Original Article

Evaluation of the PlusoptiX photoscreener in the examination of children with intellectual disabilities

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Abstract:

PURPOSE: This study aimed to determine whether the plusoptiX vision screener (PVS) can be used to detect amblyogenic risk factors (ARFs) as defined by the American Association for Paediatric Ophthalmology and Strabismus Vision Screening Committee guidelines (2013) for automated vision screening devices.

METHODS: In this cross-sectional study, children attending a special needs school underwent screening with the PVS and complete ophthalmologic examinations. Ophthalmologic examinations were used as the gold standard to compute the prevalence, sensitivity, specificity, positive predictive value (PPV), negative predictive value (NPV), and testability.

RESULTS: Forty-four children with special needs (mean age, 8.5 years; range, 4–18 years) were included. The PVS recommended referral of 31 cases (referral rate 70%). Thirty-nine of the 44 children (89%) met the referral-positive threshold for strabismus, reduced vision and/or amblyogenic factors on examination. The plusoptiX had a sensitivity of 40% (confidence interval [CI] 7%–83%), specificity of 78% (CI 55%–85%), PPV of 15% (CI 3%–46%), and NPV of 90.3% (CI 73%–97%). The PVS underestimated refractive errors by 0.67 to 0.71 D in the right ($P < 0.001$) and left eyes ($P = 0.002$). Testability was relatively low, with the PVS at 75% compared to the gold standard examination at 100%.

CONCLUSION: We found that although the plusoptiX photoscreener might be a useful tool in pediatric vision screening, it might not perform as well in children with intellectual disabilities. Utilization of the PVS as a single screening device may fail to identify a considerable proportion of young children with ARFs or amblyopia.

Keywords:

Paediatric vision screening, photoscreening, special needs, strabismus

Introduction

Although health professionals are aware of the health inequalities experienced by individuals with intellectual disabilities (ID), the health status of these individuals remains poor.[1] Reliable vision screening in children with ID is challenging but essential because ophthalmic disorders are common in this group,[2] who have a substantially higher prevalence of ocular disorders than normal children.[3-6] A systematic review that investigated the prevalence of chronic health conditions in these children revealed that the prevalence rates of refractive errors, strabismus, visual field defects, or visual impairment in this population ranged from 2.2% to 26.8%.[7] One of the contributing factors to the higher rate among children with ID is that brain injury underlies several disabilities, such as learning difficulties and sensory impairments.[8]

It is strongly believed that a minimum of five ophthalmological examinations should be offered to children with ID as per the guidelines of the International Association for the Scientific Study of Intellectual Disabilities.[9] A range of tests and approaches can be used in assessing vision in these children; however, it is essential to examine assessment tools that are noninvasive and correctly identify the visual disorder. Photoscreeners may be beneficial in screening for ophthalmic disorders in children with ID since newer versions do not highly depend on

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the cooperation of the child.[10] Due to the contradicting few reports on the validity of the use of photoscreeners in children with ID,[2,10,11] the objective of this is to further investigate the efficacy of the plusoptiX photoscreener in detecting amblyopia risk factors in children with ID.

Methods

This study was conducted in compliance with the Declaration of Helsinki. Approval to conduct this study was obtained from the local Institutional Review Board. After obtaining the approval, we invited 62 children attending a special needs school to participate in the study. We sought and obtained written, informed consent from the participants' parents. Students who attended classes during the 4-day screening period were offered a free vision screening test with the plusoptiX and full ophthalmologic evaluation in April 2018. The plusoptiX S12 photoscreener (PlusoptiX GmbH Nuremberg, Germany) was used as instructed by the manufacturer.

Two ophthalmologists and two optometrists, who had been trained in using the plusoptiX, screened the children. In cases where the photoscreener "referred" the children due to its inability to measure their amblyopia risk factors, three successive attempts were made. If measurement failed after at least three attempts, the result was documented as a "refer." In cases where the child was uncooperative, the result was considered a definitive "refer." We referred children for screening following the manufacturer's recommendations, as shown in Table 1.

Two ophthalmologists who were blinded to the screening results conducted the full ophthalmic and orthoptic assessments. These included visual acuity, slit-lamp examination, ocular alignment and motility evaluation, manual cycloplegic refraction, and fundus examination. The gold standard test used the American Association for Paediatric Ophthalmology and Strabismus Vision Screening Committee guidelines for the definition of amblyopia risk factors.[12]

We compared the testability (proportion of children who could be tested) of both methods and assessed the performance of the plusoptiX vision screener (PVS) in identifying children who screened positive on the gold standard examination. The spherical equivalent of the measurements obtained by the plusoptiX photoscreener and manual cycloplegic retinoscopy (MCR) were also computed.

