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The Cost-Effectiveness of Different Hearing Screening Strategies for 50- to 70-Year-Old Adults: A Markov Model



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ABSTRACT

Objective: To assess the cost-effectiveness of screening 50- to 70year-old adults for hearing loss in The Netherlands. We compared no screening, telephone screening, Internet screening, screening with a handheld screening device, and audiometric screening for various starting ages and a varying number of repeated screenings. **Methods:** The costs per quality-adjusted life-year (QALY) for no screening and for 76 screening strategies were analyzed using a Markov model with cohort simulation for the year 2011. Screening was deemed to be costeffective if the costs were less than €20,000/QALY. **Results:** Screening with a handheld screening device and audiometric screening were generally more costly but less effective than telephone and Internet screening. Internet screening strategies. Internet screening at age 50 years,

Introduction

Untreated hearing loss has various negative consequences, including social isolation, loneliness, psychosocial distress, anxiety, and depression [1–3]. Hearing aid fitting can increase the quality of life of people with a hearing loss [4–6]. It is a cost-effective intervention (compared with no rehabilitation) [7,8]. Nevertheless, adults generally postpone seeking help until 5 to 10 years after the onset of their hearing loss [9–11]. People mainly postpone seeking help because they are unaware or in denial of the hearing loss and believe they can still manage without hearing aids [12].

Seeking help at an early stage is important because adults who start using hearing aids early—that is, at a relatively young age or when their hearing loss is still relatively mild—do not only have more years with benefit ahead but also have greater benefit from their hearing aids during later life than do adults who start using hearing aids late [9]. This can be explained by the fact that older adults have more difficulty adapting to hearing aids and learning to use new technology because of poorer cognitive performance and poorer learning ability [13,14]. Moreover, people repeated at ages 55, 60, 65, and 70 years, was the most cost-effective strategy, costing €3699/QALY. At a threshold of €20,000/QALY, this strategy was with 100% certainty cost-effective compared with current practice and with 69% certainty the most cost-effective strategy among all strategies. **Conclusions:** This study suggests that Internet screening at age 50 years, repeated at ages 55, 60, 65, and 70 years, is the optimal strategy to screen for hearing loss and might be considered for nationwide implementation.

Keywords: adults, cost-effectiveness, hearing loss, Markov model, screening.

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with poorer cognitive performance often have difficulty in formulating their needs during a hearing aid trial, which can result in a suboptimal hearing aid fit [15].

Adult hearing screening is thought to motivate people to seek help earlier. Studies in the United Kingdom showed that screening can triple hearing aid ownership among middle-aged adults [16,17]. The only randomized controlled trial of the effectiveness of screening so far showed that screening with an objective measurement instrument can almost double the 1-year incidence of hearing aid use [18]. Adult hearing screening is found to be a cost-effective intervention to increase hearing aid use and quality of life (in comparison with no screening) [19,20]. Screening with an objective measurement instrument outperformed screening with a subjective measurement instrument (questionnaire) and screening with an objective and subjective instrument combined [19,20]. Our study is the first that assessed the costs and the effects on quality of life of telephone and Internet screening. The costs and effects were compared with those of screening with a handheld screening device (like Liu et al. [19]), screening with an audiometer (like Morris et al. [20]), and current practice in The Netherlands (no nationwide adult hearing screening program).

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Methods

Model Structure

A Markov model was constructed to evaluate the costeffectiveness of adult hearing screening from a health care perspective. All costs were in euros (€1.00 is US \$1.38 and £0.87, average 2011 conversion rates). The effectiveness measure in the model was the quality-adjusted life-year (QALY), which is a combined measure of health-related quality of life and duration of life. The model allowed simulating the lifetime course of events in 50-year-old adults who do not own hearing aids. Possible events included hearing deterioration, hearing aid uptake, hearing aid replacement, hearing aid discard, and dying. The health states of the model were based on hearing loss severity and hearing aid use (Fig. 1). For hearing loss severity, the classification of the World Health Organization was used, with a hearing threshold (pure-tone average at 0.5, 1, 2, and 4 kHz) between 25 and 40 dB in the best ear indicating mild hearing impairment and a hearing threshold of more than 40 dB in the best ear indicating moderate to severe hearing impairment. A hearing threshold of 25 dB or less in the best ear indicated either normal hearing or unilateral hearing impairment. With unilateral hearing impairment, we refer to a hearing threshold of 25 dB or less in the best ear, a hearing threshold of more than 25 dB in the worst ear, and an interaural difference of 10 dB or more at three frequencies, 15 dB or more at two frequencies, or 20 dB or more at one frequency. Approximately 27% of the people with a hearing threshold of 25 dB or less in the best ear have unilateral hearing impairment [21]. We assumed that among adults with a hearing threshold of 25 dB or less in the best ear, only adults with unilateral hearing impairment might take up hearing aids. We did not consider, however, normal hearing and unilateral hearing impairment as separate health states in our model because data on the annual probability to develop mild hearing impairment for these two groups separately were not available.

The model simulated current practice (no screening) and four types of nationwide screening:

1. Telephone screening: The target population is invited by letter to undertake the National Hearing Test by telephone. The National Hearing Test is a fully automatic adaptive speech-in-noise test that uses digit-triplets as speech material. It was developed and validated by Smits et al. [22]. There are three possible outcomes: good, insufficient, and poor hearing. People with insufficient or poor hearing are considered screen-positive. Because only one ear is tested (usually the participant's best ear), the test is unable to detect unilateral hearing impairment.

