

List of articles published on economics of Childhood Blindness

1. König HH, Barry JC, Leidl R, Zrenner E. Economic evaluation of orthoptic screening: results of a field study in 121 German kindergartens. *Invest Ophthalmol Vis Sci*. 2002 Oct; 43(10):3209-15.
2. König HH, Barry JC. Economic evaluation of different methods of screening for amblyopia in kindergarten. *Pediatrics*. 2002 Apr;109 (4):e59.
3. Schlichtherle S, Gandjour A, Neugebauer A, Russmann W, Lauterbach KW. The cost-effectiveness of screening strategies for amblyopia: a preliminary report. *Strabismus*. 2000 Dec; 8(4):291-5.
4. König HH, Barry JC, Leidl R, Zrenner E. Cost effectiveness of mass orthoptic screening in kindergarten for early detection of developmental vision disorders. *Gesundheitswesen*. 2000 Apr; 62(4):196-206.
5. Beauchamp GR, Bane MC, Stager DR, Berry PM, Wright WW. A value analysis model applied to the management of amblyopia. *Trans Am Ophthalmol Soc*. 1999; 97:349-67; discussion 367-72.
6. Snowdon SK, Stewart-Brown SL. Preschool vision screening. *Health Technol Assess*. 1997;1(8):i-iv, 1-83.
7. Simonsz HJ, Grosklauser B, Leuppi S. Costs and methods of preventive visual screening and the relation between esotropia and increasing hypermetropia. *Doc Ophthalmol*. 1992; 82(1-2):81-7.
8. König HH, Barry JC, Leidl R, Zrenner E. Cost-effectiveness of orthoptic screening in kindergarten: a decision-analytic model. *Strabismus*. 2000 Jun; 8(2):79-90.
9. Gandjour A, Schlichtherle S, Neugebauer A, Russmann W, Lauterbach KW. A cost-effectiveness model of screening strategies for amblyopia and risk factors and its application in a German setting. *Optom Vis Sci*. 2003 Mar; 80(3):259-69.
10. König HH, Walter HS, Barry JC. Resource utilisation and cost of amblyopia treatment. *Klin Monatsbl Augenheilkd*. 2003 Jul; 220(7):486-91.
11. König HH, Barry JC. Cost effectiveness of treatment for amblyopia: an analysis based on a probabilistic Markov model. *Br J Ophthalmol*. 2004 May; 88(5):606-12.

12. Konig HH, Barry JC. Cost-utility analysis of orthoptic screening in kindergarten: a Markov model based on data from Germany. *Pediatrics*. 2004 Feb;113 (2):e95-108.
13. Membreno JH, Brown MM, Brown GC, Sharma S, Beauchamp GR. A cost-utility analysis of therapy for amblyopia. *Ophthalmology*. 2002 Dec: 109 (12): 2265-71

ABSTRACTS:

1. Invest Ophthalmol Vis Sci. 2002 Oct;43(10):3209-15.

Economic evaluation of orthoptic screening: results of a field study in 121 German kindergartens.

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PURPOSE: The purpose of this study was to analyze the cost-effectiveness of an orthoptic screening program in kindergarten children.

METHODS: An empiric cost-effectiveness analysis was conducted as part of a field study of orthoptic screening. Three-year-old children (n = 1180) in 121 German kindergartens were screened by orthoptists. The number of newly diagnosed cases of amblyopia and amblyogenic factors (target conditions) was used as the measure of effectiveness. The direct costs of orthoptic screening were calculated from a third-party-payer perspective based on comprehensive measurement of working hours and material costs.

RESULTS: The average cost of a single orthoptic screening examination was 12.58 Euro. This amount consisted of labor costs (10.99 Euro) and costs of materials and traveling (1.60 Euro). With 9.9 children screened on average per kindergarten, average labor time was 279 minutes per kindergarten, or 28 minutes per child. It consisted of time for organization (46%), traveling (16%), preparing the examination site (10%), and the orthoptic examination itself (28%). The total cost of the screening program in all 121 kindergartens (including ophthalmic examination, if required) was 21,253 Euro. Twenty-three new cases of the target conditions were detected. The cost-effectiveness ratio was 924 Euro per detected case. Sensitivity analysis showed that the prevalence and the specificity of orthoptic screening had substantial influence on the cost-effectiveness ratio.

CONCLUSIONS: The data on the cost-effectiveness of orthoptic screening in kindergarten may be used by such third-party payers as health insurance or public

health services when deciding about organizing and financing preschool vision-screening programs.

