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Review Article

Health economic analysis of screening

Abstract

In this article health economic implications of screening are analysed. First, requirements screening programmes should fulfil are derived, and methodical standards of health economic evaluation are outlined. Using the example of newborn hearing screening, it is then examined if empirical studies meet the methodical requirements of health economic evaluation. Some deficits are realised: Health economic studies of newborn hearing screening are not randomised, most studies are even not controlled. Therefore, most studies do not present incremental, but only average cost-effectiveness ratios (i.e. cost per case identified). Furthermore, evidence on long-term outcomes of screening and early interventions is insufficient. In conclusion, there is a need for controlled trials to examine differences in identified cases, but particularly to examine long-term effects.

Keywords: health economics, evaluation, costs, prevention, screening, newborn hearing screening

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1 Introduction

Screening is among the central topics of prevention in Germany. The rationale for screening is to detect a target disease in an early – pre-symptomatic – phase of disease and to realise better treatment outcomes by early intervention compared to usual care in a later – symptomatic – phase.

Screening is a substantial part of SHI-authorised medical care (i.e. medical care for persons insured by the Social Health Insurance SHI) for nearly 40 years. Screening includes routine well child visits (including screening for metabolic diseases) – so-called U1 to U11. Furthermore, screening programmes for cancer (e.g., breast cancer, colon cancer, and skin cancer), cardiovascular diseases, diabetes, and renal diseases ("check-up 35") are offered to adults (each screening from a specific age of the insured person). Also, currently a nationwide mammography screening is established in Germany. And introduction of further screening programmes is expected, particularly as genetic disposition for various cancers is already or will in future be identifiable.

Screening programmes (like all health care services) not only have to prove medical effectiveness, but also economic cost-effectiveness. Over the last 20 years, decisions in the health care sector increasingly focus on health economic goals. Due to demographic ageing and technological progress the provision of health care services is characterised by an increasing shortage of resources. To limit rising health care expenses, regulatory interventions are increasingly executed.

The proof of cost-effectiveness is based on health economic evaluation studies. Health economic evaluation is to assist rational decisions in health politics. Economic evaluation provides information on costs, outcomes, and efficiency of medical technologies (in particular: screening) and supports decisions whether new technologies (in particular: screening programmes) should or should not be adopted in the health care system.

In this article screening is analysed from a health economic perspective. Section 2 describes the characteristics of screening. Section 3 presents the theoretical basis of health economic evaluation. In section 4 specific requirements for health economic analysis of screening programmes are discussed using the example of newborn hearing screening. The article ends with conclusions in section 5.

2 Screening

2.1 Definition of screening

Screening is a systematic procedure to detect a target disease in an early – pre-symptomatic – phase of disease. The aim of screening is to reduce disease specific morbidity and mortality by utilising appropriate early interventions [1]. Additional benefits of an earlier intervention compared to usual treatment in a later – symptomatic – phase are required to justify screening tests, otherwise unnecessary costs, unnecessary psychological strain, and unnecessary risks would be induced [2]. Benefits of an early treatment could even be, for example, that a less aggressive treatment is sufficient or the disease process is shorter and less serious. The following screening approaches are distinguished.

- mass screening versus selective screening
- organised screening versus opportunistic (or spontaneous) screening.

Mass screening involves the total population or at least defined large parts of the population (e.g., all children or

all women). Selective screening aims at person groups with specific risks – so-called high-risk groups – (e.g., women with BRCA gene mutations bearing an increased risk for breast cancer). The frontier between mass screening and selective screening is not unambiguous as for many diseases differences in incidence and prevalence are only explained by age. Only in some diseases high-risk groups can be defined by other determinants (an example is newborn hearing screening – see section 4).
The distinction between organised screening and opportunistic screening is more obvious. Organised screening is initiated by the health care system, i.e. subpopulations will be called on by health care providers (e.g., for screening at the workplace) or will be invited for screening by mail, telephone or public campaigns (e.g., mammography). In contrast, opportunistic screening is part of routine care. It is performed when a patient is consulting the health care system (i.e. opportunistic screening is initiated by the patient). Examples for opportunistic screening are blood pressure measurement and blood glucose measurement [1].
Screening programmes are based on two (implicit) assumptions: (1) early identification of risk factors can prevent the onset of a disease (e.g., endoscopic identification and excision of polyps in the colon) or (2) early detection and treatment of a disease provide improved health outcomes (concerning life expectancy and quality of life) (e.g., mammography to detect breast cancer).

2.2 Effectiveness criteria of screening programmes

The Word Health Organisation has presented a checklist to assess the effectiveness of a screening programme [3]

- the target disease should be a substantial health problem
- the disease should be diagnosable in a latent or pre-symptomatic phase
- the natural course of disease – including the transition from a latent to a manifest phase – should be understood
- a simple, safe and sufficiently precise screening test should exist to detect the target disease
- the screening test should be accepted in the target population
- an effective treatment should be available. Treatment outcomes should be the better the earlier the disease is detected
- it should be agreed on which persons must be treated (and which treatment is adequate)
- an adequate infrastructure for screening, diagnostics, and treatment should exist
- costs of a screening programme (including test, diagnosis and treatment) should be in a proper relation to total costs of a disease
- screening for a disease should be a continuous process (incidence screening) instead of a singular event (prevalence screening).

