# Costs and effects of conventional vision screening and photoscreening in the Dutch preventive child health care system

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**Background:** Little is known about costs and effects of vision screening strategies to detect amblyopia. Aim of this
study was to compare costs and effects of conventional (optotype) vision screening, photoscreening or a combination in children aged 3–6 years. **Methods:** Population-based, cross-sectional study in preventive child health care
in The Hague. Children aged 3 years (3y), 3 years and 9 months (3y9m) or 5–6 years (5/6y) received the conventional
chart vision screening and a test with a photoscreener (Plusoptix S12C). Costs were based on test duration and
additional costs for devices and diagnostic work-up. **Results:** Two thousand, one hundred and forty-four children
were included. The estimated costs per child screened were €17.44, €20.37 and €6.90 for conventional vision
screening at 3y, 3y9m and 5/6y, respectively. For photoscreening, these estimates were €6.61, €7.52 and €9.40
and for photoscreening followed by vision screening if the result was unclear (combination) €9.32 (3y) and €9.33
(3y9m). The number of children detected with amblyopia by age were 9, 14 and 5 (conventional screening), 6, 13
and 3 (photoscreening) and 10 (3y) and 15 (3y9m) (combination), respectively. The estimated costs per child
diagnosed with amblyopia were €1500, €1050 and €860 for conventional vision screening, €860, €420 and
€1940 for photoscreening and €730 (3y) and €450 (3y9m) for the combination. **Conclusions:** Combining photo-
screening with vision screening seems promising to detect amblyopia in children aged 3y/3y9m, whereas conven-
tional screening seems preferable at 5/6y. As the number of study children with amblyopia is small, further
research on the effects of these screening alternatives in detecting children with amblyopia is recommended.

## Introduction

Amblyopia in childhood, estimated to be present in 1–4% of children, can cause permanent vision loss.[1,2] The USPSTF[1] and AAPOS[2] recommend vision screening to detect amblyopia or its risk factors at least once in all children aged 3–5 years.[3] If treatment is started timely (<7 years), most patients have their vision restored.[3–5]

Especially in younger children, conventional vision screening using chart tests is time-consuming. Instrument-based screening is quick and requires minimal cooperation of the child. A systematic review by the USPSTF[3] reports positive likelihood ratios for instrument-based vision screening, indicating increased risk of the target condition (i.e. amblyopia, amblyopia risk factors, refractive error or reduced visual acuity) after an abnormal screening. The American Academy of Paediatrics advises instrument-based vision screening until the age at which visual acuity can be assessed reliably using optotypes (often 4 years).[6]

In the Netherlands, population-based vision screening is a task of the preventive Child Health Care (CHC) services. Aim of the current study is to establish whether vision screening in the age group 3–6 years can be improved (in terms of screening performance and costs) by the introduction of photoscreening.

## Methods

### Preventive CHC sample

From 1 September 2016 to 10 March 2017, all children aged 3–6 years without known eye or vision problems visiting CHC were recruited at five CHC locations in The Hague, The Netherlands. The recruited children received regular care, including conventional vision screening by a trained CHC nurse [3-years-olds (3y)], CHC physician [3 years and 9 months (3y9m)] or CHC assistant [5–6 years (5/6y)]. Prior to the standard vision screening, the children were screened with a handheld photoscreener (the Plusoptix S12C Mobile Vision Screener®) by a trained CHC assistant. CHC nurses and physicians were kept blind for the photoscreener results. Blinding of the CHC assistants for the measurement outcomes was not possible. Due to organizational reasons both measurements had to be performed by the same assistant at 5/6y. Therefore, at this age, conventional vision screening was performed prior to the test with the photoscreener. The study followed a within-subject design: each child had both conventional vision screening and photoscreening, allowing comparison of the results without distortion by individual differences.

### Ethics

The Medical Ethics Committee Southwest Holland judged that Medical Research (Human Subjects) Act does not apply to this study. The Committee therefore considered full evaluation not indicated. All parents were informed about the study before their visit to CHC and were asked to participate. If they agreed, they signed an informed consent form prior to inclusion of their child in the study.

