11.1 (2.7)	**
Coherence	8.7 (3.3)	11 (2.5)	*
Pragmatic Domain			
Inappropriate Initiation	7.5 (3.1)	11 (2.6)	**
Stereotyped language	6.6 (3.6)	10.3 (3.3)	**
Use of Context	6.5 (2.4)	11.2 (2.9)	**
Non verbal	4.3 (2.6)	10.9 (2.7)	**
Social Interaction Domain			
Social	5.4 (2.8)	10.4 (2.9)	**
Interests	6.5 (2.5)	9.5 (2.9)	**
General Communication Composite	61.8 (18.8)	86.8 (14)	**
(GCC)			
Social Interaction Deviance	-13.14 (7.04)	-1.5 (7.7)	N/A
Composite (SIDC)			
SCQ Lifetime Autoscore		N missing = 1	
Total score	14.3 (3.9)	4.4 (3.9)	**
Reciprocal Social Interaction	4.1 (2.3)	0.9 (0.9)	**
Communication	5.3 (2.0)	2.1 (2.0)	**
Restricted, Repetitive and	4.5 (2.0)	1.2 (2.0)	**
Stereotyped Behaviours			

Table 3: CCC-2 and SCQ – Group means and SDs

n. s. – not significant; * significant at $p \le .05$; ** significant at $p \le .01$

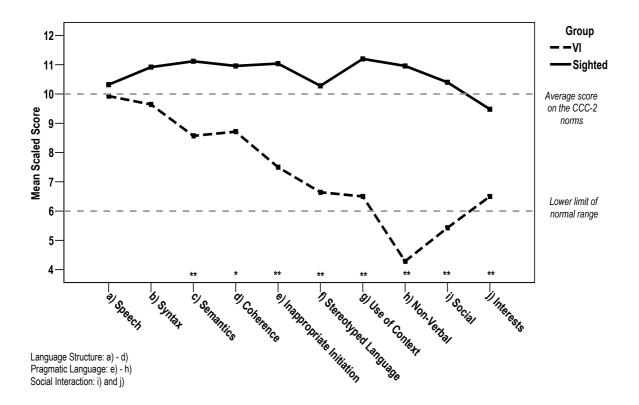


Figure 1: Differing CCC-2 profiles in the two groups (** $p \le .01$; * $p \le .05$)

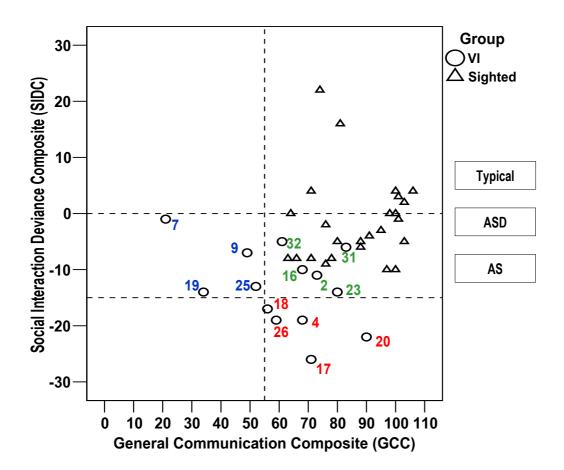


Figure 2: Individual children's CCC-2 profiles (ID numbers: children with VI)

