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Quality of Life and Cost-Effectiveness of Cochlear Implants: A Narrative Review

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Quality of life · Health utility · Costs · Savings · Cost-utility · Cost-benefit · Cost-effectiveness

Abstract
Objectives: To review evidence regarding the health-related quality of life (HRQoL) and cost-effectiveness of unilateral and bilateral cochlear implantation (CI) among children and adults with severe-to-profound hearing loss. ***Study Design:*** Narrative review. ***Methods:*** Publications related to quality of life (QoL) and costs of care in CI were acquired through searches in English-language databases. Studies were included if they had identified the HRQoL attainment, cost of care, cost-utility, or cost-effectiveness associated with CI. ***Results:*** 57 studies were critically reviewed. The QoL outcome metrics used in these articles were divided into 2 categories – generic and condition specific. In studies investigating children, many reported no significant difference in QoL attainment between CI recipients and normal-hearing peers. In adults, significant improvements in QoL after implantation and higher QoL than in their nonimplanted (hearing-aided) peers were frequently reported. Studies involving an older adult cohort reported significant improvement in QoL after implantation, which was often independent of audiological performance. Overall, the calculated cost-utility ratios consistently met the threshold of cost acceptance, indicating acceptable values for expenditures on CI. ***Conclusions:*** Considerable work has been done on the QoL attainment and health economic implications of CI. Unilateral CI across all age groups leads to reported sustained benefits in the recipients' overall and disease-specific QoL. With increased cost associated with bilateral CI, further study is needed to characterize its costs and benefits with respect to the recipients' health, well-being, and contributions to society.

Introduction

Since its introduction into clinical practice, hearing care clinicians have increasingly utilized cochlear implantation (CI) to restore auditory stimulation in selected patients with advanced sensorineural hearing loss. Over the last 15 years, cochlear implant manufacturers have consistently expanded their technologies and device so-

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phistication. In this same period, criteria for implantation in candidates of all ages have been broadened based on more refined methods of diagnosis and heightened levels of benefit observed with implantation.

The considerable direct and indirect costs associated with this intervention remain a significant barrier to patients and their families, and present an obstacle to a more widespread adoption of CI technology. However, CI also results in economically valuable quality of life (QoL) improvement [Lindemark et al., 2014; Semenov et al., 2012], enhanced academic achievement [Semenov et al., 2013], and improved vocational outcomes [Emmett and Francis, 2015; McKinnon, 2014] leading to potentially even greater clinical and nonclinical cost savings among CI recipients. As a result, the measurement of the economic benefits of implantation has become essential in making policy decisions. In the United States, one of the first economic analyses investigating CI cost-utility helped convince the Californian MediCal program to include CI in their funding schedule [Harris and Anderson, 1999]. Economic evaluation of health interventions requires the consideration of factors beyond clinical effectiveness, such as the intervention's effects on a patient's level of physical and emotional function, vitality, interpersonal communication, independence in daily living, overall satisfaction with life, and mental health.

Our objective was to perform a narrative review of the literature on the QoL attainment among CI recipients of all ages, and examine cost-effectiveness measures as related to CI. Specifically, we reviewed (1) the characteristics of the studies reporting on QoL attainment and health economic implications of CI, (2) the types of QoL measures used, (3) key results, conclusions, and limitations of the included studies, and (4) outcome differences between individuals implanted with unilateral and bilateral implants.

Methods

We designed a narrative literature review to assess the health-related QoL (HRQoL) attainment and cost-effectiveness of CI in recipients of all ages with severe to profound hearing loss. A secondary goal was to compare the health economic outcomes between unilateral and bilateral implantation. Multiple criteria were agreed upon and used in identifying studies for this review prior to inclusion or exclusion.

Study Design

Original contributions and review articles from peer-reviewed publications were identified. Both prospective and retrospective study designs, and cross-sectional and longitudinal trials were included.

Participants

Studies that enrolled participants with either prelingual or postlingual onset of hearing loss in 3 age categories included children (aged less than 18 years), adults (18–65 years), and older adults (older than 65 years). No exclusions of etiology of hearing loss, duration of hearing loss, timing of CI, or duration of follow-up were made.

Outcome Measures

Identified studies with primary outcome measures reported noted:

1. HRQoL metrics utilized in CI and overall QoL associated with CI
2. Costs and savings associated with CI
3. Cost-effectiveness, cost-benefit, or cost-utility of CI

Secondary outcomes that were also identified:

1. Types of QoL metrics
2. Consideration of bilateral versus unilateral CI

Search Methods for Identification of Studies

In order to capture the breadth of contemporary literature on all relevant studies investigating CI, the main English language electronic biomedical publication databases were utilized to identify source articles. Specifically, we mined PubMed and EMBASE for classic biomedical literature, periodicals covering audiology and communication sciences (CINAHL), psychology periodicals (PsycINFO and Web of Science), and economic periodicals (EconLit). We used variations of the term "cochlear implantation" and "cost-effectiveness" to identify potential articles of interest. While there were no restrictions on the articles' country of origin, only abstracts available in English were screened for inclusion in the review.

Study Inclusion

Identified studies using the criteria above were reviewed in 2 stages. In the first stage, titles and abstracts of all retrieved articles were reviewed for relevance. Inclusion criteria were: (1) original peer-reviewed research article or literature review published in a peer-reviewed journal, (2) study abstract available in English, (3) study participants had cochlear implants or were candidates for CI, and (4) study participant ages or age at implantation were clearly identified. After article title and abstracts had been screened, the full-length articles were reviewed. Articles were accepted for review if they met the above 4 criteria and all of the following: (1) study reported on any of the primary outcomes of this review, or (2) study included outcomes of bilateral versus unilateral implantation.

Study Analyses

For each included article, the basic characteristics of each study were extracted including details of study design, characteristics of participants, interventions, and the primary and secondary outcomes. Only data explicitly reported in the articles were included in the current review. No attempt was made to contact the investigators of included trials for any missing data. When a statistic was provided, we also examined the degree of overlap in the reported confidence intervals where appropriate. The generation of weighted averages of incremental cost-effectiveness ratios was completed by using literature-reported gains and the number of patients included in the pertinent study. If there was no overlap amongst reported statistics, this was taken to indicate the presence of heterogeneity.