### Conventional CHC vision screening

Conventional vision screening included measurement of visual acuity with the Amsterdam Picture Chart (APK) in 3y and with the Landolt-C chart at the ages of 3y9m and 5/6y.[7,8] Measurements are usually performed at a distance of 5 m. The line number per eye of smallest symbols that the child could read should be registered, but often testing was stopped as soon as a pass was obtained. Cut-off criteria for visual acuity are age-dependent. Supplementary appendix S1 shows the scores for a 'pass' (p), 'refer' (r) or 'doubtful'(d) result.[8] If the score was 'refer', the child should be referred to the clinic for diagnostic evaluation by an orthoptist. However, in practice, CHC professionals frequently re-assess children. If the score was 'doubtful', re-assessment was planned 2–3 months later. For children with more than one visual acuity assessment, results were combined to one result per child as followed: 'pass' if a 'pass' was scored on at least one visit; 'refer' after a 'refer' score or after two assessments with either a 'doubtful' score or a failed chart test (tried without a result), while none of the assessments had a 'pass' score; 'no conclusion' if neither a pass nor a refer could be concluded, e.g. only one doubtful result or no test result because child did not co-operate.

### Photoscreening

Handheld Plusoptix S12C Mobile Vision Screener® devices were used (PlusoptiX GmbH; Nuremberg, Germany). These devices measure refraction, pupil sizes and corneal reflexes (ocular alignment or gaze asymmetry) at 1 m distance. They produce a noise and light to draw attention of the child. Measurements were compared with pre-programmed cut-off criteria (Supplementary appendix S1) which rendered a 'pass' (no further action) or a 'refer' (referral for diagnostic evaluation in the clinic). When the measurement failed ('refer or try again'), the test was repeated for maximally 5 min. If the outcome remained 'refer or try again', the test results were inconclusive and the child was only referred for diagnostic evaluation if the result of the conventional screening was a refer.

### Combination of photoscreening and conventional vision screening

Results on screening tests, referrals, detection of amblyopia and costs were analyzed by age group separately for the conventional vision screening and for photoscreening. For children aged 3y and 3y9m, also an alternative procedure that combines these tests was investigated *post hoc*. In this alternative, all children were screened with the photoscreener. After a 'pass' result, no further action was needed, while after a 'refer' result immediate referral to the orthoptist for clinical consultation was prescribed. Only children with a 'refer or try again' result (i.e. no 'pass' and no 'refer') underwent conventional vision screening with a vision chart test (APK or Landolt-C). If the result on the vision chart test was 'refer', the child was referred to the orthoptist; if the result was 'doubtful', repeated vision screening at a second visit was scheduled and if the result was 'pass' no further action was needed.

### Diagnostic evaluation and target condition

At the clinic, all referred children underwent full eye examination by a trained orthoptist and ophthalmologist. Strabismus was detected with the cover–uncover test. We considered amblyopia to be the target condition of the screening programme. The diagnosis 'amblyopia' was set either after the first eye examination by an orthoptist in the clinic, or after a 13-week period of refractive adaptation. Amblyopia was diagnosed in children with a difference in best corrected vision between the left and right eye of two or more lines on the vision chart. If vision could not be determined, a unilateral amblyopia was diagnosed if there was a preference for fixating with one eye in a cover test. Bilateral amblyopia was diagnosed if the best corrected vision was 0.7 or less, on a scale from ≤0.05 (blindness) to ≥1.0 (normal vision). The diagnosis amblyopia was based on the judgement of three orthoptists. Amblyopia was treated according to professional guidelines.[9,10]

### Data extraction and analysis

Screening data from conventional vision screening as well as photoscreening were extracted from the CHC registration system, together with background variables like parental country of birth and language of the child. Detailed photoscreening results (including sphere, cylinder, axis and pupil size per eye and gaze asymmetry) were extracted from the Plusoptix devices. Clinical data were registered by the orthoptist who examined the child. All extracted data were anonymous, but had a CHC identification number. We merged all data on this number. Analyses were performed in R[11] and SPSS (IBM, version 25). Statistical analyses were performed using $\chi^2$-squared tests (Linear-by-Linear Association), paired $t$-tests for comparison of screening costs per child and McNemar tests for paired proportions.

