

Can photoscreening effectively detect amblyogenic risk factors in children with neurodevelopmental disability?

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Purpose: To analyze whether photoscreening can effectively detect amblyogenic risk factors in children with neurodevelopmental disability. **Methods:** A prospective study of 52 children attending a special school for children with neurodevelopmental disability from December 2017 to May 2018. All were initially tested with a photoscreening device: Welch Allyn® Spot® Vision Screener: model VS100 (Spot®) and further evaluated at a later date by a pediatric ophthalmologist, with a complete ocular evaluation including squint assessment, cycloplegic retinoscopy, and dilated fundus examination. The key parameters studied were demographic features, type of neurodevelopmental disability, refraction, ocular alignment, media clarity, any other ocular morbidity, and time taken for examination. The presence of amblyogenic risk factors (ARF) was analyzed as per the 2013 guidelines of the American Association for Pediatric Ophthalmology and Strabismus. **Results:** The mean age was 10.5 years (range: 1–17.5 years). Males (73.1%) outnumbered females (26.9%). The most common neurodevelopmental disability was cerebral palsy. Simple myopic astigmatism was the most common type of refractive error. Presence of ARF in our study was 73.1%. The sensitivity and specificity of photoscreening in detecting ARF were 96.5% and 63.61%, respectively, with a positive predictive value of 80% and negative predictive value of 92.31%. The predictive ability of photoscreening was 79.9% as per the area under curve. The average time taken for photoscreening was less than 60 s. **Conclusion:** Photoscreening can detect ARF with high sensitivity and reasonable specificity and is a handy, useful, and time-saving tool in screening children with neurodevelopmental disability.

Key words: Amblyogenic risk factors, neurodevelopmental disability, photoscreening, spot®vision screener

Neurodevelopmental disorders are a group of disorders arising from impairment in the developing brain and/or the central nervous system. They originate during the developmental period, i.e. during the prenatal, ante-natal, postnatal, infancy, and early childhood periods.^[1] Children with neurodevelopmental disability/delay constitute a considerable proportion of patients seen at pediatric ophthalmology clinics. Strabismus and refractive errors are the most frequent anomalies, with amblyopia being a major risk factor associated with these conditions. The prevalence of refractive errors and strabismus has been described as higher in children with developmental delay.^[2] In a considerable proportion of these children, ocular disorders were previously unknown. Children with developmental delay are often unable to provide adequate responses to subjective tests of visual acuity and do not easily cooperate for tests of ocular alignment or stereopsis. It is also very difficult for parents to bring them to higher centers for vision screening due to the presence of other comorbid conditions, behavioral problems, and difficulties in transportation. Hence, there is a need for an alternative approach to screen them in their familiar environment with

support from their caregivers, teachers, and/or parents. Knowledge of the prevalence of refractive errors, strabismus, and amblyogenic risk factors (ARF) is essential if these children are to be given optimal support in their development and learning capacity.

Photoscreening devices^[3] or photorefractors function based on the analysis of a reflected image from the patient's retina. An infrared camera contained in these devices captures the images of red reflex and images of the corneal light reflex from a child's pupil. The test is performed binocularly and is based on the reflexes; an examiner or a computer program can analyze to determine if there is strabismus and/or significant refractive error. Photoscreeners can also detect other anatomical abnormalities, including cataract, coloboma, or ptosis. The test is fast and usually takes less than a minute and can be performed on both verbal and preverbal children. Photoscreening devices can be used even in undilated pupil and is less time consuming in nature. Since it is an objective assessment device, person-to-person variability in ocular morbidity assessment is very less. Hence, even a nonmedical

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person can use the device for screening purpose and do a proper referral. These devices can increase the detection rates of visual morbidities in children and thus help in timely management to prevent amblyopia.

The aim of this study was to analyze whether photoscreening can effectively detect amblyogenic risk factors in children with neurodevelopmental disability.

Methods

The study was conducted in a prospective manner as a pilot study at a special school for children with neurodevelopmental disability from December 2017 to May 2018. Informed consent was obtained from the parents and guardians of all children prior to the study. All students of the special school were initially tested with a photoscreening device: Welch Allyn® Spot® Vision Screener [Fig. 1]: model VS100 (Spot®)^[4] in the presence of a parent or caregiver in a dark room, by a trained pediatric optometrist. Spot® gives a report at the end of the test about the ocular alignment, pupil diameter, estimated binocular refraction, anisometropia, and a referral recommendation— “all measurements in range- Pass” or “complete eye examination recommended-Fail.” Any child in whom Spot® was unable to give a report was categorized as “ Could not detect.” Apart from demographic data like age and sex, a detailed antenatal, natal history about type of delivery, birth weight, preterm, post term or full-term birth, and postnatal history regarding any birth asphyxia, neonatal seizures, and infections were taken. Visual complaints, type of neurodevelopmental disability, systemic diseases, current medical, surgical, and ocular treatment of each child were noted. Only children less than 18 years were included in this study. The study was conducted in accordance with the Declaration of Helsinki and approved by the institutional review board and ethics committee.

