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BRIEF REPORT

Testing the Hard to Test: A Pilot Study Examining the Role of Questionnaires in Eliciting Visual Behaviours in Children with Autistic Spectrum Disorder

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Abstract

Purpose There is a higher prevalence of visual problems in children with Autistic Spectrum Disorder (ASD) compared with the neuro-typical population. Issues relating to visual perception can be evident as atypical visual behaviours (ViBes). The aim of the study was to compare findings of parents and eye health professionals using questionnaires to describe visual function in young children with ASD.

Materials and methods Parents/Caregivers of patients diagnosed with ASD attending routine hospital eye clinics were invited to participate. A questionnaire to draw out presence of ViBes was completed by the parent prior to attending clinic. The clinician independently completed the same questionnaire and undertook a clinical ocular assessment.

Results 32 children were recruited (mean age 7 years, range 4–11). Most caregivers indicated they had noted at least one atypical ViBe (97%, median 5, range 0–8). Parental-reported ViBe8 scores were higher than clinician-reported scores ($p = 0.04$). The most commonly reported ViBes by both groups related to use of vision at the same time as other senses, and atypical fixation (e.g. looking away during speech or side viewing). There was weak correlation between visual acuity and parental ViBe8 score (rho = 0.364) and no correlation between refractive error and parental ViBe8 score (rho = 0.047).

Conclusion Clinicians were likely to under-report atypical visual function compared with parent. Refraction and visual acuity alone do not detect all visual problems in children with ASD. A Visual Behaviour (ViBe) questionnaire offers a structured approach and shared language to allow documentation of functional visual assessment for both parents, carers, and educational professionals. Use of the ViBe questionnaire may promote understanding between caregiver and professional and provide a baseline for visual behaviours.

Keywords Autistic Spectrum Disorder · Vision · Children · Optometry · Screening

Purpose

The prevalence of Autistic Spectrum Disorder (ASD) has increased significantly in recent decades across the world (Chiarotti, 2020). In their 2018 publication, Autism Spectrum Disorders in the EU reported the average prevalence across the EU to be 1.12%.

Several reviews have reported a higher prevalence of visual problems in children with ASD compared to the neuro-typical population (Little, 2018, Simmons, 2009). There is therefore a need for health professionals to be aware of the spectrum of visual impairments which may affect a child with ASD, including issues with visual perception, motion processing, and facial recognition.

Those caring for and teaching children with ASD often observe atypical visual behaviours (ViBes) manifesting as a sensory attentional disorder, often in conjunction with difficulties integrating central and peripheral field, and/or visual memory or facial recognition deficits (McKay, 2012; Schwarzkopf, 2014; Laycock, 2020 ; DeRamus, 2014).

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These can appear similar to the signs of Cerebral visual impairment (CVI). CVI is defined as a verifiable visual dysfunction which cannot be attributed to disorders of the anterior visual pathways or any potentially co-occurring ocular impairment (Sakki, 2018).There is an emerging understanding of the overlap in diagnostic features of ASD and CVI (Sakki, 2018). Disentangling the contributions of visual perception amongst other neurological deficits when assessing a child's atypical behaviours is a challenge. To our knowledge there has only been a single study examining the presence of CVI in those with ASD (Bhaskaran, 2018). What is currently lacking are clinical assessment tools for describing and documenting the functional visual abilities and behaviour in young children with ASD.

ViBe 8 is a questionnaire composed of eight items, derived from analysis of the most frequently recorded ViBes observed in a cohort of young children with ASD during in-school visual assessments. Caregivers are asked to state how often, if at all, their child demonstrates any of the visual behaviours described (see Appendix 1).

The CVI 5 questionnaire is composed of 5 statements relating to areas of visual function. (see Appendix 2). The questionnaire has been validated for use in identifying children for whom further CVI assessment is appropriate (Gorrie, 2019). Its use in a population of children with ASD has not been previously explored.

The aim of this study was comparing parental and clinician reported visual behaviours using CVI5 and ViBe8 questions and relate these to clinical ocular assessment findings.

Methods

All eligible children attending for routine eye exams in the additional needs clinic or hospital eye clinic between March 2021 and March 2022 were invited to participate. Children were considered eligible if aged 4–12 years and had a confirmed diagnosis of ASD (DSM-V criteria). Children were excluded if parents had not completed the questionnaire prior to clinic arrival or they had known ocular visual impairment or other systemic comorbidities (e.g., cerebral palsy, hydrocephalus).

