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Economic Evaluation of Different Methods of Screening for Amblyopia in Kindergarten

Hans-Helmut König, MD, MPH*, and Jean-Cyriaque Barry, MD‡

ABSTRACT. *Objective.* To compare the cost-effectiveness of 5 methods of screening for untreated amblyopia in kindergarten from a third-party-payer perspective: A) uncorrected monocular visual acuity testing with pass threshold ≥0.5 (20/40) and ≤1 line difference between eyes; B) same as A, but pass threshold ≥0.6 (20/32); C) same as A, plus cover tests and examination of eye motility and head posture; D) same as C, but pass threshold ≥0.6 (20/32); and E) refractive screening without cycloplegia using the Nikon Retinomax autorefractor.

Methods. A decision-analytic model was used with a time horizon until diagnostic examination. According to the model, all 3-year-old children were screened in kindergarten with 1 of the screening methods. Children with positive screening results were referred to an ophthalmologist for diagnostic examination. Children with inconclusive screening results were either referred to an ophthalmologist directly (option 1) or rescreened by the same method after 1 year and referred to an ophthalmologist if rescreening was positive or inconclusive (option 2). Screening test characteristics and costs were estimated on the basis of a field study in which 1180 3-year-old children were examined by orthoptists in 121 German kindergartens.

Results. Compared with methods A option 1 (A-1), B-1, C-1, C-2, E-1, and E-2, there was at least 1 other method that was both less costly and more effective. The average costs per detected case were lowest for method A-2 (878 Euro), followed by methods B-2 (886 Euro), D-2 (908 Euro), and D-1 (965 Euro). When these methods were compared with each other, the additional costs per extra case detected were 1058 Euro (B-2 vs A-2), 1359 Euro (D-2 vs B-2), and 13 448 Euro (D-1 vs D-2).

Conclusions. Monocular visual acuity screening with rescreening of inconclusive results had a favorable cost-effectiveness. By adding additional test items, few more cases could be detected. Because of a great proportion of false-negative, false-positive, and inconclusive results, refractive screening was less effective with an unfavorable cost-effectiveness. *Pediatrics* 2002;109(4). URL: http://www.pediatrics.org/cgi/content/full/109/4/e59; *screening, amblyopia, kindergarten, cost-effectiveness.*

ABBREVIATIONS. ICER, incremental cost-effectiveness ratio; CER, cost-effectiveness ratio.

Preschool vision screening is aimed mainly at detecting amblyopia early enough for treatment to be effective.[1,2] However, there has been concern about the lack of evidence for the effectiveness of preschool vision screening programs, and, consequently, rigorous evaluation of such programs has been called for.[3–6] Furthermore, because scarcity of resources for health care is increasing, there is a growing interest among decision makers in the cost-effectiveness of health care interventions,[7] yet little is known about the cost-effectiveness of preschool vision screening programs.[3,8,9]

In Germany, vision assessment is part of general preventive care examinations performed by general practitioners and pediatricians on children at 21 to 24 months and 42 to 48 months of age. These voluntary examinations are paid for by the social health insurance, which covers approximately 90% of the German population, as well as by private health insurance, which covers almost 10%. Although 71% to 95% of all eligible children participate in these examinations,[10] the effectiveness of vision assessment in detecting amblyopia is considered poor[11,12] because general practitioners and pediatricians lack experience with ophthalmologic tests necessary in these age groups.[13]

Screening by orthoptists has been proposed as one option to improve vision screening.[14,15] Orthoptists are specialized medical aides with a thorough training in infant and child vision assessment, ocular motility disorders examinations, and amblyopia management. In most children, monocular visual acuity can be assessed reliably by simple screening methods from age 3 onward.[16] Therefore and because clinical experience holds that treatment is most effective when administered as early as possible, the fourth year of life is considered best for vision screening programs.[17,18] From this age, children in Germany are entitled by law to be admitted to kindergarten on parents' request. As >50% of children enter kindergarten at age 3 and >80% attend kindergarten at age 4,[19] screening programs conducted in kindergarten provide easy access to children without inconveniencing parents.[20,21]