Results

Study Selection and Common Characteristics

After subjecting our initial database search to our primary exclusion criteria, 68 articles were included in the final results. Studies used both cross-sectional and longitudinal design to capture QoL and health economic outcomes following CI. The sample size of the 57 studies ranged from 8 to 908 CI recipients (Tables 1–4). Additionally, there was considerable variance in the ages at the time of CI within the 3 age categories considered in this review and the duration of implant use. Most studies did not report age ranges at implantation, but some did report on the standard deviation of age at CI.

QoL Metrics

The QoL outcome metrics used in these articles could be divided into 2 categories – generic and disease specific. They were then further divided into population-validated and ad hoc QoL instruments designed for the specific use of the given trial. Fifteen of the studies focused on presenting metrics for assessing the QoL attainment of children with cochlear implants (Table 1).

Generic Instruments in Children

Eight studies reported on generic health utility metrics, which included KINDL® [Huber, 2005; Loy et al., 2010; Warner-Czyz et al., 2009, 2011], visual analog scale (VAS) [Clark et al., 2012; Sach and Barton, 2007], Quality of Well-Being Scale (QWB) [Clark et al., 2012], Child Health and Illness Profile (CHIP) child edition [Clark et al., 2012; Meserole et al., 2014], European Quality of Life Questionnaire in 5 domains (EQ5D) [Clark et al., 2012; Sach and Barton, 2007], Satisfaction with Life Scale (SWLS) [Spencer et al., 2012], and EuroQoL VAS [Sach and Barton, 2007]. A comprehensive review of these surveys is beyond the subject of the current paper and may be found elsewhere [Froberg and Kane, 1989; Zullig et al., 2010]. KINDL® is a 24-item QoL questionnaire developed in Germany and validated for use in children from 8 to 16 years of age [Ravens-Sieberer and Bullinger, 1998]. It is composed of 6 scales: physiological and psychological well-being, self-esteem, family, friends, and functioning. Both child report and parent-proxy versions are available, and normative values have been established for comparison. As is the case for all questionnaires administered to children, results are limited by the fact that very young children cannot respond, and parents serve as proxy respondents.

The CHIP child edition is a population-validated self-reported QoL instrument for children 6–11 years old [Riley et al., 2004]. It is comprised of 5 domains: satisfaction (with self and health), comfort (emotional and physical symptoms and limitations), resilience (positive activities that promote health), risk avoidance (risky behaviors that influence future health), and achievement (of social expectations in school and with peers). The QWB is a 71-item questionnaire and measures the respondent's overall health status and well-being over the previous 3 days across 4 domains: physical activities, social activities, mobility, and symptom/problem complexes [Seiber et al., 2008].

The EQ5D is a self-administered questionnaire that assesses function in 5 socially relevant domains: mobility, self-care, usual activities, pain-discomfort, and anxiety-depression. The Euro-QoL VAS is a linear visual analog scale that accompanies the EQ5D [Gusi et al., 2010]. The VAS presents a line with grid marks from 0 ("death") to 100 and asks the respondent or a respondent's proxy to draw a line corresponding to the respondent's QoL. Lastly, the SWLS is a short 5-item instrument designed to measure global cognitive judgments of satisfaction with one's life and is not a health-specific metric [Kobau et al., 2010].

Disease-Specific Instruments in Children

Six studies reported on disease-specific QoL metrics, which include youth QoL deaf and hard of hearing edition (YQoL-DHH) [Meyer et al., 2013], Deaf Identity Scale [Spencer et al., 2012], Children with Cochlear Implants: Parental Perspectives questionnaire [Fortunato-Tavares et al., 2012; Huttunen et al., 2009], and the Nijmegen Cochlear Implant Questionnaire (NCIQ) [Necula et al., 2013]. The YQoL-DHH is a 32-item questionnaire that assesses hearing loss-related QoL across 3 domains: self-acceptance/advocacy, perceived stigma, and participation [Patrick et al., 2011]. The Deaf Identity Scale is composed of 3 subscales, including hearing identification, deaf identification, and dual identification. Each subscale consisted of 5 statements regarding the individual's desire to associate with and the individual's assumed similarity with deaf, hearing, or both groups [Weinberg and Sterritt, 1986]. The Glasgow Children's Benefit Inventory (GCBI, [Sparreboom et al., 2012]; Table 2) is used to measure and evaluate a child's health benefit retrospectively, after an otological intervention. Answers are provided on a 5-point Likert scale and converted to a 200-point scale which ranges from –100 (maximal harm) to +100 (maximal benefit). The GCBI can also be analyzed according to 4 domains, including emotion, physical health, learning, and vitality.

Table 1. Quality of life outcomes following unilateral cochlear implantation

Study	Sample size *(n)*, gender (*n* female), mean age at CI/ time of study	Study type	Primary outcome metric	Geographic origin	Findings
Studies in children					
Clark et al., 2012	*n* = 188 (98 female), undergoing CI before 5 years of age with an average age of 26.7 (14.5) months at implantation	Prospective, longitudinal study with 4 years of follow-up	VAS, VAS-D, QWB, CHIP-Child edition, and EQ5D	USA	Baseline deficits of CI relative to NH peers were larger in developmental factors than in QoL No significant QoL differences were observed in CI children after 4 years as compared to NH peers
Loy et al., 2010	*n* = 52 (29 female) implanted at 3.4 (2.1) years and 9.1 (1.1) years of age at time of study *n* = 34 (23 female) implanted at 5.8 (4.0) years and 13.7 (1.4) years of age at time of study	Cross-sectional	Kid KINDL® and Kiddo KINDL®	USA	Earlier implantation and longer use of implants resulted in higher QoL scores
Meserole et al., 2013	*n* = 129 (77 female), undergoing CI before 5 years of age with an average age of 8.0 (1.3) years at time of study	Prospective, longitudinal study with 6 years of follow-up	CHIP-Child edition	USA	QoL is comparable between CI recipients and their NH peers, while their parents reported similar scores when compared with socioeconomically comparable families of NH children
Meyer et al., 2013	*n* = 63 (31 female) implanted at 62.8 (10–180) months of age	Cross-sectional	YQoL-DHH	USA	Compared outcomes of implanted versus nonimplanted children using HA or no assistive technology Youth using no technology or using cochlear implants tended to score higher than those using HA in mainstream schools (with or without deaf and hard-of-hearing programs) and in schools for the deaf However, the no-technology group consistently outperformed implanted children in QoL attainment
Schorr et al., 2009	*n* = 37 (21 female) congenitally deaf children implanted at 3.0 (0.2–8.3) years of age and 9 (5–14) years of age at time of study	Cross-sectional	Ad hoc instrument	USA	QoL was reported as significantly improved with the cochlear implant, and increased QoL predicted better performance on the emotion identification task QoL was positively associated with age at first amplification supporting the importance of early detection and intervention in HL
Spencer et al., 2012	*n* = 41 (22 female) implanted at 7.2 (3.8) years and 21.9 (3.8) years of age at time of study	Longitudinal with over 10 years of follow-up	Satisfaction-with-Life scale, Deaf Identity scale	USA	Compared with their NH, adult-age peers, CI recipients had high educational achievement and reported a very high satisfaction with life Also, most of the individuals endorsed a dual identity, indicating they feel just as comfortable with deaf individuals as they do with hearing individuals
Warner-Czyz et al., 2009	*n* = 50 (22 female), implanted at 2.5 (1.5) years and 5.8 (1.1) years of age at time of study	Cross-sectional – CI vs. NH peers	Kiddy KINDL®	USA	Implanted children rated their overall QoL significantly more positively than did their parents, and child ratings did not differ significantly from NH children Overall, QoL correlated inversely with length of implant experience and chronologic age, but did not correlate with implantation age

