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Societal costs of hearing disorders- a systematic and critical review of literature

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Acronyms and abbreviations:

COI	Cost-of-illness
dB HL	decibel Hearing Level
FCM	Friction cost method
HCM	Human capital method
PAF	Population attributable risk
PCHI	Permanent Childhood Hearing Impairment
PTA	Pure-tone average
RR	Relative risk
WTP	Willingness to pay

Abstract

Objective

The objective of this study was to perform a critical and systematic literature review of studies on societal costs due to hearing disorders.

Design

We used pre-defined search terms and inclusion/exclusion criteria. Systematic searches were conducted in Medline, Cochrane Library, Google scholar and other relevant websites. The review included studies written in English or Swedish between 1995 and the end of January 2012.

Study sample

We identified 4 published studies and 4 reports that met the pre-defined inclusion criteria.

Results

Swedish cost studies primarily focused on costs of hearing aids. International studies with a societal perspective used different costing approaches and were limited to specific patient populations. Hearing disorders impact the social welfare system more than the medical care system. Indirect costs account for the major part and direct medical costs for a minor part of the total costs of hearing disorders.

Conclusions

There is a need for further studies estimating societal costs for all degrees of hearing disorders, in particular since a large part of the people with hearing disorders are of working age.

1. Introduction

Hearing disorders

The hearing sense is one of the main ways to receive information. The 'survival of the fittest' has in the communication age of the 21st century been redefined to a person's ability to communicate effectively and much employment today is more based on communication than on manual work (Ruben 2000). Accordingly, a hearing disorder probably not only affects the physical, mental and emotional health but also job performance and cost to the society.

In this article, a hearing disorder is defined as self-assessed hearing loss or hearing loss estimated by the pure-tone audiogram. Based on pure-tone audiogram, the Swedish National Board of Health and Welfare has estimated that approximately 2 million people in Sweden (approximately 20% of the Swedish population) have some form of hearing disorder (Socialstyrelsen 2009). The majority has a mild disorder while around 0.5 million, approximately 5% of the Swedish population, has a moderate disorder. Approximately 130,000 persons, approximately 1.5% of the Swedish population, have severe hearing disorder or total hearing loss in both ears. A mild hearing disorder is defined as a pure-tone average (PTA; frequencies 500, 1000, 2000, and 4000 Hz) 20-39 dB Hearing Level (HL), moderate disorder is defined as PTA 40-64 dB HL and severe hearing disorder is defined as PTA ≥65 dB HL. Hearing disorder is significantly increasing with age and more men than women suffer from hearing disorders (Socialstyrelsen 2009).

Another way of estimating the prevalence of hearing disorders is by using surveys with questions about self-assessed hearing disorders. Studies have found significant correlations between measured and self-assessed hearing (Pedersen and Rosenhall 1991; Karlsson and Rosenhall 1998; Nondahl, Cruickshanks et

al. 1998). According to a survey about living conditions conducted by Statistics Sweden (ULF) in 2008, over 17% of the included persons reported that they had hearing loss (Statistics Sweden 2009). However, the prevalence varies throughout the country and the highest prevalence is found in the countryside while the lowest prevalence is found in the larger cities.

Congenital hearing disorders are relatively uncommon in Sweden, but hearing disorder is one of the most common functional disabilities in the work place in Sweden. The proportion of persons under 65 years with a hearing disorder has increased during the last 20 years and in 2009 approximately 55% of all persons with hearing disorders in Sweden were in the ages 16-64, i.e. 717, 000 persons (The Swedish Association of Hard of Hearing People HRF 2009). This may have a significant effect on societal costs since the proportion of hearing impaired persons with early retirement is almost twice as large as that for the general population, 9.3% versus 5.3% (The Swedish Association of Hard of Hearing People HRF 2009). A recent study on the position of individuals with total hearing loss in both ears in the Swedish labour market found that these individuals have a higher unemployment and a lower employment rate compared to a reference population randomly chosen from the total Swedish population by Statistics Sweden. Activity/sickness compensation is almost three times higher for the population with total hearing loss than the reference population (Rydberg, Gellerstedt et al. 2010). One study, based on self-reported psychosocial work environment in a group of hearing-impaired people in Sweden, found that hearing-impaired people with highly stressful work reported poorer physical health status and lower psychological well-being than normal-hearing peers (Danermark and Gellerstedt 2004). Further, a Danish study based on a representative section of the Danish population aged 50-64 years investigated the impact of hearing disorder on the relation to the labour-market and working life (Tornhoj Christensen 2006). A hearing disorder was shown to contribute to early retirement and deterioration of the quality of working life among Danes aged 50-64. Hearing disorders in this population (50-64 years)

caused yearly productivity losses in terms of disability pension, early retirement benefits, unemployment, and change in working hours of approximately USD 470 million (DKK 2,667 million converted to 2011 USD).