The data were analyzed utilizing IBM SPSS, version 23 (IBM Corp., Armonk, NY, USA). Qualitative variables are expressed as absolute numbers and proportions or means and standard deviations. The diagnostic capacity of the plusoptiX photoscreener is presented in terms of sensitivity, specificity, positive predictive value (PPV), negative predictive value (NPV), and disease prevalence along with 95% confidence intervals (CIs). For continuous variables, a paired-sample *t*-test was used, while Wilcoxon signed-ranked test was utilized in the case of nonnormal distribution. A P value < 0.05 was the criterion to reject the null hypothesis.

Results

Demographic data

Of 62 invited children, 47 took part in this study (76%). Three students were excluded because they were adults. The remaining 44 subjects, of whom 25 were boys (57%), ranged in age from 4 to 18 years. The following diagnoses were identified in children with special educational needs: autistic spectrum disorder (n = 25), Down syndrome (n = 6), attention deficit/hyperactivity disorder (n = 5), cerebral palsy/perinatal asphyxia (n = 5), gene mutation (n = 2), and cerebral malformation (n = 1).

Photoscreening

The plusoptiX photoscreener referred 31 of 44 children (70%). Therefore, the diagnostic capacity of the plusoptiX photoscreener in predicting the gold standard assessment is presented in Table 2. Accuracy was calculated at approximately 70%, which was the overall rate that a patient would be correctly classified by the new device. Regarding refractive errors, the PVS identified emmetropia or low hypermetropia in most of the children. Testability was relatively low, with the PVS at 75% compared to the gold standard examination at 100%.

Table 1: Referral criteria for both plusoptiX vision screener and ophthalmologic assessment according to age group

Vision screener	Ophthalmologic assessment
Hyperopia	Hyperopia
4-6 years >2.50 D	>3.5 D
>6 years >2.0 D	
Myopia	Myopia
4-6 years <2.25 D	> 3.00 D
>6 years <1.50 D	
Astigmatism	Astigmatism
4-6 years <+2.25 D	>1.5 D within 10 of 90 and 180 degrees or
>6 years <+1.50 D	>1.0 D in an oblique axis
Anisometropia	Anisometropia
4-6 years <1.0 SE	>1.5 D (sphere or cylinder)
>6 years <1.25 SE	
Asymmetry >10°	Cover test
	Any manifest deviation (constant/intermittent)
Anisocoria >1.5 mm	Anisocoria >1 mm
	Visual acuity of >20/40 in either eye

SE=Spherical equivalent

Table 2: Screening results with plusoptiX photoscreener

Result	Percentage	95% CI	
		Lower limit	Upper Limit
Sensitivity	40	7	83
Specificity	72	55	84
PPV	15	3	46
NPV	90	73	97
Accuracy	68	52	81

CI=Confidence interval; NPV: Negative predictive value; PPV: Positive predictive value

Ophthalmologic evaluation

Ophthalmologic examination revealed that 39 of the 44 subjects (89%) had amblyopia risk factors. Ocular findings in decreasing prevalence were as follows: subnormal visual acuity (n = 29, 66%), refractive errors (n = 19, 43%), fundus anomalies (n = 9, 20%), significant strabismus (n = 9, 20%), abnormal head posture (n = 8, 18%), nystagmus (n = 3, 7%), extraocular motility abnormality (n = 2, 5%), and anterior segment abnormality (n = 1, 2%). Exotropia was the most prevalent misalignment in 5/44 subjects (11%), followed by esotropia in three (7%) and vertical in one (2%) Table 3 summarizes the clinical spectrum of ophthalmologic diagnoses.

Photoscreening versus ophthalmologic evaluation

Of the 31 children referred for amblyopia or manifest strabismus (70%), three were false positives and 9/31 (29%) were due to "failure of screening." We confirmed 29 children (66%) had decreased vision in one or both eyes as per the referral criteria, whereas the PVS identified 18 cases with reduced vision.

The PVS correctly identified 10 out of 19 children (53%) who had visually significant refractive errors; two out of 44 patients were referred due to incomplete testing and six were referred due to inability to perform the screening. The PVS delivered a screening outcome of "pass" in a further 13 cases (false negatives 1/44). The false-negative was related to astigmatism of > + 1.5 D. The PVS accurately identified both hyperopia cases and one out of six myopia cases. In addition, one out of two hyperopia cases and four out of six myopia cases had a screening outcome of "no reading possible–refer." The PVS measurements were 0.67 and 0.71 D lower than the MCR measurements in the right ($P < 0.001$) and left eyes ($P = 0.002$), respectively. The device did not detect the only case of anisometropia in our sample. At the screening, the PVS was unsuccessful in obtaining a refractive reading in 16 of the 44 children. On orthoptic examination, only four of those cases had large-angle strabismus.