- 2. Internet screening: The target population is invited by letter to take the Internet version of the National Hearing Test (www. hoortest.nl). The test was developed by Smits et al. [23] and validated by Leensen et al. [24]. It is greatly similar to the telephone test, except for the fact that the Internet test presents the signals to both ears simultaneously. The Internet test will not detect unilateral hearing impairment.
- 3. HearCheck screening: Members from the target population who visit the practice of the general practitioner (GP), for whatever reason, are offered a hearing screening test while waiting for a consultation with their GP. The GP assistant performs the screening using the Siemens HearCheck Navigator. The Siemens HearCheck Navigator is a handheld screening device that emits five tones: 375 (35 dB), 1000 (55 and 35 dB), and 3000 Hz (75 and 35dB). Depending on which tones are heard, the test will inform the participants whether their hearing in the tested ear is good, insufficient, or poor. Both ears are tested separately; therefore, unilateral hearing impairment can be detected. People with insufficient or poor hearing in one or both ears are considered screen-positive. The GP discusses the outcome with everyone who screened positive.
- 4. Audiometric screening: Members from the target population who visit the GP practice, for whatever reason, are offered a hearing screening test while waiting for a consultation with their GP. The GP assistant performs standard pure-tone audiometry. Because this is the criterion standard, everyone with hearing impairment is screen-positive and everyone with normal hearing is screen-negative. The GP discusses the outcome with everyone who screened positive.

In total, 76 screening strategies were included in the economic evaluation. The strategies were varied in the type of screening, the age at first screening (either 50, 55, 60, 65, or 70 years), the number of repeated screenings (up to five repetitions), and the time interval between repeated screenings (either 5 or 10 years). The model was constructed using Microsoft Excel 2010 with customized macros and had a lifetime time horizon with a cycle length of 1 year.

Model Input

In the paragraphs below we briefly describe which parameters were accounted for in the model. An elaborate description of our considerations, calculations, and assumptions with regard to the parameter estimates is given in the Appendix in Supplemental Materials found at http://dx.doi.org/10.1016/j.jval.2015.03.1789. Appendix Table A1 in Supplemental Materials found at http://dx.doi.org/10.1016/j.jval.2015.03.1789 lists the model parameter



Fig. 1 – General structure of the Markov model. The solid and dashed lines represent the transitions to other health states by hearing deterioration (bold black lines), hearing aid uptake (thin black lines), hearing aid discard (dashed lines), and dying (gray lines). Screening is offered to people without hearing aids and affects only the probability of hearing aid uptake. BEPTA, best-ear pure-tone average (0.5, 1, 2, and 4 kHz); HA, hearing aid; HI, hearing impairment; mod, moderate; sev, severe.

estimates used to calculate the costs and effects of hearing care in current practice, including base-case parameter estimates, distributions, and references to data sources. Appendix Table A2 in Supplemental Materials found at http://dx.doi.org/10.1016/j. jval.2015.03.1789 lists the additional model input for screeningspecific parameters.

Transition Probabilities

At model entrance, the cohort comprised adults without hearing aids who were distributed over the three possible starting states on the basis of the prevalence of uncorrected mild and moderate to severe hearing impairment in the general population [25]. Hearing deterioration was modeled using estimates of the 1-year incidence of mild and moderate to severe hearing impairment that were, like the prevalence data, derived from the Blue Mountains Hearing Study [25]. We used age- and sex-specific annual mortality figures from the Dutch life tables [26]. The probability of hearing aid discard was set at 6% in all strategies [27]. We modeled that screening would be offered exclusively to people without hearing aids and that it would influence only the probability of hearing aid uptake (thin black arrows in Fig. 1). The probability of hearing aid uptake was modeled separately for current practice and for each screening strategy as a function of the following parameters: the probability of screen participation, the validity of the screening test, the probability of help-seeking (first step), the probability that a help-seeker continues helpseeking, the probability that someone who continued helpseeking starts a hearing aid trial, and the probability that the trial is ended successfully and thus resulted in hearing aid purchase. The screening strategies differed in the probability of screen participation, the validity of the screening test, the probability of help-seeking, and the probability of continued help-seeking. Estimates of these screening-specific probabilities and the sources on which they were based can be found in the Appendix (pages 2 and 3 and Appendix Table A2 in Supplemental Materials). The probability that someone who continued helpseeking starts a hearing aid trial and the probability of trial success were assumed not to be influenced by screening. For adults who did not participate in screening, we used the probabilities of helpseeking and continued help-seeking from current practice.

In The Netherlands, the first step in help-seeking for hearing impairment is a consultation with either the GP or the hearing aid dispenser. Continued help-seeking may include a consultation with the ear, nose, and throat (ENT) specialist, a consultation at the audiological center, and/or an intake meeting with the hearing aid dispenser (see Appendix Fig. A1 in Supplemental Materials found at http://dx.doi.org/10.1016/j.jval.2015.03.1789 for a schematic overview of possible paths in continued help-seeking). The ENT specialist clinically examines the ear, removes earwax, diagnoses ear pathology, performs hearing tests, informs the help-seeker about treatment possibilities, and prescribes hearing aids to those who are willing to try them. Furthermore, the ENT specialist refers help-seekers to the audiological center in case of a need for additional counseling, problems in accepting hearing impairment, limited willingness to use hearing aids, severe hearing loss, poor speech discrimination, and the presence of additional sensory or mental disability. The hearing aid dispenser is responsible for hearing aid fitting and is the only hearing care professional who sells hearing aids.

Costs and Quality-of-Life Input

The following costs were included in the analysis: costs of consultations with the GP assistant, GP, ENT specialist, hearing aid dispenser, and employees of the audiological center (clinical physicist/audiologist, audiology assistant, social worker, and

speech therapist); costs of hearing aids and hearing aid batteries, maintenance, and repair; costs of the invitation letter for telephone and Internet screening; telephone costs; Web hosting costs for the Internet test; costs of the disposable ear cups for the HearCheck Navigator; and annual depreciation and maintenance costs of the HearCheck Navigator and audiometer used for screening. Price indices were used to convert costs to a 2011 price level [26].