Table 1 (continued)

Study	Sample size *(n)*, gender (*n* female), mean age at CI/ time of study	Study type	Primary outcome metric	Geographic origin	Findings
Warner-Czyz et al., 2011	*n* =140 ((72 female) implanted at 3.7 (2.8) years and 9.0 (3.2) years of age at time of study Implantation age for 3 subgroups: 2.5 (1.4), 3.4 (2.1), and 5.7 (3.9) years for youngest, middle, and oldest study groups	Cross-sectional	KINDL®, ad hoc instrument	USA	The youngest group (4–7 years: 82.8) rated generic QoL significantly more positively than older children (8–11 years: 75.3; 12–16 years: 70.4) Similar significant results emerged on the overall CI module (4–7 years: 79.8; 8–11 years: 77.8; 12–16 years: 71.3)
Edwards et al., 2012	*n* =88 (37 female) implanted at 3.8 (2.1) years and 11.1 (2.0) years of age at time of study	Cross-sectional – CI with additional comorbidities vs. no additional comorbidities	Ad hoc instrument	UK	QoL of the 42% of the children who had additional needs was rated poorer than that of the children without additional needs on 3 of the 4 subscales as well as on the total QoL rating Despite this, however, most parents reported that their child's QoL had improved "moderately" or "very much," irrespective of whether they had additional needs
Huber, 2005	*n* = 18 (6 female) implanted at 4.3 (2.0) years and 10.5 (1.4) years of age at time of study *n* = 11 (2 female) implanted at 7.6 (1.8) years and 14.5 (1.2) years of age at time of study	Cross-sectional	KINDL®	Austria	The younger group reached a significantly lower total QoL compared to NH peers, 64.6 (8.9) vs. 76.8 (8.6), respectively This result differs significantly from parent-proxy responses of 80.8 (5.4) There was no significant QoL difference between the older study group, their NH peers, and parental ratings: 72.1 (10.3) in the self-rating, 73.5 (8.8) for NH peers, and 76.3 (10.2) for parental ratings
Huttunen et al., 2009	*n* = 36 (19 females) implanted at 3.4 (2.3) years and 5 (2) years of age at time of study	Cross-sectional	CCIPP	Finland	Parents were most satisfied with improved/ expanded social relations, improved communication (the development of spoken language), general functioning with the help of hearing, and improved self-reliance of the child following CI
Necula et al., 2013	*n* = 84 (gender not reported) implanted at 5.2 (3.4) years and 7.6 (4.2) years of age at current study	Cross-sectional – young and older CI groups vs. HA peers	NCIQ	Romania	QoL was positively correlated with auditory performance and speech intelligibility, but negatively correlated with implantation age CI recipients outperformed HA users in auditory performance and speech production
Sach et al., 2007	*n* = 222 (109 female) aged 9.3 (3.6) years at time of study Children implanted at <4 years of age (108) and >4 years of age (114)	Cross-sectional	EQ5D, VAS, EuroQol VAS	UK	Regression analysis indicated lower EQ5D scores for: (1) children with an additional disability, (2) male children, (3) those with a lower level of auditory perception, and (4) those whose parents left school before the age of 18 EuroQol VAS, the mean difference between pre- and postimplantation score, was 0.14, compared to 0.35 for the QoL VAS, demonstrating that parents tended not to see HRQoL and QoL as equivalent, though both were strongly correlated
Stacey et al., 2006	*n* = 468 stratified into 6 groups by age at implantation (<5 or 5 and above) and duration of CI use (<2, 2–4, and ≥4)	Cross-sectional – CI vs. nonimplanted controls	Ad hoc instrument	UK	CI was consistently associated with advantages in auditory performance and spoken communication skills, but less consistently associated with advantages in educational achievements and QoL Only children implanted under 5 years of age and with 4 years of CI use had consistently improved QoL as compared to nonimplanted children

Table 1 (continued)