Discussion

Our analysis showed that the PVS was not effective in detecting amblyopic risk factors in our cohort; it had a sensitivity of 40% and PPV of 15%; however, it was effective in detecting cases without amblyopic risk factors with 71% specificity and 90% NPV. Moreover, the PVS missed a substantial number of cases with visually significant refractive errors (10 of 19 cases); hence, it is not reliable as a standalone vision screening tool. When compared with MCR measurements, the PVS measurements underestimated refractive errors. Further analyses suggested that the PVS had a relatively low testability compared with the gold standard examination.

We found that the prevalence of amblyogenic factors was 89%, which is approximately three-fold higher than the 31.8% reported in another cohort of children with ID.[11] However, similar to our findings, the investigators showed that children with ID comprised an intermediate-to high-risk group compared with healthy children enrolled in studies using autorefractors, where the prevalence of ocular disorders ranged from 1.9% to 2.6%.[13,14] In the current study, the PVS also underestimated the proportion of children with amblyogenic risk factors at 64%. In a previous study,[11] the plusoptiX S04 accurately identified amblyopia risk factors in 94% of children with ID. However, the sensitivity reported in our study may not be comparable with that reported by Ugurbas *et al*.[11] due to differences in our study populations. While we recruited children attending a special needs school, Ugurbas *et al*.[11] included children with ID and special needs from a pediatric ophthalmology referral practice. In addition, the authors explained that the high sensitivity in their report may be due to a high rate of large-angle strabismus, which is visible to a nonspecialist observer, prompting an automatic referral from the plusoptiX. Studies of the plusoptiX S04 conducted in a referral practice setting also reported high sensitivity rates (83%–92%).[15-17] In our study, approximately half of the failures (16/31) to photoscreening were due to incomplete/lack of screening, and 11 of 13 cases that passed the screening test had amblyopia risk factors related to subnormal visual acuity per AAPOS age-appropriate standards. Of those missed, only one patient was found to have significant strabismus and another with significant refractive error. This subnormal VA might be multifactorial and attributed to different ophthalmologic problems related to primary visual disorders, optic atrophy, or cerebral visual impairment in ID patients. This underscores the need for trained personnel to carefully assess and refer children with ID for low vision aids. Indeed, concerns have

Table 3: Summary of clinical spectrum of ophthalmologic diagnoses

Result	Vision screener	Ophthalmologic assessment	*P*
SE RE, mean±SD (range)	0.64±1.2 (−1.75-+3.875)	1.31±1.3 (−14.25-+4.25)	<0.001[a]
SE LE, mean±SD (range)	0.54±1.2 (−2.25-+3.50)	1.25±1.2 (−13.00-+5.625)	0.002[a]
IPD mm, mean±SD	53.96±4.1	54.50±4.1	0.434
Strabismus angle PD, mean±SD	7.45±16.8	6.14±12.2	0.043[a]
Hyperopia, *n* (%)	2 (7)	2 (7)	1.000
Myopia, *n* (%)	6 (20)	1 (3)	0.025[b]
Astigmatism, *n* (%)	14 (47)	9 (30)	0.059
Anisometropia, *n* (%)	1 (4)	0 (0)	0.317
Anisocoria, *n* (%)	0 (0)	0 (0)	1.000

[a]Significant using a paired sample *t*-test @ <0.05 level; bSignificant using the Wilcoxon signed-rank test @ <0.05 level. IPD=Interpupillary distance; LE=Left eye; PD=Prism diopters; RE=Right eye; SE=Spherical equivalent; SD=Standard deviation

been raised that a substantial proportion of individuals with multiple disabilities have visual impairments.[8,18] In one report, researchers used preferential looking tests to assess visual acuity and found that an unexpected 92% of clients with severe and profound multiple disabilities had visual impairments.[18] In addition, the severity of the visual impairment was related to the severity of the ID.

