# Developing a schedule to identify social communication difficulties and autism spectrum disorder in young children with visual impairment

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**ABBREVIATIONS**
CARS Childhood Autism Rating Scale
VISS Visual Impairment and Social Communication Schedule

Available observational tools used in the identification of social communication difficulties and diagnosis of autism spectrum disorder (ASD) rely partly on visual behaviours and therefore may not be valid in children with visual impairment. A pilot observational instrument, the Visual Impairment and Social Communication Schedule (VISS), was developed to aid in identifying social communication difficulties and ASD in young children with visual impairment affected by congenital disorders of the peripheral visual system (disorders of the globe, retina, and anterior optic nerve). The VISS was administered to 23 consecutive children (age range 1y 9mo–6y 11mo, mean 4y 1mo [SD 1.6]; 12 males, 11 females) with visual impairment (nine with severe and 14 with profound visual impairment). Item analysis was carried out by fit of the items to the Rasch model. Validity of the VISS was explored by comparison with the Childhood Autism Rating Scale (CARS) score, and the clinical ASD diagnosis ($n$=9). Correlation between the VISS and CARS total scores was highly significant (Spearman's $rho$=–0.89; $p$=0.01). Below threshold rating on the VISS (score of 35) showed good agreement with the clinical ASD diagnosis (sensitivity 89%, specificity 100%). This preliminary study shows the VISS to be a promising schedule to aid the identification of ASD in young children with visual impairment.

Young children with congenital severe visual impairment are at risk of early social communication difficulties; by school age (5–18y), 11 to 40% are reported to meet criteria for clinical autism spectrum disorder (ASD).[1–7] Although the mechanisms underlying the development of social communicative difficulties and ASD are not yet understood in the visually impaired population, early evidence points towards interactions of multiple factors (visual, age, sex, psychological, neurological) at different levels.[6] Because of the availability and potential importance of early intervention strategies in both visual impairment and ASD,[8,9] early accurate identification of social communication difficulties and ASD in children with visual impairment is desirable.

Available observational tools used to identify early social communication difficulties and ASD (e.g. the Autism Diagnostic Observation Schedule[10]) rely partly on visual behaviours, such as eye contact, joint attention, gesture, and are not validated for the visually impaired population. This presents a challenge for visual impairment researchers and professionals involved in diagnosing and giving advice on appropriate management of children with visual impairment with a suspected social communication disorder. Recent research studies[2,3] have used the Childhood Autism Rating Scale (CARS) in identifying ASD and some items relate to visual behaviours. We therefore aimed to develop an observational instrument, the Visual Impairment and Social Communication Schedule (VISS), to help clinicians identify early social communication difficulties and clinical ASD in preschool children with visual impairment. This paper reports on the pilot development of the VISS and initial predictive validity testing for an ASD diagnosis in a clinical sample.

## METHOD

The study was approved by the Institute of Child Health and Great Ormond Street Hospital Research Ethics Committee (data pseudo-anonymized, collected as part of clinical care, and no informed consent required). Only children with congenital disorders of the peripheral visual system (disorders of the globe, retina, and anterior optic nerve), where there is no known damage of the central nervous system, were included in order to minimize confounding factors.[6] Visual disorder diagnosis had previously been confirmed by paediatric ophthalmologists and/or neurologists. Participants were grouped according to their current visual level/degree of visual impairment at the time of assessment: profoundly visually impaired – able to detect a spinning 12.5cm light-reflecting ball at 30cm or less; severely visually impaired –

able to detect a 12.5cm spinning woollen ball at 30cm or better.[5]

Twenty-three children were referred for functional vision and developmental assessment as part of their visual impairment management; of these children, five were referred specifically because of concerns about development and behaviour. Children were assessed by a multidisciplinary team expert in the assessment and management of neurodisability and visual impairment, which included neurodisability paediatricians, clinical psychologists, and speech and language therapists. Clinical decisions regarding a diagnosis of ASD were made by the multidisciplinary team who were experienced in diagnosing ASD in the context of visual impairment; a diagnosis was later reached after collecting information from different settings and monitoring the child's development over time.

### Instrument development and administration

The VISS was designed to identify the range of social and communicative behaviours seen in visually impaired children during the developmental preschool age period;[1,3,5,6,8,11] items were also influenced by International Classification of Diseases (ICD-10) ASD diagnostic criteria.[10,12] None of the VISS items were vision dependent. All items required observational evidence of behaviours in a clinical setting. Twenty-nine items were scored: 0 (absent), 1 (partially present), 2 (definitely present); giving a possible range of 0 to 58. A high total score is indicative of more advanced social and communicative development.

The VISS was administered jointly by two experienced clinicians (ND, AS) on the basis of observed behaviours during semi-standardized clinical assessments of play behaviours and social interactions which included administration of the Reynell-Zinkin scales.[11] The CARS was administered simultaneously by another experienced clinical psychologist from the team. Items 7 and 12 were removed from the CARS as they rely on visual behaviours. The VISS and CARS were anonymized and stored separately from the medical records and not referred to during subsequent clinical visits.