Methodological issues in cost-of-illness

Cost-of-illness (COI) studies measure the economic burden of health problems and estimate the maximum amount that could potentially be saved or gained if the health problem was prevented, delayed, or eradicated. COI studies should not be used as economic evaluations but rather as sources of economic information for decision makers, researchers and the general public (Kortt, Langley et al. 1998; Drummond, Schulper et al. 2005). Methodological issues of COI include prevalence or incidence, cost definitions (external and internal costs comprising direct, indirect, and intangible components), cost measurement (database or modeling, top-down or bottom-up), the issue of attributable fraction and discount rate. Here, we report the main methodological issues and review their applicability for costs of hearing disorders.

COI research may be conducted with either prevalence or incidence based approaches (Kobelt 2002). The prevalence of a disease and its associated costs gives a moment-in-time assessment, e.g. the costs incurred during a specific year. The incidence based approach ideally provides an assessment of lifetime costs of all new cases during a specific time period. A third common approach in COI studies is a mixed incidence study that measures the costs of the prevalence of a disease at a specific moment in time and the costs that it yields over an extended time period.

To correctly measure the total burden of a disease from a societal perspective all costs should be included in the analysis. Costs consist of direct, indirect, and intangible costs. Direct costs include

resource use, such as costs of hearing aids, health care personnel and hospital facilities, but also costs of transportation to and from the hospital. Indirect costs are those resources not produced as a consequence of the disease and its treatment. They include productivity losses following increased absenteeism and a lower quality of performed work as well as increased mortality (Johannesson 1996). Intangible costs have no impact on resources and production but are related to pain and suffering resulting from a disease or treatment (Kobelt 2002).

Direct costs are estimated using the modeling or database approach. Modeling studies apply econometric methods to evaluate the impact of diseases on costs. Cross-sectional and longitudinal modeling studies may employ prevalence or incidence based approaches. Database studies stratify patients into groups of ill and healthy individuals and the costs are compared.

Indirect costs can be measured by, for example, the human capital method (HCM), the willingness to pay method (WTP) or the friction cost method (FCM) (Johannesson 1996; Drummond, Schulper et al. 2005). Human capital and its return are affected by the health status of individuals. Reduced production due to e.g. a health decline is quantified typically as the product of time absent from work and wage rate. The present value of lost incremental future earnings represents the reduced production. The HCM may be used for valuing domestic and voluntary work as well as evaluating productivity losses that are due to increased mortality. The HCM is associated with some difficulties. Firstly, labor market imperfections may lead to unemployment and thereby violating one of the assumptions of the HCM (i.e. the assumption of full employment). Secondly, the wage rates may not reflect the marginal productivity of workers since other factors may influence the wage rate, e.g. discrimination. Thirdly, COI research is often performed from a societal perspective but wage rate data reflect at best the productivity of the

working population and considerations have to be made regarding the proportion of the population that does not earn wages, e.g. homemakers (Drummond, Schulper et al. 2005).

The Willingness-to-pay (WTP), or contingent valuation, method is used for evaluating effects due to productivity changes but also for measuring health care costs and intangible costs (Drummond, Schulper et al. 2005). Hypothetical scenarios are presented to respondents and health changes are assigned values based on what the individual indicates that s/he is willing to pay or forego in order to stay at the original utility level but in a healthy state (Johannesson 1996). In such an evaluation, the focus is on the maximum amount the individual is willing to surrender so that contingency values reflecting productivity changes may be separated (Drummond, Schulper et al. 2005).