The purpose of this study was to compare the effectiveness and cost-effectiveness of different methods of screening for amblyopia conducted by orthoptists in kindergarten. The screening methods analyzed were visual acuity testing with and without cover tests and examination of eye motility and head posture, as well as refractive screening using the

Nikon Retinomax autorefractor (Nikon GmbH, Düsseldorf, Germany). The Retinomax is a portable, hand-held, monocular autorefractor that uses a fogging technique (which eases relaxation of accommodation) to measure the objective refraction.[22]

METHODS

Decision-Analytic Model

A decision-analytic model was used (Microsoft Excel spreadsheet download available as freeware for scientists at www.medizin.uni-tuebingen.de/~jcbarry/goe.html). According to the model, all 3-year-old children were screened for untreated amblyopia in kindergarten by 1 of the screening methods to be compared. Children with positive screening results were referred to an ophthalmologist for diagnosis. With respect to children with inconclusive screening results, 2 model options were distinguished: in option 1, children with inconclusive screening results were referred to an ophthalmologist directly (Fig 1); in option 2, children with inconclusive results were rescreened by the same method after 1 year and referred to an ophthalmologist if rescreening was positive or inconclusive (Fig 2).

The effects of screening were defined as the number of newly detected cases of untreated amblyopia. The costs of the screening program were defined as labor and material costs of screening examinations as well as costs of diagnostic ophthalmologic examinations resulting from referral. They were estimated from the perspective of a third-party payer in Euro for the year 2000. In that year, the average Euro to US dollar exchange rate was 0.92 US dollar per 1 Euro, and the average purchasing power adjusted conversion rate was 0.99 US dollar per 1 Euro, ie, close to parity.[23]

Screening Methods

Five different screening methods were compared (visual acuity thresholds are reported in usual 20/X notation; however, the test distance used in the study was 10 ft, not 20 ft):

A. Uncorrected monocular visual acuity testing using the Lea single optotype test (required test distance 10 ft [3 m]; Precision Vision, Villa Park, IL) with pass threshold set at ≥0.8 (20/25) monocular visual acuity in both eyes, or ≥0.5 (20/40) in both eyes and ≤1 line difference between the visual acuity of the right and left eye (L. Hyvärinen, Lea-Test Ltd, Helsinki, Finland, personal communication, 1998).
B. Uncorrected monocular visual acuity testing as described in A with pass threshold set at ≥0.8 (20/25) monocular visual acuity in both eyes, or ≥0.6 (20/32) in both eyes and ≤1 line difference between the visual acuity of the right and left eye.
C. Uncorrected monocular visual acuity testing as described in A plus cover tests and examination of eye motility and head posture.
D. Uncorrected monocular visual acuity testing as described in B plus cover tests and examination of eye motility and head posture.
E. Refractive screening without cycloplegia using the Nikon Retinomax autorefractor operated in the normal mode with pass

threshold as follows: spherical equivalent ≥−1D and ≤3D, cylindric power ≤1.5D, and spherical equivalent anisometropia ≤1D.

Screening results were classified as positive (when any screening item was abnormal), negative (when all screening items were within normal limits), or inconclusive (when cooperation was insufficient).

For each screening method, the 2 model options were analyzed, resulting in 10 method-option combinations labeled A-1 (for visual acuity screening A in model option 1) through E-2 (for refractive screening E in model option 2).

Model Parameters

The parameter values used in the decision-analytic model are shown in Table 1.

Prevalence and Test Characteristics

Data on screening test characteristics (sensitivity, specificity, proportion of inconclusive results), on the prevalence of untreated amblyopia, on the participation in rescreening, and on the compliance with ophthalmologic examinations were obtained from a field study in which 1180 3-year-old children were examined by orthoptists in 121 German kindergartens.[24] Of these children, 427 were also screened with the Nikon Retinomax autorefractor.[22] In the field study, a gold standard was established for 1114 children. The ophthalmologic criteria for a positive gold standard were any newly administered patching therapy or any newly administered spectacle therapy, if the visual acuity was ≤0.4 (20/50) in either eye, or if the difference of visual acuity between the right and the left eye was ≥3 lines.

