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Economic Evaluation of Newborn Hearing Screening: Modelling Costs and Outcomes

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Citation	Hessel, Franz, Eva Grill, Petra Schnell-Inderst, Uwe Siebert, Silke Kunze, Andreas Nickisch, Hubertus von Voß, and Jürgen Wasem. 2003. Economic evaluation of newborn hearing screening: modelling costs and outcomes. German Medical Science 1: GMS e-journal 1.
Published Version	http://en.scientificcommons.org/41888387
Accessed	February 19, 2015 12:42:43 AM EST
Citable Link	http://nrs.harvard.edu/urn-3:HUL.InstRepos:4595269
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Economic evaluation of newborn hearing screening: modelling costs and outcomes

Gesundheitsökonomische Evaluation von Neugeborenen-Hörscreening: Modellierung von Kosten und medizinischer Wirksamkeit

Abstract

Objectives: The prevalence of newborn hearing disorders is 1-3 per 1,000. Crucial for later outcome are correct diagnosis and effective treatment as soon as possible. With BERA and TEOAE low-risk techniques for early detection are available. Universal screening is recommended but not realised in most European health care systems.

Aim of the study was to examine the scientific evidence of newborn hearing screening and a comparison of medical outcome and costs of different programmes, differentiated by type of strategy (risk screening, universal screening, no systematical screening).

Methods: In an interdisciplinary health technology assessment project all studies on newborn hearing screening detected in a standardized comprehensive literature search were identified and data on medical outcome, costs, and cost-effectiveness extracted. A Markov model was designed to calculate cost-effectiveness ratios.

Results: Economic data were extracted from 20 relevant publications out of 39 publications found. In the model total costs for screening of 100,000 newborns with a time horizon of ten years were calculated: 2.0 Mio.€ for universal screening (U), 1.0 Mio.€ for risk screening (R), and 0.6 Mio.€ for no screening (N). The costs per child detected: 13,395€ (U) respectively 6,715€ (R), and 4,125€ (N). At 6 months of life the following percentages of cases are detected: U 72%, R 43%, N 13%.

Conclusions: A remarkable small number of economic publications mainly of low methodological quality was found. In our own model we found reasonable cost-effectiveness ratios also for universal screening. Considering the outcome advantages of higher numbers of detected cases a universal newborn hearing screening is recommended.

Zusammenfassung

Einleitung: Die Prävalenz von Hörstörungen bei Neugeborenen beträgt 1-3 pro 1000 Neugeborene. Entscheidend für die spätere Entwicklung der betroffenen Kinder sind möglichst frühzeitige Diagnose und Therapie. Mit BERA und TEOAE stehen risikoarme Methoden zur Frühentdeckung zur Verfügung. Obwohl ein universelles Screening empfohlen wird, wurde es in den meisten europäischen Ländern bisher nicht verwirklicht.

Ziel des Projektes war es, die wissenschaftliche Evidenz von Neugeborenen-Hörscreening zu untersuchen und die medizinischen Folgen sowie die Kosten verschiedener Programme zu vergleichen. Dabei wurden die drei verschiedenen Screening-Strategien universelles Screening (U), Risiko-Screening (R) und kein systematisches Screening (N) verglichen.

Methoden: In einem interdisziplinären Health Technology Assessment Projekt wurden alle relevanten, im Rahmen einer standardisierten ausführlichen Literaturrecherche identifizierten Studien zu Neugeborenen-Hörscreening eingeschlossen und die Ergebnisse zur medizinischen Wirksamkeit, zu Kosten und zur Kosteneffektivität extrahiert. Um die

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Kosteneffektivität verschiedener Strategien abzuschätzen, wurde ein Markov Modell entwickelt.

Ergebnisse: Aus 20 von insgesamt 39 Studien wurden ökonomische Daten ausgewertet. Im Modell wurden Gesamtkosten für eine Kohorte von 100.000 Neugeborenen in Höhe von 2,0 Mio.€ für ein universelles Screening, von 1,0 Mio.€ für Risiko-Screening und 0,6 Mio.€ für die Situation ohne systematisches Screening kalkuliert. Die Kosten pro entdecktem Fall betragen 13.395€ (U), 6.715€ (R) bzw. 4.125€ (N). Bis zum Alter von 6 Monaten wurden 72% (U), 43% (R) bzw. 13% (N) der Fälle entdeckt.