2. Pediatrics. 2002 Apr;109(4):e59.

Economic evaluation of different methods of screening for amblyopia in kindergarten

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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 \geq or $=0.5$ (20/40) and $<$ or $=1$ line difference between eyes; B) same as A, but pass threshold \geq or $=0.6$ (20/32); C) same as A, plus cover tests and examination of eye motility and head posture; D) same as C, but pass threshold \geq or $=0.6$ (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.

3. Strabismus. 2000 Dec;8(4):291-5

The cost-effectiveness of screening strategies for amblyopia: a preliminary report.

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PURPOSE: Five screening strategies for amblyopia in different age groups were compared according to a decision-analytical model from the perspective of the health insurance funds. Our findings indicate that the costs per detected case of amblyopia range from about 1200 DM to 3000 DM (613 Euro to 1534 Euro). The two most cost-effective screening strategies were to screen high-risk children up to the age of one by ophthalmologists and to screen all children up to the age of one by ophthalmologists. The screening of high-risk children identifies only about a third of all affected children in this age group, when compared with the number of cases detected by screening all children up to the age of one. However, the average cost per detected case of amblyopia among high-risk children is lower than the cost of screening all children in this age range.

4. Gesundheitswesen. 2000 Apr;62(4):196-206

Cost effectiveness of mass orthoptic screening in kindergarten for early detection of developmental vision disorders [Article in German]

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Orthoptic screening in the kindergarten is one option to improve early detection of amblyopia in children aged 3 years. The purpose of this study was to analyse the cost-effectiveness of such a screening programme in Germany.

METHODS: Based on data from the literature and own experience gained from orthoptic screening in kindergarten a decision-analytic model was developed. According to the model, all children in kindergarten, aged 3 years, who had not been treated for amblyopia before, were subjected to an orthoptic examination. Non-cooperative children were reexamined in kindergarten after one year. Children with positive test results were examined by an ophthalmologist for diagnosis. Effects were measured by the number of newly diagnosed cases of amblyopia, non-obvious strabismus and amblyogenic refractive errors. Direct costs were estimated from a third-party payer perspective. The influence of uncertain model parameters was tested by sensitivity analysis.

RESULTS: In the base analysis the cost per orthoptic screening test was DM 15.39. Examination by an ophthalmologist cost DM 71.20. The total cost of the screening programme in all German kindergartens was DM 6.1 million. With a 1.5% age-specific prevalence of undiagnosed cases, a sensitivity of 95% and a specificity of 98%, a total of 4,261 new cases would be detected. The cost-effectiveness ratio was DM 1,421 per case detected. Sensitivity analysis showed considerable influence of prevalence and specificity on the cost-effectiveness ratio. It was more cost-effective to re-screen non-cooperative children in kindergarten than to have them examined by an ophthalmologist straight-away.

CONCLUSIONS: The decision-analytic model showed stable results which may serve as a basis for discussion on the implementation of orthoptic screening and for planning a field study.

5: Trans Am Ophthalmol Soc. 1999;97:349-67; discussion 367-72

A value analysis model applied to the management of amblyopia

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PURPOSE: To assess the value of amblyopia-related services by utilizing a health value model (HVM). Cost and quality criteria are evaluated in accordance with the interests of patients, physicians, and purchasers.

METHODS: We applied an HVM to a hypothetical statistical ("median") child with amblyopia whose visual acuity is 20/80 and to a group of children with amblyopia who are managed by our practice. We applied the model to calculate the value of these services by evaluating the responses of patients and physicians and relating these responses to clinical outcomes.

RESULTS: The consensus value of care for the hypothetical median child was calculated to be 0.406 (of 1.000). For those children managed in our practice, the calculated value is 0.682. Clinically, 79% achieved 20/40 or better visual acuity, and the mean final visual acuity was 0.2 logMAR (20/32). Value appraisals revealed significant concerns about the financial aspects of amblyopia-related services, particularly among physicians. Patients rated services more positively than did physicians.

CONCLUSIONS: Amblyopia care is difficult, sustained, and important work that requires substantial sensitivity to and support of children and families. Compliance and early detection are essential to success. The value of amblyopia services is rated significantly higher by patients than by physicians. Relative to the measured value, amblyopia care is undercompensated. The HVM is useful to appraise clinical service delivery and its variation. The costs of failure and the benefits of success are high; high-value amblyopia care yields substantial dividends and should be commensurately compensated in the marketplace.

Preschool vision screening

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OBJECTIVES: To undertake a systematic review of the effectiveness of preschool vision screening. To provide evidence on which decisions about the future provision of this service can be made. To indicate areas for further research.