The friction cost method (FCM) presents the costs associated with the replacement of an ill worker (Koopmanschap, Rutten et al. 1995). The underlying concept for the use of friction costs is that production losses due to illness may not be as great as expected by the HCM because of an existing unemployment reserve that can absorb some of this lost productivity, i.e. solving the first difficulty of HCM mentioned above (Drummond, Schulper et al. 2005).

Intangible costs should also be assessed in COI studies, but the assessment is difficult and is often neglected. Health improvements do not only increase average life time but they also improve the value of life (Cutler and Richardson 1998). See (Perreira and Sloan 2002) for techniques to evaluate intangible costs associated with disability. There are quality of life measures (e.g. SF36) measuring health status, as well as utility measures that attempt to assess the individual's utility, most often on a scale of 0 to 1 where 0 is the value at death and 1 is the value of perfect health (Drummond, Schulper et al. 2005).

In COI studies, data for all costs are collected from a diverse selection of sources: national health care statistics, patient registries, cohort studies, insurance databases, patient charts or from patients themselves. Studies are made “top-down” or “bottom-up”. Top-down studies use databases and registries to estimate costs in a sample and require estimations of attributable fraction from the sample. The population attributable fraction (PAF), or attributable risk, is the proportion of the disease that is attributable to the risk factor. The PAF is closely related to the relative risk (RR) and may be estimated using aggregated RR and the prevalence (P) of the condition using the formula PAF=[P(RR-1)]/[1+P(RR-1)] (Katzmarzyk, Gledhill et al. 2000). The short-coming of the top-down approach is that data on some diseases may be lacking, and costs will thus be underestimated. In bottom-up studies, data are collected directly from patient samples, retrospectively or prospectively. Bottom-up studies do not require estimations of attributable fractions but necessitate the use of control groups to estimate the incremental costs that follow a disease. In the retrospective case, charts and questionnaires are used and in the prospective case a sample is followed over time. The challenge with this approach is to acquire an unbiased sample covering general population characteristics as closely as possible (Kobelt 2002).

Discounting is an economic method to capture an individual’s preference for income today rather than income in the future. Discounting allows calculating the present value of payments that occur in the future. The *de facto* convention has been the use of a discount rate of 5 per cent (Drummond, Schulper et al. 2005). Such a standard use facilitates comparisons across studies but may not actually reflect individual or societal preferences. Costs should be presented in such a way that discounting at other rates may be performed. Sensitivity analysis is another measure that improves the quality of studies and permits users to better evaluate the results (Drummond, Schulper et al. 2005).

Objective

The objective of this study is to perform a critical systematic literature review in order to find studies addressing societal costs due to hearing disorders for both adults and children in Sweden and internationally. The particular focus is on methodological issues and their impact on the results.

Following the introduction, Section 2 reports methods and results of hearing disorder COI studies in earlier and more recent research. The third and final section discusses the results and provides concluding remarks and suggestions for further research.

2. Methods and results of hearing disorder COI

Methods of the literature review

A systematic literature review was made to identify the costs of hearing disorders for both children and adults in Sweden and internationally. The search was conducted in January 2012. Medline (via PubMed) and Cochrane Library were used for searches of published papers. Since an initial search revealed a very limited number of relevant articles, we also, as a second step, used the search engine Google Scholar, the website of the Swedish Association of Hard of Hearing People (www.hrf.se) and the non-commercial website *hear-it (*www.hear-it.org, established and run by the organisation 'Hear-It AISBL', consisting of International Federation of the Hard of Hearing, Association Européenne des Audioprothésistes, European Hearing Industry Manufacturers Association, Knowles, Pulse and Gennumwere) in order to identify relevant unpublished literature. The first author conducted the literature search with the second author auditing the search. Borderline studies were decided on by discussions between all three authors. The scope of this review was restricted by using the following inclusion and exclusion criteria:

Studies were included in the review process if they:

- were published in the period between January 1995 and January 2012
- were written in English or Swedish
- had a comprehensive societal cost perspective, i.e. studies including both direct and indirect costs.

Studies were excluded if they:

- were a cost-effectiveness, a cost-utility, or a cost-benefit study
- focused on specific symptoms such as tinnitus (since we wanted to collect information on the effect of hearing loss rather than of specific symptoms)

The MeSH-index (Medical Subject Headings) was used to search for articles in the PubMed and Cochrane databases. The Google scholar was searched using the terms *cost and hearing* in both English (in title) and Swedish (in all text). Due to the special focus on Sweden and due to the lack of literature on societal costs of hearing disorders in Sweden, we decided also to include reports only focusing on direct costs for the searches in Swedish.