Schlussfolgerung: Es wurde eine bemerkenswert geringe Anzahl gesundheitsökonomischer Studien von überwiegend mangelhafter methodischer Qualität gefunden. Unser Modell konnte eine akzeptable Kosteneffektivitäts-Relation auch für eine universelle Hörscreening-Strategie zeigen, so dass in Anbetracht der höheren Zahl rechtzeitig entdeckter Fälle auch aus ökonomischen Gesichtspunkten die Empfehlung für ein universelles Screening auf konnatale Hörstörungen ausgesprochen wird.

Introduction

According to calculations of the WHO world-wide approximately 350 Mio. people have hearing disorders. The overall prevalence of connatal hearing disorders is 1-3 in 1,000 newborns, the prevalence in risk groups is estimated at about 10 times higher. Risk factors are e.g. early child birth, infection in early pregnancy or family history of hearing disorders [1]. For a detailed overview of the medical and epidemiological background it is referred to the full version of the German HTA-report [2] or the preceding British and French reports on NHS [3], [4]. The neurological development of hearing abilities requires an acoustic stimulation as soon as possible, latest before finishing the first two years of life. Deficits due to absent acoustic stimulation during the first years of life are nearly impossible to improve by later rehabilitation [5]. Therefore diagnosis and treatment as early as possible are necessary for a successful and effective treatment of connatal peripheral hearing disorders.

If connatal hearing disorders are detected and treated in time, most of the children are enabled to pass through a nearly normal development of speech and no special education is necessary [6], [7], [5]. For the detection of hearing disorders with TEOAE (transient evoked otoacoustic emissions) and BERA (brainstem evoked response audiometry) tests with an acceptable sensitivity and specificity are available. TEOAE is easier to perform, less time consuming, and cheaper, but shows more false positive results. BERA requires more time, but is regarded as the gold standard for the diagnosis of hearing disorders. Although for screening purposes several automated versions of BERA are available, most of the programmes are using TEOAE.

Usual treatment of connatal sensory hearing disorders consists of supply with hearing aids. If treatment with hearing aids do not improve hearing, cochlea implants should be considered.

The mean age of diagnosis of connatal hearing disorders in Germany, like in most other western countries, is 2-4

years of age depending on the severity of hearing impairment. Treatment is started on average 9 months later [8], [9], [10]. There is a marked discrepancy between these findings and the recommendation of international consensus groups.

Recommendations of the European Consensus Development Conference on Neonatal Hearing Screening [11], [12] are: diagnosis in the first 6 months of life and treatment in the first 12 months of life. More recent recommendations of the Joint Committee on Infant Hearing and the Deutsche Gesellschaft für Phoniatrie und Pädaudiologie [13], [14] even claim a detection at 3 months and treatment in the first 6 months. To achieve an early diagnosis and treatment, a universal screening for hearing disorders is recommended. But a regular screening of newborns is neither implemented in Germany nor in most other health care systems. In the last years several regional newborn hearing screening programmes were implemented in Germany [for example [15], [16], although there is no regular reimbursement of the screening tests by German statutory sickness funds.

Different tests or test combinations of BERA and TEOAE are available. At present one of the most common strategies is a so-called two-step TEOAE-strategy with a single TEOAE test in the first days of life and, if no TEOAEs are detected, a second similar test a few days later. If the first or second test is negative the children are classified as test negative. If the first and the second test is positive the tested newborns are classified as test positive.