Health-related quality of life was expressed as a utility score between 0 (death) and 1 (perfect health). Data on utility scores in relation to hearing thresholds and hearing aid fitting, measured with the Health Utility Index Mark 3 questionnaire, were collected by our research group in 2004 [28]. We reanalyzed these data to obtain age-dependent utility scores for each of the health states. Details of this analysis can be found on pages 3 and 4 of the Appendix in Supplemental Materials.

Analyses

We examined the expected costs and effects of current practice and the 76 screening strategies for adults without hearing aids using cohort simulation. Future costs were discounted at an annual rate of 4.0%, and future effects were discounted with an annual rate of 1.5% following the Dutch guidelines [29]. To account for uncertainty in the model parameter estimates, we performed probabilistic sensitivity analysis using Monte-Carlo simulation with 10,000 iterations. For each iteration, parameter values were drawn at random from the distribution around the base-case estimates. See Appendix Tables A1 and A2 in Supplemental Materials for the assigned distributions.

We sorted the strategies from lowest to highest expected costs (based on the results of the probabilistic sensitivity analysis) and calculated the incremental costs and incremental QALYs of each strategy compared with current practice (no screening). Incremental cost-effectiveness ratios (ICERs) were calculated by dividing the incremental costs by the incremental QALYs. The sequence of strategies, from lowest costs to highest, that gives the lowest ICER values forms the cost-effectiveness frontier [30]. In a cost-effectiveness plane with the incremental costs of all strategies plotted against their incremental QALYs, the costeffectiveness frontier is the line that connects the strategies that dominate or extendedly dominate the other strategies. A strategy is dominated by another strategy if it is more costly and less effective. A strategy is extendedly dominated if it is more costly, or less effective and has a higher ICER than does a more effective strategy. The optimal strategy is the strategy on the costeffectiveness frontier that has the highest ICER below an externally set cost-effectiveness threshold or ceiling ratio (λ). In The Netherlands, λ for screening is set at $\in 20,000/QALY$, which means that a screening strategy will be considered for nationwide implementation only if the costs per QALY gained are less than €20,000 [31].

To graphically illustrate the uncertainty concerning the costeffectiveness of the strategies, a cost-effectiveness acceptability frontier (CEAF) was constructed on the basis of results of the probabilistic sensitivity analysis [32]. For construction of the CEAF, the net monetary benefit of the strategies was calculated for a range of λ values using the following formula: net monetary benefit = $\lambda \times QALY - costs$. For every value of λ , the optimal strategy was identified (i.e., the strategy with the highest mean net monetary benefit for each of the 10,000 iterations). Next, the probability for the optimal strategy to be the most cost-effective strategy at a certain value of λ was calculated as the proportion of iterations in which the optimal strategy had the highest net monetary benefit. The CEAF shows the optimal strategies and their probability of cost-effectiveness for λ values between $\in O/$ QALY and $\in 20,000/QALY$.

One-Way Sensitivity Analyses

In the base-case analysis, the probability of screen participation was 0.33 for telephone screening and 0.44 for Internet screening (both estimates were derived from a study by Koopman et al. [33]). We performed a one-way sensitivity analysis to determine whether the results would have been different if the probability of screen participation for Internet screening had been 0.33 as well (one-way sensitivity analysis 1). For screen-positive adults with moderate to severe hearing impairment, the probability of help-seeking after telephone and Internet screening was 0.81 and 0.79, respectively, according to Koopman et al. [33]. Smits et al. [23] reported a probability of 0.57. A second sensitivity analysis was performed to determine whether the results would have been different if Smits et al.'s instead of Koopman et al.'s estimates had been used (one-way sensitivity analysis 2). In the base-case analysis, we assumed that the probability that someone who sought help after telephone or Internet screening continues help-seeking would be 25% higher (relative increase) than the probability of continued help-seeking in current practice. An increase is to be expected, but the size of this increase was an arbitrary choice. With a third sensitivity analysis we examined whether the results would have been different if the relative increase had been set at 10% instead of 25% (one-way sensitivity analysis 3). A rescreen after 5 years is less effective in stimulating help-seeking than the first screen [34]. In the basecase analysis, we assumed that the effect of a rescreen after a 10-year interval would be similar to the effect of a rescreen after a 5-year interval and thus be lower than the effect of the first screen. Given the large time interval, however, this assumption might not necessarily be true. We therefore reran the model, with the effect of a rescreen after a 10-year interval being adapted to equal the effect of the first screen (one-way sensitivity analysis 4). Furthermore, we examined whether the results would have been different if the future costs and effects had not been discounted (one-way sensitivity analysis 5). In addition, we performed a threshold analysis to determine the maximum costs per screen at which the telephone and Internet screening strategies would still be cost-effective compared with no screening.

Results

Base-Case Results

Table 1 presents the results of the probabilistic sensitivity analysis. The incremental costs of the screening strategies compared with no screening ranged from €4 to €59 and the incremental QALYs ranged from 0.0003 to 0.0104. The ICERs of all the screening strategies compared with current practice were below €20,000/QALY, indicating that screening is cost-effective as compared with no screening. HearCheck and audiometric screening strategies were dominated by telephone and Internet screening strategies. Telephone screening strategies were either dominated or extendedly dominated by Internet screening strategies. Only Internet screening strategies are on the cost-effectiveness frontier (Fig. 2). With an ICER of €3699/QALY, Internet screening at age 50 years, repeated at ages 55, 60, 65, and 70 years, was the most cost-effective strategy at a ceiling ratio of €20,000/QALY.

Figure 3 shows the CEAF. At λ values of €3000/QALY or lower, current practice was the optimal strategy. At λ values between €3000/QALY and €4500/QALY, the strategies that result in the highest mean net monetary benefit had a probability of being the most cost-effective between 7% and 12%. At λ values between €4,500/QALY and €20,000/QALY, Internet screening at age 50

years, repeated at ages 55, 60, 65, and 70 years, was the optimal strategy. At a threshold of \notin 20,000/QALY, this strategy was with 100% certainty cost-effective compared with current practice and with 69% certainty the most cost-effective among all strategies.