2.3 Statistical properties of a screening test

Screening tests should reliably discriminate between diseased and healthy persons [1]. The validity of a screening test depends on test sensitivity (probability of testing positive if a person is actually diseased), test specificity (probability of testing negative if a person is actually healthy), and prevalence of a target disease.
As most persons of a target population are healthy low specificity produces a large number of false-positive test results – i.e. the positive predictive value (probability that a person is actually diseased if the test result is positive) is low. The negative effects of false-positive test results are additional costs of diagnostic assessment, health risks of additional diagnostics, and the psychological strain induced by positive test results.
The prevalence of a target disease is often more important for costs and outcome of a screening programme than the test validity. Mass screening programmes are not suitable for early detection of rare diseases (with low prevalence) [2]. However, the prevalence of most diseases can be adjusted by defining age limits. Mammography, for example, is not offered to women under the age of 50 due to low prevalence. If prevalence is not adjustable, an extremely high specificity should be aimed at.
Sensitivity and specificity can be defined as a function of a diagnostic cut-off value (to dsicriminate between diseased and healthy persons) [1], i.e. a varied cut-off point is affecting sensitivity and specificity. In general, an increase in sensitivity implies a decrease in specificity, and vice versa. Thus, when the cut-off point is defined the relative importance of sensitivity and specificity should be considered (i.e. false-positive and false-negative test results should be traded off) [1].

2.4 Systematic errors

Evaluations of screening programmes should pay attention to systematic errors. For screening tests lead time bias and length time bias are particularly relevant [1].
Lead time bias: Diagnosis of a target disease is advanced by screening. Lead time denotes the interval between detection of pre-symptomatic cases at screening and usual detection of cases when clinical symptoms have been developed. Thus, the interval between diagnosis and death is prolonged by screening even if no gain in lifetime is realised (so-called unadjusted period of time). To isolate the effect of screening on survival – i.e. potential increase in life expectancy – the total interval in screened persons between diagnosis and death has to be adjusted for lead time (which is a non-trivial problem).
Length time bias: Cases of a target disease progressing more rapidly from preclinical to clinical disease will gain

less lead time from screening than cases progressing less rapidly (i.e. the preclinical phase of disease is shorter). These cases are less likely to be detected by screening than cases with a longer preclinical phase. Furthermore, it is often assumed that less rapidly progressing cases have a better prognosis (particularly assumed for cancer). Thus, cases detected by screening could have a better survival only (or at least to some extent) because cases with better prognosis are more likely to be detected at screening (length time bias).
As the effects of lead time bias and length time bias (and possible more biases) on the outcome are extremely complex, the utility of screening programmes should be proofed by randomised controlled trials (RCTs).

2.5 Parameters for the determination of screening effectiveness

Evaluation studies of screening programmes are often based on the following outcome parameters

- number of detected cases of illness (in the screened population (intervention group) versus in the not-screened population (control group))
- distribution of (cancer) stages in the intervention group versus in the control group
- survival time of the screened versus not-screened population
- disease-specific mortality.

If the evaluation is not based on RCTs, the first three outcomes are no appropriate parameters for proofing cost-effectiveness due to lead time and length time biases. Disease-specific mortality is thought to be a valid outcome parameter. Furthermore, reduction of disease-specific mortality is the central goal of many screening programmes (particularly in cancer screening).

3 Health economic evaluation

3.1 Aims of health economic analysis

Economic aims are increasingly important to the health care sector as resources are increasingly scarce due to demographic ageing and technological progress in health care. According to economic principles provision of health care services should be efficient which means that

1. only effective health care services will be provided (effectiveness)
2. health care services will be produced at the lowest possible costs (production efficiency)
3. amount, quality and structure of health care services are based on need and/or utility of the insured population (allocation efficiency).

Health economic evaluation is to assist rational decisions in health politics. Economic evaluation provides information on costs, outcomes, and efficiency of medical technologies and supports decisions whether new technologies should or should not be adopted in the health care system.

3.2 Approaches of health economic evaluation

In health economic evaluation costs, effects (outcomes) and cost-effectiveness – i.e. the ratio of effects and costs – of alternative interventions are compared. The basic approaches of health economic evaluation are [4], [5], [6], [7] (1) cost-minimisation analysis, (2) cost-effectiveness analysis, (3) cot-utility analysis, and (4) cost-benefit analysis. The evaluation approaches differ in the outcome dimension. Outcomes can be differentiated into [7], [8]

- clinical parameters
- health care outcomes
- health outcomes
- life expectancy
- quality of life (QoL)
- monetised outcomes (willingness to pay).

In cost-minimisation analyses (CMA) only costs of the intervention alternatives are considered. It is assumed that the relevant health outcomes are identical in the intervention alternatives. Identical effectiveness should be proven in clinical studies.
In cost-effectiveness analysis (CEA) outcomes are measured in real units. Possible outcomes of CEAs are clinical parameters like blood pressure or lung function capacity, health care outcomes like screened persons and detected cases of illness, and (intermediate or final) health outcomes like prevented myocardial infarctions, prevented fatalities or life years gained. CEAs are particularly appropriate when a dominant effect parameter is identified. The importance of an effect parameter should be appraised from the patients' perspective. Final outcomes tend to be particularly relevant. Clinical outcomes will be important when – based on epidemiological studies – an extrapolation is possible from clinical to final outcomes. Decision models will be constructed for the analysis of long-term final outcomes. Finally, in CEAs cost and outcome differences between intervention alternatives will be integrated to cost-effectiveness ratios.
In cost-utility analyses (CUA) multiple outcome parameters are considered. Outcomes are transferred into utility units (using a valuation algorithm), and integrated into a unique utility parameter. In general, global outcome parameters life years gained and quality of life during the remaining lifetime are integrated into an index, the so-called quality adjusted life years (QALYs). A QALY denotes numerically an additional year in complete health. CUAs support comparisons across different indications – in particular comparisons of life prolonging interventions with mere QoL improving interventions (e.g., a back school programme and a bypass operation).
In cost-benefit analyses outcomes are calculated in monetised units (similar to cost parameters). Thus multiple outcomes are considered (like in CUAs) and moreover, costs and benefits are directly comparable. A

net benefit (benefits minus costs) can be calculated, providing – at least in theory – a unique decision criterion: if benefits exceed costs (i.e. positive net benefit) the adoption of the innovative intervention is recommended. If costs exceed benefits (i.e. negative net benefits) adoption of the innovative intervention is rejected. Moreover, CBA allows comparisons of investments in health and for example investments in education or environment.
Most health economic analyses of screening programmes are cost-effectiveness analyses (costs per detected case). In long-term analyses also cost-utility analyses (particularly for cancer screening programmes) and (restricted) cost-benefit analyses are applied. In CBAs costs of the screening programme are compared to savings in direct and/or indirect costs (due to screening programme) and (monetised) benefits. Thus, screening programmes are efficient if

