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Lessons Learned from 23 Years of Experience in Testing Visual Fields of Neurologically Impaired Children

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ABSTRACT

We sought to investigate the reliability of standard conventional perimetry (SCP) in neurologically impaired (NI) children using the examiner-based assessment of reliability scoring system and to determine the difference in time to diagnosis of a visual field defect between SCP and a behavioural visual field (BVF) test. Patient records of 115 NI children were retrospectively analysed. The full field peritest (FFP) had best reliability with 44% 'good' scores versus 22% for Goldmann perimetry ($p < .001$). The mean age of NI children able to perform SCP was 8.3 years versus 4.6 years for the BVF test ($p < .001$). Use of the BVF test may significantly reduce time to diagnosis.

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Cerebral visual impairment (CVI); neurological impairment (NI); perimetry; children

Introduction

Children suffering from neurological impairment (NI) may show various visual impairments such as a decreased visual acuity, visual field defects (VFD), disorders of eye movements and disorders of higher visual processing, which may be diagnosed as cerebral visual impairment (CVI).^{1–3}

CVI, defined by Sakki et al. as “a verifiable visual dysfunction which cannot be attributed to disorders of the anterior visual pathways or any potentially co-occurring ocular impairment,”⁴ is the main cause of childhood visual disability in developed countries. It may have a pre-, peri- or postnatal origin with a prevalence between 10 and 22 cases per 10,000 births.^{2,5}

Although the need for the development and refinement of approaches which allow early detection of gross VFDs has recently been stressed by Patel et al.,⁶ retrospective studies of the visual field (VF) in a large cohort of NI children with or without CVI are lacking in the current literature.

Several techniques can be used to examine the VF in children, such as standard conventional perimetry (SCP), confrontational behavioural visual field (BVF) methods (such as the Behavioural Visual Field [BEFIE] screening test, the use of Stycar balls or double-arc perimetry^{7,8}) or eye-tracker and multifocal visual evoked potential techniques. However, despite these various options, it can still be difficult to examine the

VF in NI children^{9–12} due to a lack of concentration, short attention span, psycho-motor impairment or retardation and intolerance to the restrictions of head movement required to perform most of these tests.^{12–14}

Detection of a VFD in NI children is important because it may represent one of the first symptomatic signs¹⁵ or contribute to finding the right diagnosis in pathologies such as paediatric stroke, cerebral palsy and periventricular leukomalacia.^{16–20} It could also aid parents and caregivers to understand the child's visual behaviour, resulting in better acceptance, improved quality of life and more adequate rehabilitation strategies.^{21,22}

To the best of our knowledge a comparison of SCP with a confrontational BVF method for testing VF in NI children searching for a potential gain in time to diagnosis of a VFD has never been performed. The aim of this study is to describe the results of SCP in a cohort of NI children. Additionally, we sought to confirm a potential gain in time to diagnose a VFD by using a BVF test.

Materials and methods

Patient selection

This study retrospectively followed all NI children that underwent a confrontational BVF test before

the age of 12 and who also underwent SCP at the Utrecht University Hospital from January 1995 until June 2018. The study was approved by the institutional ethical committee of the University Medical Centre Utrecht, which also deemed that the collection of written informed consent to this study was not necessary. Publication of the child pictured in the photograph in [Figure 1](#) was authorised by obtaining written informed consent.

Data collection

The patient files were retrospectively analysed. The collected demographic and clinical characteristics included sex, age at examination and type of pathology. Data of the earliest SCP tests with best representation of VF were gathered. If multiple SCP tests were used in an individual child, our preference went initially to Goldmann perimetry. When this was lacking, data of the first performed full field Peritest (FFP) or the first performed Humphrey field analyser (HFA) including peripheral stimuli were recorded. As last resort, data on the first performed central Peritest (CP) were gathered. If scores differed per eye, the score of the best eye was included. If the scores were the same, those of the right eye were included.

SCP

The SCP tests used in our centre were manual kinetic testing on the Goldmann perimeter, semiautomatic-static testing on the Peritest (Rodenstock, Germany)²³ and automatic-static testing on the HFA.