### Estimation of costs

Costs were estimated based on the time needed for each of the screening tests (measured using a stopwatch), the CHC professional who performed the test (nurse, physician, assistant) and their hourly rates.[12] Missing time registrations were dealt with by multiple imputation. The costs of a clinical consultation (€80.87, indexed to 2017 €[13]) were added for children diagnosed at the clinic. The price of one Plusoptix S12C Mobile Vision Screener® was €6710 (excluding 21% taxes) and yearly replacement of batteries costs €21.72 including taxes. The lifetime of the devices was estimated at 7 years. Assuming an interest rate of 4.2%, yearly costs per device were €1385.[13] Use of the devices during the half year of the study was distributed uneven because in Dutch CHC children aged 3y and 3y9m visit the same CHC teams, whereas children aged 5/6y visit other teams. Thus, 1519 study children aged 3y and 3y9m were screened by five devices, while the 625 children aged 5/6y were screened by four devices. Therefore, for children aged 3y and 3y9m cost for the device per child screened was estimated at €2.29 [=(5 devices × €1385)/(1519 children in 6 months × 2)], while for children aged 5/6y this was about €4.40 [=(4 × €1385)/(625 × 2)].

The cost-effectiveness ratio (CER) of alternative screening compared with conventional vision screening was calculated by dividing the difference in costs of screening and clinical consultation by the difference in effects, i.e. number of children diagnosed with amblyopia with each screening method.

## Results

### Inclusion

At CHC, 2144 children were included: 51% were female and country of birth of at least one of the parents was non-Western in 44% (in 38% only Western lands of birth were registered, in 18% these data were lacking).

### Test results

The fractions with a pass at the first test increased with age for conventional vision screening as well as photoscreening ($\chi^2$ test, both $P < 0.001$, table 1). The fractions of referrals after one or

**Table 1** Inclusion, test and diagnostic results: all ages combined and by age group

| | All ages combined | | 3 years | | 3 years + 9 months | | 5/6 years | |
|---|---|---|---|---|---|---|---|---|
| | Number | % | Number | % | Number | % | Number | % |
| Total | 2488 | | 865 | | 847 | | 776 | |
| No parental consent | 150 | 6 | 51 | 6 | 39 | 5 | 60 | 8 |
| Excluded (glasses/clinic) | 194 | 8 | 26 | 3 | 77 | 9 | 91 | 12 |
| Included in the study | 2144 | 86 | 788 | 91 | 731 | 86 | 625 | 81 |
| **Conventional vision screening: first test** | | | | | | | | |
| Pass | 1454 | 71 | 409 | 56 | 496 | 73 | 549 | 88 |
| Refer | 289 | 14 | 104 | 14 | 151 | 22 | 34 | 5 |
| Second visit needed[a] | 295 | 14 | 218 | 30 | 37 | 5 | 40 | 6 |
| Total with test result | 2038 | 100 | 731 | 100 | 684 | 100 | 623 | 100 |
| No test result | 106 | | 57 | | 47 | | 2 | |
| **Conventional vision screening: results of all CHC visits combined[b]** | | | | | | | | |
| Pass | 1710 | 81 | 559 | 73 | 579 | 81 | 572 | 92 |
| **Refer** | **294** | **14** | **132** | **17** | **122** | **17** | **40** | **6** |
| No conclusion | 108 | 5 | 78 | 10 | 18 | 3 | 12 | 2 |
| Total with result on at least 1 test | 2112 | 100 | 769 | 100 | 719 | 100 | 624 | 100 |
| No test result | 32 | | 19 | | 12 | | 1 | |
| | | % of refer | | % of refer | | % of refer | | % of refer |
| Arrived at clinic | 206 | 70 | 83 | 63 | 92 | 75 | 31 | 78 |
| | | % of arrived | | % of arrived | | % of arrived | | % of arrived |
| Amblyopia diagnosed | 28 | 14 | 9 | 11 | 14 | 15 | 5 | 16 |
| Amblyopia missed | 5 | 2 | 2 | 2 | 2 | 2 | 1 | 3 |
| Pass | 4 | | 2 | | 1 | | 1 | |
| No conclusion | 1 | | – | | 1 | | – | |
| **Photoscreening (1 test)** | | | | | | | | |
| Pass | 1789 | 84 | 620 | 79 | 620 | 85 | 549 | 88 |
| **Refer** | **140** | **7** | **49** | **6** | **47** | **6** | **44** | **7** |
| Refer or try again | 206 | 10 | 114 | 15 | 62 | 9 | 30 | 5 |
| Total with test result | 2135 | 100 | 783 | 100 | 729 | 100 | 623 | 100 |
| No test result | 9 | | 5 | | 2 | | 2 | |
| | | % of refer | | % of refer | | % of refer | | % of refer |
| Arrived at clinic | 92 | 66 | 26 | 53 | 34 | 72 | 32 | 73 |
| | | % of arrived | | % of arrived | | % of arrived | | % of arrived |
| Amblyopia diagnosed | 22 | 24 | 6 | 23 | 13 | 38 | 3 | 9 |
| Amblyopia missed | 11 | 12 | 5 | 19 | 3 | 9 | 3 | 9 |
| Pass | 4 | | 1 | | 1 | | 2 | |
| Refer or try again | 6 | | 3 | | 2 | | 1 | |
| No data | 1 | | 1 | | – | | – | |