Searches

The literature search is presented in Table 1. In total, the search in PubMed generated 485 hits. Adding "Sweden" to the PubMed search generated an additional 2 hits. Articles that did not fit the inclusion criteria were excluded. Only 4 articles fulfilled the inclusion criteria and were left for review. Several of the excluded articles were cost-effectiveness or cost-utility studies evaluating hearing aids, hearing screening strategies, or cochlear implants. The Cochrane search generated 101 hits in all Cochrane products but only one fulfilled the inclusion criteria. The search using Google Scholar generated 41 hits in English and 1150 hits in Swedish each identifying one relevant report. The searches at the website of the Swedish Association of Hard of Hearing People and at the website *hear-it.org* generated 2 additional documents/reports of interest, one in English, and one in Swedish.

Table 1. Literature search, Cost of Illness and hearing disorders

Database	Search terms	Number of results	Abstracts selected	Articles/reports included
PubMed	(Costs and Cost Analysis"[Mesh] OR "cost"[tiab] OR "economic burden"[tiab]) AND ("Hearing Disorders"[Mesh] OR hearing impairment[tiab] OR "Hearing Impaired Persons"[Mesh]) AND (English[lang] OR Swedish[lang]) Date range: Jan 1995-Jan 2012	485	15	4
Cochrane (all Cochrane products)	(Costs and Cost Analysis"[Mesh] OR "cost"[tiab kw] OR "economic burden"[tiab kw]) AND ("Hearing Disorders"[Mesh] OR hearing impairment[tiab kw] OR "Hearing Impaired Persons"[Mesh]) Date range: Jan 1995-Jan 2012	101	1	1
Google Scholar (English)	Cost AND Hearing Date range: Jan 1995- Jan 2012	In document: 431 000 In title: 41	Restricted for “in title”	1
Google Scholar (Swedish)	Kostnad OCH hörsel Date range: Jan 1995-Jan 2012	In document: 1150 In title: 0	Search in all results	1
Search on Web sites: Swedish Association of Hard of Hearing People and *hear-it*		n/a	n/a	2

tiab=Title/Abstract

Results

Cost-of-illness studies of hearing disorders

The included studies were divided into two groups, i.e. one group with Swedish studies and one group with studies from other countries. The studies were also divided according to publication status, i.e. whether they were published in peer-reviewed journals or not. The characteristics of the Swedish and international studies are presented in Table 2 and Table 3. All cost results from the reviewed literature

are converted and Consumer Price Index adjusted to USD 2011 (with the original cost and currency in parenthesis).

Table 2. Cost studies of hearing disorders in Sweden (not peer-reviewed)

Study, Year of publication	Costing year	Sample Size	Cost items	Results
Hörapparat för vuxna (*Hearing aids for adults*), 2003	2002	58,000 adults	Direct costs for diagnosis and hearing aid fitting.	Estimated to $66.8 mn (SEK 562 mn) in 2002. Average cost per person was $1,188 (SEK 10,000)
Nationella medicinska indikationer: Hörselrehabilitering till vuxna (*Hearing rehabilitation for adults*), 2008	2006	National estimate Adults	Direct purchase costs of hearing aids	Estimated to $55 mn (SEK 390 mn)
Kostnader och effekter vid hjälpmedelsförskrivning (*Costs and effects of prescriptions of hearing aids*), 2010	2009	Adults. Data from 4 Swedish counties	Direct cost for hearing aid and aid fitting.	Average cost per prescription was $ 685 (SEK 5,209)