The Peritest used a measure point in the fovea to determine the sensitivity threshold. Stimuli could be presented that were 2, 4, or 6 decibel supra-luminal. Light intensity was adjusted for the sensitivity decrease in the periphery. The CP protocol consisted of either 75 or 150 points within the inner 25 degrees of the VF. The FFP protocol consisted of the central Peritest with an additional 55 points above 25 degrees of the VF.

All children that were tested with the Goldmann perimeter and the HFA, were tested with the V4 isopter and the 120-point protocol respectively.

Due to the retrospective nature of this study, exact protocol times were not listed. All SCP tests took approximately 30–40 minutes with breaks included. As soon as SCP was possible for a child, they were tested with the CP or an SCP test with periphery, if it were possible. The treating ophthalmologist chose between Goldmann, FFP and HFA.



Figure 1. The behavioural visual field screening test. Equipment includes a rod with a level attached to it used for positioning (1), a graded semicircular black metal arc with a white stimulus at the end (2), and a white fixation target on a rod (3).

Confrontational behavioural measurement

The confrontational BVF method used in our centre is the BEFIE screening test (see [Figure 1](#)), a simple kinetic BVF test, designed in our institution with the aim of testing children of preverbal ages or NI children.⁷

The BEFIE test, which requires an examiner and an observer, is easy to apply in clinical practice and creates a high intrinsic motivation for the child to cooperate due to the game-like interaction between examiner, observer and patient. With this test, peripheral VFDs such as hemianopic, quadrantanopic or concentric VFDs can be detected in children as young as four months of age.²⁴

SCP test reliability in NI children

To gain insight into the reliability of SCP tests in children with NI, the examiner-based assessment of reliability (EBAR) scoring system was used.²⁵ To score tests more objectively, cooperation and fixation were dichotomised using the test results with comments made by the examiner. The scores were made by matching the descriptions of the EBAR scoring system with our retrospectively gathered results and comments. SCP tests were rated “good” when cooperation and fixation both had a score of “+”. “Poor” ratings were given if cooperation and/or fixation were rated as “-” and an SCP test was scored as “fair” when cooperation and fixation were intermediate. Patients were excluded when no comment was given on the test result.

Additionally, follow-up data of all children that underwent a second SCP test at this institution were gathered to compare whether the first and second SCP test were similar or different. If there were any apparent reasons for deterioration of disease and/or VFD, patients were excluded for this sub-analysis.

Average age

The children’s age during the earliest SCP test with best representation was compared with their age during the earliest reliable monocular BEFIE test. This analysis was performed in order to find the average age at which NI children are able to perform perimetry tests and to examine the potential

gain in time to diagnose a VFD should the BEFIE test be routinely incorporated into ophthalmological practice.

After their first full ophthalmological and orthoptic investigation including the first BEFIE test, the NI children obtained a regular follow-up using the BEFIE test until SCP was possible. All BEFIE tests were performed by the same examiner (GP).

Reasons for exclusion for this analysis were: no monocular, but only binocular BEFIE tests, BEFIE and SCP tests performed on same day (for example before epilepsy surgery in accordance with local protocol) or age above 12 during the first BEFIE test.

Statistical analysis

Reliability was calculated using the chi-square test. Due to the large sample size and the normality of data in the BEFIE test age group, the analysis for average age difference between SCP and BEFIE was calculated using the paired sample t-test. All statistical tests were performed using IBM SPSS Statistics for Macintosh, version 25 (IBM Corp., Armonk, NY, USA).

Results

In total 138 children were eligible for this study of which 115 NI children (69 boys) were included after implementation of the exclusion criteria. The mean age in which the NI children could perform an SCP test was 8.3 (range 4.5–17.4 standard deviation [SD] 2.5) years. The majority of the children suffered from neoplasms or stroke/haemorrhage. Most children had normal VFs or suffered from (partial) hemianopias or quadrantanopias.

Of the SCP tests used, 43% were FFP, 23% were Goldmann perimetry, 30% were CP and 4% were HFA tests. For more details on the pathologies present see [Table 1](#).