One-Way Sensitivity Analyses

Lowering the probability of screen participation for Internet screening from 0.44 to 0.33 decreased the incremental costs and QALYs of Internet screening as compared with no screening. The QALY gain by Internet screening at age 50 years with repetition at ages 55, 60, 65, and 70 years became lower than the QALY gain by the audiometric screening at age 50 years with repetition at ages 55, 60, 65, and 70 years, but the difference was only 0.0001 QALY, while the incremental costs of the audiometric screening strategy were twice as high as the incremental costs of the Internet screening strategy (see Appendix Table A3 in Supplemental Materials found at http://dx.doi.org/10.1016/j.jval.2015.03.1789). Besides, the CEAF showed that Internet screening still had the highest probability to be cost-effective (see Appendix Fig. A2.1 in Supplemental Materials found at http://dx.doi.org/10.1016/j.jval. 2015.03.1789). The results of sensitivity analysis 2 to 4 were similar to the results of the base-case analysis (see Appendix Tables A4-A6 and Fig. A2.2-A2.4 in Supplemental Materials found at http://dx.doi.org/10.1016/j.jval.2015.03.1789). Without discounting future costs and effects, the cost-effectiveness frontier was still formed by Internet screening strategies only but strategies starting at age 50 years outperformed strategies that started later. Internet screening at age 50 years, repeated at ages 55, 60, 65, and 70 years, was still the optimal strategy (see Appendix Table A7 and Fig. A2.5 in Supplemental Materials found at http://dx.doi. org/10.1016/j.jval.2015.03.1789).

In the base case, the costs of the telephone screening were set at €1.23 per screen (3.5 minutes at €0.35/min). Threshold analysis on the screen costs showed that all telephone scenarios would still be cost-effective compared with no screening when the costs would have been €83.74 per screen. At €271.66 per screen, none of the telephone screening scenarios was cost-effective compared with no screening. In the base case, the annual costs of the Internet screening were set at €144.47 (Webhosting). Depending on the size of the target population and the participation rate, the costs per screen were between € 0.0013 (for screening offered to people aged 50 years) and €0.0026 (for screening offered to adults aged 70 years). Threshold analysis showed that if the costs per screen would have been €92.73, all Internet screening scenarios would still be cost-effective compared with no screening. At €320.49 per screen, none of the Internet screening scenarios was cost-effective compared with no screening.

Conclusions

Adult hearing screening was cost-effective compared with current practice (no screening). Internet screening strategies were the most cost-effective, closely followed by telephone screening strategies. Internet screening probably outperformed telephone screening because of its lower test costs and higher test sensitivity and screen participation rate. Internet and telephone screening because of their lower test costs and accessibility for people who do not annually visit the GP. Internet access hardly forms a barrier for Internet screening nowadays in The Netherlands because 98% of adults aged between 45 and 55 years, 94% of adults aged between 55 and 65 years, and 85% of adults aged between 65 and 75 years have access to Internet at their home [26].

Table 1 – Cost-effectiveness of adult hearing screening.									
Screening strategy		Costs (€)		QALY		Compared with no screening			Conclusion
Туре	Age (y)	Mean	95% CI	Mean	95% CI	Δ Costs	Δ QALY	ICER	
No screening	-	1054.95	684.56– 1613 25	17.6879	17.3809– 17 9877				
HearCheck	70	1059.42	690.00– 1618.54	17.6888	17.3816– 17.9893	4.47	0.0009	4938	Extendedly dominated
HearCheck	60	1059.74	689.07– 1619.68	17.6884	17.3813– 17.9886	4.79	0.0005	9187	Dominated
HearCheck	55	1059.81	689.17– 1620.30	17.6882	17.3813– 17.9882	4.87	0.0003	14143	Dominated
HearCheck	65	1059.97	690.14– 1619.09	17.6887	17.3815– 17.9892	5.03	0.0008	6157	Dominated
Telephone	70	1060.07	689.37– 1618.11	17.6895	17.3820– 17.9909	5.13	0.0017	3028	Extendedly dominated
HearCheck	50	1060.56	689.66– 1620.90	17.6882	17.3812– 17.9885	5.61	0.0003	17466	Dominated
Telephone	65	1061.03	689.94– 1619.08	17.6897	17.3822– 17.9915	6.09	0.0018	3349	Extendedly dominated
Telephone	60	1061.04	688.86– 1619.41	17.6895	17.3821– 17.9909	6.09	0.0016	3694	Dominated
Telephone	55	1061.54	688.96– 1620.84	17.6895	17.3818– 17.9912	6.60	0.0016	4014	Dominated
HearCheck	60, 70	1062.81	693.30– 1621. 47	17.6889	17.3819– 17.9898	7.86	0.0011	7315	Dominated
Internet	70	1062.89	692.15– 1617.75	17.6905	17.3827– 17.9914	7.94	0.0026	3024	Cost-effective
HearCheck	65, 70	1063.03	693.91– 1620.83	17.6892	17.3820– 17.9903	8.09	0.0014	5921	Dominated
Telephone	50	1063.04	690.15– 1624.46	17.6897	17.3819– 17.9918	8.10	0.0019	4340	Dominated
HearCheck	60, 65	1063.32	693.47– 1621.96	17.6889	17.3819– 17.9896	8.37	0.0010	8263	Dominated
HearCheck	55, 65	1063.38	693.45– 1622.29	17.6887	17.3817– 17.9891	8.43	0.0008	10063	Dominated
HearCheck	55, 60	1063.45	692.22– 1622.84	17.6885	17.3816– 17.9886	8.50	0.0007	12911	Dominated
Internet	60	1064.05	691.93– 1623.02	17.6903	17.3826– 17.9922	9.10	0.0025	3667	Dominated
HearCheck	50, 60	1064.17	692.88– 1624 19	17.6885	17.3815-	9.23	0.0006	14495	Dominated
Internet	65	1064.27	692.74– 1620.34	17.6906	17.3828-	9.32	0.0028	3337	Extendedly
HearCheck	50, 55	1064.42	692.98– 1624.46	17.6884	17.3814-	9.48	0.0005	17906	Dominated
Internet	55	1064.60	692.05- 1625.29	17.6903	17.3827-	9.65	0.0024	3975	Dominated
Telephone	65, 70	1065.05	692.75-	17.6910	17.3830-	10.11	0.0031	3215	Extendedly
Telephone	60, 70	1065.07	692.65- 1623.21	17.6908	17.3830-	10.13	0.0030	3398	Dominated
Telephone	60, 65	1065.76	692.82- 1624.45	17.6909	17.3830-	10.82	0.0030	3551	Dominated
Telephone	55, 60	1066.18	692.20- 1628.35	17.6907	17.3829-	11.24	0.0029	3913	Dominated
Telephone	55, 65	1066.28	693.41– 1627.31	17.6909	17.3829-	11.34	0.0030	3722	Dominated
HearCheck	60, 65, 70	1066.39	697.19 to	17.6894	17.3824 to	11.44	0.0016	7319	Dominated
Internet	50	1066.69	693.17- 1628.83	17.6906	17.3828-	11.75	0.0027	4298	Dominated
			1020.03		17.3332			сс	ntinued on next page