All enrolled participants were further evaluated at a later date by a pediatric ophthalmologist at the special school itself in the presence of a parent or caregiver. A complete ocular evaluation, including vision and squint assessment, ocular motility, media clarity, cycloplegic retinoscopy with Homatropine 2% and Tropicamide 1%, handheld slit-lamp examination, dilated fundus examination, and any other ocular morbidity evaluation was done. Intraocular pressure recording with tonometer (Reichert-Tono-Pen XL) was done in suspicious cases (e.g. microcornea, megalocornea, previous cataract surgery/glaucoma surgery, etc.). Those who could not complete the entire examination schedule or follow-up were excluded. Time taken for Spot® and clinical examination were compared. The presence of ARF was analyzed as per the 2013 guidelines [Table 1] of the American Association for Pediatric Ophthalmology and Strabismus^[3] separately in the two groups: Spot® and clinical evaluation, respectively.

Spherical equivalent (SE) of refractive error of each eye calculated by Spot® was compared with SE of refractive error of each eye obtained by cycloplegic refraction. Anisometropia of >1.5D were taken as significant and those detected by Spot® were compared with anisometropia calculated from cycloplegic refraction. Presence or absence of squint in Spot® was confirmed with clinical evaluation. The Spot® results of Pass or Fail were compared with clinical evaluation results of Pass or Fail, respectively. Sensitivity, specificity, positive

predictive value (PPV), and negative predictive value (NPV) of the Spot® were determined. The study was conducted in accordance with the Declaration of Helsinki and was approved by the appropriate Institutional Review Board and Ethics committee.

Results

There were 52 children in our study and males (73.1%) outnumbered females (26.9%). The mean age was 10.5 years (range: 1–17.5 years). The different types of neurodevelopmental disability [Fig. 2] were cerebral palsy (9), autism (4), attention deficit hyperactive disorder (2), mental retardation with delayed milestones (10), cortical visual impairment (1), seizure disorder (1), and syndromes (9) like down syndrome, Smith–Mageson syndrome, Beckwith–Wiedmann syndrome with cerebral palsy being the most common (48.07%). There were many children with more than one disability. The presence of ARF in our study was 73.1 [Fig. 3]. Spot® was able to screen 48 children (92.3%) out of the total 52. In the 4 children (7.69%) who were grouped as “could not detect,” 3 had ARF in the form of refractive error and squint, which was detected by clinical evaluation. One child in “could not detect” group did not have ARF but had poor vision and wandering eye movements, secondary to optic atrophy.

Statistical analysis was done with SPSS Version 16 software. The sensitivity and specificity of photoscreening in detecting ARF were 96.5% and 63.61%, respectively, with a PPV of 80% and NPV of 92.31% [Tables 2 and 3]. The predictive ability of

Table 1: Amblyopia risk factors targeted with automated preschool vision screening

Age, months	Refractive risk factor targets ^a			
	Astigmatism	Hyperopia	Anisometropia	Myopia
12-30	>2.0 D	>4.5 D	>2.5 D	>-3.5 D
31-48	>2.0 D	>4.0 D	>2.0 D	>-3.0 D
>48	>1.5 D	>3.5 D	>1.5 D	>-1.5 D
All ages	Nonrefractive amblyopia risk factor targets ^b			
	Manifest strabismus >8 PD in primary position			
	Media opacity >1 mm			

D, dioptres; PD, prism dioptres. ^aAdditional reporting of sensitivity to detect greater-magnitude refractive errors is encouraged. ^bFor all ages.

Table 2: Amblyogenic risk factor detection

Spot® Judgement	Clinical Evaluation		Total
	Clinical evaluation: Fail	Clinical evaluation: Pass	
Spot® Judgment: Fail	28	7	35
Spot® Judgment: Pass	1	12	13
Could not detect	3	1	4
Total	32	20	52

SPOT® vs clinical evaluation: Out of the 32 subjects who failed in the clinical evaluation, 28 were failed, 1 passed, and in 3, no report could be given by the Spot®. Out of the 20 who passed the clinical examination, 7 were failed, 12 passed, and in 1, no report could be given by the Spot®

Table 3: Sensitivity and specificity of SPOT® in amblyogenic risk factor detection*