Reliability of SCP tests in NI children

All 115 children were included to measure the reliability of SCP tests, but due to the small number of children tested with central and full field HFA tests (1 and 3 respectively), results obtained using these two methods were excluded for the statistical analysis.

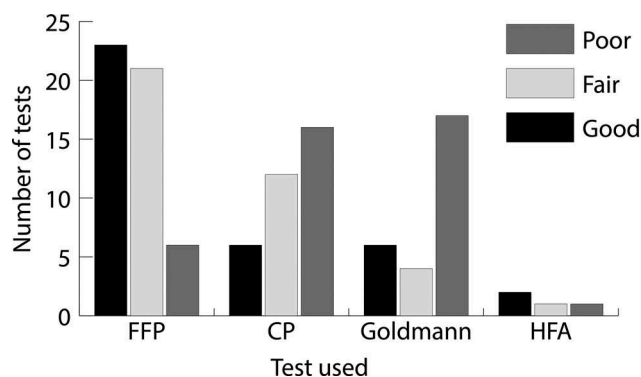
Table 1. Baseline table showing the different pathologies present in the neurologically impaired children.

Pathologies	n (%)
Neoplasm	36 (31)
Stroke/haemorrhage	34 (30)
Raised intracranial pressure	13 (11)
Cyst	8 (7)
Asphyxia/periventricular leukomalacia	8 (7)
Lesion/dysplasia	6 (5)
Epilepsy	5 (4)
Unknown	3 (3)
Trauma	2 (2)
Total	115 (100)

In total, 37 were rated “good”, 38 as “fair”, 40 as “poor”. The ratings for the different SCP tests are shown in Figure 2 and Table 2. Among the FFP tests, 46% (23) had “good” reliability, 42% (21) were rated as “fair” and 12% (6) as “poor”. For Goldmann perimetry, 22% (6) had “good” reliability, 15% (4) were rated as “fair” and 63% (17) as “poor”. Among the CP tests 18% (6) had “good” reliability, 35% (12) were rated as “fair” and 47% (16) as “poor”. The difference between SCP tests was significant with a $p < .001$.

Out of 115 children, 45 children (84 eyes) underwent more than one SCP test. The mean age at which these children were able to perform the first SCP test per method was: 7.2 (range 5.3–9.8, SD 1.2) years for the CP, 7.5 (range 4.5–10.7, SD 1.5) years for the FP and 8.6 (range 5.6–14.6, SD 2.6) years for Goldmann perimetry. The mean age at which they performed their second test was 9.0 (range 5.0–16.5, SD 2.5) years.

86.9% of all second SCP results were congruent with those of the first test. For CP, FFP and Goldmann perimetry percentage of congruence

**Figure 2.** Results of the EBAR scoring system per SCP test: full field Peritest (FFP); central Peritest (CP); Goldmann perimetry; and Humphrey Field Analyser (HFA).**Table 2.** Number of Standard Conventional Perimetry (SCP) tests used for the cohort of neurologically impaired children. Examiner-based assessment of reliability (EBAR) scores (i.e. *Good, Fair or Poor*) per SCP type is shown. Gender percentages calculated horizontally of total in subgroup. Percentages of total calculated vertically.

Standard Conventional Perimetry	Male (%)	Female (%)	Total (%)
Full Field Peritest	28 (56)	22 (44)	50 (43)
<i>Good</i>	11	12	23
<i>Fair</i>	14	7	21
<i>Poor</i>	3	3	6
Central Peritest	25 (74)	9 (26)	34 (30)
<i>Good</i>	4	2	6
<i>Fair</i>	10	2	12
<i>Poor</i>	11	5	16
Goldmann perimetry	13 (48)	14 (52)	27 (23)
<i>Good</i>	2	4	6
<i>Fair</i>	4	0	4
<i>Poor</i>	7	10	17
Humphrey Field Analyser	3 (75)	1 (25)	4 (4)
<i>Good</i>	1	1	2
<i>Fair</i>	1	0	1
<i>Poor</i>	1	0	1
Total	69 (60)	46 (40)	115 (100)

was 89.7%, 81.3% and 91.3% respectively. Nine eyes showed deterioration of VF and two showed improvement (Figure 3).