Table 1 – continued									
Screening strategy		Costs (€)		QALY		Compared with no screening			Conclusion
Туре	Age (y)	Mean	95% CI	Mean	95% CI	Δ Costs	∆ QALY	ICER	
HearCheck	55, 60, 65	1067.02	697.00-	17.6890	17.3822-	12.08	0.0012	10501	Dominated
HearCheck	50, 60, 70	1067.24	697.33– 1628.37	17.6890	17.3823-	12.29	0.0012	10331	Dominated
Telephone	50, 60	1067.70	693.51– 1630.87	17.6910	17.3826– 17.9933	12.75	0.0031	4117	Dominated
Telephone	50, 55	1067.99	693.45– 1631.63	17.6909	17.3826– 17.9933	13.04	0.0031	4266	Dominated
HearCheck	50, 55, 60	1068.06	695.99– 1630.07	17.6887	17.3819– 17.9889	13.11	0.0008	15549	Dominated
Telephone	60, 65, 70	1069.77	696.77– 1629.46	17.6922	17.3837– 17.9945	14.83	0.0044	3394	Extendedly dominated
HearCheck	55, 60, 65, 70	1070.09	700.55– 1630.28	17.6896	17.3827– 17.9906	15.14	0.0017	8907	Dominated
Internet	60, 70	1070.11	697.62 to 1627.88	17.6924	17.3838 to 17.9956	15.17	0.0045	3369	Extendedly dominated
Internet	65, 70	1070.29	698.70– 1626.66	17.6927	17.3839– 17.9956	15.34	0.0048	3197	Cost-effective
Telephone	55, 60, 65	1070.90	696.47– 1633.20	17.6921	17.3836– 17.9950	15.96	0.0043	3739	Dominated
Internet	60, 65	1071.06	697.93– 1629.75	17.6924	17.3838– 17.9959	16.12	0.0046	3512	Dominated
Internet	55, 60	1071.32	697.04– 1633.83	17.6921	17.3838– 17.9956	16.37	0.0043	3850	Dominated
Audiometry	55	1071.57	697.85– 1636.87	17.6893	17.3821– 17.9904	16.63	0.0014	11607	Dominated
HearCheck	50, 55, 60, 65	1071.63	700.55– 1632.74	17.6892	17.3825– 17.9896	16.69	0.0013	12500	Dominated
Internet	55, 65	1071.65	697.69– 1631.41	17.6924	17.3839– 17.9962	16.70	0.0045	3674	Dominated
Telephone	50, 60, 70	1071.73	696.84– 1634.65	17.6923	17.3836– 17.9952	16.79	0.0044	3791	Dominated
Audiometry	60	1072.15	698.35– 1635.88	17.6900	17.3826– 17.9911	17.21	0.0022	7871	Dominated
Telephone	50, 55, 60	1072.62	696.72– 1636.92	17.6921	17.3833– 17.9953	17.68	0.0043	4127	Dominated
Audiometry	70	1072.90	700.53– 1632.68	17.6916	17.3830– 17.9930	17.96	0.0037	4845	Dominated
Internet	50, 60	1073.44	697.78– 1638.90	17.6924	17.3839– 17.9963	18.50	0.0046	4051	Dominated
Audiometry	50	1073.54	699.17– 1644.11	17.6892	17.3822– 17.9901	18.59	0.0013	14162	Dominated
Internet	50, 55	1073.69	697.65– 1640.50	17.6923	17.3837– 17.9964	18.75	0.0045	4188	Dominated
Audiometry	65	1074.42	701.10– 1634.80	17.6913	17.3830– 17.9924	19.48	0.0034	5702	Dominated
HearCheck	50, 55, 60, 65, 70	1074.70	704.20– 1634.19	17.6897	17.3829– 17.9905	19.75	0.0019	10479	Dominated
Telephone	55, 60, 65, 70	1074.91	700.72– 1635.46	17.6934	17.3838– 17.9970	19.97	0.0056	3572	Extendedly dominated
Internet	60, 65, 70	1077.08	702.83– 1637.42	17.6944	17.3850– 17.9982	22.13	0.0066	3358	Cost-effective
Telephone	50, 55, 60, 65	1077.34	700.93– 1640.30	17.6935	17.3838– 17.9974	22.39	0.0057	3944	Dominated
Internet	55, 60, 65	1078.32	702.41– 1641.64	17.6942	17.3848– 17.9981	23.38	0.0064	3678	Dominated
								C	continued on next page