- net savings are realised (i.e. discounted savings in direct/indirect follow-up costs overcompensate programme costs) and health outcomes do not worsen (restricted CBA)
- health outcomes like life years or QALYs are gained at relative lower costs (CEA and CUA).

3.3 Incremental analysis

In health economic analysis a comparison of treatment alternatives is performed. The comparison is based on the fundamental concept of incremental analysis. According to the incremental concept, cost and outcome (e.g., health effect or quality-adjusted life year) differences between two or more alternatives are derived (i.e. incremental costs and outcomes). Incremental costs and incremental effects or quality-adjusted life years form the core result of a health economic evaluation, the incremental cost-effectiveness ratio (see Table 1).
According to guidelines for health economic analysis, it is crucial to select appropriate comparators. Relevant comparators are (1) the dominant intervention (or an intervention mix based on market share weights), (2) the most cost-effective alternative and (3) the cost-minimal or do-nothing alternative (if relevant) [5], [6].

3.4 Perspectives of the health economic analysis

Health economic evaluation can be performed from different perspectives, e.g., the societal perspective, the payers' perspective or the providers' perspective. The different perspectives must clearly be distinguished. They determine how costs and health effects of the alternatives are defined, measured and valuated.
The societal perspective is the broadest perspective. All costs are included, regardless of who will incur them: health insurance, public sectors (e.g., education in special schools for hearing-impaired children), the patients and their families (e.g., time and travel expenses), or the rest of the society (e.g., production losses). International guidelines recommend to use a societal perspective in all health economic evaluations [5], [8], [9], [10].
In addition, other perspectives can be used, e.g. the payers' perspective [11], the providers' perspective or the patients' perspective. Health technologies should also be evaluated from the decision-maker's viewpoint to check whether the decision maker's appraisal is or is not consistent with the societal appraisal.

3.5 Costs

Cost estimation is based on a four-step process:

1. identification of the relevant cost items
2. measuring resource use
3. valuation of resource units
4. calculating total costs of the intervention alternatives.

Costs are defined as valuated resource consumption. Resource use is measured in quantity units, and valuation is based on opportunity costs (see below). From a societal perspective, costs in health economic evaluations are commonly classified into [5], [12] (see Table 2):

- direct medical costs
- direct non-medical costs
- indirect costs.

Direct costs refer to the resource consumption in the provision of health care interventions. They encompass the entire current resource use (e.g., the costs of a mammography screening programme) as well as future resource use attributable to the programme (e.g., validation of test results and diagnostics, costs or cost savings associated with breast cancer or prevented breast cancer). Future costs can span a lifetime in some indications. Direct costs are differentiated into direct medical and direct non-medical costs. Direct medical costs refer to the resource consumption in the health care sector associated with the production of health interventions. Resource consumption includes, for example, the costs of hospital stays, outpatient visits, pharmaceuticals and devices. Direct non-medical costs refer to resources supporting the medical production in the health care sector. These are, for example, transportation costs to medical interventions, child care costs for an ill parent, time of patients in the co-production of medical interventions, and time of family members (or volunteers) in informal care for ill or disabled patients.
For the quantification of resource use a range of costing approaches exists with micro-costing and gross-costing defining the ends of the range [1], [3], [4], [6], [12], [13]. In gross-costing, composite intermediate products and services (e.g. inpatient days) will be identified and measured. Micro-costing, on the other hand, starts with a detailed identification and measurement of services (e.g. a hospital stay will be split into components like consultation, operation, medication, diagnostics, nursing, housing, food, cleaning, overheads etc) and determines the required resource use (personnel, material, equipment, building, overhead etc.).

Table 1: Approaches of health economic evaluation

Approach	Evaluation criterion
Cost-minimisation analysis	$\min[C1, C2]$
Cost-effectiveness analysis	$\frac{C1 - C2}{E1 - E2}$
Cost-utility analysis	$\frac{C1 - C2}{U1 - U2}$
Cost-benefit analysis	$(B1 - C1) - (B2 - C2)$
C Costs	
E Effects	
U QALYs	
B Monetised benefit	
1 New technology	
2 Comparator	

Table 2: Items of resource use and health economic evaluations

Cost categories	Resource consumption
Direct medical costs	Outpatient visits - general practitioner - specialist Procedures and diagnostics - tests - diagnostic imaging - surgical interventions Pharmaceuticals Physiotherapy Medical devices Hospital stays Rehabilitation Services - home care - nursing care
Direct non-medical costs	Patient time - in treatment - in health activities - time expenditure due to illness Informal care Services Devices and investments Transportation

The valuation of health care interventions should be based on opportunity costs. Opportunity costs refer to the benefit of resources from the next best alternative use. As is shown in micro-economic theory, opportunity costs are reflected in the market prices of a perfectly competitive market. From a societal perspective valuation based on micro-costing is preferred as most health care resources are negotiated on competitive markets (though an efficient resource use is not guaranteed). Prices for health care services are often regulated by public institutions (e.g, Uniform Value Scale for outpatient services and DRGs for inpatient services) or negotiated between associations of providers and payers (e.g. outpatient budgets). Nevertheless, these prices can be used as an approximation of opportunity costs – with some well-founded adjustments where required [14].