References

- Andersen, E. S., Dunlea, A., & Kekelis, L. S. (1984). Blind children's language: Resolving some differences. *Journal of Child Language*, *11*(3), 645-664.
- Bishop, D. V. M. (2003). *The Children's Communication Checklist (CCC) 2.* London: The Psychological Corporation.
- Bishop, M., Hobson, R. P., & Lee, A. (2005). Symbolic play in congenitally blind children. *Development and Psychopathology*, *17*(2), 447-465.
- Botting, N., & Conti-Ramsden, G. (2003). Autism, primary pragmatic difficulties, and specific language impairment: can we distinguish them using psycholinguistic markers? *Developmental Medicine and Child Neurology*, *45*(8), 515-524.
- Brown, R., Hobson, R. P., Lee, A., & Stevenson, J. (1997). Are there 'autistic-like' features in congenitally blind children? *Journal of Child Psychology and Psychiatry*, *38*(6), 693-703.
- Cass, H. D., Sonksen, P., & McConachie, H. R. (1994). Developmental Setback in Severe Visual Impairment. *Archives of Disease in Childhood*, *70*(3), 192-196.
- Chess, S. (1971). Autism in children with congenital rubella. *Journal of Autism and Developmental Disorders*, 1(1), 33-47.
- Conti-Ramsden, G., Botting, N., & Faragher, B. (2001). Psycholinguistic Markers for Specific Language Impairment (SLI). *Journal of Child Psychology and Psychiatry*, 42(6), 741-748.
- Dale, N., & Sonksen, P. (2002). Developmental outcome, including setback, in young children with severe visual impairment. *Developmental Medicine and Child Neurology*, 44(9), 613-622.
- Dennis, M., Lazenby, A. L., & Lockyer, L. (2001). Inferential language in high-function children with autism. *Journal of Autism and Developmental Disorders*, *31*(1), 47-54.
- DSM-IV-TR. (2000). *Diagnostic and Statistical Manual of Mental Disorders Text Revision* (4 ed.). Washington DC: American Psychiatric Association.
- Dyck, M. J., Farrugia, C., Shochet, I. M., & Holmes-Brown, M. (2004). Emotion recognition/understanding ability in hearing or vision-impaired children: do sounds, sights, or words make the difference? *Journal of Child Psychology and Psychiatry*, *45*(4), 789-800.
- Fraiberg, S. (1977). Insights from the blind. London: Souvenir.
- Green, S., Pring, L., & Swettenham, J. (2004). An investigation of first-order false belief understanding of children with congenital profound visual impairment. *British Journal of Developmental Psychology*, 22(1), 1-17.
- Hobson, R. P. (1993). Autism and the development of mind. Hove: Earlbaum.
- Hobson, R. P., Lee, A., & Brown, R. (1999). Autism and congenital blindness. *Journal of Autism and Developmental Disorders*, 29(1), 45-56.
- Hull, T., & Mason, H. (1995). Performance of Blind Children on Digit-Span Tests. *Journal of Visual Impairment and Blindness*, 89(2), 1666-1169.
- James, D. M., & Stojanovik, V. (2007). Communication skills in blind children: a preliminary investigation. *Child: Care, Health and Development, 33*(1), 4-10.
- Klin, A. (2000). Attributing Social Meaning to Ambiguous Visual Stimuli in Higher-functioning Autism and Asperger Syndrome: The Social Attribution Task. *Journal of Child Psychology and Psychiatry*, *41*(7), 831-846.
- Landau, B., & Gleitman, L. R. (1985). *Language and Experience: Evidence from the Blind Child*. Cambridge: Harvard University Press.
- Lewis, V., Norgate, S., Collis, G. M., & Reynolds, R. (2000). The consequences of visual impairment for children's symbolic and functional play. *British Journal of Developmental Psychology*, *18*, 449-464.