Bold lines facilitate comparison of the percentage of refers between both screening methods.

a: Second visit needed: lines read on chart indicated doubtful vision or chart test was tried without a result: another chart test is needed.

b: Pass: 'pass' (p) on at least one visit. Refer: 'refer' (r) or 2× 'doubtful'(d)/tried without result, AND not 'pass'. No conclusion: neither pass nor refer, e.g. only one doubtful result or no test result because child did not cooperate.

more conventional screening tests were higher than after photoscreening at age 3y and 3y9m (McNemar test, both $P < 0.001$).

## Detection rate

In total, 53–78% of children with a 'refer' arrived at the clinic (table 1). Not all 33 known children diagnosed with amblyopia were detected by both methods: with conventional vision screening 5 children were missed (4 had a 'Pass' and 1 'No conclusion'), whereas 11 were missed with photoscreening (4 'Pass', 6 'Refer or try again', 1 'No data' (not measured)). Detection rate improved if both ways of screening were combined: at both 3y and 3y9m one child was still missed (table 3).

## Time

Mean time needed for the screening tests decreased with age. At each age, the Plusoptix test took on average less than half of the time needed for the vision chart tests (table 2).

## Costs per child screened

Costs per child screened are presented including and excluding the costs of diagnostic consultation (table 3 and figure 1). Costs of conventional vision screening are higher than photoscreening at the ages of 3y and 3y9m, but lower at 5/6y (paired *t*-tests, all $P \leq 0.006$, see table 3 for more statistics). In this older age group, the vision chart test takes less time than at younger age, and more often results in a 'pass' and thus no need for a follow-up visit at CHC or a referral for diagnostics, which reduces costs at 5/6y. Also, at 5/6y the Plusoptix screening devices had higher costs per child screened than at age 3y and 3y9m (see Methods section).

With the combination of photoscreening and conventional vision screening, costs per child screened are lower than for conventional vision screening alone at 3y and 3y9m (paired *t*-tests, all $P < 0.001$, table 3). Also, with combining tests, in comparison with conventional vision less children would need a referral to the clinic for diagnosis [85 (11%) at 3y and 61 (8%) at 3y9m instead of 132 (17%) and 122 (17%), table 1, $P < 0.001$ for both ages].

## Costs per child diagnosed with amblyopia and CER

The estimated costs per child diagnosed with amblyopia and CER are uncertain as the number of children diagnosed with amblyopia per age group is small. At age 3y, estimated costs per child diagnosed with amblyopia are €1500 for conventional screening, €860 for photoscreening and €730 for the combination (table 3). A similar pattern is found at 3y9m (€1050, €420 and €450, respectively), but at

**Table 2** Mean and standard deviation (SD) of time needed (in minutes) for the different screening tests by age group

| | 3 years | | 3 years and 9 months | | 5/6 years | |
|---|---|---|---|---|---|---|
| | Mean | SD | Mean | SD | Mean | SD |
| First conventional vision screening | 5.8 | 2.7 | 5.2 | 2.1 | 3.1 | 1.1 |
| Second conventional vision screening | 6.0 | 2.6 | 6.2 | 2.9 | 4.8 | 1.8 |
| Photoscreening | 2.2 | 1.5 | 2.0 | 1.4 | 1.1 | 0.9 |

Calculations are based on imputed data for time measurements that are lacking.[a]

a: Time measurements were available for 70% of first conventional vision screenings, 22% of second conventional vision screenings and 96% of photoscreening tests.

5/6y this pattern is reversed (€860 for conventional vision screening, €1940 for photoscreening).

At 3y and 3y9m, both costs and number of children with amblyopia diagnosed were less for photoscreening compared with vision screening, resulting in CER estimates of €2840 and €9400 saved per child with amblyopia missed. At 5/6y, photoscreening had higher costs but less children with amblyopia diagnosed, and was thus dominated by conventional vision screening. However, the combination of photoscreening and conventional screening dominated conventional vision screening at 3y and 3y9m, as costs were less while the number of children with amblyopia diagnosed was higher for the combination of tests (table 3).