Table 3. International studies of societal costs of hearing disorders

Study, Year of publication, Country	Costing year	Perspective	Time frame	Costing approach	Sample size	Cost items	Discount rate	Results
The societal costs of severe to profound hearing loss in the United States, 2000 (Mohr, Feldman et al. 2000) US	1998	Societal	Lifetime	Incidence-based. Cohort-survival Markov model	15,400 incident cases of severe to profound hearing loss in 1991	Direct costs and indirect costs (productivity loss).	3%	Total lifetime costs $6.4 billion. Lifetime cost per person $410,000. 67% of costs indirect. Magnitude of costs directly related to age of onset of hearing loss.
Economic costs associated with mental retardation, cerebral palsy, hearing loss and vision impairment – United States 2003 (Honeycutt, Dunlap et al. 2004) US	2003	Societal	Lifetime	Incidence based	Persons born in year 2000, estimated as 1.2/1,000 children times live-born infants in 2000	Direct (medical and non-medical) and indirect costs.	3%	$2.3 billion in total and $468,000 per person. Indirect costs largest part (69%).
The economic costs of congenital bilateral permanent childhood hearing impairment (Schroeder, Petrou et al.	2003	Societal	One year (preceding year of life)	Prevalence-based	120 children, 7-9 years, with bilateral permanent hearing	Health and social care costs, education costs, direct non-medical		Mean societal cost in the preceding year of life $27,729(£14 092.5) compared to $8,277(£4,206.8) for normal-hearing

2006) UK					impairment	costs and indirect cost (lost productivity by parents).		children.
The economic effect of age-related hearing loss: national, state, and local estimates 2002 and 2030 (Stucky, Wolf et al. 2010) US	2002 and 2030	Societal		Prevalence-based simulation model	≥ 65 year old persons i.e. in 2002: 6.4 mn and in 2030: 12.8 mn	Direct medical costs and costs from lost productivity.		In total $12 billion in 2002 and $75.5 billion in 2030. Direct medical cost $10.2 billion and lost productivity $1.75 billion (per person $1,897) in 2002. Direct medical cost $64.2 billion and lost productivity $11 billion (per person $5,913) in 2030.
Not peer-reviewed studies								
The economic impact and cost of hearing loss in Australia (CRC HEAR/Access Economics 2006) Australia	2005	Societal		Prevalence-based	3.55 mn Australians	Direct medical costs, direct non-medical costs, informal care and lost productivity.		Total financial cost $10.49 billion ($2,960 per person) out of cost for productivity loss 57%. Cost for loss of wellbeing (based on DALYs) $10.1 billion.

Studies based on Swedish data

Previous studies of hearing disorders in Sweden have primarily focused on prevalence (Jonsson and Rosenhall 1998; Johansson and Arlinger 2003; Widen and Erlandsson 2004; Hietanen, Era et al. 2005; Muhr, Rasmussen et al. 2007; Hasson, Theorell et al. 2010; Muhr and Rosenhall 2010). We have not identified any peer-reviewed study estimating the total societal costs for hearing disorders in Sweden. We have identified three Swedish reports which were not published in journals with peer-review. These Swedish cost studies have primarily focused on costs for hearing aids which will be presented here.

The Swedish Council on Technology Assessment in Health Care (SBU) published a report on the utility and costs of hearing aids (The Swedish Council on Technology Asessment in Heatlh Care 2003). The aim of the report was to conduct a systematic review of the scientific literature on the utilities and risks of using hearing aids. The report concluded that there is lack of published studies on costs in relation to the utility of hearing aids compared with other health care interventions. The direct costs for diagnosis and hearing aid fitting were estimated to USD 66.8 million (SEK 562 mn) in 2002. Based on an estimation of 58,000 persons prescribed with hearing aids in 2002, the average cost per person was less than USD 1,188 (SEK 10,000). The SBU report also concludes that there are significant differences between the Swedish counties in the number and types of the prescribed hearing aids. These differences are explained by regional differences in the counties' praxis.

In another report, the direct cost for hearing aids only was estimated to USD 55 million (SEK 390 million) for the year 2006 (Arlinger, Danermark et al. 2008). Another study has estimated the average cost of hearing aid and aid fitting to USD 685 (SEK 5 209) per prescription in 2009 (Handikappförbunden 2010).

Published studies based on data from other countries

Several studies have examined the cost-utility and cost-effectiveness of interventions such as screening programmes and hearing aids (Cheng, Rubin et al. 2000; Barton, Stacey et al. 2006; Uus, Bamford et al. 2006; Chao and Chen 2008; Bond, Mealing et al. 2009; Bond, Elston et al. 2010; Summerfield, Lovett et al. 2010) but few studies provide comprehensive estimations of total societal costs for hearing disorders. Only four published studies were identified; three from the US and one from the UK, although some of them were limited to specific patient populations.