Table 1 – continued									
Screening strategy		Costs (€)		QALY		Compared with no screening			Conclusion
Туре	Age (y)	Mean	95% CI	Mean	95% CI	Δ Costs	∆ QALY	ICER	
Internet	50, 60, 70	1079.50	703.63-	17.6944	17.3852-	24.56	0.0066	3729	Dominated
Internet	50, 55, 60	1080.40	702.33-	17.6942	17.3849–	25.45	0.0063	4042	Dominated
Telephone	50, 55, 60, 65, 70	1081.34	704.84– 1642.77	17.6949	17.3845–	26.40	0.0070	3772	Extendedly
Audiometry	55, 60	1081.38	705.12-	17.6907	17.3832– 17.9918	26.44	0.0028	9324	Dominated
Audiometry	60, 70	1082.33	708.54– 1646.09	17.6924	17.3838– 17.9943	27.39	0.0046	5975	Dominated
Audiometry	55, 65	1082.67	708.39– 1649.31	17.6915	17.3835– 17.9925	27.73	0.0036	7607	Dominated
Audiometry	50, 55	1082.89	706.58–	17.6901	17.3832–	27.95	0.0022	12544	Dominated
Audiometry	50, 60	1083.13	706.49– 1655 95	17.6906	17.3833– 17.9913	28.19	0.0027	10338	Dominated
Audiometry	60, 65	1083.47	709.07– 1647 56	17.6922	17.3838– 17.9942	28.52	0.0044	6515	Dominated
Internet	55, 60, 65, 70	1084.33	707.71–	17.6962	17.3860-	29.38	0.0084	3516	Cost-effective
Audiometry	65, 70	1084.54	711.28-	17.6936	17.3842–	29.60	0.0058	5119	Dominated
Internet	50, 55, 60, 65	1087.39	707.29–	17.6963	17.3862-	32.44	0.0084	3865	Extendedly
Audiometry	55, 60, 65	1092.64	715.32-	17.6929	17.3844–	37.69	0.0050	7495	Dominated
Audiometry	50, 55, 60	1092.73	713.99–	17.6915	17.3843– 17.9926	37.78	0.0036	10408	Dominated
Audiometry	50, 60, 70	1093.26	715.71 to 1664.86	17.6930	17.3853 to 17.9946	38.31	0.0051	7473	Dominated
Internet	50, 55, 60, 65, 70	1093.39	712.99– 1665.27	17.6982	17.3878– 18.0048	38.45	0.0104	3699	Cost-effective
Audiometry	60, 65, 70	1093.65	717.87–	17.6946	17.3850-	38.70	0.0067	5736	Dominated
Audiometry	55, 60, 65, 70	1102.81	724.24-	17.6953	17.3863-	47.86	0.0074	6468	Dominated
Audiometry	50, 55, 60,	1103.96	723.53-	17.6937	17.3861-	49.02	0.0058	8417	Dominated
Audiometry	50, 55, 60, 65, 70	1114.12	734.06–	17.6960	17.3878– 17.9995	59.18	0.0082	7222	Dominated
CI, confidence interval; ICER, incremental cost-effectiveness ratio; QALY, quality-adjusted life-year.									

Morris et al. [20] reported an ICER of £1461/QALY (€1680/QALY) for an audiometric screening at age 60 years, repeated at ages 65 and 70 years, compared with current practice in the United Kingdom. In our study, the ICER of this strategy was €5736/QALY (dominated by Internet screening at age 50, 55, 60, 65, and 70 years). The difference in the ICER might be caused by differences in model input estimates. For example, the costs of a hearing aid trial and a hearing aid are much higher in The Netherlands than in the United Kingdom, and the probability of hearing aid uptake might be lower because Dutch patients have to pay a substantial amount of money for a hearing aid (on average €600 in 2011), whereas most patients in the United Kingdom receive hearing aids for free. Differences in model structure probably contribute to the discrepancy between the ICERs as well. We modeled

screening programs for the detection of a best-ear pure-tone average hearing loss (at 0.5, 1, 2, and 4 kHz) of 25 dB or more, whereas Morris et al.'s screening program aimed to detect a hearing loss of 30 dB or more. And unlike Morris et al. [20], we included the costs of care given to people who consult a hearing care professional or even start a hearing aid trial but do not take up hearing aids. Our study showed that in a more expensive hearing care system, audiometric screening is cost-effective compared with no screening. Internet screening, however, is the most cost-effective approach.

Liu et al. [19] reported that adult hearing screening using a handheld screening device is cost-effective. They based their conclusion on a randomized controlled trial that showed that screening led to a significant increase in hearing aid use 1 year



Fig. 2 – Cost-effectiveness plane. The incremental costs and quality-adjusted life-years of telephone screening (stars), Internet screening (circles), HearCheck screening (triangles), and audiometric screening (squares) compared with no screening are presented. The line represents the costeffectiveness frontier. The figure is based on the results of the probabilistic sensitivity analysis.

later (2.8%). The incremental costs of screening per additional hearing aid user were US \$1439 (€1040). It cannot be concluded from this trial, however, whether screening is indeed cost-effective because it is unknown how much society is willing to pay for one additional hearing aid user. So far, it has only been assessed how much society is willing to pay for QALYs gained. Other limitations of this trial are the short follow-up period and the fact that it included particularly male veterans, a study sample that may not be representative for the total adult population. By using a Markov model we were able to calculate the lifetime costs and effects of screening for a general population cohort. And because we measured the effectiveness in terms of QALYs gained, we could draw conclusions with regard to the cost-effectiveness of a large number of screening strategies.