Parameter	Area Under Curve	Sensitivity	Specificity	Positive Predictive Value	Negative Predictive Value
Spot® Vision	0.799	96.50	63.61	80.00	92.31

*4 children in “Could not detect” group were not included



Figure 1: Welch Allyn spot vision screener: model VS100 (Spot)

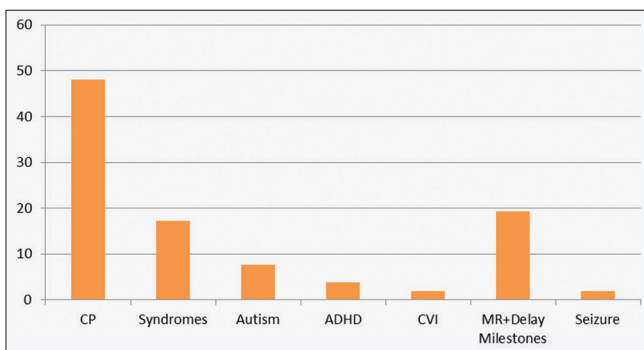


Figure 2: Type of neurodevelopmental disability detected

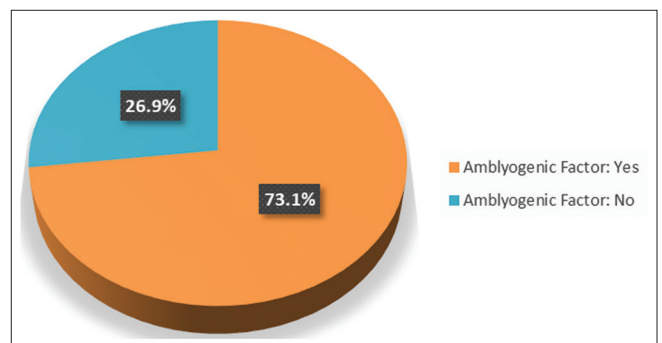


Figure 3: Distribution of amblyogenic risk factors

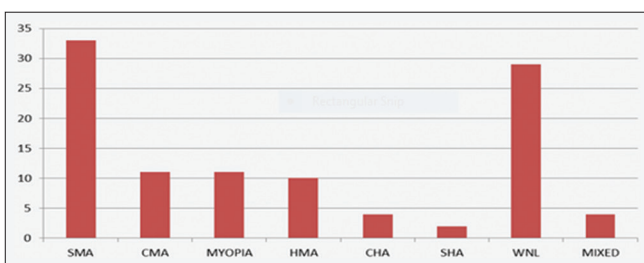


Figure 4: Types of refractive error detected

photoscreening was 79.9% as per the area under curve. The sensitivity and specificity of photoscreening in detecting ARF changed to 96.8% and 60%, respectively, when the “could not detect” group of 4 children were automatically included in the referrals. Simple myopic astigmatism was the most common type of refractive error noted (31.73%) among the 104 eyes of 52 patients [Fig. 4]. Twenty-five out of the 35 children (71.45%) who failed Spot® screening had refractive errors, 7 (20%) had gaze anomalies, and 3 (8.57%) had both. The average SE of right eye on Spot® was -0.44 as compared to -0.21 on clinical evaluation and the average SE of refractive error of left eye on

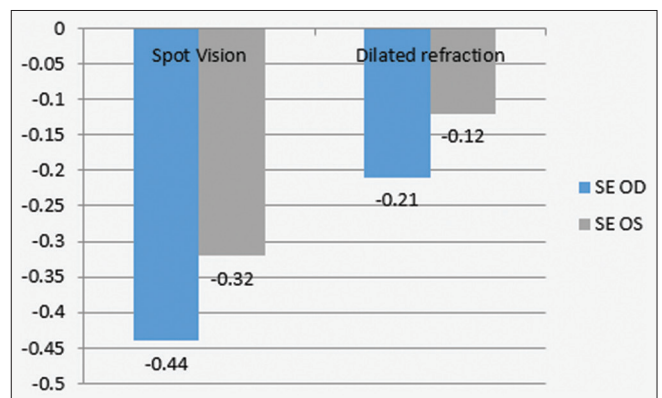


Figure 5: Spherical equivalent of Right eye and Left eye across two groups

Spot® was -0.32 as compared to -0.12 on clinical evaluation, respectively [Fig. 5]. The average SE of right eye and left eye [Fig. 6] on Spot® (-0.38) was compared with average SE of right eye and left eye on clinical evaluation (-0.16) and *P* value calculated using Mann-Whitney U test was not found to be significant (*P* = -0.213). Spot® was able to detect squint in 15

children and could not detect in 4 children, as compared to 24 children with squint in the clinical evaluation group [Table 4]. Sensitivity and specificity of Spot® in Squint detection [Table 5] were noted to be 68.18% and 100%, respectively, excluding the “could not detect” group. There were 2 children with visually significant media opacities out of which Spot® was able to detect one and the other child belonged to the “could not detect” group. The average time taken for Spot® was 47.5 s as compared to 1.75 h in clinical evaluation. After identification of the appropriate ARF and ocular morbidity, 37 children were given glasses, 8 were given occlusion therapy, 3 underwent surgery (2

squint, 1 cataract), 1 patient was started on antiglaucoma medication, and all were advised regular follow-up.