Average age

For the comparison between the average age during the first BEFIE test and during the first SCP test, 104 out of the total 115 children were included. For this subgroup, the mean age in which the first monocular BEFIE test was possible for children with NI was 4.5 (range 0.7–11.8, SD 2.4) years. The mean age at which SCP was possible for children with NI was 8.2 (range 4.5–17.4, SD 2.5) years. The mean total difference between the first BEFIE test and the first SCP test was 3.7 years (95% confidence intervals 3.15–4.21, $p < .001$).

Comparison between BEFIE and SCP results

58.8% of the children had the same results on BEFIE and SCP tests. 17.1% had similar results without clinical difference. In 24.2% the results were different, either due to one test showing a VFD whereas the other did not (20.7%), or due to both showing different VFDs (3.5%) (Table 3).

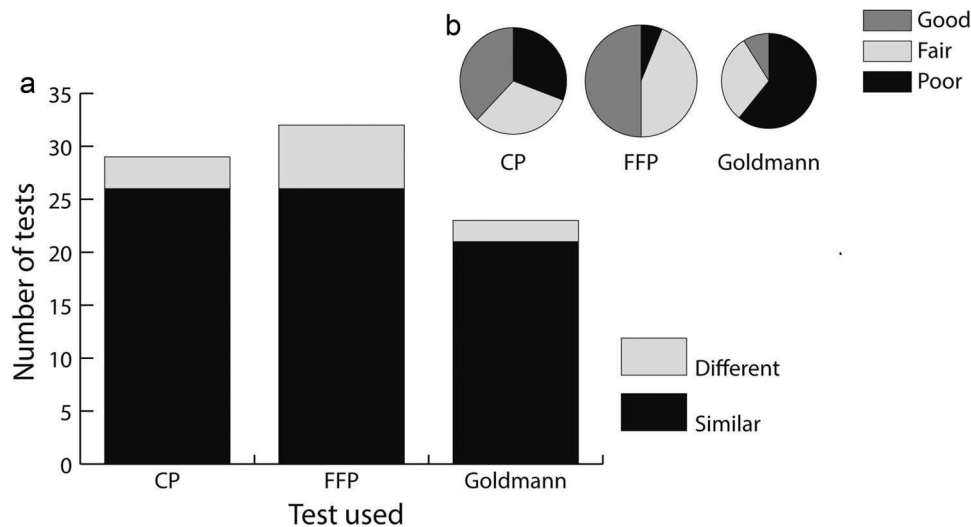


Figure 3. Comparison between the first and second SCP tests for the central Peritest (CP), the full field Peritest (FFP) and the Goldmann perimetry for all eyes of children that underwent a second SCP test.

A shows the number of tests that had clinically similar or different results per SCP test. **B** shows the distribution of the EBAR scoring system results per SCP test for this subcohort.

Table 3. Comparison of visual field defects detected by the behavioural visual field (BEFIE) test and standard conventional perimetry (SCP) per eye (with some missing datasets). Results scored per eye. Note that percentages do not always exactly add up to 100% due to rounding of numbers.

Visual Field Defect	BEFIE n (%)	SCP n (%)
No defect	134 (65)	92 (42)
Hemianopia	26 (13)	33 (15)
Partial hemianopia	20 (10)	10 (5)
Quadrantanopia	1 (1)	8 (4)
Partial quadrantanopia	12 (6)	22 (10)
Spread scotomas	0 (0)	23 (10)
Peripheral defect	0 (0)	13 (6)
Concentric defect	12 (6)	12 (5)
Centrocaecal scotoma	0 (0)	8 (4)
Total	205 (100)	221 (100)

Discussion

With data gathered over a period of 23 years, this is the longest and, as far as we know, the only retrospective study of the testing of VFs in such a large cohort of NI children with or without CVI. This is probably due to the commonly underestimated importance of examining VFs in this group and the well-known methodological difficulties,^{9–12} such as a lack of concentration, short attention span, psycho-motor impairment or retardation and the intolerance to the restrictions of head movement required to perform most SCP tests.^{12–14} Furthermore, there is little evidence regarding the reliability of SCP in children with NI, a group of children that is at higher risk of developing a VFD.⁶