## Discussion

Main findings of our study are that costs per screened child as well as the costs per detected child with amblyopia are substantially lower for photoscreening than for conventional vision screening for ages 3y and 3y9m, but higher for age 5/6y. Neither conventional vision screening nor photoscreening detected all children with amblyopia. At 3y and 3y9m, far less children needed referral with photoscreening than with conventional screening, but photoscreening also detected less children with amblyopia. Most children with amblyopia that remained undetected with photoscreening had a 'Refer or try again' result and would therefore be detected with a follow-up conventional screening. Combining methods at 3y and 3y9m (photoscreening, followed by conventional vision screening only in children with a 'refer or try again') resulted in detection of almost all known children with amblyopia, and seems to be an interesting scenario, although further investigation is needed to evaluate if these *post hoc* results are valid.

Photoscreeners only detect risk factors for amblyopia (refractive errors and strabismus) rather than lost visual acuity from amblyopia itself. This may be the reason that photoscreening is usually only recommended until an age at which children can participate reliably in optotype-based vision screening.[6,14] However, our findings indicate that with use of photoscreening at 3y and 3y9m similar numbers of children with amblyopia are detected with far fewer unnecessary referrals and at much lower costs, and thus may be preferable to conventional vision screening only. In fact, Groenewoud[15] showed that amblyopia remained undetected despite conventional vision screening at 3y, 3y9m and 5/6y (and other eye examinations at ≤24 months) in 0.6% of Dutch 7-years-olds, i.e. 17% of all cases with amblyopia were missed.

Amblyopia screening costs were €3.73–9.93 per child (table 3), which is less than for some other Dutch screening programmes (e.g. neonatal hearing screening (NHS) by CHC (€20–40[16]), newborn bloodspot screening (NBS, €99 for 20 congenital disorders[17]), €4.82 for adding SCID to NBS[18], all excluding diagnostic costs). Costs per case detected were €420–1500 for amblyopia at 3–4y, while e.g. for NHS estimates were €35 000–60 000 per case,[16] for cystic fibrosis €32 000,[19] and for SCID €419 000.[18] However, to select the most optimal preventive programmes, also future costs and savings need to be known. For example, early detection and treatment of cystic fibrosis may result in savings in treatment costs later in life.[19] Also, effectiveness should be taken into account, preferably by assessing benefits in terms of both quality and duration of life by using quality-adjusted life years. Unfortunately, there is only scarce scientific evidence regarding the utility impacts of amblyopic monocular visual loss,[20–23] limiting comparison of cost-effectiveness of screening for amblyopia with other screening programmes.

The advantage of photoscreeners to prevent amblyopia was already shown by Carlton et al.,[20] who calculated that screening programmes that included autorefraction dominated screening programmes without autorefraction. Their analyses based on the cost per case of amblyopia prevented showed that screening at either 3 or 4 years prevented additional cases at a low absolute cost (£3000–6000 in 2006, i.e. ±€5000–10 000 in 2017). These costs are much higher than our estimates per child diagnosed with amblyopia (table 3). Reasons for this large difference are that part of children diagnosed remain amblyopic despite treatment, and treatment costs were not included in our estimates. For conventional vision screening, costs per child screened in our study are comparable with those in Carlton et al.,[20] but higher than in Rein et al.[21] (US$3–7, i.e. €2–€5). For photoscreening, our cost estimates are less that reported in the literature (€5.35–7.20).[24,25] Reported time measurements in other studies are similar to ours (mean 2.5 min for photoscreening and 5.9 min for conventional vision screening in preschoolers;[26] 1.7 min for autorefraction and 5.4 for conventional vision screening in children aged 3–6y,[27] and table 2). Hence, differences in cost estimates may be explained by differences in hourly rates of health personnel performing the screening.

### Study limitations
### Reliability of cost estimates

For the interpretation of the study results, it should be noticed that the estimated mean costs per screened child are based on a large number of children (600–800 per age group). Differences in costs between screening methods are statistically significant as well as substantial (€3–13 per child screened, table 3). Thus, there is strong evidence that alternative ways of screening reduce costs in children aged 3–4 years, but not at age 5/6 years. Note however that the costs for the photoscreeners depend on the number of children screened per device, which will likely vary per CHC location and age group. In contrast, the cost estimates per child diagnosed with amblyopia and CERs are quite variable, as the number of children diagnosed with amblyopia is small, and only provide a rough indication of these costs in practice.

