

Subnormal visual perception in school-aged ex-preterm patients in a paediatric eye clinic

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Abstract

Purpose The aim of this study was to assess visual perception at school age of children born preterm with known lesions to the posterior visual pathways or with ophthalmologic signs that might indicate such lesions.

Methods The study group consisted of 91 patients born before the 37th gestational week. Visual perception was assessed using the TVPS-R (Test of Visual Perceptual Skills – Revised) and a structured interview. In addition, ophthalmologic and orthoptic examinations were performed.

Results On the test of visual perception, 67% of the patients had results below the third percentile of the American reference group. This is to be compared with 10% of Swedish full-term controls. Scores below the third percentile were observed in 87% of the patients with known brain lesions, 48% of those with strabismus without known brain lesion, and 86% of those with reduced visual acuity in the absence of strabismus and known brain lesion.

Conclusions Reduced visual perception is common among children born preterm who have strabismus and/or reduced visual acuity, as well as in those with known brain lesions. This study emphasises the need to find tools to identify and assess those patients who have visual perceptual problems that may restrict their ability to meet the demands of daily life.

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Introduction

Preterm birth is associated with increased morbidity of the eyes and visual pathways. In the neonatal period, retinopathy of prematurity (ROP) may cause severe visual impairment due to retinal detachment. Children born preterm have been found to have increased frequency of strabismus, amblyopia, refractive errors, optic atrophy and reduced contrast sensitivity, colour vision, and visual acuity (VA).^{1–6} In addition, a reduction of the neuroretinal rim area of the optic disc and increased tortuosity of the retinal vessels, especially arterioles has been observed.⁷ The posterior visual pathways may be injured by periventricular leukomalacia (PVL) and/or hydrocephalus secondary to intraventricular haemorrhage (IVH). PVL develops between 24 and 34 gestational weeks and may affect the corticospinal tracts causing cerebral palsy (CP) and/or the optic radiation causing cerebral visual impairment (CVI). Children with PVL have been reported to have ocular sequelae in the form of large excavations in normal-sized discs.⁸ While the eye offers the opportunity to view the changes of ROP directly, the diagnosis of IVH and PVL depends on indirect neuroimaging techniques such as cerebral ultrasonography (US), computed tomography (CT), and magnetic resonance imaging (MRI) of the brain.

In the industrialised world, screening for ROP is widespread as is neonatal cranial US. Unfortunately, cases with less severe lesions to the periventricular white matter are often missed by US.^{9–11} The method of choice for diagnosis of PVL is MRI, which is expensive, time consuming, and usually requires that the child be anaesthetised. Thus, only a small portion of children at risk for PVL are examined by MRI. CVI may affect the same visual

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functions as eye diseases and, in addition, produce significant impairment of visual perception, that is, the ability to extract information from what is seen. Impaired recognition of faces and forms, orientation, depth perception, and simultaneous perception have been described in children with CVI. Affected children may also have crowding, that is, difficulties identifying one object surrounded by others. Movement perception may be impaired in some children, while others see moving objects better than stationary ones. Vision may be variable, and paradoxes in visual behaviour that are difficult to understand are common.^{12–15} Visual perceptual problems may cause serious visual impairment despite good VA and normal visual fields.

As more immature and sick neonates survive, an increase in incidence of CVI is anticipated. In Sweden, CVI is now the leading cause of visual impairment in children, as defined by VA (< 0.3).¹⁶ There is reason to believe that many children who are visually impaired due to perceptual problems are not being identified, because they do not fulfill the criteria for visual impairment of the World Health Organization (WHO). Visual perception is not usually tested in eye clinics, but is evaluated by neuropsychologists as part of neurodevelopmental assessment. In a population-based study of 50 children born before 29 weeks of gestation, we found that 40% had results below the third percentile of the American reference group on a test of visual perception (TVPS-R, Test of Visual Perceptual Skills — Revised), compared with 10% in a Swedish group of age-matched full-term controls.⁵ The aim of the present study was to investigate the extent to which prematurely born patients in a paediatric eye clinic have visual perceptual problems.