Indirect costs denote the production losses due to

- unfitness for work (in the case of illness)

- early retirement/incapacity for work (in the case of long-term illness or disability)
- premature death.

For the valuation of productivity costs, there are two fundamental methods, the human capital approach and the friction cost approach. The human capital approach (HCA) suggests, that health care interventions are a kind of investment in an individual's human capital (similar to, for example, education). HCA rests on neoclassical theory of the firm. According to neoclassical theory, profit-maximising firms expand their labour input until marginal revenue product of labour equals unit labour costs (gross wage plus payroll related costs) – assuming diminishing marginal productivity of labour. Thus, according to the human capital approach (HCA), valuation of production losses is based on labour costs. All future productivity losses (up to retirement age) will be considered in HCA.
The friction cost approach (FCA) was developed to overcome some unrealistic assumptions of the HCA, particularly the assumption of perfectly competitive labour markets, which implies existence of full employment (in contrast to empirical experience of substantial unemployment in many countries) [15]. FCA suggests that, for long-term incapacity to work, costs of production loss are limited to a so-called friction period – i.e. until a patient will be replaced with a previously-unemployed individual and the former production level will be restored (which needs time for searching and training the previously-unemployed). Costs in the FCA encompass production loss in the friction period and transaction costs (searching and training the previously-unemployed individual). For short-term unfitness to work (within the friction period), FCA argues that part of the workload might be performed by colleagues of the patient or made up by the patient after return to work. Thus, short-term production losses might be less than labour costs (according to HCA). Empirical studies in the Netherlands found that short-term costs are about 80 percent of labour costs [16]. Thus, the cost difference between HCA and FCA may be small for short-term absence from work while it will increase markedly for long-term absence from work as HCA considers all future productivity losses, while FCA is limited to the friction period.
There is an ongoing debate in the literature whether HCA or FCA is better representing productivity costs. As mentioned above, the human capital approach rests on some unrealistic assumptions (particularly full employment in the labour market). HCA rather shows potential then real production losses. In contrast, FCA is focused on real production losses. Nevertheless, FCA has been criticised, too. In particular, the assumption of zero opportunity costs of labour after the friction period has implications on the calculation of direct medical costs. Then, it is argued, opportunity costs of labour inputs in health care are nearly zero, too (because health care workers could be substituted by a previously-unemployed individual at nearly zero cost – only transaction costs for searching and training the previously-unemployed individual would accrue). In conclusion, it might be argued that HCA is overestimating and FCA is underestimating opportunity costs of paid work. Thus, according to international guidelines sensitivity analyses are recommended [5].

3.6 Quality of life and quality-adjusted life years

Health-related quality of life (HRQL) is an increasingly important outcome parameter in health economic evaluation. In the concept of health-related quality of life, the general concept of quality of life (QoL) (including wealth, liberty, education, culture and religion, amongst others) is limited to health relevant dimensions. The definition of health-related quality of life is derived from the WHO-definition of health [17] – and comprises

- physical health
- mental health or emotional wellbeing
- social integration.

Approaches for measuring HRQL can be grouped into indication specific measures, generic profile measures (e.g., SF-36), and preference-based measures (e.g., EQ-5D) [5]. Preference-based approaches provide a quality of life index (i.e. a score reflecting HRQL numerically). Like generic profile measures they can be used for comparisons across indications. Preference-based approaches are less differentiated and less sensitive than generic profile measures (and even less sensitive than specific measures). But – providing a QoL-index – the preference-based measures allow unique comparisons of intervention alternatives and thus are the only suitable approach for cost-utility analyses.
The concept of quality-adjusted life years (QALYs) integrates health-related quality of life and (remaining) life expectancy into a one-dimensional outcome parameter [18], [19]. The QALY approach is focused on the subjective health of patients. With quality of life and life expectancy the most relevant dimensions of health (from the perspective of the individual) are considered.
The QALY approach allows comparisons across different indications – in particular comparisons of life prolonging interventions with mere QoL improving interventions (i.e. not life prolonging interventions). HRQL scores are measured on a cardinal scale ranging from 0 (death) to 1 (complete health). A QALY denotes numerically one year in complete health – or for example two years in a health state valued 0.5. The QALY measure corresponds to the integral of the QoL-function (over time) or – graphical – the area under the QoL-index curve (in Figure 1). In a discrete approach (with time as discrete parameter) QALYs are the product of QoL of a health state and the period the health state is realised.

3.7 Time horizon of the analysis

The time frame should be long enough to capture all relevant cost and outcome differences between the programmes compared in the health economic analysis. For