Study	Sample size *(n)*, gender (*n* female), mean age at CI/ time of study	Study type	Primary outcome metric	Geographic origin	Findings
Fortunato-Tavares et al., 2012	*n* = 10 (5 female) implanted at 4.5 (2.2) years and 6.2 (2.4) years of age at time of study	Cross-sectional	CCIPP	Brazil	CI had a positive impact on QoL of implanted children and their families; there is a direct relationship between oral communication and QoL
Studies in adults					
Chung et al., 2012	*n* = 283 (163 female) implanted at 52.9 (15.1) years of age	Prospective, longitudinal study with 1 year of follow-up	SF-36	Canada	Significant improvement in QoL was observed before and after implantation across 5 of 8 domains of SF-36 (vitality, physical role functioning, mental health, emotional role functioning, and social functioning) Younger implant recipients outperformed older recipients in several domains
Cohen et al., 2004	*n* = 24 (16 female) implanted at 62.8 (7.3) years and 67.2 (8.3) years of age at study	Cross-sectional – CI vs. HA controls	NCIQ	USA	CI users showed twice as much overall QoL Significantly greater QoL benefit in CI than HA users across physical, psychological, and social subdomains of the questionnaire
Damen et al., 2007	*n* = 37 (20 female) implanted at 45.2 (5.4) years and 55.1 (16.0) years of age at time of study *n* =22 (14 female) implanted at 57.0 (13.4) years and 61.5 (13.1) years of age at time of study	Longitudinal, with at least 6 years of follow-up – compared 2 CI (younger and older) groups with nonimplanted peers	NCIQ, HUI3, SF-36	The Netherlands	Beneficial impact of implantation on QoL, effect remained stable across 6 years Implanted adults attained higher QoL than nonimplanted study participants
Fuller et al., 2013	*n* = 22 (16 female) implanted at 41.2 (14.3) years and 47.4 (15.0) years of age at time of study	Cross-sectional	Ad hoc instrument regarding perception and enjoyment of music, SSQ, NCIQ, CIFI	The Netherlands	Mean total scores on QoL of implanted adults were: NCIQ – 72 (44–92), CIFI – 11 (4–19), and SSQ – 4.4 (0.6–7.6) No significant correlations were shown between music perception and QoL scores
Harris and Anderson, 1999	*n* = 9 (3 women), age range 23–59 years	Longitudinal	CES-D, SLA, QWB	USA	Overall, there was no significant change in scores on the CES-D, but a measured increase in quality of life metrics on the SLA and QWB
Klop et al., 2007	*n* = 8 (5 female) pre-lingually hearing-impaired adults implanted at 36.0 (21–55) years of age	Longitudinal	HUI2, NCIQ, VAS	The Netherlands	Compared QoL before and after implantation Significant improvement was measured at 4 months afterimplantation, but no additional significant changes occurred thereafter
Klop et al., 2008	*n* = 44 (29 female) implanted at 54.7 (15.5) years of age	Longitudinal with 1 year of follow-up	HUI3, NCIQ	The Netherlands	Significant improvement in QoL following implantation, with the largest improvement in categories concerning physical functioning (hearing) Most QoL benefit observed within 4 months of implantation HUI2 increased from 0.68 (0.14) to 0.83 (0.12), and NCIQ increased from 44.5 (12.7) to 70.3 (10.0)
Kuthubutheen et al., 2015	*n* = 30 (16 female) 56 years of age at time of study Mean duration of unilateral CI use was 4 years	Cross-sectional	HUI3, EQ5D, TTO, VAS	Canada and USA	Individuals with unilateral CI scored consistently higher on all measures of QoL attainment than nonimplanted controls: gaining HUI3 (0.270), EQ5D (0.140), VAS (0.130), and TTO (0.170)

Table 1 (continued)

Study	Sample size *(n)*, gender (*n* female), mean age at CI/ time of study	Study type	Primary outcome metric	Geographic origin	Findings
Lassaletta et al., 2006	*n* = 30 (18 female) implanted at 48.0 (8–66) years of age	Cross-sectional	GBI and 2 ad hoc instruments	Spain	GBI revealed a positive effect from CI in 93% of patients CI significantly enhanced discrimination ability, telephone use, and self-confidence A high degree of satisfaction was achieved in all situations except when there was background noise, and 96% of patients would recommend the operation to a friend
Mo et al., 2005	*n* = 27 (15 female) at 57.6 (14.5) years of age at time of study The mean duration of deafness before CI was 8.5 (10.3) years	Longitudinal with 12–15 months of follow-up	PQLF, HSCL-25, SF-36, IRQF	Norway	Scores were significantly improved following CI compared to preimplantation baseline on HSCL-25, in 4 of 6 categories of the PQLF and in 4 of 5 categories of the IRQF In the SF-36, only 1 of 8 scales showed significant improvement These improvements were largest in categories concerning communication, feelings of being a burden, isolation, relations to friends and family, and improvement in relatives' daily lives
Olze et al., 2011	*n* = 43 (31 female) implanted at 51.7 (16.9) years of age	Cross-sectional	NCIQ	Germany	Significant increase in disease-specific QoL following implantation: mean NCIQ scores increased from 39.3 (15.1) to 60.3 (13.1) Highly significant improvements were observed in all 6 subdomains of the NCIQ In addition to improvements in hearing, speech understanding, and disease-specific QoL, psychological comorbidity was reduced, and coping strategies were improved following CI surgery
Rembar et al., 2009	*n* =74 (44 female) 56.2 (15.2) years of age at time of study and with 2.1 (1.5) years since CI	Cross-sectional	Ad hoc qualitative instrument	Norway	Recipients reported that they had received "new life" with the implant and that their overall psychological well-being was improved
Rembar et al., 2012	*n* = 53 (18 female) implanted at 53.9 (16.2) years of age with 1.5 (0.5) years since CI	Cross-sectional	PGWB	Norway	No difference in mean PGWB index was found between CI users and the general population
Zhao et al., 2008	*n* = 24 (17 female) 53.9 (12.9) years of age at time of study with a mean of 4.2 years since CI	Longitudinal with 4 years of follow-up	VAS, ad hoc instrument	UK	Changes in rated QoL of all patients were significantly associated with changes in specific complaints (ability to communicate, feelings of isolation, telephone use, etc.) After implantation, improvements in communication abilities, reduced psychological problems, and improvements in abilities of daily life were the key determinants of QoL improvement of individual patients
Hawthorne et al., 2004	*n* = 34 (16 female) 49 (13) years of age at time of study with a mean of 6 months since CI	Prospective, longitudinal	AQoL, HPS	Australia and New Zealand	Implantation resulted in significant improvements from preimplantation to 3- and 6-month postimplantation scores on AQoL (0.36–0.50 and 0.64, respectively) and HPS (0.48–0.64 and 0.68, respectively) Those in the top socioeconomic group obtained greatest gains
Looi et al., 2011	*n* = 94 (58 female) implanted at 51.9 (15.3) years and 51.5 (14.5) years of age at time of study	Prospective, longitudinal	Modified NCIQ, CISQ	New Zealand	Implanted adults had significantly higher QoL across all 6 subdomains of NCIQ than their nonimplanted hearing-impaired peers on the waiting list for a CI (mean QoL of 69.97 and 41.24, respectively)

Table 1 (continued)