Our study has several limitations. Bias may be introduced by the fact that the model parameter estimates were derived from several sources, none of which was a randomized controlled trial. The probabilities of screen participation for telephone screening, Internet screening, and screening at the GP, and some of the probabilities of help-seeking after screening, however, were all derived from one study [33]. The assumptions we had to make about some of the parameter estimates due to a lack of evidence

may have biased the results as well. With probabilistic sensitivity analysis, however, we accounted for the uncertainty in the estimates by assigning wide distributions to the model parameters and one-way sensitivity analyses showed that the results were insensitive to the assumptions we were most uncertain about. A second limitation is that the costs of management, quality control, and monitoring of the screening strategies were not taken into account. Because these costs will be similar for all screening strategies, inclusion of these costs would not have changed the strategy ranking. Inclusion of these costs, however, would have resulted in higher ICERs of the screening strategies compared with no screening. Post hoc threshold analysis of strategy costs revealed that the strategy with Internet screening at age 50, 55, 60, 65, and 70 years would still be cost-effective if the incremental costs of the strategy (compared with no screening) would increase from €38 to €208. Based on this, it also seems plausible that when the costs of management, quality control, and monitoring are taken into account, the screening strategy will be cost-effective. A third limitation of the study is that the model disregarded rehabilitation options other than conventional hearing aids, such as communication training or assistive listening devices. In most previous studies on adult hearing screening, rehabilitation was limited to hearing aid fitting [35]. Consequently, data on the effect of screening on the uptake of other rehabilitation options were insufficient to incorporate in the model, as were data on quality-of-life gain by other rehabilitation options. Future research to address this topic is recommended.

The utility estimates used in the model were based on the Health Utility Index Mark 3 questionnaire. This is the recommended instrument for measuring utility scores to assess healthrelated quality of life in hearing impairment because of its good validity and responsiveness [36]. Other multiattribute utility instruments, such as the Health Utilities Index Mark 2, the EuroQol five-dimensional questionnaire, and the six-dimensional health state short form (derived from short-form 36 health survey), are less sensitive to changes in utility scores after hearing aid fitting or even unable to measure a change at all [28,37,38]. If another multiattribute utility instrument had been used, adult hearing screening may not have been cost-effective. Although we used the most sensitive utility instrument, the screening strategies led to only a little gain in quality of life (0.0003-0.0104 QALYs). QALY gains from population screening programs generally tend to be quite modest [39,40], probably because the effect of treatment is diluted by the large number of healthy people in the target population for whom no quality-oflife gain can be achieved.

To what extent our results can be generalized and transferred to other countries depends on international similarities and differences with regard to health care system and culture. For example, hearing aid uptake after screening may be higher in



Fig. 3 - Cost-effectiveness acceptability frontier. QALY, quality-adjusted life-year.

countries where hearing aids are provided free of charge, as in the United Kingdom or Denmark. And in countries where patients' financial contribution to hearing aids is higher than in The Netherlands, for example, in Belgium, screening will probably be less cost-effective because the high hearing aid costs may withhold screen participants from taking up hearing aids. Also, the likelihood to participate in hearing screening differs between countries. Koopman et al. [33] found that compared with Dutch adults, British and German adults were much more likely to do a telephone test (50% and 76% vs. 33%) or an Internet test (65% and 73% vs. 44%). And although Dutch and British adults were found to be equally likely to opt for additional hearing testing after a positive screen (~80%), the Germans were much more likely to do so (~95%) [33]. Furthermore, the effectiveness of Internet screening will depend on the proportion of adults in the target population who have Internet access at home. The Netherlands is leading in Europe, with 95% of the households having Internet access, followed by Luxembourg (94%), Denmark, and Sweden (both 93%). Internet access in Europe is lowest in Bulgaria (54%), Greece (56%), and Romania (58%) [41].

In conclusion, this study suggests that nationwide adult hearing screening programs are (in comparison with no screening) cost-effective interventions to increase the quality of life of adults with hearing loss, with Internet screening at age 50 years, repeated at ages 55, 60, 65, and 70 years, being the optimal strategy. Because at a threshold of \notin 20,000/QALY this strategy was with 100% certainty cost-effective compared with current practice and with 69% certainty the most cost-effective among all strategies, policymakers might consider nationwide implementation. Telephone screening is a good alternative for people without Internet access.

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Supplemental Materials

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REFERENCES

- Nachtegaal J, Smit JH, Smits C, et al. The association between hearing status and psychosocial health before the age of 70 years: results from an internet-based national survey on hearing. Ear Hear 2009;30:302–12.
- [2] Monzani D, Galeazzi GM, Genovese E, et al. Psychological profile and social behaviour of working adults with mild or moderate hearing loss. Acta Otorhinolaryngol Ital 2008;28:61–6.
- [3] Gopinath B, Wang JJ, Schneider J, et al. Depressive symptoms in older adults with hearing impairments: the Blue Mountains Study. J Am Geriatr Soc 2009;57:1306–8.
- [4] Chisolm TH, Johnson CE, Danhauer JL, et al. A systematic review of health-related quality of life and hearing aids: final report of the American Academy of Audiology Task Force on the Health-Related Quality of Life Benefits of Amplification in Adults. J Am Acad Audiol 2007;18:151–83.
- [5] Vuorialho A, Karinen P, Sorri M. Effect of hearing aids on hearing disability and quality of life in the elderly. Int J Audiol 2006;45:400–5.
- [6] Chia EM, Wang JJ, Rochtchina E, et al. Hearing impairment and healthrelated quality of life: the Blue Mountains Hearing Study. Ear Hear 2007;28:187–95.
- [7] Joore MA, van der Stel H, Peters HJ, et al. The cost-effectiveness of hearing-aid fitting in the Netherlands. Arch Otolaryngol Head Neck Surg 2003;129:297–304.