Also, there is little consensus on how to approach these measurements, both in healthy and in NI children. In a recent study, Goldmann perimetry and Humphrey field tests, the two most common perimetric tests used in children, have shown to be reliable in healthy children using the EBAR scoring system.²⁵ Patel et al. measured the reliability of the discontinued Goldmann and Octopus perimeters in children with NI, while other authors limited their measurements to confrontation methods or the Amsler test.^{6,26,27}

Pathologies

Out of all NI children who were able to perform both a BEFIE test and a SCP test, the majority suffered from neoplasm (30.9%) or stroke/haemorrhage (26.6%). This could suggest that when these are the causes of NI, children may have a higher chance to be able to perform an SCP test, due to plasticity of the brain.²⁸ However, these data could be biased by a high prevalence of these pathologies in our centre. Therefore, more research is needed to support these data and hypotheses.

Although this study did focus on the various brain pathologies, it did not include their localisations. Therefore, no assumptions can be made about which SCP test is more reliable for each

localisation. An assumption about which SCP test is more reliable for each pathology group cannot be made, due to the small number of children in each group.

Perimetry tests used

Only 24.6% of NI children managed to complete Goldmann perimetry and only 26.6% completed a CP, whereas the majority of all participants (44.9%) managed to complete an FFP. Therefore, a Peritest could be suggested when the commonly used Goldmann perimetry is likely to fail.¹⁰

Unfortunately, internationally the Peritest is nowadays not routinely used, even though it was reported to perform well in the few studies that described it^{23,29,30} and the same will probably happen to the discontinued Goldmann perimeter.

Currently, the most commonly used perimetry test in children is the HFA, although recent studies have opted for Octopus perimetry as a replacement for the Goldmann perimeter.^{6,25,31} In a recent study comparing Goldmann with Octopus perimetry, broad agreement was found and these tests were recommended for children over eight-years-old with neuro-ophthalmological disease.⁶

We believe that eye tracking applications might prove useful when testing VFs in children in the future.³²⁻³⁴ Furthermore, predicting VFDs using OCT seems to be possible in children with a developmental age of 3–6 years.³⁵

In our centre the Octopus perimeter is not available and the HFA is sparsely used in children due to the extensive and positive experience of staff in testing children with or without NI using the Peritest.^{36,37} Hence, a comparison of Octopus perimeter and HFA was not possible in our retrospective study. Therefore, we suggest prospective studies using the two above-mentioned, more widely used, VF tests for a conclusive comparison.

Reliability of SCP tests in NI children

Due to the retrospective nature of this study, the results of the EBAR analysis might not be perfectly representative. Although a prospective study could incorporate the score definitions more accurately, in our opinion the data obtained give a fairly accurate representation of SCP reliability in children with NI.

Although both kinetic and static perimetry should be considered in the NI child, the FFP, a static VF test, has a significantly higher reliability score for the first measurement of VF in NI children.

The Goldmann perimeter, a kinetic VF test and one of the more commonly used SCP tests in clinical practice for testing children,^{6,10,14} has been shown to be highly reliable in healthy ones.²⁵ It showed only 22% “good” reliability in the NI children in our cohort, probably due to a prolonged learning curve in comparison with the Peritest.

Pathology, localisation and severity can predict which SCP method could be more suited or might have a higher chance of a successful measurement of VF. For instance, in damage to the periventricular matter or to the parieto-occipital region, the sensitivity of movement perception might be reduced, consequently rendering the kinetic perimetry less applicable.²⁶

The CP had high numbers of “poor” ratings. This is probably due to more severe pathology, as this sample of NI children were only able to perform this shorter test of the central field and not more difficult tests.

When looking at the results of the comparison between the first and second SCP tests, there are a few limitations. Although the numbers give a fair perspective as to what age a clinically significant VFD can be detected, a prospective study with a correct set-up would be needed to accurately determine specificity and sensitivity of these tests. Furthermore, only a small portion of the cohort performed more than one SCP test. The reasons for the paucity of visual field testing during follow-up remain unknown in our retrospective analysis. Also, even though the most apparent reasons for deterioration, e.g. surgery, were excluded, differing results between the first and the second test could still originate from progression of disease or neuronal plasticity.³⁸ Lastly, all SCP tests used are subjective tests, complicating efforts to perfectly replicate a previous test.