Study	Sample size *(n)*, gender (*n* female), mean age at CI/ time of study	Study type	Primary outcome metric	Geographic origin	Findings
Studies in older adults					
Chung et al., 2012	*n* = 283 (163 female) implanted at 52.9 (15.1) years of age	Prospective, longitudinal study with 1 year of follow-up	SF-36	Canada	Significant improvement in QoL was observed before and after impantation across 5 of 8 domains of SF-36 (vitality, physical role functioning, mental health, emotional role functioning, and social functioning) Younger implant recipients outperformed older recipients in several domains
Di Nardo et al., 2013	*n* = 20 (13 female) implanted at 67.6 (5.5) years and 72.5 (5.6) years of age at time of study	Cross-sectional	SF-36 and Questionnaire for Self-Evaluation of CI Benefit with SADL scale modification	Italy	No significant differences were found between the study population and control (younger CI patients) on all subdomains of SF-36 and the Questionnaire for Self-Assessment of CI Benefit However, a significant difference was noted between the 2 populations in overall satisfaction derived from the implant (greater reported satisfaction in the older group)
Olze et al., 2012	*n* = 20 (10 female) implanted at 74.4 (4.6) years of age with a mean of 1.8 (1.6) years since CI	Longitudinal	NCIQ, SF-36	Germany	Elderly patients benefited from CI to a higher extent than younger implanted controls on disease-specific quality of life (NCIQ): 31.3 (16.6) to 68.0 (12.4) for the elderly group and 39.7 (14.8) to 61.1 (14.3) for the younger group before and after implantation, respectively Baseline QoL did not differ significantly between elderly and younger controls on SF-36 After CI, the elderly group experienced a significant increase in social functioning and mental health and a significant decrease in physical functioning, and physical component summary subscales of the SF-36
Orabi et al., 2006	*n* = 34 (15 female) implanted at 69.8 (0.8) years of age	Longitudinal	GBI, GHSI	UK	Statistically significant improvements after implantation were found in word and sentence recognition scores in quiet and noise, and 82% of patients were completely satisfied with their cochlear implants Patients judged that the implantation had restored half the loss of QoL that they had experienced as a result of severe-to-profound deafness, with a highly significant improvement in overall QoL after implantation
Ramos et al., 2013	*n* = 26 (9 female) implanted at >60 years and 69.0 (6.6) years of age at time of study	Longitudinal	GBI, SQ	Spain	Patients experienced significant improvement in their QoL in all areas, especially in general health, with a smaller improvement in social interaction Age, duration of deafness, and years wearing the process were statistically related to QoL
Vermiere et al., 2005	*n* = 25 (not provided) implanted at >70 years	Longitudinal	HHIA, GBI	Belgium	QoL improvement following CI for the elderly was similar to that of implanted younger adults

AQoL, Assessment of Quality of Life; CCIPP, Children with Cochlear Implants: Parental Perspectives; CES-D, Center for Epidemiologic Studies Depression Scale; CHIP, Child Health and Illness Profile; CIFI, Cochlear Implant Function Index; CISQ, Cochlear Implant Satisfaction Questionnaire (a combination of Satisfaction in Daily Life Questionnaire and Client Satisfaction Questionnaire-8); EQ5D, EuroQoL-5D; GBI, Glasgow Benefit Inventory; GHSI, Glasgow Health Status Inventory questionnaire; HA, hearing aid; HHIA, Hearing Handicap Inventory – Adults; HL, hearing loss; HPS, Hearing Participation Scale; HSCL-25, Hopkins Symptom Check List – 25 items; HUI, Ontario Health Utilities Index; IRQF, Index Relative Questionnaire Form; KINDL®, questionnaire for measuring health-related quality of life in children and adolescents including Kiddy (4–7 years), Kid (8–11 years), and Kiddo (12–16 years); NCIQ, Nijmegen Cochlear Implant Questionnaire; NH, normal hearing; PGWB, Psychological General Well-Being index; PQLF, Patient Quality of Life Form; QoL, quality of life; QWB, Quality of Well-Being Scale; SADL, Satisfaction in Daily Life Questionnaire; SLA, Satisfaction with Life Areas Scale; SF-36, Short Form-36 questionnaire; SSQ, Speech, Spatial, and Qualities questionnaire; TTO, time trade-off; VAS, visual analog scale; VAS-D, visual analog scale development; YQoL-DHH, Youth Quality of Life – Deaf and Hard of Hearing; SQ, Specific Questionnaire.

Table 2. Quality of life outcomes following bilateral cochlear implantation

Study	Sample size (n), gender (*n* female), mean age at CI/time of study	Study type	Primary outcome metric	Geographic origin	Findings
Studies in children					
Perez-Mora et al., 2012	*n* = 30 (14 female) 8 years (2–16 years) of age at time of study with 3.5 years (1–9 years) since implantation	Cross-sectional	KINDL®	Spain	No difference in QoL was found between bilaterally hearing-impaired children with (1) CI and contralateral HA, (2) bilateral HA, and (3) normal-hearing peers
Sparreboom et al., 2012	*n* = 30 children with prelingual HL implanted at 1.8 years and 5.3 years of age for first and second CI, respectively	Prospective, longitudinal	Generic instruments: VAS, HUI3, PedsQL Disease-specific instruments: GCBI, SSQ, NCIQ	The Netherlands	No significant gain in generic QoL associated with sequential bilateral CI Significant QoL gain of 10.42, 0.62, and 0.79 for GCBI, SSQ, and NCIQ, respectively, at 24 months after implantation Unlike children with unilateral CI, QoL measures continued to improve with longer durations of bilateral implant use Age at second implantation had no significant impact on QoL gain
Studies in adults					
Kuthubutheen et al., 2015	*n* = 30 (19 female) 53 years of age at time of study Mean duration of bilateral CI use was 2 years	Cross-sectional	HUI3, EQ5D, TTO, VAS	USA and Canada	Individuals with bilateral CI scored consistently higher on all measures of QoL attainment than those with a unilateral implant and nonimplanted controls Bilateral vs. unilateral CI: HUI3 (0.035), EQ5D (0.040), VAS (0.070), and TTO (0.120) Bilateral vs. nonimplanted: HUI3 (0.305), EQ5D (0.180), VAS (0.200), TTO (0.290)
Buhagiar and Lutman, 2010		Cross-sectional	Outcomes from Bilateral Cochlear Implantation – Adults questionnaire	UK	While the first CI had the greatest perceived effect on the daily lives of families and friends of CI recipients, the second cochlear implant had more of an advantage on psychological and lifestyle effects than on changes in speech perception The second implant improved confidence levels and increased social participation
Lovett et al., 2010	*n* = 30 (14 female) implanted at 1.3 (2.9) and 3.8 (4.6) years for the 1st and 2nd CI, respectively, with 4.1 (3.1) years of CI use at time of study	Cross-sectional	HUI3	UK	No significant health utility difference between bilateral and unilateral CI: 0.83 vs. 0.78
Olze et al., 2012	*n* = 40 (29 female) implanted at 50.3 (14.5) and 53.8 (14.0) years for 1st and 2nd CI, respectively	Longitudinal	NCIQ	Germany	The first CI resulted in a statistically significant increase in disease-specific QoL as measured by the NCIQ: 39.3 (12.3) to 65.4 (12.7) The second implantation induced a further increase in the NCIQ scores to 71.3 (12.7)

For explanations of abbreviations, see Table 1.