Costs of Screening Examinations

The costs of screening examinations were estimated on the basis of results of the field study in which the costs of orthoptic screening and device-based screening were calculated on the basis of comprehensive and detailed measurement of working time, material costs, and travel costs.[25] Working time was divided into office time for organizing the screening program (organization time) and time for visiting the kindergartens. The latter was subdivided into travel time, time for preparing the examination site in the kindergarten, and examination time. Material costs included costs for orthoptic materials, stationery, parent information leaflets, postage, and telephone. Estimates of the costs of the screening methods analyzed in this study were derived as follows: costs of methods C and D were assumed to be equal to the costs of the orthoptic examination calculated in the field study. For visual acuity testing (methods A and B), it was assumed that examination time would be 25% less than for the orthoptic examination, all other working times being equal. For the autorefractor (method D), it was assumed that examination time was equal to what was measured for device-based screening, all other working times being equal to the other screening methods analyzed. Furthermore, investment costs and maintenance costs of the Nikon Retinomax autorefractor were included in the cost calculation. For calculating labor costs, working time was valued at 23.31 Euro/h
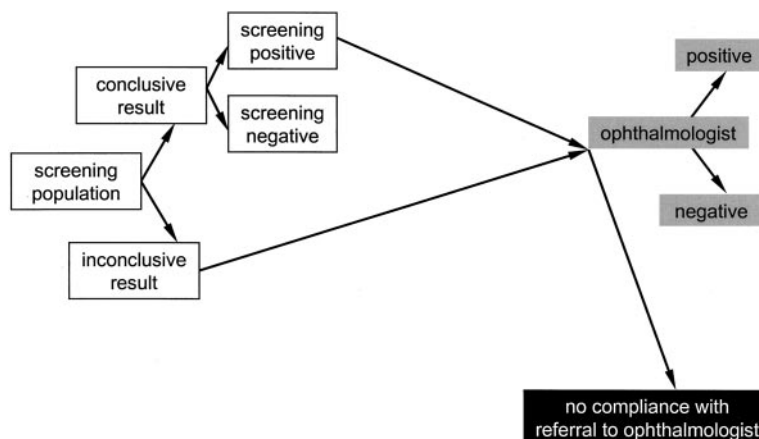

Fig 1. Screening model option 1.
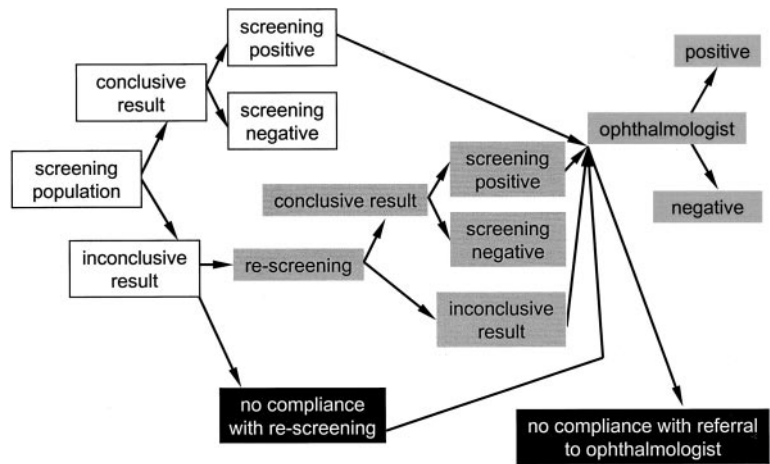
Fig 2. Screening model option 2.