STUDY SELECTION: The Centre for Reviews and Dissemination guidelines for systematic reviews were used. The research questions were formulated using the Wilson and Jungner criteria for evaluating screening programmes. They concerned prevalence, natural history, disability, treatment and screening in relation to three target conditions: amblyopia, refractive errors and squints which are not cosmetically obvious. Studies were considered for inclusion according to pre-determined criteria for the age group studied, the outcomes measured and the study design. The following types of study design were considered: cross-sectional studies of prevalence, cohort studies of natural history, any type of study (e.g., cross-sectional surveys, case-series, qualitative studies) of disability attributable to a target condition, controlled trials, observational studies and audits of screening programmes, and prospective controlled trials of treatment.

DATA SOURCES: The following electronic databases were searched: Biological Abstracts, CINAHL, Embase, ERIC, IAC Health Periodicals, IAPV, Medline, Psychlit, Science Citation Index, System for Information on Grey Literature in Europe, DHSS-Data, Faculty of Public Health Medicine Database of Dissertations, Index of Scientific and Technical Proceedings, Dissertation Abstracts, Index of Theses, NHS Research Register, Public Health Information Sharing Database. A limited amount of handsearching was undertaken. Reference lists were scanned to identify other relevant studies, and requests for unpublished data were made to people working in the field.

DATA EXTRACTION: Data was extracted by the first author and then checked by the second.

DATA SYNTHESIS: Quantitative analysis was undertaken where possible. Qualitative analysis was performed where studies were too heterogeneous for the data

to be combined, or for research questions that were not suitable for quantitative synthesis.

RESEARCH FINDINGS: The electronic search yielded over 5000 references, and over 500 abstracts were downloaded from the databases for further scrutiny. A total of 85 studies were included in the main analysis.

PREVALENCE: No studies were found with the primary aim of establishing the prevalence of visual defects in preschool children. Data from studies of screening programmes report a range of yields for all the target conditions combined of 2.4-6.1%.

NATURAL HISTORY: No studies designed with the intention of documenting the natural history of the target conditions in children aged 3 or 4 years were found. Other studies that provide some natural history data suggest that mild degrees of amblyopia may resolve spontaneously. In the absence of information about natural history it is impossible to estimate the effect of treatment from studies without a control group that was not treated.

DISABILITY: A total of 21 studies exploring disability in relation to the target conditions were included. The literature provides a reasonable basis for generating plausible hypotheses about the ways in which the target conditions might disable people, but is insufficient to draw any firm conclusions about their impact on quality of life. The research to date is not sufficient to determine appropriate outcomes for controlled trials of treatment.

TREATMENT: Five randomised controlled trials of treatment and six prospective controlled trials without randomisation were found. No studies compared treatment with no treatment. Most of the studies were methodologically flawed.(ABSTRACT TRUNCATED)

7. Doc Ophthalmol. 1992;82(1-2):81-7

Costs and methods of preventive visual screening and the relation between esotropia and increasing hypermetropia

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Atkinson has shown that early correction of hypermetropia reduces the incidence of esotropia. If esotropia is reduced by prescribing glasses early, the rate of esotropia-induced amblyopia can be similarly reduced; this would have important economic consequences. We have studied (1) how costs compare to benefits in early visual screening, (2) how videorefraction as used by Atkinson compares to retinoscopy, and (3) whether esotropia is more likely to occur in children who have increasing as opposed to decreasing hypermetropia. The costs of the study so far have been high. It was exceedingly difficult to get all infants invited, come to the clinic and examined. Videorefraction did not compare favourably with retinoscopy in terms of costs and precision, whereas the amount of skill and time needed was approximately equal. The third question, whether esotropia is more likely to occur in children who have increasing as opposed to decreasing hypermetropia, arose from the controversy whether, in the general population, refraction increases or decreases during the first years of life. We found that papers reporting a decrease of hypermetropia in early childhood were studies of large cross-sections of the general population, whereas papers that reported an initial increase originated from ophthalmological practices or strabismus departments. These conflicting results could be reconciled by assuming a population bias: if esotropia is more likely to occur in children with increasing hypermetropia, children with increasing hypermetropia will preferentially be seen by ophthalmologists. It seems natural that children with increasing hypermetropia are more likely to squint, because additional accommodation, needed to overcome increasing hypermetropia, will inevitably confer additional convergence. This relationship has meanwhile been confirmed by others.

8. Strabismus. 2000 Jun;8(2):79-90

Cost-effectiveness of orthoptic screening in kindergarten: a decision-analytic model

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PURPOSE: The purpose of this study was to analyze the cost-effectiveness of orthoptic screening for amblyopia in kindergarten.