The CCIPP is a cochlear implant-specific closed-set questionnaire that assesses the following dimensions of a child's experience with the implant: decision to implant, process of implantation, positive effect of the implant, communication, supporting the child, self-reliance, well-being and happiness, social relationships, education, and pre- and postoperative services provided by the implant center [O'Neill et al., 2004]. The NCIQ is another CI-specific QoL instrument covering 6 subdomains: basic sound perception, advanced sound perception, speech production, self-esteem, activity and social interactions [Hinderink et al., 2000].

Ad hoc Instruments in Children

Four articles used ad hoc instruments, which are designed for the purposes of an individual study [Edwards et al., 2012; Schorr et al., 2009; Stacey et al., 2006; Warner-Czyz et al., 2011]. These questionnaires provide considerable information on a child's experience with an implant and use one or a combination of open-ended questionnaires, semistructured interviews, or quantitative instruments. Three studies used quantitative QoL questionnaires [Edwards et al., 2012; Schorr et al., 2009; Stacey et al., 2006; Warner-Czyz et al., 2011] and one used a combination qualitative and quantitative instruments [Schorr et al., 2009]. While providing considerable detail and additional insights that are not often assessed in more established QoL instruments, these questionnaires have not been population validated and are not generalizable across different study groups.

Summary of Results for Children

Overall, there was no significant difference in QoL attainment between CI recipients and their normal-hearing peers [Clark et al., 2012; Huber, 2005; Meserole et al., 2014; Meyer et al., 2013]. Factors associated with poorer QoL outcomes included use of hearing aids (rather than a CI) [Meyer et al., 2013; Necula et al., 2013], shorter experience with an implant [Loy et al., 2010], older age at implantation [Necula et al., 2013; Schorr et al., 2009; Stacey et al., 2006; Warner-Czyz et al., 2011], additional developmental comorbidities [Sach and Barton, 2007; Edwards et al., 2012], male gender [Sach and Barton, 2007], lower auditory perception [Sach and Barton, 2007], lower parental educational level [Sach and Barton, 2007], and reduced oral communication by the child [Fortunato-Tavares et al., 2012].

QoL Outcomes in Adults

Fourteen of the 57 studies focused on presenting metrics for assessing the QoL attainment of adults with cochlear implants. The studies were comprised of participants from 9 countries with a sample size ranging from 8 to 283 study participants. The mean age at implantation ranged from 43 to 67.2 years (Table 1).

Generic Instruments in Adults

Ten studies reported on generic health utility metrics, which included the Short Form-36 (SF-36) [Chung et al., 2012; Damen et al., 2007; Mo et al., 2005], Ontario Health Utilities Index (HUI) Marks II and III [Damen et al., 2007; Faber and Grøntved, 2000; Klop et al., 2007, 2008; Sparreboom et al., 2012], the EQ5D, the VAS from the EQ5D, the time trade-off technique (TTO), the Australian Quality of Life (AQoL) instrument [Hawthorne et al., 2004], the Hopkins Symptom Check List 25 (HSCL-25) [Mo et al., 2005], the Psychological General Well-Being (PGWB) Index [Rembar et al., 2009], and the Glasgow Benefit Inventory (GBI) [Lassaletta et al., 2006]. Although none of these measures are specific to hearing or the effects of CI, some do incorporate hearing ability as part of the measure, as discussed below.

The SF-36 health questionnaire is a generic, validated, 36-item tool developed to evaluate HRQoL of a medical or surgical intervention. The multi-item scale assesses 8 different health domains, including physical functioning, role limitations due to physical problems, social functioning, bodily pain, general mental health, role limitations due to emotional problems, vitality, and general health perceptions [Chung et al., 2012]. The PGWB is a questionnaire that has been used to compare general psychological well-being of CI recipients with the general population. It has also been used to study general psychological well-being in other patient groups treated in audiology practices [Rembar et al., 2012].

The HUI3 is the latest version of the Canadian multi-attribute utility instrument. It scores in 8 dimensions, including vision, speech, hearing, ambulation, dexterity, emotion, cognition, and pain. Although a generic instrument, it does include an assessment of the impact of sensory impairments [Kuthubutheen et al., 2015]. The EQ5D and associated VAS are described above. The TTO technique requires subjects to hypothetically trade years of life with normal hearing while maintaining the same health status in other aspects. In one example, subjects were instructed to choose between living in a state of hearing loss for 30 years or forfeiting a portion of the next 30 years beyond their current age for normal hearing

Table 3. Savings and cost-utility associated with unilateral cochlear implantation

Study	Sample size *(n)*, gender (*n* female), mean age at CI/time of study	Primary benefit	Cost analysis	Geographic origin
Studies in children				
Cheng et al., 2000	*n* = 78 (48 female) 7.5 years of age at time of study	Statistically significant increases in QoL before and after CI: TTO: 0.75–0.97 VAS: 0.59–0.86 HUI3: 0.25–0.64	Highly favorable cost-utility ratios: TTO: USD 9,029 per QALY VAS: USD 7,500 per QALY HUI3: USD 5,197 per QALY	USA
Francis et al., 1999	*n* =35 prelingually hearing-impaired children (gender not provided) implanted at 5.2 (3.0) years of age	Children with >2 years of CI experience were mainstreamed at greater than twice the rate of age-matched nonimplanted children with profound hearing loss and were placed less frequently in self-contained classrooms, utilizing fewer hours of special education support	Pediatric CI results in educational cost savings ranging from USD 200,000 to 30,000 from kindergarten to 12th grade when compared to school for the deaf and partial mainstream classroom integration, respectively	USA
Koch et al., 1997	*n* = 42 between 4 and 11 years of age at time of study		Net present value of CI at a mean age of 4 years, cost of USD 53,098, and average annual educational cost savings of USD 5,986 was highly favorable at greater than USD 40,000	USA
Semenov et al., 2013	*n* = 175 children implanted under 5 years of age	Statistically significant projected lifetime QoL (HUI3) gains among CI recipients by age at implantation: <18 months: 10.7 QALYs 18–36 months: 9.0 QALYs >36 months: 8.4 QALYs	Highly favorable cost-utility ratios with increasingly favorable ratios at younger ages: <18 months: USD 14,996 per QALY 18–36 months: USD 17,859 per QALY >36 months: USD 19,173 per QALY	USA
Barton et al., 2003	*n* = 199		Per-child average discounted costs of CI were: 1-year horizon: EUR 42,972 15-year horizon: EUR 73,763 73-year horizon: EUR 95,034 Cost of maintaining implanted children was estimated to account for 22% of total CI expenditures in 2000/2001 and was predicated to rise to 63% by 2015/2016	UK
Barton et al., 2006a	*n* = 338		Mean 2000/2001 annual educational costs: CI: EUR 28,058 nonimplanted with moderate HL: EUR 15,745 nonimplanted with severe HL: EUR 28,058	UK
Barton et al., 2006b	*n* = 338		Cumulative economic cost incurred by the families of implanted children between implantation and age 16 years as compared to families of nonimplanted children was EUR 3,533 These costs are 3% of the incremental health sector costs of implantation	UK
Barton et al., 2006c	*n* = 403	QoL (HUI3): a profoundly hearing-impaired child implanted by 6 years of age is expected to gain 2.23 QALYs over 15 years after implantation	Cost of CI: EUR 57,359 Cost-utility ratio: EUR 26,629 per QALY Cost-effectiveness more favorable when estimated over a child's lifetime rather than 15 years, when implanting a child with greater degree of hearing loss, and with implantation at younger ages	UK