Interestingly, even though the majority of Goldmann tests were made with a ‘poor’ EBAR reliability score, 91.3% of the children showed similar VFDs at follow-up. Though the EBAR scoring system has proven to be a useful tool to determine whether a visual field test is executed

reliably,^{6,25} in our cohort a lower EBAR score did not seem to correlate with a lower chance of finding a clinically significant VFD. The reason for this disparity in results is unknown to the authors, but it could be due to the use of stricter EBAR protocols, resulting in a bias towards more 'poor' scores. Another explanation may be that reliability indices for the EBAR scoring system contribute little to test-retest reliability, much like traditional perimetry indices.³⁹

Mean age

An important finding in this study is that the mean age at which a NI child can perform an SCP test is 8.3 years. Furthermore, this study has shown that a monocular BEFIE test for testing the peripheral VF can be successfully performed on average 3.7 years earlier than an SCP test. This is due to the adaptations at the psycho-motor impairment of the NI child and the game-like interaction between the child and the examiner and observer.²⁴ These characteristics make it very suitable for healthy children of preverbal ages as well. This finding highlights the importance of a wider clinical application of this behavioural test, especially when considering that Koenraads et al. already showed a specificity of 98% and sensitivity of 60% for this test, which increased to 80% when only absolute PVF defects at SCP are taken into account.²⁴ From these results and the congruence found in our study, we can conclude that using a behavioural visual field test, like the BEFIE screening test, leads to a high probability of diagnosing a clinically significant VFD in children affected by cortical damage. Please note that the BEFIE test is unable to diagnose central and relative VFDs, whereas a percentage of the NI children in this study were proven to have these defects. If those were to be excluded, the congruence could potentially be higher.

Considering that VFDs may represent one of the first symptomatic signs of CVI in children,¹⁵ such a considerable time gain of 3.7 years in the diagnosis of a VFD using the BEFIE test could help to drastically lower the delay in diagnosing CVI in children. Furthermore, it could help parents and caregivers to understand the child's behaviour, resulting in better acceptance, improved quality of life and more adequate treatment or rehabilitation strategies.^{21,22}

In addition, as 29% of all NI children tested were able to have only their central visual field tested with SCP, while in all of them it was possible to test the peripheral VF using the BEFIE test, the BEFIE test could be a useful complementary test in addition to SCP.

Limitations

This study has several limitations; first of all those associated with a retrospective study.

In addition, this study reports the experience of a single centre cohort, in which only one examiner (the ophthalmologist GP) performed all of the BEFIE tests, helped by different observers, who were all orthoptists. No inter-user data of the BEFIE test was hence gathered, while the SCP tests were performed by different technicians.

The BEFIE test requires a trained observer and examiner and it has the limitation of only testing the peripheral VF. Therefore, we strongly suggest the development of a reliability scoring system for the BEFIE test prior to widespread implementation in ophthalmology departments. Alternatively, we recommend the development of a better BVF test or ultimately an objective measurement of VF in children, less influenced by a lack of co-operation, attention or psychomotor impairment.

Our centre has extensive experience using the Peritest for testing children, resulting in a larger cohort of NI children that performed the Peritest than HFA, which is nowadays considered the state-of-the-art when testing VF in children.⁴⁰

A prospective study, without the above-mentioned limitations, could further clarify which SCP test is best suited for NI children.

Conclusion

In conclusion, this retrospective study from 23 years of experience in testing VFs of NI children showed that the FFP was the most reliable VF screening test. A BVF test, such as the BEFIE test led to a significant gain in time to diagnose a peripheral VFD of 3.7 years. We emphasise the importance of an early diagnosis of a peripheral VFD by means of any available BVF test in clinical practice as it can lead to better care for NI children.

Declaration of interest statement

The authors report no conflicts of interest.

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Data availability statement

The data that support the findings of this study are available upon request from the corresponding author. The data are not publicly available due to their containing information that could compromise the privacy of research participants.

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