TABLE 1. Parameter Values Used in Model

Parameter	Visual Acuity Test		Visual Acuity Test/Cover Tests/Motility Test		Autorefractor
	VA Threshold ≥0.5	VA Threshold ≥0.6	VA Threshold ≥0.5	VA Threshold ≥0.6	
Screening population					
Prevalence of untreated amblyopia	-- 2.2%* ---------------------				
Participation in rescreening (option 2)	-- 78.2% ---------------------				
Compliance with referral to ophthalmologist					
After positive screening	-- 97.3% ---------------------				
After inconclusive screening (option 1)	-- 90.0% ---------------------				
After lacking rescreening (option 2)	-- 79.3% ---------------------				
After inconclusive rescreening (option 2)	-- 64.7% ---------------------				
Screening test characteristics					
Sensitivity	86.4%	90.9%	90.9%	95.5%	77.8%
Specificity	94.8%	91.9%	92.4%	90.9%	84.4%
Inconclusive screening results	--------- 11.2% ---------		---------11.3% ---------		31.1%
Inconclusive rescreening results (option 2)	--------- 15.4% ---------		---------16.3% ---------		31.1%
Costs per single examination					
Screening test	------ 11.79 Euro ------		------ 12.58 Euro -------		12.49 Euro
Ophthalmologic examination	---36.40 Euro ---------------------				

VA indicates visual acuity; dashed line indicates that number applies to all respective columns.
* The prevalence measured in the field study among 1114 children with gold standard was 26/1114 = 2.3%. For a conservative estimate, a prevalence of 2.2% was used, assuming that there were no additional cases among those children for whom no gold standard could be obtained.

(0.39 Euro/min). This amount corresponds to the tariff class Vb of the German Federal Employee Tariff (Bundesangestelltentarif), which applies to salaried orthoptists working in the public health sector in the year 2000 and includes all ancillary wage costs.

Costs of Ophthalmologic Examinations

Costs of ophthalmologic examinations were calculated to be 36.40 Euro, based on the German social health insurance's relative value scale for outpatient physician services (Einheitlicher Bewertungsmaβstab für Ärzte[26]), which states point volumes for each service. A conversion factor (point value) of 0.041 Euro was used. For those covered by the social health insurance, there is no copayment for physician services.

Effectiveness and Cost-Effectiveness Ratios

Effectiveness was defined as the proportion of detected cases of untreated amblyopia in all cases of untreated amblyopia among the participating children. Average cost-effectiveness ratios were calculated for each screening method by dividing the total costs (for screening tests and ophthalmologic examinations) of the respective screening program by the number of newly detected cases of untreated amblyopia. Incremental cost-effectiveness ratios (ICER) were calculated by dividing the additional costs of a screening method compared with another screening method by the additional units of effects of this screening method compared with the other screening method: ICER = (costs$_{method\ X}$ − costs$_{method\ Y}$)/(effects$_{method\ X}$ − effects$_{method\ Y}$).

Sensitivity Analysis

Sensitivity analysis was performed to test the influence of uncertainty in screening method–specific model parameters on the cost-effectiveness. For this purpose, screening method–specific parameter values for sensitivity and specificity were varied according to their 95% confidence intervals found in the field study. For the parameter "costs per single screening test," the baseline value was decreased and increased by 25% each.

RESULTS

Costs of Screening Examinations

The costs per single screening examination were similar for the screening methods analyzed ranging from 11.79 Euro for visual acuity testing (methods A and B) to 12.58 Euro for visual acuity testing plus cover tests and examination of eye motility and head posture (methods C and D; Table 2). Labor was the most important resource used, accounting for approximately 80% of total costs. Time spent on organization accounted for approximately 50% of labor