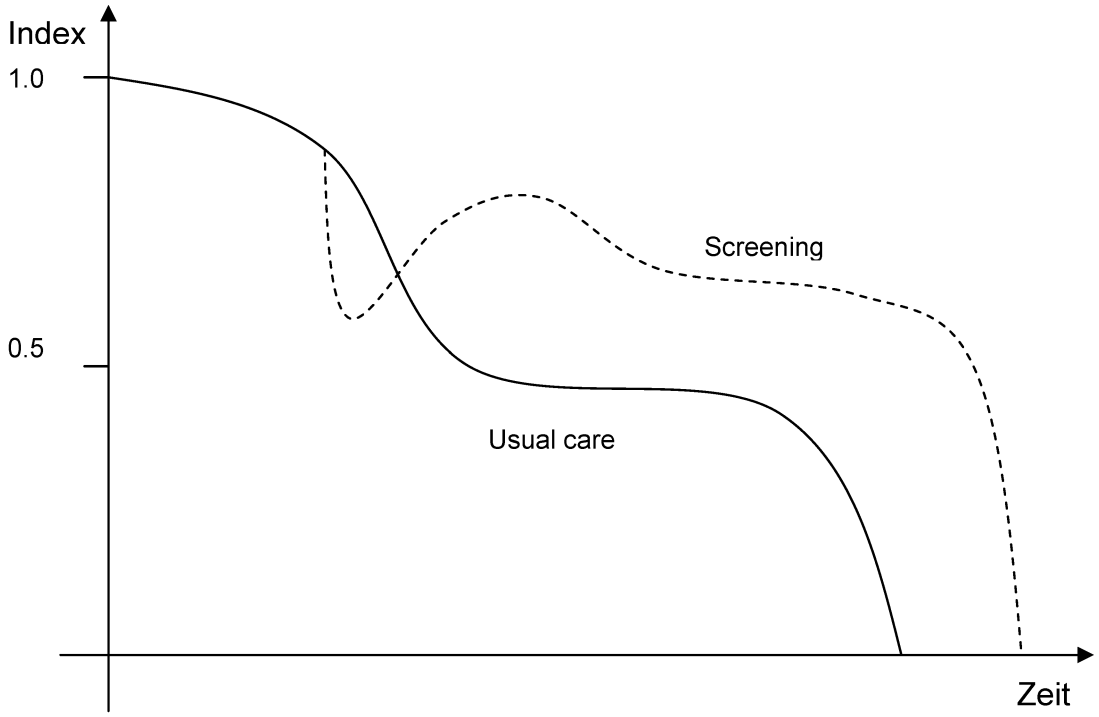

Figure 1: Quality of Life and QALYs: Screening versus usual care

chronic conditions, a time horizon spanning to a lifetime often is required (particularly if lifespan gains are expected).
Often, the appropriate time horizon exceeds the available primary data (from prospective clinical studies), because prospective studies mostly examine intermediate outcomes (e.g., reduction of blood pressure or improvement of lung function capacity). Then it is appropriate to use several time horizons in economic evaluations: a short-term analysis that is based only on data from prospective studies (e.g., detected cases in a mammography screening programme) and a long-term analysis that includes modelling data – based on epidemiologic studies (e.g., effect of a sooner treatment on mortality, quality of life und health care costs).

3.8 Discounting

There is a broad consensus in the literature, that costs (and outcomes) should be discounted to their present value to adjust for differential timing [20]. Discounting reflects the positive time preference of individuals, i.e. individuals prefer present to future benefits (as it leaves more options). Reasons for a positive time preference are (1) diminishing marginal utility of consumption, combined with expected increasing future incomes (assuming positive economic growth) and (2) risk of lifespan, i.e. risk whether future consumptions will be available. As is shown in neoclassical theory, assuming perfect markets with certain information about future (i.e. no risk) and absence of taxation, the time preference will equal the market interest rate (i.e. the opportunity cost of capital) – indicating the social discount rate. Individuals will only forgo present consumption, if future consumption (based on investment) is exceeding present consumption.
Usual discount rates range from 0 to 10 percent, often rates of 5 or 3 percent are suggested in international guidelines (for both, costs and outcomes). The discussion of the so-called Keeler-Cretin paradox [20] resulted in the recommendation of health economic guidelines to discount costs and effects at an identical discount rate [5].

3.9 Efficacy versus community effectiveness

Randomised controlled trials (RCTs) form the most appropriate study design to identify effects of an intervention. Thus, Randomised controlled trials form the analytic gold standard in health economic evaluation studies – like in clinical studies. Though, health economic analysis is less interested in efficacy (i.e. effects under strictly controlled study conditions), but more in community effectiveness (i.e. effects under everyday life conditions) of health care services. RCTs often are characterised by specific limitations. Patients are included into the study by restrictive inclusion and/or exclusion criteria (e.g., having no other diseases). Furthermore, the clinical personnel are highly motivated and the patients are particularly compliant.
Evaluation of community effectiveness will often not be based on RCTs, but on observational studies with control groups (where the allocation of patients to intervention or control group is not randomised but depends on decisions of physicians and/or patients). Using appropriate multivariate statistics, ex-post comparableness of intervention and control group is supported. Often the out-

comes of RCTs will be extrapolated to everyday life conditions using epidemiologic models.

3.10 Analysis of uncertainty

In empirical evaluation studies the determination of cost and outcome parameters is based on uncertain data bases. Uncertainty rests on [5], [21]

- sample variations of patient-based data in prospective studies
- point estimates of prices for health care services and resource use. Point estimates are based on data of health care providers, health insurances and/or expert opinion
- extrapolation of intermediate outcomes to final outcomes (e.g., blood pressure to life expectancy) in decision models.

Health economic evaluation studies should report uncertainty of cost and outcome parameters. Methods for controlling parameter uncertainty due to sampling variations in prospective studies are statistical methods of hypothesis testing and bootstrapping [6]. The effects of other uncertain assumptions – price estimates of services and resources and extrapolations – are examined in (deterministic and stochastic) sensitivity analyses, i.e. variations of parameters which are particularly relevant for the study results.

4 Health economic evaluation of newborn hearing screening

In this section the methodical process of health economic evaluation is illustrated using the example of newborn hearing screening. Specific requirements in the empirical analysis of screening programmes are discussed.