A study by Mohr and colleagues provides a comprehensive, national estimation of the economic burden of hearing impairment in the US (Mohr, Feldman et al. 2000). It was an incidence-based cost study, where the lifetime medical and other costs were estimated for newly diagnosed cases. Diagnosis was based on self-reported hearing loss. Incident cases of severe to profound hearing loss in the United States population in 1991 were estimated to approximately 15,400. A cohort-survival Markov model with five age cohorts was developed. The five cohorts were prelingual (0-2 years), prevocational (3-17 years), early working age (18-44 years), later working age (45-64 years) and retirement age (65 years and older). The principal data source was a national representative household survey which was complemented with literature and an expert panel when data were missing. Indirect costs were estimated using the HCM. The life time cost for a person with severe to profound hearing loss was estimated to USD 410,000 (converted to 2011 USD) (297,000 in 1998 USD). The majority of the costs were indirect costs due to reduced work productivity (67%). Based on this, persons who experience severe to profound hearing loss before retirement age were expected to lose between USD 300,000 and USD 610,000 (USD 220,000-440,000) in earnings during their working life, i.e. 50-70% of their normal hearing peers. Medical costs were a relatively small

proportion of the total costs. The authors concluded that in contrast to many other clinical conditions, severe and profound hearing loss largely impacts the social welfare system rather than the health care system. Further, the magnitude of costs was directly related to age of onset of the hearing loss, i.e. persons with a hearing loss onset during childhood incurred the largest costs. The lifetime costs for a child with prelingual onset of hearing loss were estimated to exceed USD 1.4 million (USD 1mn) while the lifetime costs for a person acquiring hearing loss as an adult were expected to be USD 59,000 (USD 43,000), on average. In addition, the study used an attributed risk methodology in order to find any association between hearing loss and other health conditions and premature mortality. However, no significant differences were seen in physician visits, depression/anxiety, traumatic injury or mortality. In summary, the total societal costs amounted to USD 6.35 billion (USD 4.6 billion) during the lifetime for the estimated 15,400 persons with profound hearing loss. The authors showed that this estimate of lifetime costs for profound hearing loss compares with that of schizophrenia and is twice those reported for stroke.

Honeycutt and colleagues estimated direct and indirect costs associated with four developmental disabilities in the US (Honeycutt, Dunlap et al. 2004). One of these (defined as a chronic condition that is manifested during the development period, 0-18 years of age) was hearing loss for which the prevalence was estimated to 1.2 per 1,000 children aged 5-10 years based on a development disabilities surveillance program. The size of the cohort was then estimated by multiplying the prevalence by the number of live-born infants in the US in 2000. The estimation of direct medical and non-medical costs (e.g. physician visits, inpatient stays, assistive devices, home and automobile modifications) was based on data from multiple national surveys and reports. The indirect costs were calculated through estimation of productivity losses in workplaces and households due to premature death or that the person was unable to work or limited in amount or type of work. Based on this, the lifetime cost for all persons with hearing loss born in year 2000 was estimated to USD 2.3 billion in

2011 dollars (1.9 billion in 2003 USD). The average lifetime costs per person with a hearing disorder were estimated to USD 468,000 (USD 383,000). Indirect costs accounted for the largest share of the total costs, i.e. USD 1.6 billion (USD 1.3 billion). The total direct costs were estimated to USD 735 million (USD 601 mn) where the nonmedical costs accounted for USD 573.4 million (USD 469 mn) and the direct medical costs for USD 161.4 million (USD 132 mn). Among the total direct costs, special education caused a significant part (Honeycutt, Dunlap et al. 2004).

Another American study demonstrated the economic effect of age-related hearing loss in the US for the years 2002 and 2030 (Stucky, Wolf et al. 2010). In 2002, approximately 6.4 million people aged 65 and older reported some form of hearing loss, based on the U.S. Census. The prevalence for 2030 was estimated to 12.8 million people and accounted for increased life expectancy and the prevalence of existing hearing problems today for people 45-64 who will reach age 65 by 2030. The analysis included direct medical costs and costs from lost productivity in adults with hearing loss aged 65 years or older. These findings suggest that the financial and societal burden of treating age-related hearing loss will increase significantly in the coming years. In 2002, the direct medical costs for the first year treatment of hearing loss in adults aged 65 and older was estimated to USD 10.2 billion (8.2 in 2002 USD), i.e. USD 1,615 per person, and USD 64.2 billion in 2030 (USD 51.4 billion), i.e. USD 5,035 per person. The lost productivity attributable to hearing loss for this age group was estimated to approximately USD 1.75 billion in 2002, i.e. USD 281 per person and approximately USD 11 billion in 2030, i.e. USD 878 per person, based on extrapolated data from (Mohr, Feldman et al. 2000).