TABLE 2. Costs of Screening Tests

Resource	Costs per Unit (a)	Visual Acuity Test			Visual Acuity Test/Cover Tests/Motility Test			Autorefractor		
		Quantity per Kindergarten (b)	Quantity per Examination (c = b/9.88)	Costs per Examination (a*c)	Quantity per Kindergarten (d)	Quantity per Examination (e = d/9.88)	Costs per Examination (a*e)	Quantity per Kindergarten (f)	Quantity per Examination (g = f/9.88)	Costs per Examination (a*g)
Labor										
Organization	0.39 Euro/min	127 min	12.9 min	5.00 Euro	127 min	12.9 min	5.00 Euro	127 min	12.9 min	5.00 Euro
Travel	0.39 Euro/min	45 min	4.6 min	1.78 Euro	45 min	4.6 min	1.78 Euro	45 min	4.6 min	1.78 Euro
Site preparation	0.39 Euro/min	28 min	2.8 min	1.10 Euro	28 min	2.8 min	1.10 Euro	28 min	2.8 min	1.10 Euro
Examination	0.39 Euro/min	59 min	6.0 min	2.33 Euro	79 min	8.0 min	3.11 Euro	61 min	6.2 min	2.40 Euro
Total labor		260 min	26.3 min	10.20 Euro	279 min	28.3 min	10.99 Euro	262 min	26.5 min	10.28 Euro
Materials				1.04 Euro*			1.06 Euro*			1.66 Euro†
Travel	0.27 Euro/km	20 km	2.0 km	0.54 Euro	20 km	2.0 km	0.54 Euro	20 km	2.0 km	0.54 Euro
TOTAL				11.79 Euro			12.58 Euro			12.48 Euro

* Includes patch that costs 0.48 Euro per piece.
† Includes investment costs for Retinomax of 0.78 Euro per examination and maintenance costs of 0.35 Euro per examination on the basis of the following data: costs of Retinomax including transportation box 11 510 Euro; useful life 5 years; maintenance costs 1023 Euro per year; 300 visits to kindergarten per year; 9.88 examinations per kindergarten.

time. Only approximately one quarter of labor costs was caused by the examination itself, which on average lasted between 6 minutes (methods A and B) and 8 minutes (methods C and D) per child.

Effectiveness

The effectiveness of screening ranged from 75.1% (autorefractor in model option 2) to 92.5% (visual acuity testing plus cover tests and examination of eye motility and head posture in model option 1; Table 3).

Cost-Effectiveness

The average costs per newly detected case of untreated amblyopia ranged from 878 Euro for visual acuity testing in model option 2 (A-2) to 1514 Euro for refractive screening in model option 1 (E-1). Because of the comparatively low specificity and high rate of inconclusive results of the autorefractor, a large proportion of children were referred to an ophthalmologist after refractive screening, causing additional costs: 39.4% in option 1 and 24.4% in option 2. For the other screening methods, the respective proportion ranged between 9.4% (visual acuity testing in option 2 [A-2]) and 19.7% (visual acuity testing plus cover tests and examination of eye motility and head posture in model option 1 [D-1]).

Figure 3 illustrates for a screening population of 1180 children that for 6 of the analyzed method-option combinations (A-1, B-1, C-1, C-2, E-1, E-2), there was at least 1 other option associated with both lower costs and greater effects. Thus, these 6 method-option combinations were so-called "dominated" and could be excluded from additional analysis. Among the remaining method-option combinations, visual acuity testing in option 2 (A-2) had the best average cost-effectiveness ratio of 876 Euro per detected case. By increasing the pass threshold for monocular visual acuity to $\geq$0.6 (20/32) in method B-2, additional costs of 1176 Euro would arise and 1.11 calculated additional cases would be detected. The ICER of B-2 versus A-2 would be 1058 Euro per additional case detected (1176 Euro/1.11), which is reflected by the slope of the line between A-2 and B-2 in Fig 3. Analogously, the ICER of D-2 versus B-2 and of D-1 versus D-2 would be 1359 Euro and 13 448 Euro, respectively.