Materials and methods

Patients

Children born preterm who were patients of the authors at the Department of Pediatric Ophthalmology at the Queen Silvia Children's Hospital, in or after 1997, were invited to participate in the study if they had been born before the 37th week of gestation, had a known brain lesion (CP and/or PVL) and/or strabismus and/or reduced VA (<0.8 with the best eye at the age of 5 years). They also had to be more than 5 years old and able to participate in testing of visual functions. The study was approved by the Ethics Committee of Göteborg University, and informed consent was obtained from each child's parent/guardian. The patients were divided into three subgroups. The 'brain lesion group' contained all patients with known CP and/or PVL; the 'strabismus group' all children with strabismus without

known CP/PVL, and the 'low VA group' the children with reduced VA without known CP/PVL or strabismus. Fewer than 10 of the children who were invited did not participate. Of the 91 children who participated, 65% had been referred to the eye clinic for ROP screening, 19% for strabismus, 11% for reduced vision, 3% because of known brain lesions and single cases (2%) for other reasons. In total, 10 of the children born before 29 weeks of gestation had participated in another study.⁵

A total of 31 children had brain lesions. Four of these 31 children had PVL without CP. In all, 46 children had strabismus without known brain lesions, and 14 had reduced VA in the absence of known brain lesions and strabismus. Median gestational age was 29 weeks (range, 24–36 weeks) and median birth weight was 1290 g (range, 390–3220 g). The median age at examination was 6.7 years (range, 5.1–12.3 years).

A total of 64 children (70%) had been examined by ultrasound in the neonatal period and 28 (31%) had had CT and 11 (12%) MRI performed at least once. The results of neuroimaging are presented in Table 1. ROP had been diagnosed in 33/91 children and eight of them had been treated with cryotherapy. Seven children were operated once and four children twice for strabismus.

Tests

Visual perception was examined with the TVPS-R.¹⁷ This is a nonmotor test for children aged between 4 and 12 years and 11 months of age. It consists of seven subtests of different visual functions; (1) *discrimination*: identification of forms; (2) *memory*: ability to remember single forms individually; (3) *spatial relations*: determination of the correct direction of forms; (4) *form constancy*: recognition of the same form when it varies in

Table 1 Results of neuroimaging

Study subgroup, result on neuroimaging	Neuroimaging		
	US	CT	MRI
<i>Known brain lesion (n = 31)</i>			
Abnormal	20	18	8
Normal	3	4	0
No information	8	9	23
<i>Strabismus (n = 46)</i>			
Abnormal	3	2	0
Normal	25	3	3
No information	18	41	43
<i>Low visual acuity (n = 14)</i>			
Abnormal	6	0	0
Normal	7	1	0
No information	1	13	14

size from the stimulus; (5) *sequential memory*: ability to remember a number of forms in a series; (6) *figure ground*: finding a form when it is hidden among other forms; (7) *closure*: determination of a whole form from parts of this form. Each subtest consists of 16 items and the number of correct responses yields a 'raw score' from 0 (all failed) to 16 (all correct). For each subtest and age (at 3 months intervals) the 'raw scores' are converted to 'scaled scores', the sum of which is the basis for a visual perceptual quotient (VPQ) and a percentile score for the whole test. Subnormal visual perception was defined as a result on the test below the third percentile of the reference material for the test corresponding to a VPQ of <70. A structured interview with the parents and children concerning five areas of visual perceptual problems reported in CVI (recognition, orientation, perception of depth and motion, and simultaneous perception) was performed using a questionnaire.¹²

VA, with best possible correction, was tested with the KM-Boks chart (Ortho KM), which is an arithmetically based letter-matching chart with seven different letters (C D E F K N V) of equal readability.¹⁸

Distance VA was tested binocularly with best correction at a distance of 3 m with a linear chart and with single symbols. A better VA with single than linear optotypes may be a manifestation of crowding. *Close range VA* was tested binocularly with a linear KM-Boks chart at a distance of 0.33 m. If children were found to need new spectacles after refraction in cycloplegia, they were rescheduled for repeated acuity testing with the new spectacles. *Visual fields* were tested with the standard Goldmann perimeter, and the largest brightest stimulus V4 was used to delineate the outer limits of the visual fields. In three patients an arc perimeter was used. As visual field testing at this age is difficult to evaluate, only larger defects like hemi- or quadrantanopias were looked for.