The objectives of this interdisciplinary economic health technology assessment project were to compare the costs, effects and cost-effectiveness ratios of three different strategies:

1. Universal screening of all hospital born newborns
2. Risk screening of all hospital born newborns with risk factors
3. No regular screening

Cost-effectiveness ratios to be calculated were:

1. Costs per screened child
2. Costs per case detected (case defined as hearing loss >40dB on better ear)
3. Costs per case detected in time (in time defined as in the first 6 months of life)

Methods

According to the methodological recommendation for health technology assessment projects in Germany [17], cost and cost-outcome calculations based on published literature data combined with actual item costs were performed. An externally reviewed literature search for publications on newborn hearing screening from 1990 to September 2001 of relevant electronic databases Medline, Embase, Evidence based Medicine, Healthstar, Current Contents, DARE, NEED, Cochrane Library, ERIC, PsycLIT, PsycINFO, INSPEC, and SOMED was performed. If costs or cost calculations were mentioned in the title, abstract or keywords, publications were included in the economic part of the report. "Gray literature" like conference booklets, relevant internet homepages or publications not listed in literature databases was scanned by hand. A detailed description of the literature search strategy is documented in the full version of the report [2] and can be obtained from the authors on request. Detected publications were scored according to an established standardized questionnaire [2] and included respectively excluded for further evaluation. Data from included studies were converted to € using the purchasing power parities (PPPs) of the Organization for Economic Co-operation and Development (OECD) and standardized to the year 1999 according to the German health sector specific inflation rates given by the Federal Statistical Office Germany (Statistisches Bundesamt Deutschland). Overall nearly 800 publications dealing with hearing screening were found. For the numbers of economic publications please see result section.

Cost Calculations

As possibly relevant cost components were defined:

Direct medical costs:

- Costs for the implementation of the screening programme
- Costs for screening tests
- Costs for the organization of the screening programme
- Costs for tracking
- Costs for further diagnostic procedures to detect (true and false) screening positives
- Treatment for detected cases of hearing disorder
 - • Regular controls
 - • Treatment with hearing aids (supply, controls, batteries etc.)
 - • Treatment with cochlear implants (pre-operative tests, device, operation procedure, rehabilitation)

Direct non-medical costs:

- Transportation costs for diagnostic procedures and treatment
- Additional education costs for special institutions for children with hearing disorders

Indirect costs:

- Work time loss for parents
- Work time loss for adults with hearing disorders
- Income loss due to hearing disorders
- Productivity loss due to premature mortality

Markov Model

To estimate the long-time costs and outcomes of newborn hearing screening a Markov model [18] was designed. Markov models in medical decision analysis are considered as explicit and quantifying approach for decisions between alternatives under uncertainty. The decision is made according to the trade off between medical risks, benefits and costs. As all models Markov models are not able to reflect all aspects of clinical reality, but the most relevant structures and parameters are demonstrated and offered for discussion.

For design and calculation of the Markov model the software DATA Treeage was used. The literature search was extended to publications dealing with direct or indirect costs of hearing disorders, costs, cost-effectiveness, and long-time outcomes of children supplied with hearing aids or cochlear implants independent from newborn hearing screening. A two-step TEOAE test strategy as described above was chosen.

In the cost calculations costs of TEOAE-tests and of further diagnostic procedures were included. Because of the lack of outcome and cost data, costs for medical treatment and education of children as well as indirect costs could not be included.

"Universal screening" (U), "risk screening"(R), and "no screening"(N) as described above were defined as alternative strategies.

Predicted outcomes were defined as:

1. Number of true positively detected cases of connatal hearing disorder at 6 months
2. Number of "detected child months" at 6, 12, and 120 months
3. Costs per 100,000 screens

The outcome "Number of detected child months" (2.) is described as the amount of months in the defined time frame of 6, 12, or 120 months in which a hearing disorder is known. For example, in a time horizon of 6 months a child with hearing disorder detected at birth is equivalent to 6 detected child months. Detection with 4 months is equivalent to 2 detected child months. This outcome measure was chosen in addition to the classical outcome "Number of cases detected" (1.) to underline the importance of early detection. As cost-effectiveness outcomes were calculated:

1. Costs per case of hearing disorder detected
2. Costs per detected child month

A health care system's perspective was chosen for the cost calculations. To reflect the time dependence of