METHODS: A decision-analytic model was used. In this model all kindergarten children in Germany aged 3 years were examined by an orthoptist. Children with positive screening results were referred to an ophthalmologist for diagnosis. The number of newly diagnosed cases of amblyopia, amblyogenic non-obvious strabismus and amblyogenic refractive errors was used as the measure of effectiveness. Direct costs were measured from a third-party payer perspective. Data for model parameters were obtained from the literature and from own measurements in kindergartens. A base analysis was performed using median parameter values. The influence of uncertain parameters was tested in sensitivity analyses.

RESULTS: According to the base analysis, the cost of one orthoptic screening test was 7.87 euro. One ophthalmologic examination cost 36.40 euro. The total cost of the screening program in all kindergartens was 3.1 million euro. A total of 4,261 new cases would be detected. The cost-effectiveness ratio was 727 euro per case detected. Sensitivity analysis showed considerable influence of the prevalence rate of target conditions and of the specificity of the orthoptic examination on the cost-effectiveness ratio.

CONCLUSIONS: This analysis provides information which is useful for discussion about the implementation of orthoptic screening and for planning a field study.

9. Optom Vis Sci. 2003 Mar;80(3):259-69

A cost-effectiveness model of screening strategies for amblyopia and risk factors and its application in a german setting

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PURPOSE: To develop a general setting-independent decision-analytical model that determines the costs, effectiveness, and cost-effectiveness of four screening strategies to detect amblyopia or amblyogenic factors in pre-school children and to apply the model in a German setting.

METHODS: The general setting-independent decision-analytical model was developed from the perspective of society and the statutory health insurance was developed. Outcomes were the total number of newly detected true positive cases of amblyopia and the costs per newly detected true positive case of amblyopia. Strategies were screening of high-risk children up to the age of 1 year (ophthalmologists), screening of all children up to the age of 1 year (ophthalmologists), screening of all children aged 3 to 4 years (pediatricians or general practitioners), and screening of children aged 3 to 4 years visiting kindergarten (orthoptists). For the application example in a German setting, data from the published medical literature were used.

RESULTS: In the base-case analysis of the application example, screening high-risk children by ophthalmologists had the lowest average cost per case detected but became dominated (less effective and more costly than an alternative) if a low (5.3%) probability of familial clustering of strabismus was assumed. Considering the various assumptions tested in the sensitivity analysis, screening of all children up to the age of 1 year by ophthalmologists was the only strategy not dominated by others. Detection rates, including cases detected before screening, were between 72% and 78% for the strategies that screen for all children.

CONCLUSIONS: The model suggests that in Germany, both from a cost-effectiveness and a pure effectiveness point of view, screening all children up to the age of 1 year by ophthalmologists is the preferred strategy to detect amblyopia or amblyogenic factors. All strategies left a significant portion of children undetected.

10. Klin Monatsbl Augenheilkd. 2003 Jul;220(7):486-91

Resource utilisation and cost of amblyopia treatment [Article in German]

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BACKGROUND: The cost-effectiveness of screening for amblyopia is a controversial issue of international debate. The purpose of this study was to estimate the cost of amblyopia treatment to be used as a component for modelling the cost-effectiveness of prevention programmes. Cost was calculated from the perspective of the German social health insurance in the year 2002.

MATERIALS AND METHODS: A standardised detailed survey was conducted in writing among 13 experienced experts in amblyopia treatment from various offices and strabismological units in Germany. Average volumes of treatment items were estimated for a maximum treatment period of nine years. Cost was calculated using administrative prices (based on the social health insurance's uniform fee schedule for physician services and reference prices for therapeutic aids) and market prices.

RESULTS: The questionnaires were fully completed by 12 of the 13 experts. The mean total cost of treatment was estimated at 2.472 Euro (95 %-CI: 1.171 - 3.774) for strabismic amblyopia and 2.051 Euro (95 %-CI: 426 - 3.675) for amblyopia of refractive origin. About 70 % of the total cost was caused by the therapeutic aids (e. g. glasses, patches). The price of the patches had a marked impact on the total treatment cost. **CONCLUSIONS:** The results may be used for modelling the cost-effectiveness of screening programmes for the prevention of amblyopia.

11. Br J Ophthalmol. 2004 May;88(5):606-12

Cost effectiveness of treatment for amblyopia: an analysis based on a probabilistic Markov model

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AIMS: To estimate the long term cost effectiveness of treatment for amblyopia in 3 year old children.