For the *orthoptic status*, cover-uncover tests were performed for close range and distant fixation. Strabismus was defined as a heterotropia that was manifest always or intermittently. Motility was tested grossly for paresis of the extraocular muscles.

The *anterior segment of the eye* was examined with a slitlamp and any visible nystagmus was noted. *Indirect ophthalmoscopy* was used for examination of the ocular fundus through dilated pupils and cycloplegic autorefractometry was performed 45 min after a single instillation of mixture of cyclopentolate (0.85%) and phenylephrine (1.5%). Significant refractive errors were defined as a spherical equivalent of myopia >0.5 diopters (D), hyperopia >2.5D, or anisometropia of >1.0D, or astigmatism >1.0D. Examinations were performed between April 1997 and April 2001. Before the data were analysed the hospital files of the children were

reviewed for information on neuroimaging and neuropsychological evaluation.

Ocular fundus photographs were taken and analysed concerning optic disc variables using a specially designed computer-assisted digital mapping system.¹⁹ The outlines of optic disc and the cup were marked by a cursor and their areas were calculated. The neuroretinal rim area was obtained by subtraction of the cup area from the disc area. When photographs of sufficient quality were available for both eyes the mean values were used. If only one eye had a sufficiently good photograph taken, the values of that eye were used.

Results

A total of 61 (67%) of the 91 patients had scores under the third percentile of the American reference material on TVPS-R. Of the 31 patients with known brain lesions 27 (87%) had scores under the third percentile. Scores under the third percentile were also observed in 22 of the 46 (48%) with strabismus without known brain lesions, and in 12 of 14 (86%) with reduced VA without strabismus or known brain lesion. The results of TVPS-R in relation to binocular VA are plotted in Figure 1. VA could be tested in all children but in two, only monocular but not binocular VA was tested and, therefore, their results are not plotted in the figure. Four children had not been tested with single symbols. Good VA did not preclude very poor results on the TVPS-R. In all, 86 patients and their parents were interviewed using the questionnaire. The proportion of patients for whom any problems were reported in none, one, two, three, four, or all of the five areas of visual perception (recognition, orientation, depth perception, motion perception, or simultaneous

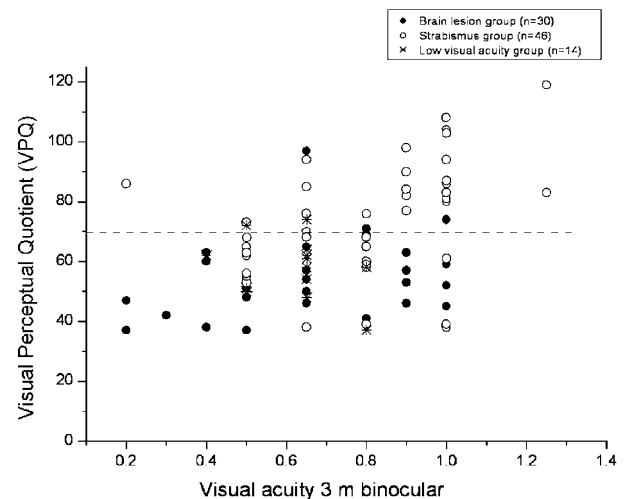


Figure 1 VPQ in relation to VA in preterm patients. The dotted line depicts VPQ <70 corresponding to results below the third percentile.