Table 1: Relevant parameters and assumptions of Markov model

Parameter	Baseline (Range) %
Prevalence of connatal hearing disorder	0,15 (0,09-0,3) ¹
Prevalence of ≥ 1 risk factor	20 (10-30) ²
Prevalence of hearing disorders	
In children with risk factor	0,38 ^{3,4}
In children without risk factor	0,09
Prevalence of risk factor in children with hearing disorder	50 (46-56) ⁵
Screening S-TEOAE/S-ABR	
Sensitivity	96 (96-100) ⁶
Specificity	89 (77-96)
Further diagnostic:	
Sensitivity	98
Specificity	98
Participation in screening	90 (85-95)
Follow-up after screening	80 (75-85)
Probability of (false) suspect of hearing disorder in healthy child	0,1 (0-0,5) ⁷
Discount rate	3 per year (0-5)
Probability of „natural“ detection of hearing disorder	Empirische Wahrscheinlichkeitsfunktion ⁸ Mediane Entdeckungszeit 18 Monate

¹ [31, 33, 34, 37, 8, 42, 43, 47, 51, 52, 29]² [30, 36, 42, 43]³ [39, 56]⁴ no range because depending on prevalence of hearing disorders and risk factors⁵ [35, 45, 8]⁶ [32, 40, 53]⁷ Own calculation based on expert statements: 15% of all children are tested before 6 years because of delay in development of speech, one third is test positive, 20% of test positives due to connatal hearing disorders⁸ Total count of all diagnosed cases of the regions of Schwaben und Oberbayern from 1st January 1998 to 31st December 1999 of three centers in Munich (Klinikum München-Großhadern der Ludwig-Maximilians-Universität München, Kinderzentrum München, Klinikum Rechts der Isar Technische Universität München), unpublished data from Dr. Bernhard Bornschein, School of Public Health and Epidemiology Ludwig-Maximilians-University München

subjective preferences costs and outcomes were discounted with a rate of 3%. A Markov-cycle length of 1 month and a total time of 10 years were chosen for modelling. Sensitivity analyses were performed on relevant parameters. All assumptions made and all parameters used are shown in Table 1.

Results

Overall 39 economic publications on newborn hearing screening were found. Because of a lack of transparent cost data 19 publications were excluded. For further calculations 20 publications remained: 16 journal articles, three health technology assessment reports [4], [3], [19]

and one conference abstract. A series of three publications [20], [21], [22] reporting the results of the same study was considered as one. Overall 15 different primary studies on newborn hearing screening with economic components were included. For references please see Table 2.

In general most of the economic publications showed a relatively poor methodological quality according to international recommendation for economic evaluation studies of health care programmes [23], [24], [25]. Only one single study included costs of treatment and education and chose a time horizon of more than one year. The cost calculations were not transparent, references for resource uses and valuation as well as the perspective of the calculation often remained unclear.

Table 2: Costs and cost-effectiveness according to published studies, adjusted to € of year 1999

Study	Test strategies	Costs per newborn (€ 1999)	Costs per case detected (€ 1999)	Long-time cost-effectiveness ratio
Friedland et al. 1996 [38]	BERA	26.90-100.56 according to setting	13411-49434 according to setting	-
Heinemann & Bohnert 2000 [40]	BERA TEOAE	7.05-22.07 according to test method	-	-
Kemper & Downs 2000 [41]	BERA TEOAE	Risk group: 1.59 Universal: 9.93	Risk group: 3097 Universal: 11564	-
Kezirian et al. 2001 [44]	BERA	13.05-25.45 according to test method	5170-9575 according to test method	-
Markowitz 1990 [46]	BERA	54.82-90.91 according to test method	3083-4712 according to test method	-
Mason & Herrmann 1998 [47]	BERA	28.59	19768	-
Maxon et al. 1996 [48]	TEOAE	32.30	5428	-
Mehl & Thompson 1998 [49]	BERA TEOAE	27.84	total: 10692 bilateral: 13699	Savings: 4.2 Mio by 54000 newborns/a over 12 years
Messner et al. 2001 [50]	BERA	27.71	-	-
Stevens et al. 1998 [54]	BERA TEOAE	17.73-36.07 according to test method	-	-
Turner 1991/1992 [20, 21, 22]	BERA	47.53-187.38 according to test method and setting	6517-96404 according to test method and setting	-
Verkerk & Boshuizen 1998 [55]	subjective testing	24.61-27.85 according to test method	34852-59902 according to test method	-
Vohr et al. 2001 [57]	BERA	16.33-25.23 according to test method	8149-11666 according to test method	-
Watkin 1996 [59]	TEOAE	17.83	8913	-
Weirather et al. 1997 [60]	TEOAE	8.68	-	-