### Item analysis and internal reliability

For analysis of internal item consistency and reliability, Rasch analyses of the VISS produced by the WINSTEPS (winsteps.com, Beaverton, OR, USA) software (partial credit model) was used for this study.[13] In order to determine whether or not the items of the instrument 'fit' the measurement model, Rach analysis uses 'fit' statistics. 'While there are various types of fit statistics and various criteria to determine if items fit the measurement model, for purposes of this study, infit mean square 0.4 to −1.5 and standardized z-score less than 2.0 were used.[13] The WINSTEPS software also produces person and item measures (and their associated error). Since person and item measures are placed on the same scale, person ability can be directly compared to item difficulty. The unit of measurement for these values, logits, represents an interval-based measure. Logits range from negative values to positive values, but can easily be converted to other scales (e.g. 0–100)'.[14] The sample size range needed to have 95% confidence that no item calibration is more than 1 logit away from its stable value using Rasch analysis is 16 to 36.[15]

**What this paper adds**

- The Visual Impairment and Social Communication Schedule is potentially a useful observational tool that helps clinicians make an autism spectrum disorder diagnosis in young children with visual impairment.

### Validity testing

The validity of the VISS total score was investigated: by comparison (1) to the concurrent CARS total score in the total sample; and (2) with the later ICD-10 clinical ASD diagnosis in the subgroup of children who were followed up subsequently. The five children who were referred because of concerns about their development and behaviour were excluded from the predictive validity analysis, leaving a sample of $n$=18. An independent investigator analysed the VISS and CARS questionnaires separately and then compared the VISS total score with the child's clinical ASD diagnosis (yes/no) which was obtained from their medical record. Descriptive, non-parametric, internal reliability and correlation statistics were analysed using SPSS v17.0 (IBM, New York, NY, USA) statistical software.

## RESULTS

Data were available for 23 children (12 male, 11 female). At the time of VISS completion, children's ages ranged from 1 year 9 months to 6 years 11 months (mean 4y 1mo [SD 1.6], median 4y 2mo). Nine children had severe visual impairment and 14 children had profound visual impairment. Thirteen children had a developmental quotient for sensorimotor skills on the Reynell-Zinkin scales of less than 70.

The summary fit statistics of the scale on the Rasch measurement model (for measured items and persons) showed acceptable fit statistics (infit mean square 0.4–1.5 and standardized z-score <2.0). The fit of the individual items to the Rasch measurement model is shown in Table I. Twenty-five of 29 items showed acceptable fit statistics (with the exception of items 5, 6, 12, 13). A further indication of the appropriateness of the VISS items was the point measure correlations which indicated that all items, except for the misfitting ones, correlate well with the overall scale. The higher 'item measures' (column 2 in Table I) depict a higher degree of item difficulty. Internal reliability of the VISS was acceptable with a Cronbach's alpha coefficient of 0.80.

Concurrent validity data were available from 23 children. VISS scores ranged from 4 to 58 (median 31, interquartile range 41). Correlation between the VISS and CARS total scores was highly significant (Spearman's $rho$=−0.89; $p$<0.01). A partial correlation whilst controlling for age and cognitive level did not lower the strength of the correlation between the VISS and CARS scores ($r$=−0.87; $p$<0.001).

Predictive validity for ASD diagnosis was carried out with a subgroup ($n$=18) of whom nine children (50%) received a later clinical diagnosis of ASD (5/9 with profound visual impairment and 3/9 with severe visual impairment). ROC analysis reveals that below a VISS score of 35 there is good agreement

**Table I:** Visual Impairment and Social Communication Schedule item psychometrics (note: item definitions necessary for administration not included)