Few reports have investigated the utility of photoscreeners in children with ID.[10,19] In previous reports investigating the diagnostic performance of PVS in detecting amblyopia risk factors, investigators reported sensitivity rates ranging from 88% to 100%,[20-24] showing that the sensitivity of PVS in our study is considerably low. On the contrary, the specificity rate of the PVS in our study is close to the 82%–88% reported in previous studies.[11,20,21] However, specificity rates as low as 37%–50% have been reported with the plusoptiX photoscreener.[11,23,24] In one report, the PVS identified approximately 6% of the children with potential vision problems, indicating moderate sensitivity at 44% but high specificity at 100%.[25] Other investigators[26] reported that the sensitivity of both the medical technology and innovations (MTI) and Visiscreen (VR) photoscreeners were significantly higher than the PVS at 93%, but the specificity of these autorefractive devices was significantly lower than that of the PVS (35% and 55% for the MTI and VR, respectively) in children with Down syndrome. Although we found a lower PPV for the PVS compared with the MTI (66%) and VR (69%), the NPV of the PVS in our study is higher compared with MTI (78%) and VR (81%).[26]

The PVS measurements in our cohort were significantly lower than the MCR measurements in both the left and right eyes. Evidence shows that autorefractors are likely to overestimate myopia and underestimate hyperopic refractive errors. Thus, refined screening values are warranted to optimize performance.[20,27] A comparison between the PVS and MCR results in our study highlighted a major shortcoming of the PVS–the device was unsuccessful in identifying a large proportion of cases with significant refractive errors. We found that the PVS only accurately identified one out of six myopia cases. However, the small proportion of cases with anisometropic amblyopia and visually significant hyperopia in our cohort does not permit us to make relevant conclusions. Furthermore, to give appropriately sensitive results, photoscreeners may require further adjustment of pass/refer criteria in children with disorders such as neurodevelopmental delay, hypermetropia, strabismus, cataract, and hypo-accommodation, which are relatively more common in those with ID.[11]

An accuracy rate of approximately 70% for the PVS is acceptable or borderline; however, the accuracy of the device should improve with an increase in sample size. We believe the PVS could add to the range of orthoptic tests for pediatric vision testing if its accuracy in detecting visually significant refractive errors was improved. In our cohort, the PVS had relatively low testability compared with the gold standard examination. One hundred percent testability was achieved by the gold standard testing, although most of the children had behavioral problems and nine failed the screening test due to hyperactivity. Compared with other reports,[11,25] testability was much higher in our study.

One of the main shortcomings of monocular visual acuity testing used traditionally by general practitioners is that this test can occasionally fail to detect vision disorders in children with ID due to poor cooperation.[11] Most of the children in our cohort could not communicate during the monocular testing of their visual acuity. In another study, only 64% were able to be tested under the monocular reading of optotypes in a pediatric ophthalmology setting at a teaching hospital.[11] In practice, testability with traditional tools could be much lower, resulting in over-referrals for full ophthalmological examinations.[11] Many cases of amblyopia are undetected due to ineffective screening.[13] Thus, parents of children with ID should be educated on the importance of having their children screened for visual disorders and having at least five ophthalmologic evaluations during childhood.[9]

This study has several limitations. One major limitation is the relatively low sample size of this study. Unfortunately, nine of the parents/guardians failed to respond and six rejected participation for various reasons. An increase of the sample size would have allowed for narrower CIs to more appropriate ranges. This limitation renders our findings tentative and in need of further investigation on a larger scale. Another limitation is that the PVS does not simply detect strabismus. Therefore, a proportion of false negatives can be reported in populations where the incidence of strabismus is up to 21%.[28] Nine of 44 cases in this study (20%) had strabismus. Screening could not be performed in eight of the nine subjects with the plusoptiX photoscreener. The plusoptiX will trigger a referral automatically in the case of moderate or large-angle manifest strabismus since it most likely will be unable to obtain a reading. A limitation due to the study design is the number of observers included. It is possible that interobserver variability was introduced because two orthoptists and two ophthalmologists performed screening. To circumvent this, we set out clear instructions for each test at the beginning of the study. On the other hand, this is one of the few reports which assesses the performance of photoscreeners in children with ID.[11,21,23] Due to the lack of vision screening programs of children in Saudi Arabia and those with ID specifically, exploring the performance of such noninvasive, easy-to-use screening tools could prove to be beneficial due to its simplicity and cost-effectiveness.

Conclusion

Based on our results, the plusoptiX is less sensitive, although more specific at detecting amblyopic risk factors in children with ID. The plusoptiX photoscreener did not appear to be beneficial as an alternative to a full examination as it did not decrease the need for a full ophthalmological examination

in this category of patients. Failure of the PVS to identify patients with significant refractive errors renders the test an inaccurate screening tool by itself. While plusoptiX is a useful tool in pediatric vision screening, its performance might be decreased in children with ID. Nevertheless, we recommend further studies with larger sample size.

Patient consent

Consent was obtained from the parents.

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Conflicts of interest

There are no conflicts of interest.

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