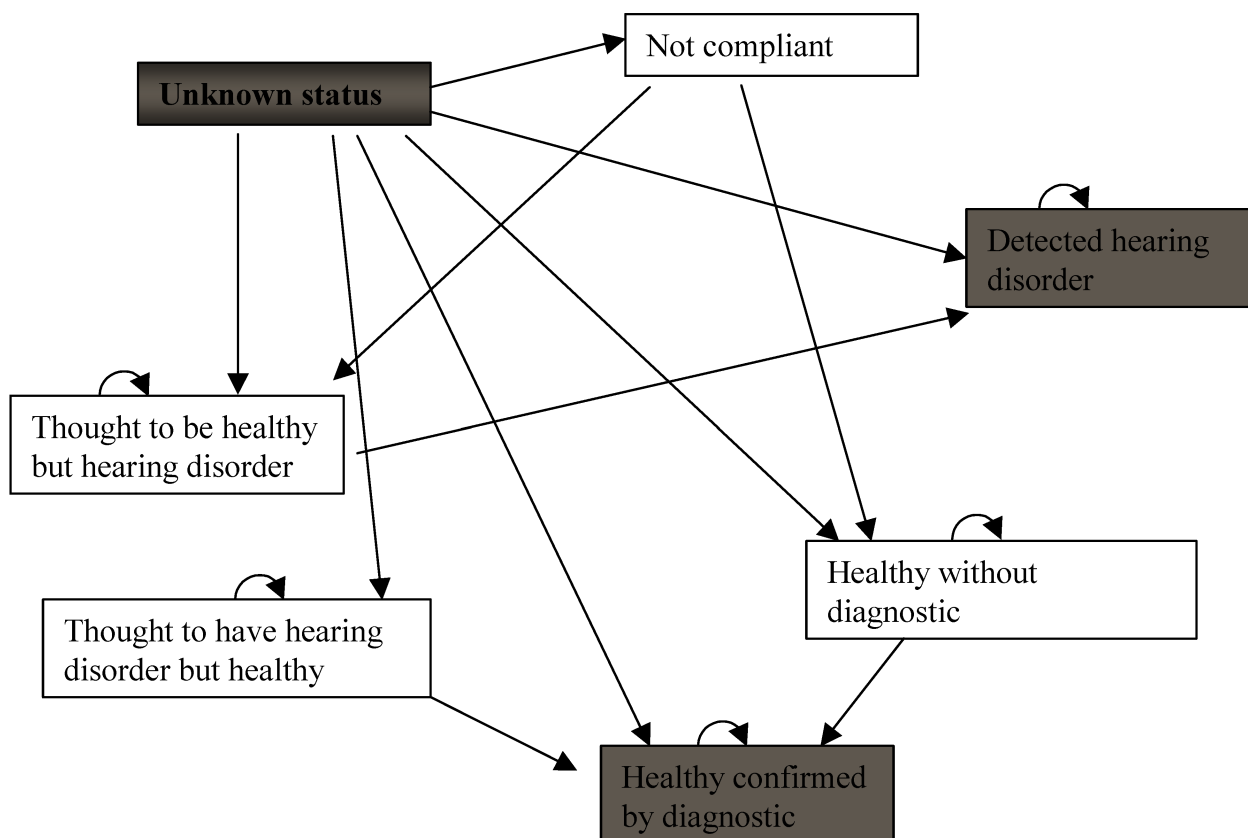
Besides the intervention costs for the screening test itself most of the studies limited the calculation to the costs for tracking and further diagnostic of hearing disorders. One study included the costs for implementation of the screening programme. The costs for further medical treatment of detected cases and special education were included in one study. None of the studies considered other direct non-medical costs or indirect costs. Also no studies with utility measures or health related quality of life were found (see also Table 2).

