Developmental Cognitive Neuroscience 27 (2017) 10–18

Contents lists available at ScienceDirect

Developmental Cognitive Neuroscience

journal homepage: www.elsevier.com/locate/dcn

Event-related potential response to auditory social stimuli, parent-reported social communicative deficits and autism risk in school-aged children with congenital visual impairment

Joe Bathelt[a,*], Naomi Dale[b,c], Michelle de Haan[c]

[a] MRC Cognition & Brain Sciences Unit, University of Cambridge, Cambridge, United Kingdom
[b] Developmental Vision Clinic, Great Ormond Street Hospital for Children NHS Foundation Trust, United Kingdom
[c] Developmental Neurosciences, UCL Great Ormond Street Institute of Child Health, United Kingdom

ARTICLE INFO

Keywords:
Social development
Childhood
Visual impairment
Autism spectrum disorder
Event-related potentials

ABSTRACT

Communication with visual signals, like facial expression, is important in early social development, but the question if these signals are necessary for typical social development remains to be addressed. The potential impact on social development of being born with no or very low levels of vision is therefore of high theoretical and clinical interest. The current study investigated event-related potential responses to basic social stimuli in a rare group of school-aged children with congenital visual disorders of the anterior visual system (globe of the eye, retina, anterior optic nerve). Early-latency event-related potential responses showed no difference between the VI and control group, suggesting similar initial auditory processing. However, the mean amplitude over central and right frontal channels between 280 and 320 ms was reduced in response to own-name stimuli, but not control stimuli, in children with VI suggesting differences in social processing. Children with VI also showed an increased rate of autistic-related behaviours, pragmatic language deficits, as well as peer relationship and emotional problems on standard parent questionnaires. These findings suggest that vision may be necessary for the typical development of social processing across modalities.

1. Introduction

The ability of adults to infer the mental state of others by interpreting fleeting contractions of facial muscles is an amazing feat of our species. Developmental studies show that infants are intuitively drawn to faces (Johnson et al., 1991, 2000) and engage in reciprocal communication with their caregivers through facial expressions from just a few months of age (Leppänen and Nelson, 2008). Non-verbal signals, like facial expression, eye gaze, and posture, are a rich source of interpersonal communication signals. Reduced visual attention to these cues has been found in disorders of social development and some theories suggest a causal link between attending to facial communication cues and social deficits in autism spectrum disorder (ASD) (Campbell et al., 2006). The potential impact on social development of being born with no or very low levels of vision is therefore of high theoretical interest and clinical concern. Without visual interpersonal abilities, children with congenital visual impairment (VI) may be at higher risk of social deficits. They also provide a natural experiment to study the effects of absent vision from birth on the development of social cognition and behaviour. The current study, therefore, sets out to investigate if differences in social responses and behaviour, including social communicative/ASD risks, are found between children with congenital VI and typically-sighted controls, with a particular focus on neural responses toauditory social stimuli and their associations with parent-reported measures of social communicative and behavioural function and risk.

Children with congenital VI have been found to deviate from the typical trajectory of social development observed in typically-sighted children. Firstly, children with VI pass social cognitive Theory of Mind tests at a later age compared to typically-sighted children (Brambring and Asbrock, 2010; Peterson et al., 2000; Minter et al., 1998; Green et al., 2004; McAlpine and Moore, 1995). Secondly, a relatively large proportion of children with VI display behaviours that are commonly associated with autism, like stereotyped movement, echolalic speech, and lack of engagement with caregivers and peers (Preisler, 1991; Brown et al., 1997; Hobson and Bishop, 2003; Tadic et al., 2009). Difficulties in social processing persist in mid-childhood and potentially into adolescence. For instance, a study in a specialist secondary school indicated that 22% of students with VI met clinical criteria for ASD (Mukkades et al., 2007, also see Jure et al., 2016). Analyses of clinically

* Corresponding author.
E-mail address: joe.bathelt@mrc-cbu.cam.ac.uk (J. Bathelt).

http://dx.doi.org/10.1016/j.dcn.2017.07.003
Received 8 February 2017; Received in revised form 27 May 2017; Accepted 10 July 2017
Available online 19 July 2017

referred samples suggest an even higher prevalence (Absoud et al., 2010; Williams et al., 2013; Rogers and Newhart-Larson, 2008). However, Hobson et al. (2010) provided some evidence of later 'recovery' from early symptomatology This potential change in the trajectory of social processing makes mid-childhood a particularly interesting period to investigate if qualitative differences in the processing of social stimuli can be detected.

Behavioural studies of social function in VI to date have some significant methodological limitations. First, many previous studies included congenital visual disorders that also involve central brain structures or included children with low intellectual functioning. It is therefore not clear if VI, low intellectual ability, or brain damage is affecting social development. In this study, we addressed these limitations by investigating social development in VI in a group of children with relatively pure disorders of the anterior visual system, who also scored in the normal range for verbal IQ.

Another potential limitation of previous studies concerns the behavioural assessment methods. Standard assessments of social cognition and Theory of Mind for sighted children rely predominantly on visually presented materials, e.g. recognising facial expressions or inferring the mental state of actors or puppets. Some attempts have been made to adapt materials for children with VI (Brambring and Asbrock, 2010), but these assessments have not been sufficiently tested and standardised in a representative sample − probably due to the rarity of isolated VI disorders. Other studies rely on verbally presented materials (Pijnacker et al., 2012) that are potentially confounded by differences in language development in children with VI, such as linguistic delays and difficulties in semantic and pragmatic comprehension (Andersen et al., 1993; Mulford, 1988; Wakefield et al., 2006). Therefore, the current study used a more direct measurement of auditory social stimulus processing in an electrophysiological experiment, which was not dependent on vision or language ability or verbal working memory.