### Study design

Children who passed both screening tests were not referred, implying that some cases may have been missed. Also, only about 70% of referred cases arrived for diagnosis. Sensitivity could therefore not be determined, but comparison of test methods is possible. Our study is a within-subjects cross-sectional study performed in three age groups. Therefore, we can only compare photoscreening with conventional screening at a certain age. For the choice of an optimal

**Table 3** Screening costs (in 2017 €) by age group

| | 3 years | | | 3 years 9 months | | | 5/6 years | |
|---|---|---|---|---|---|---|---|---|
| | Vision screening | Photo screening | Combination[a] | Vision screening | Photo screening | Combination[a] | Vision screening | Photo screening |
| Costs per child screened (excl. clinical consultation) (SD) | 8.83 (SD 5.48) | 3.92[b] (SD 1.09) | 5.17[c] (SD 4.14) | 9.93 (SD 5.85) | 3.73[b] (SD 1.00) | 4.52[c] (SD 3.54) | 2.75 (SD 1.47) | 5.35[b] (SD 0.68) |
| Costs per child screened (incl. clinical consultation) (SD) | 17.44 (SD 27.16) | 6.61[b] (SD 14.53) | 9.32[c] (SD 19.17) | 20.37 (SD 28.55) | 7.52[b] (SD 17.16) | 9.33[c] (SD 19.85) | 6.90[b] (SD 18.35) | 9.40[b] (SD 17.66) |
| Number with amblyopia diagnosed | 9 | 6 | 10 | 14 | 13 | 15 | 5 | 3 |
| Number with amblyopia missed | 2 | 5 | 1 | 2 | 3 | 1 | 1 | 3 |
| Costs per child diagnosed with amblyopia (incl. clinical consultation) | 1500 | 860 | 730 | 1050 | 420 | 450 | 860 | 1940 |
| **Totals per age group** | | | | | | | | |
| Costs of screening and clinical consultation for whole age group | 13 700 | 5200 | 7300 | 14 900 | 5500 | 6800 | 4300 | 5900 |
| Difference with vision screening: costs | | −8500 | −6400 | | −9400 | −8100 | | 1600 |
| Difference with vision screening: number with amblyopia diagnosed | | −3 | 1 | | −1 | 1 | | −2 |
| Cost-effectiveness ratio compared with vision screening: savings per child with amblyopia missed | | 2840 | Dominant | | 9400 | Dominant | | Dominated |

a: Combination of both ways of screening, investigated *post hoc* for children aged 3y and 3y9m. All children are first screened with the photoscreener, and all children with a 'refer' result are referred to the clinic for diagnosis. Only children with a 'refer or try again' result (i.e. no 'pass' or 'refer' result could be obtained) get a vision chart test.

b: Comparison vision screening and photoscreening:
At 3y, mean difference in costs per screened child (vision screening minus photoscreening) was €4.89 (95% CI 4.44 - 5.34; $t=21.3$, $p<0.001$) excluding clinical consultation and €10.70 (95% CI 8.81-12.60; $t=11.1$, $p<0.001$) including clinical consultation.
At 3y9m, mean difference in costs per screened child (vision screening minus photoscreening) was €6.20 (95% CI 5.65 - 6.74; $t=22.4$, $p<0.001$) excluding clinical consultation and €12.86 (95% CI 10.85 - 14.87; $t=12.5$, $p<0.001$) including clinical consultation.
At 5/6y, mean difference in costs per screened child (vision screening minus photoscreening) was -€2.61 (95% CI -2.77 - -2.45; $t= -31.9$, $p<0.001$) excluding clinical consultation and -€2.47 (95% CI -4.22 - -0.73; $t= -2.7$, $p=0.006$) including clinical consultation.