The different studies did not use or calculate similar single item costs. After adjustment to € of 1999 the costs for the same test showed an up to five time range regarding the costs for screening tests, tracking and further diagnostic, e.g. the test costs for a two-step TEOAE-ABR-screen varied from 7.76€ to 26.88€. The highest single item costs with more than 500,000€ were calculated for long-time treatment and special education of children with hearing disorders. In contrast to the importance of the long-time outcome these costs were only included in one study.

As all total costs presented in the different studies were based on single item bottom-up cost calculations the total costs per screened child and the costs per case detected also showed a wide range. Table 2 shows the results of all included studies. If only recent publications and comparable settings and calculation methods are considered, the range can be narrowed to about 7-36€ per screened child and 3,000-13,000€ per case detected. None of these calculations included the costs or savings for long-time treatment or education. There was no systematic difference or trend towards a definite test-method.

Markov Model

Graph 1 shows the structure of the Markov model. All children begin in the starting state "status unknown" and end up in one of the absorbing states "detected hearing disorder" or "healthy, confirmed by diagnostic". The children are screened with a certain probability. This probability is 1.0 in strategy U (universal screening), according to the prevalence of at least one risk factor 0.2 in strategy R (risk screening) and 0.0 in strategy N (no screening).



Graph 1: Structure of Markov model

Further diagnostic procedures are performed in all screening positive children. The children with hearing disorders, who were not screened, are detected with a "natural" detection rate, based on an empirical function taken from the register of hearing disorders of the area of Munich. Concerning the extent of a non-systematic screening this area is considered as representative for Germany. Therefore, strategy N does not represent a situation without any screening at all but without a systematic screening.

For the calculation presented here a two-step TEOAE-test strategy was chosen. According to the specific sensitivity and specificity of a two-step TEOAE-screening some children are screening positive but healthy, some are screening negative with undetected hearing disorder. A drop out either at screening (10%) or at follow up after positive screening test (20%) was included in the model calculation.

With the assumed prevalence of 0.15 % in a cohort of 100,000 newborns 150 cases of congenital hearing disorder are present. At 6 months of life with a universal screening strategy 108 cases (72%) are detected, with a risk screening 64 (43%), without regular screening 20 (13%). Out of possible 900 detected child months with a universal screening 630 months, with a risk screening 354 months and without screening 78 detected child months were achieved.

The costs for screening 100,000 newborns using TEOAE are calculated with about 2.0 Mio.€ for a universal screening, 1.0 Mio.€ for screening of risk groups. The

costs for a strategy without regular screening were 0.6 Mio.€. This leads to costs per newborn of 20€ (U), 10€ (R) respectively 6€ (N). The costs per case of congenital hearing disorder detected were calculated with 13,395€ (U), 6,715€ (R) and 4,125€ (N) (see also Table 3).

Sensitivity analyses of all relevant parameters and assumptions were performed. Similar for all strategies the strongest influence on outcomes was seen if prevalence was varied. The variation of discount rate had little influence; the model was insensitive to test parameters (sensitivity and specificity) and loss to follow up. The costs were strongly influenced by variation of test costs, test parameters especially the number of false test positives, and the probability to be detected without screening. Prevalence and discount rate did not affect the costs.

Table 3: Results of the Markov model, base case, discount rate 3%

Outcome-Parameter	Universal Screening	Risk Screening	No Screening
	U	R	N
Detected child months at 6 months	630	354	78
Detected child months at 12 months	1,298	801	304
Detected child months at 120 months	13,926	12,063	10,201
Cases detected at 6 months (per 100,000)	108	64	20
Cases detected at 120 months (per 100,000)	150	150	150
Difference in detected child months comparing strategies	U vs. R: 44 U vs. N: 88	R vs. N: 44	-
Total costs (per 100,000; over 120 months)	2,009,281 €	1,007,297 €	618,677 €
Costs per child	20.09 €	10.07 €	6.18 €
Costs per case detected	13,395 €	6,715 €	4,125 €
Costs per detected child months	144 €	84 €	61 €

Discussion

As part of a German interdisciplinary health technology assessment project the economic consequences of newborn hearing screening were investigated. In a detailed literature search overall nearly 800 publications dealing with hearing screening were found. Despite this relatively large number only 15 studies with own economic calculations were detected.