A questionnaire composed of 13 items (combined ViBe8 and CVI5) was sent by post to parents to complete at home prior to their scheduled attendance at the eye clinic. The questionnaire employed a Likert scale for parent/care giver to indicate how often they had observed specific atypical visual behaviours; always; often; sometimes; rarely; never; not applicable. A response of 'always' or 'often' is coded as a positive result indicating the presence of atypical visual behaviour. Missing data or 'not applicable' responses were coded as a zero response (Gorrie, 2019). The same questionnaire was completed by a clinician independently, i.e., without sight of the parent/carer questionnaire based on their observations during the clinic visit.

Clinical assessment comprised visual acuity measurement (with spectacles if worn), ocular alignment, ocular movements, stereopsis (Lang II or Frisby), confrontational visual field, refraction and ophthalmoscopy.

An observational approach was taken when assessing the saccadic type of eye movements. Two small targets (of interest) were used, and the child was instructed to alternately fixate between the two targets, in the vertical and horizontal planes. Both accuracy and latency were estimated and categorised as follows: Accurate/no delay in response; Accurate/delay in response; Inaccurate/no delay in response; Inaccurate/delay in response.

Wilcoxon signed rank tests were used to test the difference between the groups, as the data was nonparametric, and a small sample size. Correlation coefficients were used to explore the relationships between different variables e.g.: ViBe8 responses and visual acuity.

Ethical approval for the study was granted from CHI @ Temple Street (National Paediatric Hospital, Dublin, Ireland) and Cardiff University, Wales, U.K.

Results

There were 32 children whose caregivers agreed to participate. There were 21 male and 11 were female participants with a median age of 7 years (range 4–11 years).

Table 1 describe the number of positive responses to each of the ViBe8 questions.

Thirty-one (97%) caregivers indicated at least one positive response on the ViBe8 questionnaire. The median number of positive responses per child based on clinician reporting was 3 (range 1–8) and the median number of positive responses per child based on parental reporting was 5 (range 1–8). Three (9.4%) caregivers indicated at least one 'not applicable' response, with a median of one.

Table 1 Comparison of parental and clinican positive responses to each of the ViBe8 questions

ViBe8 question	Parental positive response %	Clinician positive response %
Bump into/walk through object	25	6
Hold items close to examine	47	58
Side view	50	48
Distracted by periphery	56	29
Trail hand	62	23
Rock or flap	53	58
Look away during speech	72	58
Use other senses	62	74

The most frequently parental reported atypical ViBe was 'looks away while speaking'. The most frequently clinician reported atypical ViBe related to observing the child using other senses instead of vision. Clinicians underreported items relating to movement around the environment compared to parents.

A Wilcoxon signed rank test indicated there was significant difference between parental and clinic total scores for ViBe 8 (z=-2.858, $p=0.04$). It would seem ViBe8 score based on parental reporting does not reflect that of the clinician observed score, with parents indicating a greater number of issues.

Thirty (93.7%) caregivers indicated at least one positive response on the CVI5 questionnaire. The median number of positive responses per child based on clinician reporting was 1.5 (range 0–5) and the median number of positive responses per child based on parental reporting was 2 (range 0–5). Eight (25%) caregivers indicated at least one 'not applicable' response (median 0, range 0–2). Parental scores were higher than clinical observed scores, indicating the presence of atypical visual function present at home which was not evident in clinic.

Wilcoxon signed rank test (non-parametric data) indicated there was significant difference between parental and clinic total scores for CVI5 (z=-1.548, $p=0.122$). A higher number of respondents replied not applicable to one or more CVI5 questions ($n=8$; 25%) compared with ViBe8 not applicable responses ($n=3$, 9%).

To compare the CVI5 and ViBe8 questionnaires, the percentage of total possible scores was calculated for each participant as there were a different number of questions in CVI5 and ViB8. There appears to be a weak positive correlation (rho = 0.331) between the ViBe8 and CVI5 based on clinic scores. There is a slightly stronger positive correlation (rho = 0.389) between ViBe8 and CVI5 based on parental scores. This does not suggest that we can predict one from another but suggests that those that score highly on ViBe8 are more likely to score high on the CVI5 questions.

Visual acuity was normal in all but one patient. The mean spherical equivalent refraction was + 0.9DS. The most common atypical clinical findings related to eye movements; 40.6% ($n=13$) demonstrated abnormal saccadic type movement; 31.25% ($n=10$) had anomalous smooth pursuit. Eight of the participants demonstrated visual field inattention in one or more quadrant. Stereoacuity of 85 s or better was only achieved by 22% of the participants.

There appears to be a weak correlation between visual acuity and the total ViBe8 score (rho = 0.364) and a very weak correlation between visual acuity and the total CVI5 score based on parental observations (rho = 0.096). There appears to be no correlation between the requirement for glasses and CVI5 or ViBe8 responses (rho = 0.047, rho = 0.08) based on parental responses.