Sensitivity Analysis

Variation of screening method–specific model parameters for visual acuity testing with and without cover tests and examination of eye motility and head posture had moderate influence on cost-effectiveness ratios (CERs). The resulting intervals for average CERs were similar for these screening methods, ranging from 705 Euro to 1100 Euro per detected case in model option 2 (Fig 4). For refractive screening (method D), the respective intervals ranged from 1237 Euro to 2053 Euro. Thus, the costs per detected case were always higher for refractive screening than for the other screening methods analyzed. Because of the wide confidence interval of the sensitivity of refractive screening, which was based on gold stan-

TABLE 3. Effectiveness and Average CER

Screening Test	Model Option	Effectiveness*	Proportion of Children Receiving Ophthalmologic Examination After Screening	Average CER (Euro/Case)
A. Visual acuity test (VA threshold ≥0.5)	1	84.7%	16.2%	943
	2	83.6%	9.4%	878
B. Visual acuity test (VA threshold ≥0.6)	1	88.6%	18.7%	948
	2	87.9%	12.1%	886
C. Visual acuity test/cover tests/motility test (VA threshold ≥0.5)	1	88.6%	18.3%	982
	2	87.8%	11.7%	924
D. Visual acuity test/cover tests/motility test (VA threshold ≥0.6)	1	92.5%	19.7%	965
	2	92.1%	13.2%	908
E. Autorefractor	1	80.1%	39.4%	1514
	2	75.1%	24.4%	1471

VA indicates visual acuity.

* Proportion of newly detected cases of untreated amblyopia in all cases of untreated amblyopia among participating children.

dards in 404 children, the uncertainty with respect to sensitivity had substantial influence on the CER.

DISCUSSION

The decision-analytic model showed similar CERs of visual acuity testing with and without cover tests and examination of eye motility and head posture and for both pass thresholds for visual acuity analyzed. The average CER was approximately 900 Euro per detected case when children with inconclusive screening results were rescreened after 1 year (option 2). The effectiveness of screening could be increased slightly by using the more demanding pass threshold for visual acuity of ≥0.6 (20/32) instead of ≥0.5 (20/40) and by adding the cover tests and examination of eye motility and head posture. Both changes caused more children to screen positive, among whom were both true positives (which increased sensitivity) and false positives (which decreased specificity). The increase in effectiveness resulting from these changes was approximately 4% each.

Refractive screening led to many false-positive and inconclusive results, thus inducing additional ophthalmologic examinations that resulted in an unfavorable cost-effectiveness. Furthermore, the comparatively low sensitivity of refractive screening brought about the lowest effectiveness of the screening methods compared.

For all screening methods, it was more cost-effective to rescreen children with inconclusive results than to refer them to an ophthalmologist directly. It may be sufficient to perform rescreening after 1 year because treatment started in the fifth year of life still is considered to be effective.[27]

Incremental analysis showed that only 4 method-option combinations were not dominated by other combinations, which means that compared with these 4 combinations, no other combinations were both less costly and more effective and thus clearly preferable. When these 4 combinations were compared, ICERs ranged from 1058 Euro to more than 13 000 Euro per additional case detected. On the basis of this information, a decision maker may
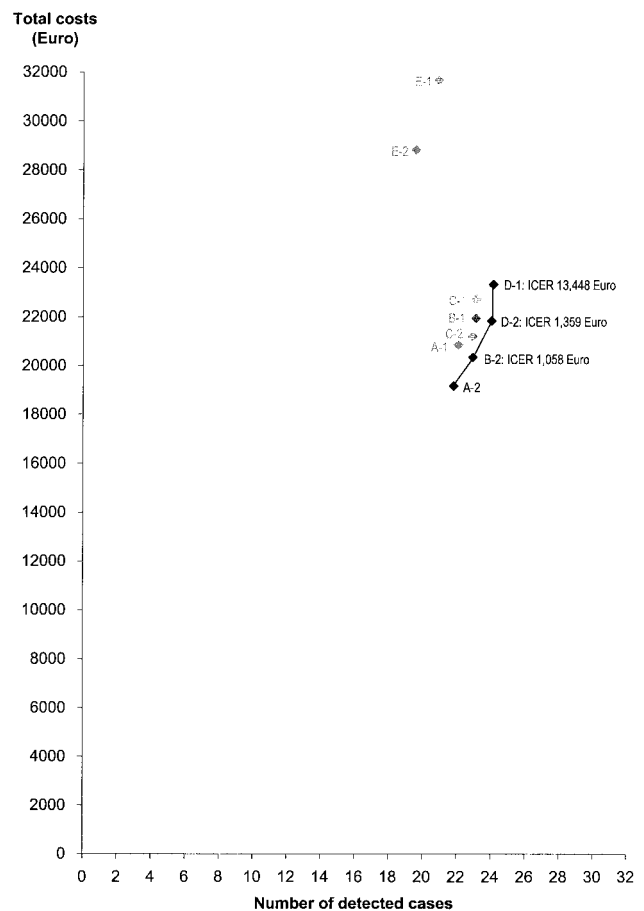