Possibly also due to the different methodological approaches the costs per screening test and the costs per case detected showed - although adjusted and standardised to one currency and one year - a wide range without a clear tendency to a definite test method.

The results of the published studies are insufficient to answer the policy question regarding the economic consequences in a sense of costs and cost-effectiveness of different strategies for detection of connatal hearing disorders. There is a lack of convincing studies presenting results on a high level of evidence according to recommendations of evidence based medicine and the need for further research, especially randomised controlled trials with a sufficient follow up, must be underlined.

The results of the assessment of the studies on the medical effectiveness of newborn hearing screening are presented in detail elsewhere. Most of the authors conclude from the results of their studies that a newborn hearing screening should be recommended, although there is an ongoing discussion if the present scientific evidence can be considered as sufficient concerning the maximum age of diagnosis and treatment [26], [27], [28], [29].

To estimate the costs and outcomes of newborn hearing screening a Markov model was designed. The model

presented here considers the aspect that, on one hand all cases of connatal hearing disorders are detected sooner or later, but on the other hand they should be diagnosed as early as possible. Therefore in addition to the outcome "number of cases detected by screening", which was used in all newborn hearing screening studies found, we included the date of detection in our outcome measure "detected child months".

On the outcome side expressed in the number of detected cases and the number of detected child months the model shows clear advantages for newborn hearing screening compared to no regular screening. At the crucial date of 6 months without screening only 9% of the cases are detected, with a screening of children with one or more risk factors the detection rate is 40%, with a universal screening about 70%. A universal screening strategy shows a higher rate of cases detected in time, and a percentage of 40% detected with a risk screening does not seem to be sufficient in considering the importance of early diagnosis and treatment. Combining the mentioned advantages in medical outcome with still acceptable and reasonable costs a universal hearing screening is recommended.

We chose a two-step TEOAE-strategy for the model calculation presented here, but the model could be easily adjusted for other test strategies.

The cost-effectiveness ratios are not directly comparable to those of other health care technologies. But with costs per case detected of 14,000€ a universal screening strategy seems to be reasonable if the lifelong benefits are taken into account. If it is assumed in a conservative calculation that half of the children benefit from earlier detection and their quality of life improves by 10% over 50 years, 2.5 QALYs are gained. Based on our cost calcu-

lations the costs per quality adjusted life year (QALY), a possible utility measure for the comparison of different health care strategies, would be less than 10,000€.

Because of the lack of literature data on health care resource use and the percentage of children, who are able to visit a regular school after treatment with hearing aid or cochlea implant - the existing studies on cochlea implantation used a different population only of children with severe hearing disorders - the cost calculations are incomplete. Any savings connected to a better outcome because of an earlier detection and treatment were not included. If these savings as well as indirect costs e.g. estimated as the avoided loss of income due to hearing disorders, would be included in the calculation, there might well be an overcompensation of screening costs. Further studies will have to answer these questions. Nevertheless, the present data on medical and economic outcomes suggest a recommendation of a universal hearing screening with TEOAE or other test strategies.

Acknowledgements

The study is part of a project of the German Agency for Health Technology Assessment DAHTA@DIMDI and was supported by the German Ministry of Health.

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Please cite as

Hessel F, Grill E, Schnell-Inderst P, Siebert U, Kunze S, Nickisch A, von
Voß H, Wasem J. Economic evaluation of newborn hearing
screening: modelling costs and outcomes. *Ger Med Sci.* 2003;1:Doc09.

This article is freely available from

<http://www.egms.de/en/gms/2003-1/000009.shtml>

Received: 2003-06-02

Published: 2003-12-15

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