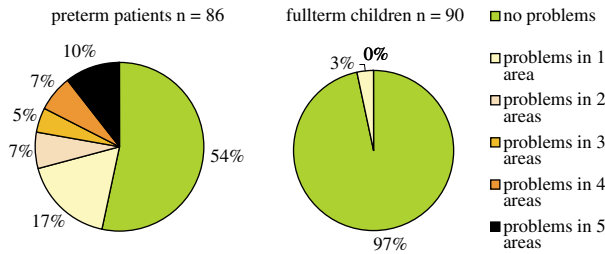


Figure 2 Proportion of patients ($n=86$) who exhibit either no problems with visual perception or who have problems in one, two, three, four or five areas (recognition, orientation, depth perception, motion perception, and/or simultaneous perception) according to the questionnaire. For comparison, the results of a reference group of 90 healthy full-term children of approximately the same age (median 8.3 years, range 5.1–12.1 years) are shown (Andersson-Gronlund M, Department of Pediatric Ophthalmology, the Queen Silvia Childrens Hospital, Göteborg, Sweden. Personal communication).

perception) is presented in Figure 2. For comparison, the results of the structured interview concerning 90 age-matched children born at term are shown. Problems were reported for 17 (61%) of the 28 in the brain lesion group, 17 (38%) of the 45 in the strabismus group and six (46%) of the 13 in the low-vision group in at least one area of visual perception. Problems in one area were reported for 3% of the full-term controls.

Three children had visual impairment according to the definition of the WHO, with a VA of less than 0.3. Median binocular VA at 3 m was 0.65 (range, 0.2–1.0) in the group with known brain lesions, 0.8 (range, 0.2–1.25) in the strabismus group and 0.65 (range, 0.4–0.8) in the low VA group. Linear binocular VA at 3 m is plotted against VA with single symbols in Figure 3. Better VA with single than linear optotypes was common. In total, 84 children had their visual fields examined. One child appeared to have a nasal hemianopia in a cryo-treated myopic eye with a VA of light perception only, otherwise no hemi- or quadrantanopias were observed.

The frequency of strabismus and reduced VA in each subgroup is presented in Table 2. Three patients in the strabismus subgroup, two with intermittent exotropia, and one with fully accommodative esotropia, presented no manifest strabismus at the examination of the study.

Examination of the anterior segment revealed ectropion uveae, coloboma of the iris, anomaly of the iridocorneal angle, iridodonesis, and prominent iris vessels in one child each. Nystagmus at slit-lamp examination was found in 13 of 31 children in the brain lesion group, in nine of 46 in the strabismus group and in one of the 14 in the subnormal VA group.

At ophthalmoscopy, anomalies of the optic nerve head were common. One optic nerve coloboma and one optic pit were observed. Three eyes had crowded discs and in 31 eyes of 16 patients the optic discs were considered

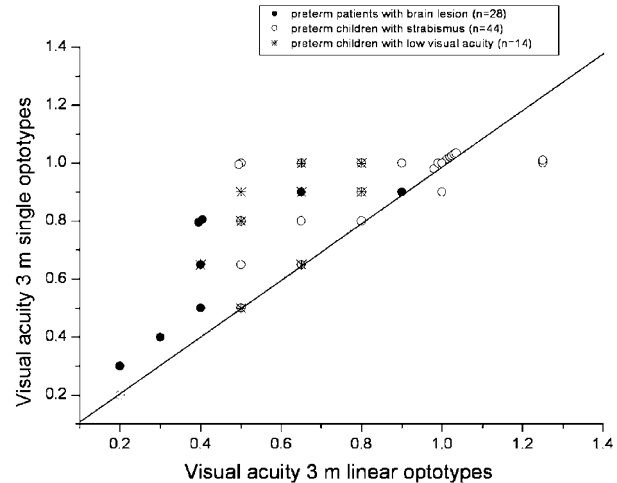


Figure 3 Linear binocular VA at 3 m in relation to binocular VA with single optotypes at the same distance.

Table 2 Frequency of strabismus and subnormal VA at the examination of the study

Study subgroup	Strabismus	Binocular visual acuity <0.8
Known brain lesion ($n=31$)	26	17
Strabismus ($n=46$)	43 ^a	16
Low visual acuity ($n=14$)	0	12 ^b

^aAt the examination of the study, two children with earlier diagnosed intermittent exotropia had exophoria but no apparent tropia and one child with fully accommodative esotropia was esophoric with her glasses on.