- [8] Chao TK, Chen TH. Cost-effectiveness of hearing aids in the hearingimpaired elderly: a probabilistic approach. Otol Neurotol 2008;29:776–83.
- [9] Davis A, Smith P, Ferguson M, et al. Acceptability, benefit and costs of early screening for hearing disability: a study of potential screening tests and models. Health Technol Assess 2007;11:1–294.
- [10] Leegwater E, Lammerts van Bueren W. Hearing in the Netherlands [in Dutch]. Amsterdam: TNS NIPO, 2005.
- [11] Meister H, Walger M, Brehmer D, et al. The relationship between prefitting expectations and willingness to use hearing aids. Int J Audiol 2008;47:153–9.
- [12] Van Thiel L. Hearing in the Netherlands [in Dutch]. Amsterdam: TNS NIPO, 2010.
- [13] Humes LE. The contributions of audibility and cognitive factors to the benefit provided by amplified speech to older adults. J Am Acad Audiol 2007;18:590–603.
- [14] Janacsek K, Fiser F, Nemeth D. The best time to acquire new skills: agerelated differences in implicit sequence learning across the human lifespan. Dev Sci 2012;15:496–505.
- [15] Lunner T. Cognitive function in relation to hearing aid use. Int J Audiol 2003;42:S49–58.
- [16] Davis A, Stephens D, Rayment A, Thomas K. Hearing impairments in middle age: the Acceptability, Benefit and Cost of Detection (ABCD). Br J Audiol 1992;26:1–14.
- [17] Stephens SD, Callaghan DE, Hogan S, et al. Hearing disability in people aged 50-65: effectiveness and acceptability of rehabilitative intervention. BMJ 1990;300:508–11.
- [18] Yueh B, Collins MP, Souza PE, et al. Long-term effectiveness of screening for hearing loss: the Screening for Auditory Impairment – Which Hearing Assessment Test (SAI-WHAT) randomized trial. J Am Geriatr Soc 2010;58:427–34.
- [19] Liu CF, Collins MP, Souza PE, Yueh B. Long-term cost-effectiveness of screening strategies for hearing loss. J Rehabil Res Dev 2011;48:235–43.
- [20] Morris AE, Lutman ME, Cook AJ, Turner D. An economic evaluation of screening 60- to 70-year-old adults for hearing loss. J Public Health (Oxf) 2013;35:139–46.
- [21] Duijvestijn JA, Anteunis LJ, Hoek CJ, et al. Help-seeking behaviour of hearing-impaired persons aged ≥55 years: effect of complaints, significant others and hearing aid image. Acta Otolaryngol 2003;123:846–50.
- [22] Smits C, Kapteyn TS, Houtgast T. Development and validation of an automatic speech-in-noise screening test by telephone. Int J Audiol 2004;43:15–28.
- [23] Smits C, Merkus P, Houtgast T. How we do it: the Dutch functional hearing-screening tests by telephone and internet. Clin Otolaryngol 2006;31:436–40.
- [24] Leensen MC, de Laat JA, Dreschler WA. Speech-in-noise screening tests by internet, part 1: test evaluation for noise-induced hearing loss identification. Int J Audiol 2011;50:823–34.
- [25] Mitchell P, Gopinath B, Wang JJ, et al. Five-year incidence and progression of hearing impairment in an older population. Ear Hear 2011;32:251–7.
- [26] Statistics Netherlands. StatLine database. Available from: http://statline.cbs.nl/StatWeb. [Accessed April 11, 2014].
- [27] Van den Brink RH, Wit HP, Kempen GI, van Heuvelen MJ. Attitude and help-seeking for hearing impairment. Br J Audiol 1996;30:313–24.
- [28] Grutters JP, Joore MA, van der Horst F, et al. Choosing between measures: comparison of EQ-5D, HUI2 and HUI3 in persons with hearing complaints. Qual Life Res 2007;16:1439–49.
- [29] Hakkaart-van Roijen L, Tan SS, Bouwmans CAM. Manual for costing research [in Dutch]. Diemen: College voor Zorgverzekeringen, 2010.
- [30] Drummond MF, Sculpher MJ, Torrance GW, et al. Methods for the Economic Evaluation of Health Care Programmes. (3rd ed.). New York: Oxford University Press, 2005.
- [31] Van den Berg M, de Wit GA, Vijgen SM, et al. Cost-effectiveness of prevention: opportunities for public health policy in the Netherlands. Ned Tijdschr Geneeskd 2008;152:1329–34.
- [32] Fenwick E, Claxton K, Sculpher M. Representing uncertainty: the role of cost-effectiveness acceptability curves. Health Econ 2001;10:779–87.
- [33] Koopman J, Davey E, Thomas N, et al. How should hearing screening tests be offered? Int J Audiol 2008;47:230–7.
- [34] Van den Berg PJ, Prins A, Verschuure H, Hoes AW. Effectiveness of a single and a repeated screen for hearing loss in the elderly. Audiology 1999;38:339–40.
- [35] Pronk M, Kramer SE, Davis AC, et al. Interventions following hearing screening in adults: a systematic descriptive review. Int J Audiol 2011;50:594–609.
- [36] Yang Y, Longworth L, Brazier J. An assessment of validity and responsiveness of generic measures of health-related quality of life in hearing impairment. Qual Life Res 2013;22:2813–28.
- [37] Joore MA, Brunenberg DE, Chenault MN, Anteunis LJ. Societal effects of hearing aid fitting among the moderately hearing impaired. Int J Audiol 2003;42:152–60.

- [38] Barton GR, Bankart J, Davis AC, Summerfield QA. Comparing utility scores before and after hearing-aid provision: results according to the EQ-5D. HUI3 and SF-6D. Appl Health Econ Health Policy 2004;3:103–5.
- [39] Kim LG, Thompson SG, Briggs AH, et al. How cost-effective is screening for abdominal aortic aneurysms? J Med Screen 2007;14:46–52.
- [40] Maeda K, Shimbo T, Fukui T. Cost-effectiveness of a community-based screening programme for chronic atrial fibrillation in Japan. J Med Screen 2004;11:97–102.
- [41] Eurostat. News Release 199/2013: Internet Access and Use in 2013. Luxembourg: Eurostat Press Office, 2013.