We hypothesised that differences in the processing of social stimuli in children with VI would be apparent in responses to auditory social stimuli. The 'subjects-own-name' (SON) paradigm was considered particularly suitable as it assesses responses to a basic social stimulus in the auditory domain, ie. hearing one's own name spoken. Own-name stimuli have been shown to be processed preferentially, automatically, and without conscious control in the typically-sighted child from as early as 5 months of age (Alexopoulos et al., 2012; Parise et al., 2010; Perrin et al., 2006; Pfister et al., 2012). Differences in processing of own-name stimuli, compared with typically developing children, have been associated with deficits in mentalizing ability in children with high-functioning autism (Cygan et al., 2014; Lombardo et al., 2009). In the event-related response, own-name stimuli consistently elicit an early negative deflection with a maximum over frontal channels followed by later positive deflection similar to the orienting-P3 (P3a) in typical adults (Tateuchi et al., 2012, 2015). Manipulations of presentation frequency indicate that the early SON-related negativity is distinct from the orienting P3 (Eichenlaub et al., 2012). Further, comparisons of responses to names of close family members indicate that the SON-related negativity is specific to the subject's own name rather than personally familiar names in general (Tateuchi et al., 2015). Neural generators of SON-related processing are likely to involve a network of areas, including the right superior temporal sulcus (STS), medial prefrontal cortex (PFC), and inferior parietal sulcus (IPS) (Holeckova et al., 2006; Perrin et al., 2005; Kampe et al., 2003). Using the event-related response in the SON paradigm, we anticipated that there may be a reduction in amplitude of the SON-related response in children with VI similar to effects observed in other groups with suspected deficits in social processing (Cygan et al., 2014; Nadig et al., 2007).

A school-aged sample of children with congenital disorders of the peripheral visual system (CDPVS), aged 8–13 years of normal verbal intelligence, were recruited for the investigation with matched controls of typical sight. Parent-reported questionnaires that are validated on children with typical sight for social communication difficulties, pragmatic language disorder, autism risk, and behavioural strengths and difficulties were included to assess the level of difficulties in the social domain. It was hypothesised further that individual differences between children with VI on the questionnaire measures (Sonksen and Dale 2002; Tadic et al., 2009) would be associated with differences in the ERP response to SON.

2. Materials and methods

2.1. Participants

The assessments presented here were part of a wider study on the neural and cognitive sequelae of congenital VI during mid-childhood. This study was performed in accordance with the Declaration of Helsinki. The study was approved by Bloomsbury Research Ethics Committee − approval: 12/LO/0939. All parents, guardians or next of kin provided written informed consent and children provided verbal assent to participate in this study.

A prospective cross-sectional study was undertaken with eighteen children with VI aged between 8 and 13 years. Congenital disorders of the peripheral visual system with severe VI are rare with an estimated prevalence of less than 2–3 per 10,000 children (UK) raising challenges for recruitment and sampling (Rahi, Cable, BCVISG, 2003). Children were therefore recruited through national specialist clinics at Great Ormond Street Hospital for Children NHS Foundation Trust and Moorfields Eye Hospital NHS Foundation Trust. Inclusion criteria: i) children with congenital VI in the moderate to profound range with primary diagnosis of 'potentially simple' congenital disorders of the peripheral system (CDPVS), that is disorders affecting the globe of the eye, retina, or anterior optic nerve up to the optic chiasm, with no known brain disorder indicated by the paediatric or ophthalmological diagnosis (Sonksen and Dale, 2002), ii) between 8 and 13 years, iii) good verbal functioning (verbal IQ at the last assessment > 75 or attending mainstream school at age-appropriate level), iv) English as their first language. Children with indications of additional neurological or endocrine abnormalities in their clinical records were excluded. Recruitment was undertaken through initial identification through clinical databases of children who had attended a tertiary paediatric specialist clinic at the hospital research site and open recruitment call through voluntary agencies associated with VI.

Children in the typically-sighted control group were recruited to match the same age range and were included if they fit the following criteria: attend mainstream school at the age-appropriate level, have no known neurological or psychiatric conditions, have either normal or corrected-to-normal vision, and have English as a first language. Sample characteristics are presented in Tables 1 and 2.

The full sample consisted of 18 children with VI (9 female) between 8 and 13 years of age (mean age: 10.76, age SD: 1.39, age range: 8.27-13.32) and a control group of 18 typically-sighted children (8 female, mean age: 10.62, age SD: 1.44, age range: 8.73-12.92). Verbal comprehension was assessed using verbal subtests of the Wechsler Intelligence Scale for Children 4th edition (WISC-IV) (Wechsler, 2004). Verbal subtests of previous and current editions of the WISC have also been used with children with VI (Greenaway et al., 2016; Dekker, 1993; Tillman, 1973; Tillman and Bashaw, 1968; Witkin et al., 1968). The administered subtests included all items of the Verbal Comprehension composite score (Vocabulary, Similarities, Comprehension). Two items were altered that required direct visual experience: The WISC-IV first practice item on the Similarities subtest which includes colour was not administered. The Comprehension question that asks about a situation in which 'you see thick smoke' was changed to 'you smell thick smoke'. These alterations were used for the whole sample, including the typically-sighted control group. All other items were administered verbatim according to the WISC-IV administration manual (Wechsler, 2004). There was no significant difference in verbal IQ composite scores