Fig 3. Cost-effectiveness of screening methods. VA, visual acuity. Results refer to a screening population of 1180 3-year-old children. A-1, visual acuity test (VA threshold ≥0.5) model option 1; A-2, visual acuity test (VA threshold ≥0.5) model option 2; B-1, visual acuity test (VA threshold ≥0.6) model option 1; B-2, visual acuity test (VA threshold ≥0.6) model option 2; C-1, visual acuity test/ cover tests/motility test (VA threshold ≥0.5) model option 1; C-2, visual acuity test/cover tests/motility test (VA threshold ≥0.5) model option 2; D-1, visual acuity test/cover tests/motility test (VA threshold ≥0.6) model option 1; D-2, visual acuity test/cover tests/motility test (VA threshold ≥0.6) model option 2; E-1, autorefractor model option 1; E-2, autorefractor model option 2. Screening methods in gray are dominated by other screening methods. Slope of lines reflects ICER.
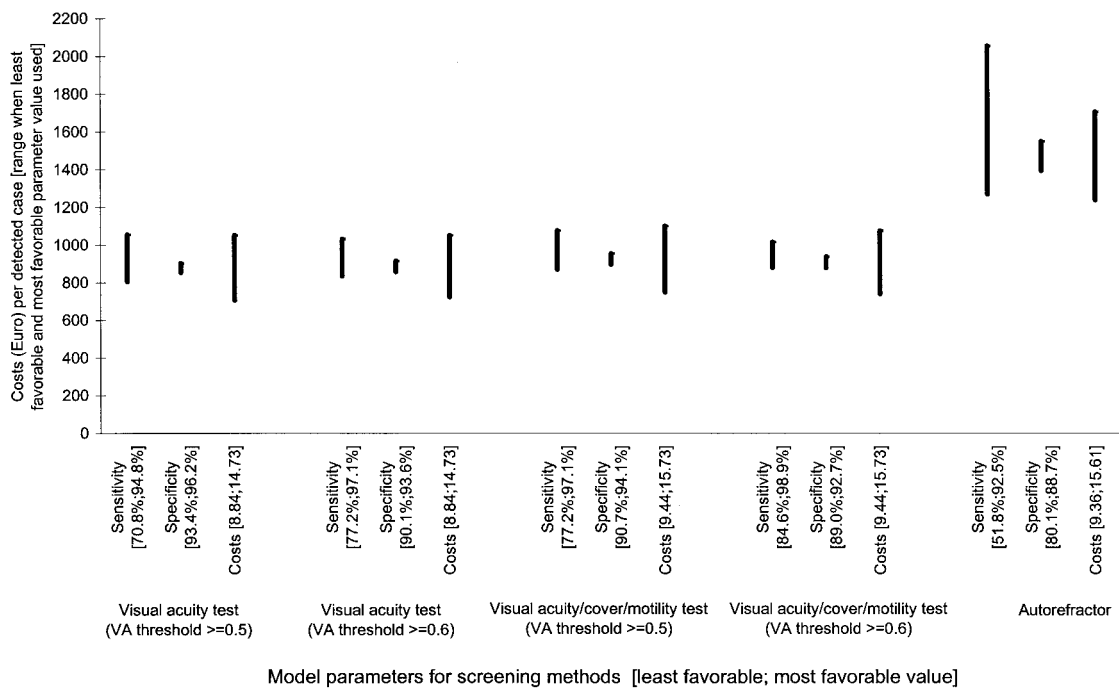
Fig 4. Results of sensitivity analysis for screening method–specific parameters in model option 2.

choose the most suitable method-option combination taking into account the maximum limit he or she is willing to spend to detect an additional case.