Table 3 (continued)

Study	Sample size *(n)*, gender (*n* female), mean age at CI/time of study	Primary benefit	Cost analysis	Geographic origin
O'Neill et al., 2000		CI enables a profoundly hearing-impaired child to function at the level of a severely hearing-impaired child using HA with associated differences and cost-savings in classroom placement Estimated annual QoL gain of 0.23	Cost-utility of CI vs. HA: USD 16,546 per QALY across a child's projected lifetime	UK
O'Neill et al., 2001			Favorable cost-utility ratios: USD 12,000–18,000 per QALY	UK
Sach et al., 2003	*n* = 98 (45 female) implanted at 5.0 (1.8–15.5) years of age		Total cost was negatively related to year of implant and positively related to number of hours of rehabilitation The overall cost-effectiveness improved over time, suggesting a learning curve in CI use	UK
Schulze-Gattermann et al., 2002	Pediatric CI recipients stratified by age at implantation: 0–1.9 (youngest), 2.2–3.9 (middle), and 3.4–6.9 (oldest) years	Educational savings were used to measure benefits of CI up to 16 years of age (completion of compulsory education)	Total costs: CI: USD 113,000–152,000 (youngest to oldest) HA: USD 138,000 Age at implantation has significant implications on the overall favorability of pediatric CI: children under 2 years of age having most favorable cost-benefit ratios as compared to nonimplanted HA peers	Germany
Studies in adults				
Monteiro et al., 2012	*n* = 637		CI was significantly associated with an increase in median yearly income compared to that before implantation (USD 42,672 vs. 30,432), not only improving QoL, but also translating into significant economic benefits for patients and the overall economy, which appear to exceed overall costs of implantation	Canada
Harris and Anderson, 1999	*n* = 9 (3 women), age range 23–59 years	The QWB increased 7.1% in 26 years, therefore a "well-years" benefit of 1.85 The estimated well-years benefit was 0.418 after discounting 5% per year	Cost-benefit/utility ratio can be calculated to be USD 22,380 – 9,125 = USD 13,255/ 0.418 = USD 31,711 per well-year	USA
Kuthubutheen et al., 2015	*n* = 30 (16 female) 56 years of age at time of study. The mean duration of unilateral CI use was 4 years	Statistically significant QoL differences between unilateral CI vs. no intervention HUI3: 0.765 vs. 0.495 EQ5D: 0.890 vs. 0.750 VAS: 0.810 vs. 0.680 TTO: 0.820 vs. 0.650	Highly favorable cost-utility ratios: HUI3: USD 9,425 EQ5D: USD 18,178 VAS: USD 19,576 TTO: USD 14,970	Canada and USA
Palmer et al., 1999	*n* = 46 (25 female) implanted at 54.0 (15.0) years of age	QoL (HUI): baseline QoL was not statistically different between CI recipients and nonimplanted HA controls CI recipients significantly outperformed nonimplanted controls at both 6 months and 12 months afterimplantation attaining health utility scores of 0.76 (0.18) versus 0.57 (0.18) and 0.78 (0.17) versus 0.58 (0.23), respectively	Cost-utility ratio: USD 14,670 per QALY over a 22-year life expectancy Over 90% of the benefit realized in the first 6 months after implantation	USA

Table 3 (continued)

Study	Sample size *(n)*, gender (*n* female), mean age at CI/time of study	Primary benefit	Cost analysis	Geographic origin
Wyatt et al., 1996	*n* = 229 (106 female) implanted at 57.1 years of age with a 4.6-year experience with CI	QoL (HUI3): the health utility of the implanted group was significantly higher than that of their nonimplanted peers awaiting CI by 0.204	Cost-utility ratio: USD 15,928 per QALY with a range from USD 12,000 to 30,000 per QALY	USA
Manrique et al., 2006	*n* = 677		Cost of implantation: postlingual HL (adult): EUR 36,912–37,048, prelingual HL (child): EUR 37,689–44,273	Spain
UK Cochlear Implant Study Group, 2004	*n* = 311 (159 female) implanted at 50.8 (49.1–52.5) years of age	QoL measured using HUI3	Cost-utility ratio: EUR 27,142 (24,532–30323) per QALY Cost-utility varied with age at implantation: <30 years: EUR 19,223 per QALY >70 years: EUR 45,411 per QALY A greater than 40-year duration of deafness prior to implantation yielded unfavorable cost-utility ratios given minimal health utility benefit from implantation	UK
Lee et al., 2006	*n* = 11 (4 female) implanted at 49.6 (10.9) years of age with 5.6 (4.8) years of CI use	QoL pre- and post-CI gain: VAS: 0.3 (0.3–0.6) HUI: 0.4 (0.3–0.7) EQ5D: 0.3 (0.5–0.8) QWB: 0.2 (0.5–0.7)	Cost of CI (discounted): USD 22,320 Cost-utility ratios: VAS: USD 19,223 per QALY HUI: USD 17,387 per QALY EQ5D: USD 26,064 per QALY QWB: USD 40,474 per QALY	South Korea
Studies in older adults				
Francis et al., 2002	*n* = 47 implanted at 63.4 (8.6) years and 66.5 (8.9) years of age at time of study	Significant QoL (HUI3) pre- to post-CI gain: overall population: 0.24 postlingual HL: 0.35 prelingual HL: 0.25 (not significant)	Cost-utility ratio: USD 9,530 per QALY	USA