4.1 Screening for congenital hearing loss

The prevalence of moderate to profound bilateral congenital hearing loss is about 1 to 3 per 1,000 newborns [22]. When mild hearing impairment is included, prevalence is about 5 to 6 per 1,000 newborns [23], [24]. 1 to 3 per 100 newborns is affected in the subgroup of high-risk children [25], [26].
On average, congenital hearing impairment is diagnosed at the age of 31 months (if no newborn hearing screening exists) [27], and initial provision of hearing aids takes four more months. Moderate hearing impairment is identified at the age of 46 months and mild hearing impairment at the age of 48 months [27], [28], [29].
Delayed identification of hearing impairment implies that a crucial period (ranging from 6 to 30 months of age [30]) for language acquisition and verbal communication abilities is missed – a period when neuronal structures for language acquisition (which are high-sensitive to auditory stimuli) are developed [31], [13]). Hearing impairment results in deficits in language acquisition and verbal communication competence. Delayed identification also produces subsequent impairments of cognitive, intellectual, emotional und psychosocial development. Furthermore, access to education and vocation is more difficult. Thus, it is required that infant hearing loss is identified and treated in due time. Most experts agree that hearing impairment should be identified before the age of 3 months and appropriate interventions (hearing aid or cochlear implant, early language promotion) should be initiated before the age of 6 months. Newborn hearing screening is a detection strategy to identify hearing impairment in due time. Two alternative tests methods for newborn hearing screening exist: otoacoustic emissions (OAE) and auditory brainstem response (ABR).

4.2 Questions

The following questions are addressed by health economic analyses of newborn hearing screening

1. How high are the costs of newborn hearing screening?
2. Is newborn hearing screening cost-effective compared to non-screening (i.e. usual treatment in a later – symptomatic – phase of a target disease)?
3. Is a universal hearing screening cost-effective compared to selective hearing screening for high-risk infants?
4. Which institutional access for universal newborn hearing screening is more cost-effective – hospitals or paediatric practices?
5. Which access is reaching more children for screening?
6. Which long-term outcomes are realised by (universal) newborn hearing screening?

These questions are discussed in the following sections.

4.3 Methods

Nine health economic studies of newborn hearing screening (since 1998) were identified ([22], [16] and own literature search). Most studies restricted their analysis to screening test and confirmation. Only one study [32] described long-term costs and outcomes.

4.4 Costs of newborn hearing screening

There are several studies calculating costs of newborn hearing screening [32], [33], [34], [35], [36], [37], [38], [39]. In Germany one cost-analysis was conducted under everyday life conditions (the so-called Hannover Model Project) [40].
The Hannover Model Project evaluated the introduction of a universal hearing screening in the Hannover region. Screening was executed in all maternity clinics (well babies) and neonatal intensive care units (NICU) (high-risk infants) of the Hannover region. For ambulant births and home births screening was delegated to a limited number of office-based otorhinolaryngologists. The screening test method applied was OAE. Between 07/2000 and

12/2002 17,920 newborns were screened by the hearing screening programme.
In the Hannover Model Project a micro-costing approach was applied for calculating screening costs. The following programme components were included

- programme development
- programme implementation
- monitoring
- initial OAE-measurement in clinics
- confirmation of test-positive results (in selected otorhinolaryngologist's practices).

The micro-costing approach is based on quantification and valuation of resource use – personnel, materials, equipment, building and overheads [41]. Labour utilisation formed a dominant part of screening costs. Thus, labour utilisation in the screening programme was precisely calculated, using time records, oral interviews and documentations. Valuation of labour utilisation was based on employers' labour costs.
Programme implementation included information, instructions (how to administer the screening test), motivation of and assistance to hospitals and participating practices during the introduction of the screening programme in the Hannover region. The monitoring process aimed at giving prompt feedbacks to hospitals on their screening rates (i.e., screened children as percentage of all births in a clinic). For both, implementation and monitoring, resource use (particularly personnel) of the screening coordination unit (located at the Hannover Model Project), and of clinics and practices was quantified.
OAE-devices (depreciation and interest) and personnel (information of parents, measurement, documentation of measurement results, and organisation of the initial screening test) were the dominant cost components of the initial screening test for hearing loss conducted in clinics. Time expenditure for complete OAE-measurement per child (including repetitions if necessary) was about 10 minutes, and costs of measurement per child were about 10 euro. Confirmation of test-positive screening results was conducted by certificated otorhinolaryngologist's practices. Costs of confirmation were estimated to 23.1 euro per false-positive case and 80 euro per true-positive case (due to larger expenditures as the total process of confirmation must be applied). Screening costs (for programme development and implementation, monitoring and initial OAE-measurement, but not confirmation) amounted to about 23 euro per child screened (see Table 3).
As the Hannover Model Project evaluated the introduction of a newborn hearing screening programme, substantial experience-based cost savings are expected (e.g., for instruction, assistance and monitoring of clinics and practices) when the screening programme is run longer. For example, in industrial production experience curve effects are verified by empirical studies. Average cost reductions are estimated to about 20% of unit production costs if cumulated output is doubled [42]. Furthermore, costs per child screened for programme development and implementation is decreasing the longer the screening programme is existing (i.e., the more newborns are screened) as implementation costs only accrue during the introduction period. In the present calculation implementation costs are about 36% of total screening costs. Thus, substantial reductions of screening costs are expected in the long run. A model analysis calculated long-term costs of about 16 euro per child screened (including confirmation costs). The average costs per case identified are about 15,500 euro.
In international studies screening costs range from 14 to 25 euro per child screened [32], [33], [34], [36], and costs per case detected range from 5,000 to 31,000 euro [32], [34], [36], [39]. Thus, German screening costs are in the range of international screening costs.