METHODS: A cost utility analysis was performed using decision analysis including a Markov state transition model. Incremental costs and effects during the children's remaining lifetime were estimated. The model took into account the costs and success rate of treatment as well as effects of unilateral and bilateral visual impairment caused by amblyopia and other eye diseases coming along later in life on quality of life (utility). Model parameter values were obtained from the literature, and from a survey of experts. For the utility of unilateral visual impairment a base value of 0.96 was assumed. Costs were estimated from a third party payer perspective for the year 2002 in Germany. Costs and effects were discounted at 3%. Uncertainty was assessed by univariate and probabilistic sensitivity analysis (Monte-Carlo simulation).

RESULTS: The incremental cost effectiveness ratio (ICER) of treatment was euro2369 per quality adjusted life year (QALY). In univariate sensitivity analysis the ICER was most sensitive to uncertainty concerning the utility of unilateral visual impairment-for example, if this utility was 0.99, the ICER would be euro9148/QALY. Monte-Carlo simulation yielded a 95% uncertainty interval for the ICER of euro710/QALY to euro38 696/QALY; the probability of an ICER smaller than euro20 000/QALY was 95%.

CONCLUSION: Treatment for amblyopia is likely to be very cost effective. Much of the uncertainty in results comes from the uncertainty regarding the effect of amblyopia on quality of life. In order to reduce this uncertainty the impact of amblyopia on utility should be investigated.

12. Pediatrics. 2004 Feb;113(2):e95-108

Comment in:

- [Pediatrics. 2004 Feb;113\(2\):404-5.](#)

Cost-utility analysis of orthoptic screening in kindergarten: a Markov model based on data from Germany

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OBJECTIVE: To estimate the long-term cost-effectiveness of a hypothetical screening program for untreated amblyopia in 3-year-old children conducted by orthoptists in all German kindergartens in the year 2000.

METHODS: A cost-utility analysis was performed for which a decision tree was combined with a Markov model. Incremental costs and effects during the children's remaining lifetime were estimated. The model took into account the probability of treatment without screening, age-specific treatment success rates, costs of screening and treatment, as well as effects of unilateral and bilateral visual impairment caused by amblyopia and other eye diseases coming along later in life on quality of life (utility). Model parameter values were obtained from a field study of orthoptic screening in kindergarten, from the literature, and from expert interviews. Costs were estimated from a third-party payer perspective. Uncertainty was assessed by univariate and probabilistic sensitivity analysis (Monte Carlo simulation).

RESULTS: The incremental cost-effectiveness ratio (ICER) of orthoptic screening was 7397 Euro (euro) per quality-adjusted life year (QALY) when costs and effects were discounted at 5%. In univariate sensitivity analysis, the ICER was sensitive to the uncertainty regarding the utility of unilateral visual impairment and to the discount rate for effects; besides uncertainty regarding the prevalence of untreated amblyopia, the odds ratio of success of treatment when started late, and the probability of treatment without screening had a noticeable but much smaller effect. Monte Carlo simulation yielded a 90% uncertainty interval for the ICER of 3452 euro/QALY to 72 637 euro/QALY; the probability of an ICER <25 000 euro/QALY was 84%.

CONCLUSIONS: The ICER of orthoptic screening seems to fall within a range that warrants careful consideration by decision-makers. Much of the uncertainty in results comes from the uncertainty regarding the effect of amblyopia on quality of life. To reduce this uncertainty, the impact of amblyopia on utility should be investigated.

13: Ophthalmology. 2002 Dec;109(12):2265-71

A cost-utility analysis of therapy for amblyopia

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OBJECTIVE: Evaluation of the incremental cost-effectiveness of therapy for amblyopia. **DESIGN:** Cost-utility reference-case analysis.

METHODS: A cost-utility analysis was performed from a third-party insurer perspective by using decision analysis, evidence-based data from the literature, and patient preference-based time trade-off utility values.

DATABASE: Patient-derived time trade-off ocular utility values and the American Academy of Ophthalmology Preferred Practice Pattern guidelines for the treatment of amblyopia.

INTERVENTION: Treatment of childhood amblyopia using medical and surgical therapies per the American Academy of Ophthalmology Preferred Practice Pattern.

MAIN OUTCOME MEASURE: Dollars (year 2001 nominal U.S. dollars) expended per quality-adjusted life-year (\$/QALY) gained. **RESULTS:** Treatment for amblyopia resulted in a \$/QALY gained of \$2281 with a discount rate of 3% for costs and outcomes. Sensitivity analysis, varying costs and utility values by 10%, resulted in a \$/QALY gained range from \$2053 to \$2509.

CONCLUSIONS: When compared with other interventions in health care, therapy for amblyopia seems to be highly cost-effective. This information is increasingly important for health care policy makers.