^bTwo children who had visual acuities of <0.8 at the age of 5 years had a binocular VA of 0.8 at the time of the examination of the study.

pale or pathologically excavated. The areas of the optic discs, cups, and neuroretinal rims are presented in Figure 4. A reduced neuroretinal rim area was present in some of the children with known brain lesions. The maculas of two eyes of two cryo-treated patients were grossly abnormal. Both had pigment disturbances and one had a retinal fold and the other lacked the annular reflex. In 23 eyes of 13 patients, the maculas looked somewhat hypoplastic with either a shallow appearance or a small annular reflex. In addition, one boy with PVL and pale optic discs lacked visible macular structures.

Refraction in cycloplegia for right and left eyes, respectively, is plotted in Figure 5. The majority of the children with refractive errors had hyperopia and/or astigmatism, while few had myopia. Significant anisometropia was found in 15 patients.

Discussion

In this study, we observed a high frequency of poor performance on a test of visual perception among

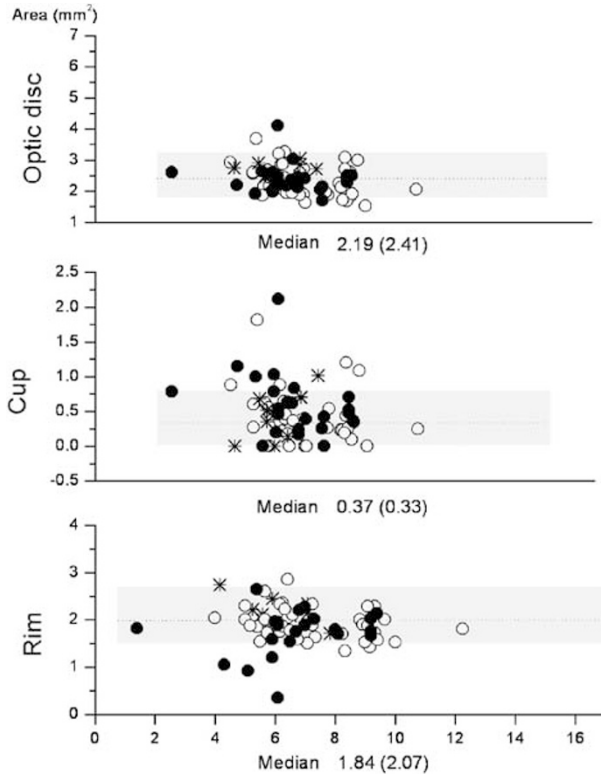


Figure 4 Areas of the optic disc, the cup and the neuroretinal rim of the patients in relation to reference material from full-term healthy children. The shaded area depicts the fifth and 95th percentiles of a reference material.²⁰

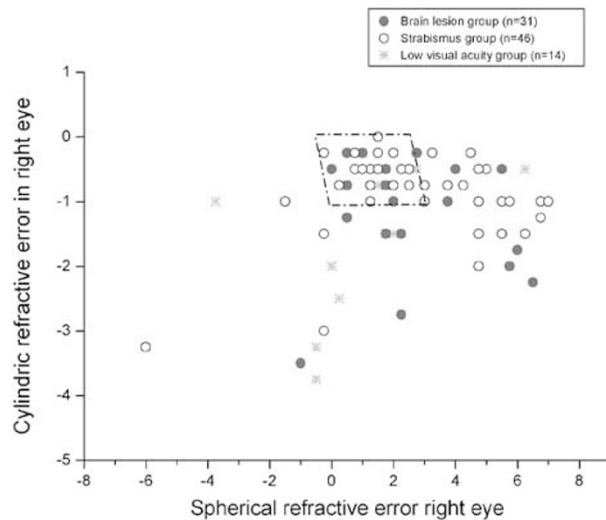


Figure 5 (a) Refraction of the right eye of each patient. (b) Refraction of the left eye. Values outside the marked areas are defined as significant refractive errors.

prematurely born ophthalmologic patients both with and without known brain lesions. A large proportion of these children were also reported to have perceptual problems.