c: Comparison vision screening and combination:
At 3y, mean difference in costs per screened child (vision screening minus combination) was €3.60 (95% CI 3.13 - 4.09; $t=14.7$, $p<0.001$) excluding clinical consultation and €7.94 (95% CI 6.19 - 9.70, $t=8.9$, $p<0.001$) including clinical consultation.
At 3y9m, mean difference in costs per screened child (vision screening minus combination) was €5.38 (95% CI 4.82 - 5.94; $t=18.9$, $p<0.001$) excluding clinical consultation and €10.92 (95% CI 9.00 - 12.84; $t=11.1$, $p<0.001$) including clinical consultation.
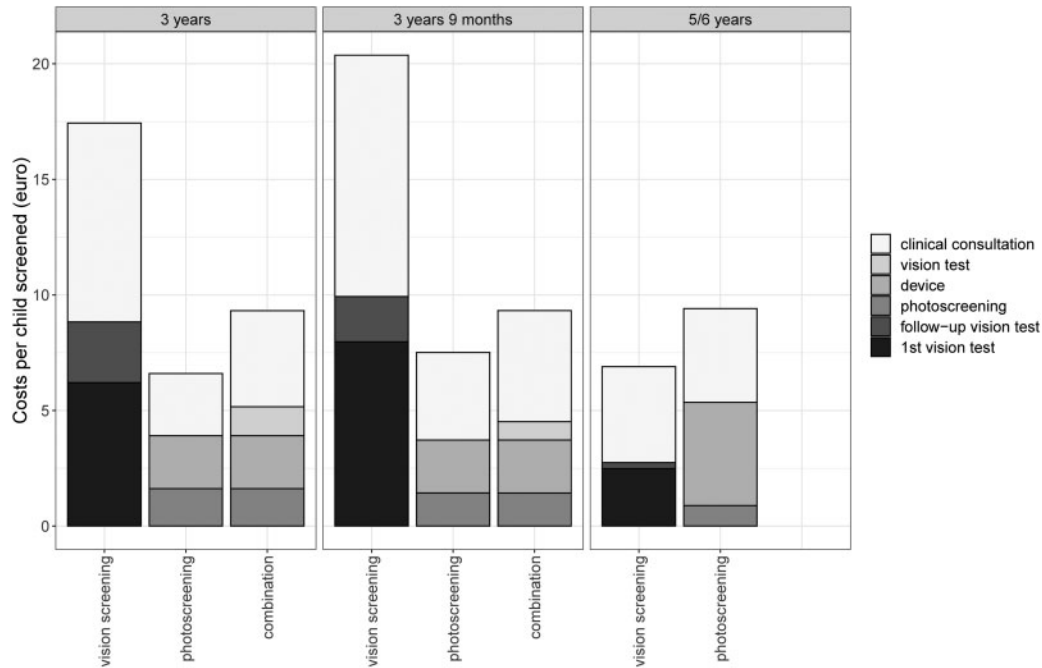
**Figure 1** Mean costs per child screened (in 2017 euro) by age group, split up by cost components (of performing the various tests, device costs and clinical consultation)

screening programme consisting of testing in more than one age group a longitudinal cohort design is necessary.

## Cut-off values

For conventional vision screening as well as photoscreening cut-off values needed to be determined upon which referrals to the clinic were indicated (Supplementary appendix S1). Our results depend on these values. E.g. changing the cut-off for conventional vision screening from 'pass' to 'doubtful' at certain scores will result in more second visits needed and more referrals to the clinic. Similarly, lower cut-off values for photoscreening will also result in more referrals. It would have been interesting to assess at what costs more patients could be detected by changing the cut-off values, but especially because of the small number of children diagnosed with amblyopia in our study, it was not possible to reliably further optimize cut-off values.

## Conclusions

At age 3y and 3y9m photoscreening followed by vision screening if the result was unclear, seems to be more cost-effective than conventional vision screening, whereas at age 5/6y conventional vision screening seems preferable. As the number of study children with amblyopia is small, further large longitudinal studies on the effectiveness of these promising screening alternatives in detecting children with amblyopia are recommended.

## Supplementary data

Supplementary data are available at *EURPUB* online.

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**Key points**

- Vision screening to detect amblyopia or its risk factors is traditionally performed by time-consuming chart tests.
- Photoscreening is quick and requires minimal cooperation of the child.
- Among 2144 children, we found that photoscreening or a combination, i.e. photoscreening followed by vision screening if the result was unclear, resulted in lower costs and less referrals than conventional vision screening at the ages of 3y and 3y9m, whereas conventional screening had better results at 5/6 years.
- The number of children with amblyopia diagnosed was small, but neither conventional nor photoscreening detected all children with amblyopia.
- Photoscreening followed by vision screening if the result was unclear seems to improve detection of amblyopia with less false-positive tests, and seems cost-effective compared with conventional screening in children aged 3–4 years, whereas at 5/6 years conventional screening seems recommendable.

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