4.5 Cost-effectiveness analysis of newborn hearing screening – comparison of universal screening, screening of high-risk children, and non-screening

Average costs per identified case (as presented in section 4.3) are not relevant to health economic decisions. Decisions are rather based on comparative health economic evaluations calculating cost and outcome (e.g., health effect or quality-adjusted life year) differences between two or more relevant intervention alternatives. Thus, in the present analyses incremental costs and outcomes of a screening programme (compared to intervention alternatives) are determined. The incremental cost-effectiveness ratio of a screening programme describes the additional costs per additional case identified (compared to relevant intervention alternatives).
Only one study (of nine health economic studies of newborn hearing screening identified in a systematic literature search (see section 4.3)) is performing a comparison of screening to non-screening [32]. Keren et al. compare (1) a universal newborn hearing screening, (2) newborn hearing screening of high-risk children, and (3) non-screening. Even if no screening programme is implemented some cases are identified in due time – particularly children with profound hearing loss. Then, costs of identification and confirmation must be considered.
In the model analysis by Keren et al., 116 cases (of 128 expected cases from a birth cohort of 80,000 children) are identified during the first six months of age if a universal hearing screening is implemented, 62 cases if screening is restricted to high-risk newborns, and still 30 cases if no screening is implemented. The average costs per identified case amount to 2,300 euro for non-screening, 10,100 euro for screening high-risk children, and 21,700 euro for universal screening.
Keren et al. calculate incremental cost-effectiveness ratios for screening high-risk infants compared to non-screening and (2) for universal screening compared to screening high-risk infants. (1) Incremental costs amount to 16,600 euro per additional identified case for screen-

Table 3: Screening costs ((without confirmation costs)

Cost component	*Cost per child*	*Number of screened children*	*Total Costs*
I. Implementation costs	8.28 €	19,703	163,140 €
II. Monitoring costs	3.22 €	19,703	63,444 €
III. Cost of initial measurement			
- maternity clinics	10.27 €	16,251	166,898 €
- neonatal intensive care units	10.27 €	1,669	17,140 €
- Practices	23.10 €	1,783	41,187 €
Costs of Screening			451,809 €
Cost per child screened			22.93 €

ing high-risk newborns compared to non-screening (detecting 32 additional cases in the first six months of age) and (2) to 45,000 euro per additional identified case for universal screening compared to screening high-risk children (detecting 54 additional cases).
The analysis demonstrates that incremental costs increase compared to average costs (45,000 euro versus 21,700 euro for universal hearing screening and 16,600 euro versus 10,100 euro for screening high-risk children).

4.6 Analysis of participant rates and costs of different institutional access to newborn hearing screening

Based on the empirical results of the Hannover Model Project (see section 4.4) model analyses of newborn hearing screening for Germany (in 2001) were performed. Alternative institutional access – screening in hospitals (model 1) versus screening in paediatric practices (model 2) – were compared regarding participation rates and programme costs. A similar comparison of hospital and ambulant screening was modelled for the UK health care system [43].

Model 1 ("inpatient newborn hearing screening")

In model 1 newborn hearing screening is performed in maternity clinics (well babies) and neonatal intensive care units (high-risk infants). For ambulant and home births screening was delegated to a limited number of office-based otorhinolaryngologists. This institutional screening structure of model 1 corresponds to the screening programme of Hannover Model Project.

Model 2 ("ambulant newborn hearing screening")

In model 2 newborn hearing screening is performed in paediatric practices during routine well child visits (so-called U3 at 4 to 6 weeks of age) and neonatal intensive care units (high-risk infants).
The assumptions of the model analyses are summarised in Table 4. Prevalence rates, validity (sensitivity and specificity), distribution of children to the screening institutions (maternity clinics, neonatal intensive care units, practices) in model 1, and screening rates of these screening institutions (i.e. screened children as percentage of all births in an institution) are based on the empirical results of the Hannover Model Project.
Models 1 and 2 differ in screening rates and specificity of the screening test. The Hannover Model Project showed differences in test specificity between clinics and practices (about 90% in clinics and 80% in practices). The lower specificity is due to worse screening conditions in practices (e.g., narrow time period for screening). However, the estimated screening rate is much higher in paediatric practices of model 2 (98%) than in maternity clinics of model 1 (93.5%). The assumed screening rate in paediatric practices corresponds to the nationwide participation rate of U3 routine well child visits. The screening rate in maternity clinics is lower because a substantial part of newborns leaves hospital before valid test results can be produced (i.e. before 2 to 3 days after birth).
In model 1 (inpatient newborn hearing screening) about 50,000 newborns (of 735,000 births per year in Germany) miss screening. 710 hard of hearing children are detected and 93 hearing-impaired newborns are not identified by the inpatient screening programme. About 73,000 false-positive cases must undergo confirmation diagnostics.
In model 2 (ambulant newborn hearing screening) about 16,000 children are not screened. 741 hearing-impaired newborns are detected and 62 hard of hearing children are not identified by the ambulant screening programme. About 136,000 false-positive cases must undergo confirmation diagnostics. Thus, ambulant screening (model 2) identifies 31 more hearing-impaired newborns than inpatient screening (model 1). However, significantly (i.e. about 63,000) more test-positive children than in model 1 need confirmation diagnostics. Cost differences between models 1 and 2 are due to

- higher investments in OAE-devices and higher time expenditures for monitoring in model 2 (ambulant screening) as there are much more paediatric practices than maternity clinics

Table 4: Parameters of model analysis

General data		
Births	735,000	
Prevalence		
- Well babies	0.65/1,000	
- High-risk children	4.47/1,000	
Screening test		
- Sensitivity	95%	
- Specificity	90% (clinic)	
	80% (practice)	
Model 1		
	Percentage of all births	Screening rates
Maternity clinics	82%	93.5%
NICU	12%	92.7%
Practice	6%	90.5%
Model 2		
	Percentage of all births	Percentage of all births
Paediatric practice	88%	98.0%
NICU	12%	92.7%

- higher expenditures for confirmation diagnostics in model 2 (ambulant screening) as significantly more false-positive test results are produced
- larger number of screened infants in model 2 (ambulant screening).