One study by Schroeder and colleagues estimated the economic costs of bilateral permanent childhood hearing impairment (PCHI) (i.e. ≥40-dB HL) in the preceding year of life for children aged 7 to 9 years in selected areas of England (Schroeder, Petrou et al. 2006). The study results are based on 120 hearing impaired children with mean age of 7.9 years (divided into moderate, severe and

profound PCHI), their parents, and a group of 63 normal-hearing children. The mean societal cost in the preceding year of life at 7 to 9 years of age was USD 27,729 (£ 14 092.5, 2003 prices) for children with PCHI. The corresponding cost for normal-hearing children was USD 8,277(£ 4 206.8). The study adopted a broad societal perspective including direct medical, direct non-medical, and indirect costs. It is clear from the study that the costs were mainly driven by special education services (approximately 77%). The study also compared costs among children with PCHI who were born in areas with or without universal new born screening (UNS). After adjusting for severity and other potential confounds, the mean societal costs was 15% lower for children in the UNS cohort, although the difference was not statistically significant. Based on an incidence of 840 hearing impaired children in the UK, the total yearly cost at 7.9 years of age was estimated to be between USD 8 and 28 million (£4 -14 mn) with an average of about USD 16.4 million (£8.3 mn) per year.

Unpublished studies based on data from other countries

One report from Australia was identified as having a comprehensive societal cost approach of hearing disorders. Using a prevalence based approach, the Australian report estimated, the financial costs and the loss of wellbeing from hearing loss in Australia for the year 2005 (CRC HEAR/Access Economics 2006). One in six Australians (3.55 million) is affected by hearing loss and due to an ageing population hearing loss is estimated to increase to one in four Australians by 2050. Of the estimated 3.55 million with hearing loss, 0.3 % was estimated to be children aged up to 14 years, almost 50% were of working age (15-64 years), and 37% aged 70 years or older. It was not clear from the report whether the currency used was AUD or USD. However, we assumed that the currency used was AUD and have thus converted the figures into 2011 USD. The financial cost of hearing loss was estimated to USD 10.5 billion (11.75 billion in 2005 AUD) or 1.4% of GDP which corresponds to an average cost per year of USD 2,956 (AUD 3,314) per person with hearing loss. The cost for productivity loss accounted for the largest part, 57%, of the financial costs, i.e. USD 6 billion (AUD 6.7 billion). Nearly

half of the people with hearing loss are of working age, 15-64 years, and it was estimated in 2005 that 158, 676 people were not employed due to their hearing loss. The cost of informal care (informal careers assist with e.g. making telephone calls, taking notes in a meeting at work or in a classroom) was 27% of total financial cost. Direct health care costs included costs for diagnosis, treatment, and management of hearing loss and accounted for USD 601 million (AUD 674 mn) (6% of total financial costs). Devices were the largest cost item of the direct health care costs, i.e. hearing aids and cochlear implants which cost USD 336 (AUD 376.7 mn) and USD 8.9 million (AUD 10 mn), respectively per year. Other direct health care costs amounted to USD 221 million (AUD 247.5 mn), i.e. USD 62 (AUD 70) per person with hearing loss. Education and support services comprise 1.6% of the financial costs. The quality of life impact in the Australian study is measured in terms of Disability Adjusted Life Years (DALYs). Ninety-five thousand and five DALYs are estimated to be lost in 2005 due to hearing loss which is valued to USD10.1 billion (AUD 11.3 billion). In terms of disability weighting, mild hearing loss is comparable to mild asthma, moderate hearing loss to chronic pain resulting from a slipped disc, and severe hearing loss is comparable to having pneumonia on an on-going basis.