| Items | Item measure | Error | Mean square infit | Standardized z-score | Point measure correlation |
|---|---|---|---|---|---|
| Social interaction | | | | | |
| 1. Makes social approach | 43.4 | 4.2 | 0.56 | −1.4 | 0.83 |
| 2. Makes social response | 41.7 | 4.2 | 0.4 | −2.1 | 0.85 |
| 3. Has a social smile | 57.5 | 4.2 | 1.25 | 0.8 | 0.74 |
| 4. Responds to voices | 36.5 | 4.2 | 0.50 | −1.7 | 0.78 |
| 5. Responds selectively to voices | 29.5 | 4.2 | **0.26** | **−3.2** | 0.18 |
| 6. Holds arms up to be lifted | 57.5 | 4.2 | **3.60** | **4.8** | 0.44 |
| 7. Enjoys social touch and being held | 31.3 | 4.2 | 0.79 | −0.6 | 0.69 |
| 8. Positive acceptance of social approach | 34.7 | 4.2 | 0.48 | −1.8 | 0.78 |
| 9. Directs attention of other | 57.5 | 4.2 | 0.84 | −0.4 | 0.84 |
| 10. Directs adult's attention to own activity | 61.1 | 4.3 | 0.88 | −0.3 | 0.83 |
| 11. Joins in activities of others | 63.0 | 4.3 | 0.92 | −0.1 | 0.82 |
| 12. Enjoys social play | 46.9 | 4.2 | **0.39** | **−2.2** | 0.4 |
| Communication and language | | | | | |
| 13. Responds to being called by name | 53.9 | 4.2 | **2.34** | **3.0** | 0.58 |
| 14. Uses or responds to gestures | 66.8 | 4.5 | 1.10 | 0.4 | 0.72 |
| 15. Communicates need for help by vocalisation or gesture | 57.5 | 4.2 | 1.00 | 0.1 | 0.80 |
| 16. Use of language for social chat | 55.7 | 4.2 | 0.83 | −0.4 | 0.82 |
| 17. Use of language for communication | 48.7 | 4.2 | 0.94 | 0.0 | 0.79 |
| 18. Expresses emotion | 57.5 | 4.2 | 0.92 | −0.1 | 0.81 |
| 19. Uses conventional words and meanings | 48.7 | 4.2 | 0.94 | 0.0 | 0.79 |
| 20. Spontaneous and meaningful use of referential language | 50.4 | 4.2 | 1.48 | 1.3 | 0.73 |
| 21. Appropriate wide range of topics of interest | 52.2 | 4.2 | 0.92 | −0.1 | 0.83 |
| Play | | | | | |
| 22. Engage in spontaneous play | 48.7 | 4.2 | 0.64 | −1.1 | 0.84 |
| 23. Engage in functional play | 50.4 | 4.2 | 0.81 | −0.5 | 0.82 |
| 24. Engage in symbolic play | 59.3 | 4.3 | 1.03 | 0.2 | 0.82 |
| 25. Engage in imaginative play | 46.9 | 4.2 | 0.8 | −0.5 | 0.81 |
| Routines, behaviours, and interests | | | | | |
| 26. Has appropriately wide repertoire of actions with objects | 50.4 | 4.2 | 0.83 | −0.4 | 0.82 |
| 27. Has range of interests in different objects | 48.7 | 4.2 | 0.71 | −0.8 | 0.83 |
| 28. Uses hands and body in functional manner | 45.2 | 4.2 | 0.77 | −0.6 | 0.80 |
| 29. Willing to be redirected to new activity or focus of attention | 48.7 | 4.2 | 1.30 | 0.9 | 0.70 |

Items in bold show misfit statistics (infit mean square <0.4 or >1.5 and standardized z-score >2.0). The higher item measures (second column) depict a higher degree of difficulty.

with the clinical ASD diagnosis (sensitivity 89%, specificity 100%, PPV=100%, NPV=90%).

## DISCUSSION

Preliminary development of the VISS is an important initial step toward a clinical tool to aid early ASD diagnosis for children with visual impairment, which can subsequently lead to appropriate early management and intervention. Clinicians may understandably be reluctant to make an early diagnosis of ASD in the absence of validated objective measures to support clinical indices of suspicion. A schedule that takes into account the overall social communicative skill profile in a group of visually impaired children aged 2 to 6 years has been shown to be of value for identifying social communication difficulties and ASD. The significant correlation relationship of VISS with CARS showed that the VISS has construct validity for identifying visually impaired children with social communication difficulties. Predictive validity analysis showed that children with visual impairment who have a below threshold rating on the VISS (score of 35) are at significant risk of developing ASD.

The Rasch mathematical model is increasingly being used when assessing clinical measurement and quality of life tools.[14,16] Analysis showed acceptable fit of the scale and of the items to the Rasch model, except for four of the items (responds selectively to voices [5]; holds arms up to be lifted [6]; enjoys social play [12]; responds to being called by name [13]). Items 6 and 13 were underfitting (too unpredictable according to the Rasch model) and items 5 and 12 were overfitting (too predictable). Progressive item difficulty demonstrated its basis as a developmental schedule for this age range. These results will inform future VISS redesign and item reduction for future studies of the scale.

Our findings are considered within the limitations of retrospective analysis and applied clinical research design. Sources of possible bias include the referral pattern to a specialist service that accepts UK-wide referrals. Although the study group may be representative of the clinical visually impaired population, the extent to which these findings can be applied in general clinical practice are limited at present. The vision groups varied in size with the profoundly visually impaired group being the largest, and most likely to show disordered social communication.[5,6] Interrater reliability was also not assessed as consensus rating was the preferred method. In the future, following item reduction, the VISS will require test–retest and interrater reliability testing with independent raters and fur-

ther validation in larger samples of children with visual impairment. Because of the subtlety of social communicative signs in young children with visual impairment,[1,4] reliability testing might reveal the need for a training model to be developed for the VISS (as in the Autism Diagnostic Observation Schedule). Despite the limitations, this preliminary study shows the VISS to be a promising developmental schedule, which in the future has the potential to support clinicians' assessment of early social communicative difficulties and ASD diagnosis in preschool-aged children with visual impairment.


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