- Lloyd, H., Paintin, K., & Botting, N. (2006). Performance of children with different types of communication impairment on the Clinical Evaluation of Language Fundamentals (CELF). *Child Language Teaching and Therapy*, 22(1), 47-67.
- McConachie, H. R., & Moore, V. (1994). Early Expressive Language of Severely Visually-Impaired Children. *Developmental Medicine and Child Neurology*, *36*(3), 230-240.
- Mills, A. E. (1993). Visual handicap. In D. Bishop & K. Mogford (Eds.), *Language development in exceptional cicumstances* (pp. 150–164). Hove: Earlbaum.
- Minter, M., Hobson, R. P., & Bishop, M. (1998). Congenital visual impairment and 'theory of mind'. *British Journal of Developmental Psychology, 16*, 183-196.
- Minter, M., Hobson, R. P., & Pring, L. (1991). Recognition of Vocally Expressed Emotion by Congenitally Blind-Children. *Journal of Visual Impairment & Blindness*, 85(10), 411-415.
- Mulford, R. (1983). Referential development in blind children. In A. E. Mills (Ed.), *Language Acquisition in the Blind Child: Normal and Deficient.* London: Croom Helm.
- Mulford, R. (1988). First words of the blind child. In M. Smith & J. Locke (Eds.), *The emergent lexicon: The child's development of a linguistic vocabulary* (pp. 293–338). New York: Academic Press.
- Norgate, S., Collis, G. M., & Lewis, V. (1998). The developmental role of rhymes and routines for congenitally blind children. *Cahiers De Psychologie Cognitive-Current Psychology of Cognition*, 17(2), 451-477.
- Pérez-Pereira, M., & Conti-Ramsden, G. (1999). *Language development and social interaction in blind children*. Hove: Psychology Press.
- Peterson, C. C., Peterson, J. L., & Webb, J. (2000). Factors influencing the development of a theory of mind in blind children. *British Journal of Developmental Psychology*, *18*(3), 431-447.
- Preisler, G. M. (1991). Early patterns of interaction between blind infants and their sighted mothers. *Child: Care, Health and Development, 17*(2), 65-90.
- Pring, L. (2005). Autism and Blindness: Research and Reflections. London: Whurr.
- Reynell, J. (1978). Developmental patterns of visually handicapped children. *Child: Care, Health and Development, 4*(5), 291–303.
- Roch-Levecq, A. (2006). Production of basic emotions in children with congenital blindness: Evidence for the embodiment of Theory of Mind. *British Journal of Developmental Psychology*, 24, 507-528.
- Rutter, M., Bailey, A., & Lord, C. (2003). *The Social Communication Questionnaire (SCQ)*. Los Angeles: Western Psychological Services.
- Semel, E., Wiig, E. H., & Secord, W. (2000). *Clinical Evaluation of Language Fundamentals (CELF)* (UK, 3 ed.). London, UK: The Psychological Corporation.
- Smits, B. W. G. M., & Mommers, M. J. C. (1976). Differences between blind and sighted children on WISC verbal subtests. *New Outlook for the blind, 70,* 240-246.
- Sonksen, P., & Dale, N. (2002). Visual impairment in infancy: impact on neurodevelopmental and neurobiological processes. *Developmental Medicine and Child Neurology*, 44(11), 782-791.
- Sonksen, P., Petrie, A., & Drew, K. J. (1991). Promotion of Visual Development of Severely Visually-Impaired Babies - Evaluation of a Developmentally Based Program. *Developmental Medicine* and Child Neurology, 33(4), 320-335.
- Tadić, V., Pring, L., & Dale, N. (2008). Attentional processes in young children with congenital visual impairment. *British Journal of Developmental Psychology*. In press (available online).
- Tomasello, M., & Farrar, M. J. (1986). Joint Attention and Early Language. *Child Development*, 57(6), 1454-1463.
- Tröster, H., Brambring, M., & Beelmann, A. (1991). Prevalence and situational causes of stereotyped behaviors in blind infants and preschoolers. *Journal of Abnormal Child Psychology*, *19*(5), 569-590.

- Urwin, C. (1978). The development of communication between blind infants and their parents. In A. Lock (Ed.), *Action, gesture and symbol: The emergence of language* (pp. 79-108). London: Academic Press.
- Urwin, C. (1983). Dialogue and cognitive functioning in the early language development of three blind children: Normal and deficient. In A. E. Mills (Ed.), *Language Acquisition in the Blind Child* (pp. 142-161). London: Croom Helm.
- Wechsler, D. (1992). Wechsler Intelligence Scale for Children (WISC) (UK, 3 ed.). London, UK: The Psychological Corporation.