Discussion

The study has demonstrated the role of questionnaires in understanding elements of visual function in a child with ASD which are not captured by a routine community eye test. The higher parental reported ViBe scores (indicating more atypical visual behaviours) were in domains which are difficult to capture in a clinical setting, namely use of vision in the wider environment. It is of interest that while clinicians did not report these behaviours on the questionnaire, an allied clinical feature (e.g. visual field inattention, atypical eye movements) was elicited. The interplay between ASD, intellectual disability and CVI is drawing more attention, with considerable overlap in symptomatology, aetiology and proposed neurobiological anomalies (Chokron, 2020). The British Childhood Visual Impairment Study 2 group (BCVIS2) report suggests that paediatric non-eye health specialists should be educated on the symptoms of visual disorders and be able to undertake simple assessments of vision (Solebo, 2022). The types of activities which are included in the CVI5 questionnaire may be less easily noted or may be beyond the cognitive ability of this cohort than those within the ViBe8, as reflected in the number of 'not applicable' responses (25% vs. 9.4% of respondents).

The results from this small cohort are consistent with findings from other groups in that refractive error and visual acuity alone are inadequate to fully capture a child's visual behaviours and demonstrate visual function difficulties in a young child with autistic spectrum disorder. All of the participants demonstrated atypical visual function in one or more domains; the most frequently affected areas of visual function were atypical eye movements (40%) and visual field inattention (25%). Previous studies have reported hypometric saccades in up to 50% of adults with ASD (Bast, 2021; Vinuela-Navarro, 2018). Visual field inattention, however, is an emerging area of visual dysfunction which may merit further investigation.

There are several limitations to the study we present: as a pilot study the number of participants recruited was low and may not be representative of the population. Whilst all parents/caregivers of children with ASD in the age range were invited to participate, there is the potential for inclusion bias, with only those parents who had visual concerns choosing to attend, or those with co-existing ophthalmological diagnoses already being under the care of the hospital eye service. The recruitment criteria excluded children with dual diagnosis of ASD and another cause of developmental delay, particularly those with motor delay. Future studies

would look to understand the validity of the testing protocol in a more diverse and complex needs cohort.

The study benefits from demonstrating a novel approach to vision assessment which could be adopted by existing services without costly investment. Questionnaires are a non-invasive approach to understanding how the child uses their vision in the real world, which crucially do not require the child to engage in a battery of clinical tests. They may support the clinician in prioritising areas of visual testing (for example, to establish the presence of a visual field defect).

The approach facilitates the parent in having an equal voice within the consultation which can be difficult if there is limited time available and/or the focus is on maintaining the child's attention and supporting them in a new and often challenging environment. It offers the clinician a broader awareness of the child's visual strengths and weaknesses which can be limited by a snapshot obtained in a clinic appointment. In an era of transforming outpatient care, utilising telephone or video consultations to support parents in reporting visual behaviours may have a role to play in reducing the barriers to accessing care for families of children with ASD.

Our findings have demonstrated that a Visual Behaviour questionnaire offers a structured approach and shared language which facilitates the documentation of important aspects of visual functional assessment in a child with ASD which may not be present in the clinic environment but will be relevant for the wider health care and educational teams. A useful next step would be to understand if community paediatric teams are able to utilise the tool to identify children with ASD who might benefit from a more detailed eye assessment, to exclude co-existing ocular impairment, and support parents in better understanding their child's visual function.

Appendix 1 ViBe8 Questions

1. How often does your child walk into or through large objects when moving about e.g.: whilst moving across a room to retrieve a favourite object/toy.
2. How often does your child hold objects very close to inspect them in detail/become visually stuck on an object and seem to stop looking.
3. How often does your child turn their face to the side/look at something from the corner of their eye?
4. How often is your child distracted by people or objects moving alongside them e.g.: turning from an activity they are enjoying watching a moving car /family pet/new person entering the room.
5. How often does your child move about a room trailing their hand along surfaces/picking up objects/touching everything in a new environment.
6. How often does your child rock or spin their whole body or flap their hands in front of their eyes?
7. How often does your child look away from you when they are speaking/when you are speaking to them?
8. How often does your child seem more engaged with other sensory activities e.g.: humming/chewing/biting/touching objects rather than using their vision.

Appendix 2 CVI5 Questions

1. How often does your child have difficulty walking downstairs?
2. How often does your child have difficulty seeing things that are moving quickly such as small animals or children?.
3. How often does your child have difficulty seeing something that is pointed out in the distance?
4. How often does your child have difficulty locating an item of clothing in a pile of clothing or a favourite toy in a toy box?
5. How often does your child find copying words or drawing lines time consuming and difficult?

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Declarations

Competing Interests JMW has a financial interest in the Cardiff Acuity Test. RFP is co-creator of the Bradford Visual Function Box.

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