In model 1 (inpatient screening) total screening costs (for implementation, monitoring, initial measurement, and confirmation) are estimated to 11 million euro if the screening programme is extended to Germany. The average costs per screened newborn are 16.1 euro (with about 685,000 screened children), and average screening costs per case identified amount to 15,500 euro (with 710 hearing-impaired children detected).
In model 2 (ambulant screening) total screening costs are estimated to 24.2 million for Germany. The average costs per screened newborn are 33.8 euro (with about 718,000 screened children), and average screening costs per case detected amount to 32,700 euro (with 741 hearing-impaired newborns identified).
Thus, programme costs of ambulant screening model 2 are about 13.2 million euro more expensive than programme costs of inpatient model 1. However, ambulant screening model 2 identifies 31 more hearing-impaired newborns than inpatient screening model 1. The incremental cost-effectiveness ratio – i.e. additional costs per additional case identified of ambulant screening compared to inpatient screening – is 426,000 euro (13.2 million euro/31 cases). Whether model 2 is cost-effective must be decided by decision makers (e.g., Federal Joint Committee for Germany). Long-term outcomes should be considered when a decision is made on the nationwide introduction of newborn hearing screening.

4.7 Long-term outcomes of newborn hearing screening

Most health economic studies of newborn hearing screening analyse costs per identified case which is an indicator for short-term cost-effectiveness. However, the time frame should be long enough to capture all relevant cost and outcome differences between intervention alternatives compared in health economic analyses. To evaluate interventions for hearing-impaired children, a time horizon spanning to a lifetime is required. The cost calculation includes

- health care costs
- cost of education and
- loss of human capital (due to lower education).

The small number of long-term health economic analyses is due the small number of long-term effectiveness studies analysing screening and/or early interventions (e.g., hearing aid or cochlear implant).
Only two studies (of nine health economic studies of newborn hearing screening identified in a systematic literature search (see section 4.3)) evaluate the long-term effects of newborn hearing screening [44], [45]. More studies examine how long-term outcomes depend on detection age of hearing loss and/or intervention age (e.g., hearing aid or cochlear implant, early language promotion) [46], [47], [48], [49], [50], [51], [52], [53], [54], [55]. The following long-term outcome parameters are considered

- language acquisition
- cognitive development
- pre-school and school performance (education costs)
- professional career (human capital)
- quality of life.

Most studies examine (receptive or expressive) language development [44], [45], [46], [47], [48], [49], [50], [51], [52], but only few studies analyse school performance and professional career [53], [54], [55] as long-term observations of at least 6 and 16 years are required, respectively. Furthermore, most evaluations focus on cochlear implant supply for children with profound hearing loss [48], [49], [50], [51], [52]. Though most gain in time (and benefit) of newborn hearing screening is expected for infants with moderate hearing loss.

The studies' results on long-term outcomes can be summarised as follows: Universal hearing screening shows significantly positive effects on receptive language development of hearing-impaired children [44], [45], but only non-significant (positive) effects on expressive language development [45]. Similar results are found for earlier compared to later diagnosis and intervention [46].

In the subgroup of children with profound hearing loss, cochlear implants show better effects on general language development than hearing aids. This is particularly true if the cochlear implant is supplied before the age of 4 years [56]. Language acquisition is the better the sooner cochlear implants are supplied [50], [52]. Studies of school performance and professional career exist only for deaf children. Again, children with earlier cochlear implantation are more likely to visit regular kindergarten and regular schools, respectively [53], [54].

In most studies confounding parameters (like education and income of parents, and family support) are not controlled. Thus, the existing studies of long-term outcomes are insufficient to prove long-term cost-effectiveness of newborn hearing screening.

5 Discussion and conclusion

This article examined health economic implications of screening. Requirements screening programmes should fulfil were developed. Main requirements for the implementation of screening programmes are

- the target disease should be a substantial health problem
- an effective treatment should be available. Treatment outcomes should be the better the earlier the disease is detected
- the disease should be diagnosable in a latent or presymptomatic phase
- a simple, safe and sufficiently precise screening test should exist to detect the target disease
- an adequate infrastructure for screening, diagnostics, and treatment should exist.

Requirements the evaluation of screening programmes should fulfil are as follows

- systematic errors should be avoided. In the evaluation of screening tests lead time bias and length time bias are particularly relevant. The effects of these two biases should be controlled by randomised controlled trials
- the time frame of a health economic analysis should be long enough to capture all relevant cost and outcome differences between the intervention alternatives. Often, long-term effects are presented in decision models
- health economic evaluations are based on a comparison of relevant intervention alternatives (e.g., screening versus non-screening). The main health economic criterion is the incremental cost-effectiveness ratio (e.g., additional costs per additional case identified).

An application of these requirements to the example of newborn hearing screening shows the following results

- health economic studies of newborn hearing screening are not randomised, most studies are even not controlled
- thus, most studies do not present incremental costs per additional case identified by newborn hearing screening (compared to non-screening), but only average costs per identified case
- most studies focus on short-term outcomes of newborn hearing screening (i.e. identified cases). Only one model analysis examines long-term outcomes
- the evidence on long-term outcomes is not sufficient to develop health economic decision models.

Obviously, the empirical studies of newborn hearing screening do not meet all requirements health economic evaluation of screening should meet. However, empirical studies cannot fulfil all requirements. Particularly, a randomised-controlled trial of universal newborn hearing screening is not feasible. It would need about 100 study regions with each 1 million inhabitants to find significant outcome differences. Recruitment of control regions for a controlled trial is difficult as control regions are not motivated to participate in the trial. Furthermore, at least unsystematic hearing screening is provided in most regions. Thus, an improvement of outcomes (e.g., an increase in identified cases) of screening compared to non-screening cannot be proved. Nevertheless, there is a need for controlled trials (to examine differences in identified cases, but particularly to examine long-term effects).

In the end, (short-term) outcomes of a screening programme depend on programme infrastructure, process management including established control routines, and capacities for screening and confirmation diagnostics.

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