Sensitivity analysis of screening method–specific parameters showed reasonably stable results. Only uncertainty with respect to the sensitivity of refractive screening had substantial influence on the CER, yet even when the most favorable parameter value was used, refractive screening was less cost-effective than all other methods analyzed.

It is important to point out that the average CER is almost inversely proportional to the prevalence rate of untreated amblyopia in the screening population. If, for example, in a different population the prevalence were twice as high as assumed in the model, then the average CERs for all screening methods would approximately be cut by half. Interested readers may test the influence of the prevalence rate as well as other model parameters on the CER by using a Microsoft Excel spreadsheet provided as freeware by the authors (download available as freeware for scientists at www.medizin.uni-tuebingen.de/~jcbarry/goe.html).

The data on screening test characteristics used in the decision-analytic model were obtained from a field study in which screening was performed by orthoptists. The most important screening test item was visual acuity testing. The high sensitivity and specificity of visual acuity testing was most likely attributable to the orthoptists' training and experience in vision assessment of children. In many European countries, such as Germany and Great Britain, orthoptists are trained in sufficient numbers, and thus an orthoptic screening program could probably be implemented easily. In other countries, such as the United States, orthoptists may not be available, which raises the question of whether screening personnel without orthoptic training could achieve similar results. In Sweden, visual acuity screening has been performed successfully by nurses specialized in screening children,[27] and similar screening test characteristics have been reported.[2,28] The authors know from experience that nurses and other medical aides can perform visual acuity testing according to ophthalmologic standards in preschool children provided that they have received adequate training and perform visual acuity testing routinely and frequently. Thus, it can be expected that visual acuity testing performed by trained personnel other than orthoptists may be similarly effective and cost-effective.

Other visual acuity tests with standardized optotypes suitable for children[5] are likely to yield similar results if used strictly according to their manuals. The authors chose the Lea single optotype test because it combines high testability, high sensitivity for amblyopia, and reduced test time compared with line tests.[29] However, if other tests are used, then the thresholds may have to be adjusted to achieve similar test characteristics.

In this study, costs were calculated from a third-party-payer perspective on the basis of resource prices from the German health care system. Because there was no copayment by the parents, neither for the screening nor for the diagnostic ophthalmologic examinations, calculated costs include all medical costs. Obviously, resource prices may be different in other countries, such as the United States. The use of gross domestic product purchasing power parities for currency conversion adjusts for differences in the general price level of different economies. In 2000, the purchasing power of 1 Euro in Germany was almost equal to the purchasing power of 1 US dollar in the United States. However, gross domestic product purchasing power parities do not account for differences in relative prices of health care compared

with the prices of all goods and services. According to the most recent data available for 1996,[23] relative prices of health care in the United States were approximately one third higher than in Germany, which could cause cost figures for the United States to be higher in this order of magnitude.

The CER of visual acuity screening in kindergarten with and without cover tests and examination of eye motility and head posture found in the study falls within a range likely to be acceptable for decision makers. Because visual acuity testing is a simple and accurate screening method, comparable results can be expected in other health care systems provided that access to children is easy and that the prevalence of untreated amblyopia is not fundamentally different.

Like many published cost-effectiveness analyses of screening programs and diagnostic tests in other areas of medicine, this study considered only costs and effects that occurred up the clinical endpoint "diagnosis." It provides information on how efficient different methods are in achieving the target of early detection of amblyopia. However, diagnosis of amblyopia is only an intermediate target. A more comprehensive evaluation of vision screening would also take into account the cost-effectiveness of subsequent treatment. Such an analysis would have to compare the costs and long-term effects, including the impact on quality of life, associated with treated and untreated amblyopia. At this time, such a comprehensive evaluation is not possible because sufficient data on the effectiveness of treatment and on the disability caused by amblyopia are not available.[30]

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