Another report presented results from a literature review of the social and economic effects of hearing loss in Europe (Shield 2006). The review revealed a nonexistence of studies on socio-economic costs or hearing impairment in Europe. The review also identified the US based Mohr study (Mohr, Feldman et al. 2000) on societal costs of severe to profound hearing impairment but concluded that there is a lack of studies on costs to society of less severe hearing impairment.

3. Discussion, concluding remarks and future research

Several studies have examined the cost-effectiveness of interventions such as screening programs and hearing aids (e.g. Cheng, Rubin et al. 2000; Barton, Stacey et al. 2006; Uus, Bamford et al. 2006; Chao and Chen 2008; Bond, Mealing et al. 2009; Bond, Elston et al. 2010; Summerfield, Lovett et al. 2010) but studies estimating the total societal costs for hearing disorders are limited. This literature review found few studies addressing the societal costs of hearing disorders. The identified cost studies for Sweden primarily focused on costs for hearing aids and no study estimated the total societal costs for hearing disorders in Sweden. The international studies with a societal cost perspective identified in this review used different starting points, costing approaches, and were limited to different patient populations which make them difficult to compare. Two studies were incidence-based and the others were prevalence-based. Some examined hearing disorder in adults; other focused hearing disorder in children, while some concentrated on patients with severe to profound hearing disorder and some on age-related hearing disorders.

The most comprehensive study estimating the societal costs of hearing disorders identified in this review was the one by Mohr et al (Mohr, Feldman et al. 2000). This incidence-based study used a lifetime societal perspective including the hearing impairment´s impact on earnings and use of educational and medical resources. The authors estimated that severe to profound hearing loss in the US costs society approximately USD 410,000 during the lifetime for an individual, comparable to the lifetime cost of schizophrenia. Most of the costs were caused by productivity loss. The study also considered an attributed risk methodology. A limitation of the study is that it only focused on severe and profound hearing disorders. The second incidence-based study, also with a comprehensive societal perspective, but with the limitation that it only regarded hearing disorder defined as a developmental disability, estimated a lifetime cost of USD 468,000 per person (Honeycutt, Dunlap et al. 2004). Similar to the study by Mohr and colleagues, most of the costs were due to productivity

losses. The only study incorporating all degrees of hearing loss and all ages in their cost estimate was the Australian report from 2005. This report also revealed the indirect costs as the largest cost component. An interesting aspect in this report was the cost of informal caregivers, which was shown to be considerable. The results from the international studies indicate that the societal costs for hearing disorders are considerable, e.g. 1.4% of GDP or, considering only profound hearing loss, comparable to the cost for schizophrenia or twice the cost of stroke.

This literature review has identified some key issues in estimating societal costs related to hearing disorders. It seems that hearing disorders impact the social welfare system more than the health care system. The direct medical costs account for a minor part of the total costs of hearing disorders. Further, the literature has pointed at special education as an important factor in the direct non-medical costs. Thus, the results of a study on costs of hearing disorders will depend on whether taking a child and/or an adult perspective. When calculating costs of hearing disorders, the onset of hearing disorder is essential, i.e. whether the hearing disorder is congenital, or if the hearing loss is acquired. Another methodological issue to consider is whether the hearing disorder is identified by self-assessment or through measurement by audiometry. The use of self-assessed or measured hearing disorder may influence the estimation of the societal costs since the prevalence of hearing disorder could differ depending on use of method. It is clear from this literature review that indirect costs (e.g. cost of lost productivity) are major components of the societal cost of hearing disorders. It would be of interest to further explore the attributed risk and costs of other health problems, e.g. depression, for patients with hearing disorder compared to the general population.

Although there are a few comprehensive studies addressing the societal costs of *severe* hearing disorders there is little information available on the costs of *less severe* hearing disorders. Also, the studies identified dealt with the US, the UK, and Australia, while no study with a societal perspective has been found for Sweden. Since the Swedish cost studies only focused on the direct medical costs of hearing disorders, such as cost of diagnosis, hearing aid fitting and hearing aids, it is clear that a

large part of the costs of hearing disorders in Sweden has not yet been highlighted. Thus, there is a need for further studies exploring societal costs for all degrees of hearing loss especially since a large part of the people with hearing loss are of working age, and for studies including all cost components as well as studies in other countries than the ones reviewed here.

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