Many of those with reduced VA had better results with single than linear optotypes, a possible indicator of perceptual problems. However, in the absence of normal values for crowding in this age group this finding cannot be evaluated. Hyperopia was common in the children with known or suspected brain lesions. Interestingly, associations between early hyperopia and neonatal ultrasound abnormalities²¹ as well as visuocognitive and visuomotor developmental deficits have been reported recently.²² Impaired visual perception is a consequence of dysfunction of visual processing by the brain, not a symptom of ocular disorders. Perceptual problems have been observed in patients with lesions of the periventricular white matter such as PVL and hydrocephalus, but there is reason to believe that in addition to the primary visual pathways, associative areas, not easily visualised with neuroimaging, might be involved. Jacobson *et al*²³ have used the term cerebral visual dysfunction (CVD) to describe the visual consequences of PVL. The term might be used to describe CVI in which the impairment is severe enough to fulfill the WHO criteria for visual impairment (VA <0.3) and, in addition, other forms of dysfunction of vision, including visual perceptual problems, that might influence the ability to cope with activities of daily life.

The relationship between radiological findings and visual functions in children with perinatal lesions of the posterior visual pathways has been studied mainly in children with large lesions detected by ultrasound and in neurologically impaired children. An association has been observed between extensive cystic PVL and CVI.^{24,25} Lanzi *et al*.²⁶ found a correlation between reduction of VA and reduction of the peritrigonal white matter as well as the extent of calcarine atrophy. Reduction of the volume of the peritrigonal white matter on MRI has been reported to correlate with impairment of visual perception in children with spastic diplegia.²⁷ Also, a negative correlation has been observed between visuoperceptual ability and the ratio of the areas of the posterior horns of the lateral ventricles to the anterior horns in children with spastic diplegia and spina bifida with hydrocephalus.^{28,29}

Lesions of the periventricular white matter visible by MRI are common in children who were born prematurely and PVL is also reported to be common in apparently healthy preterm children.^{30–33} Few studies have been conducted on the visual functions of children with PVL in the absence of CP. It seems likely that many of these children are not identified as having PVL, as few children who do not have CP undergo CT or MRI. No direct correlation has been determined between radiological findings and pattern or severity of visual difficulties in children who sustain cerebral insults preterm. Therefore, each child requires individual assessments of multiple

aspects of vision.^{34,35} There is evidence that some difficulties related to prematurity disappear with age. In a study of teenagers born preterm, periventricular white matter lesions were found to be common, but teenagers with these lesions did not differ in neuropsychological outcomes from those without such lesions.³⁴ The last few years, there has been an increased awareness among paediatric ophthalmologists about lesions of the posterior visual pathways and accompanying visual perceptual problems especially among children born preterm. There are, however, many questions to be answered: What do we tell parents about the visual prognosis in children who have been found to have PVL on neuroimaging? Should the possibility of future visual problems be raised in the neonatal period or later? Should we as ophthalmologists screen for visual perceptual problems? If so, how? Are the tests that are available appropriate for finding visual perceptual problems that impair activities of daily life? At what age should these tests be applied? To provide optimal education, teachers and parents need to be aware of the existence of perceptual problems before the child begins school. Which children should be assessed and for what? It seems reasonable to examine visual functions, including visual perception, in all children with a diagnosis of CP, hydrocephalus, and PVL, but how do we identify those children who do not exhibit a neurological handicap and in whom insufficient neuro-imaging has been performed to set the diagnosis?

Disability in preterm children is often expressed as the proportion of children with major handicaps such as CP and/or visual impairment. We believe it is important to monitor defects that are less evident, such as visual perception, to obtain a more comprehensive measure of the outcome of the pre- and perinatal care. Children with profound visual perceptual problems need an early diagnosis, to assure appropriate support. Our results highlight the high frequency of reduced visual perception in preterm children with CP, PVL, strabismus and/or subnormal VA. We recommend that the possibility of visual perceptual problems is considered and evaluated in all preterm children with these symptoms and/or signs.

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