Discussion

Automated screeners have been validated in the pediatric population and have been recommended for screening for amblyopia risk factors (ARF) in children <5 years of age and those unable to cooperate with optotype screening.^[3] The Spot® Vision Screener, Welch Allyn®, Skaneateles Falls, NY, has been reported to have good sensitivity in pediatric patients.^[6-10]

Spot® was able to screen 92.3% of the total children with neurodevelopmental disability in our study with a sensitivity and specificity of 96.5% and 63.61%, respectively. The PPV of 80% implies that most of the patients who were failed by Spot® had ARFs and NPV of 92.31% indicates that most of those who were passed by Spot® were not having ARFs. Spot® was also able to complete the screening in considerably less time (<60 s). In a study of 128 European Caucasian children with autistic spectrum disorder, Anketell *et al.*^[10] found increased prevalence and magnitude of astigmatism. In our study too, we found simple myopic astigmatism to be the most common type of refractive error (31.73%).

Marzolf *et al.*^[11] found a 38% prevalence of ARF in their study of 100 children with developmental disability (average age, 5.7 years; range, 2.2–9.2 years) using Spot® Vision Screener, which was higher than the 15%–20% prevalence of ARF in the general pediatric population.^[12] The sensitivity of the Spot® in detecting ARF was 84% and the specificity was 62% with a PPV of 58% and NPV of 86%, respectively. We got a greater prevalence (73.1%) of ARF in our study. However, sensitivity and specificity of photoscreening in detecting ARF in our study were 96.5% and 63.61%, respectively, which were comparable; however, the PPV of 80% and NPV of 92.31% were higher than the study by Marzolf *et al.* [Table 6]. The possible reason for large differences could be Marzolf *et al.* screened children with developmental disability presenting for ophthalmologic examination in an outpatient clinic, whereas we did our study at a special school for children with neurodevelopmental disability. There could have been some bias in screening as we could have screened the ones with severe comorbidities and vision impairment who normally would not have presented to an ophthalmic OPD due to difficulties in transportation.



Figure 6: Average spherical equivalent across two groups

Table 4: Squint detection

Spot®: Squint Detection	Clinical Evaluation: Squint Detection		Total
	Yes	No	
Yes	15	0	15
No	7	26	33
Could not detect	2	2	4
Total	24	28	52

SPOT® vision vs clinical evaluation: Out of the 24 subjects who were confirmed to have squint (Yes) by clinical evaluation, 15 were detected to have squint, 7 were not detected to have squint and in 2, no report could be given by the Spot®. Similarly, out of the 28 subjects confirmed to have no squint (No) in the clinical evaluation, 26 were cleared by Spot® and in 2, no report could be given by the Spot®

Table 5: Sensitivity and specificity of SPOT® in squint detection**

Parameter	Area Under Curve	Sensitivity	Specificity	Positive Predictive Value	Negative Predictive Value
Squint Detection	0.841	68.18	100	100	78.79

**“Could not detect” was not included

Table 6: Comparison with other published studies

	No of patients	Average age	Amblyogenic Risk Factor	Sensitivity	Specificity	Positive Predictive Value	Negative Predictive Value
Marzolf ^[11]	100	5.7	38%	84%	62%	58%	86%
Our study	52	10.5	73.1%	96.5%	63.61%	80%	92.31%

Advantages of our study were the use of a novel screening technique in a high-risk population in their familiar environment, with support from their caregivers, understanding their comorbid conditions, identifying ARF, performing relevant investigations, administering appropriate interventional therapy in the form of glasses, medications, surgery, occlusion, and regular follow-up. We believe that recommendations given after our analysis of child's visual functions would help teachers to adapt materials, environment, and methods, which, in our opinion, would further improve students' skills and optimally support their development and learning capacity.

Limitations of our study were the smaller study population and difficulties in fixation and getting reliable readings, intervention, and regular follow-up.

Conclusion

Photoscreening can detect ARF with high sensitivity and reasonable specificity and is a handy, useful, and time-saving tool in screening children with neurodevelopmental disability.

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Conflicts of interest

There are no conflicts of interest.

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