AQoL, Assessment of Quality of Life; CCIPP, Children with Cochlear Implants: Parental Perspectives; CHIP, Child Health and Illness Profile; CIFI, Cochlear Implant Function Index; CISQ, Cochlear Implant Satisfaction Questionnaire (a combination of Satisfaction in Daily Life Questionnaire and Client Satisfaction Questionnaire-8); EQ5D, EuroQoL-5D; GBI, Glasgow Benefit Inventory; GHSI, Glasgow Health Status Inventory questionnaire; HA, hearing aid; HHIA, Hearing Handicap Inventory – Adults; HL, hearing loss; HPS, Hearing Participation Scale; HSCL-25, Hopkins Symptom Check List – 25 items; HUI, Ontario Health Utilities Index; IRQF, Index Relative Questionnaire Form; KINDL®, questionnaire for measuring health-related quality of life in children and adolescents including Kiddy (4–7 years), Kid (8–11 years), and Kiddo (12–16 years); NCIQ, Nijmegen Cochlear Implant Questionnaire; NH, normal hearing; PGWB, Psychological General Well-Being index; PQLF, Patient Quality of Life Form; QALY, quality-adjusted life years; QoL, quality of life; QWB, Quality of Well-Being Scale; SADL, Satisfaction in Daily Life Questionnaire; SF-36, Short Form-36 questionnaire; SSQ, Speech, Spatial, and Qualities questionnaire; TTO, time trade-off; VAS, visual analog scale; VAS-D, visual analog scale development; YQoL-DHH, Youth Quality of Life – Deaf and Hard of Hearing; SQ, Specific Questionnaire.

[Chung et al., 2012]. The TTO is noted to be a difficult task for subjects to complete. Of the 4 instruments – HUI3, EQ5D, VAS, and TTO – that have been used in many recent studies, HUI3 is the only tool that measures health utility directly and has been shown to be the most conservative measure of health utility [Chung et al., 2012].

The AQoL measure is a multiattribute utility HRQoL instrument that comprises 5 dimensions measuring illness, social independence, social relationships, sensory abilities, and psychological well-being [Hawthorne et al., 2004]. The HSCL-25 is a self-report, psychological inventory with 58 items, scoring on the 5 underlying dimensions of somatization, obsessive-compulsive, interpersonal sensitivity, anxiety and depression. It has been shown to be a good indicator of emotional distress [Mo et al., 2005]. The GBI is a questionnaire developed to as-

sess communication abilities and QoL after otolaryngology procedures [Lassaletta et al., 2006]. It has been validated over a wide range of procedures and found to be sensitive to changes in health after the procedure. QoL is measured in 3 domains, social, general, and physical.

Disease-Specific Instruments in Adults

Nine studies reported on disease-specific QoL instruments in adults, most specific to the benefits of CI. These include the Nijmegen Cochlear Implant Questionnaire (NCIQ) [Cohen et al., 2004; Damen et al., 2007; Fuller et al., 2013; Klop et al., 2007; Looi et al., 2011; Olze et al., 2011], the Cochlear Implant Function Index (CIFI) [Fuller et al., 2013], the Speech, Spatial and Qualities Questionnaire (SSQ) [Fuller et al., 2013], Patient Quality of Life Form (PQLF) [Mo et al., 2005], Index Relative Questionnaire Form (IRQF) [Mo et al., 2005], the Hearing Participation Scale (HPS) [Hawthorne et al., 2004], and the Complete Intelligibility Spatiality Quality (CISQ) [Looi et al., 2011].

The NCIQ is a validated, CI-specific HRQoL questionnaire, composed of 3 categories in 6 domains: physical functioning: sound perception – basic, sound perception – advanced, speech production; social functioning: activity, social functioning; and psychological functioning: self-esteem. The CIFI is a tool developed to assess the auditory related function of CI users, in 6 fields of auditory functioning: reliance on visual assistance, telephone use, communication at work, "hearing" in noise, hearing in groups, and hearing in large room settings. The SSQ is a validated environmental and spatial hearing questionnaire that was developed to quantify the abilities of hearing-impaired people and CI users, particularly for speech perception and spatial hearing [Fuller et al., 2013].

The PQLF and IRQF are related instruments, both developed at the House Ear Institute as disease-specific instruments to assess patients' ability to cope with their hearing loss, adaptation to the CI, and emotional alterations since implantation. The PQLF is completed by the hearing-impaired individual, while the IRQF reflects a relative's experience with that individual, including the effect of the hearing handicap on their daily activities, and the hearing-impaired individual's adaptation to the implant [Mo et al., 2005].

The HPS is an 11-item instrument that measures self-esteem, social handicap, and hearing handicap [Hawthorne et al., 2004]. The CISQ was created to enable a rapid and simple instrument to evaluate the benefits of a hearing aid. This 36-item questionnaire assesses an individual's hearing abilities relative to spatiality and quality of signal, intelligibility in silence, background noise intelligibility, averseness, and reverberation [Looi et al., 2011].

Ad hoc Instruments in Adults

Four reports included ad hoc QoL instruments [Fuller et al., 2013; Lassaletta et al., 2006; Rembar et al., 2009; Zhao et al., 2008]. Of these, 2 studies used quantitative QoL questionnaires [Fuller et al., 2013; Zhao et al., 2008], 1 used a qualitative instrument [Rembar et al., 2009], and 1 used a combination qualitative and quantitative instrument [Lassaletta et al., 2006].

Summary of Findings for Adults

Overall, implanted adults showed significant improvement in QoL after implantation [Chung et al., 2012; Hawthorne et al., 2004; Klop et al., 2007, 2008; Mo et al., 2005; Olze et al., 2011; Zhao et al., 2008] and attained significantly higher QoL than their nonimplanted (hearing-aided) peers [Cohen et al., 2004; Looi et al., 2011]. There were no differences in QoL between implanted adults and normal-hearing peers [Rembar et al., 2012]. Most of the QoL benefit of CI was observed within the first 4 months following surgery [Klop et al., 2007, 2008], with the gain sustained for at least 6 years after CI [Damen et al., 2007]. Younger age at implantation was associated with improved QoL outcomes [Chung et al., 2012].

QoL Outcomes in Older Adults

Six of the 57 studies focused on presenting metrics for assessing the QoL attainment of older adults with cochlear implants. The studies were comprised of participants from 6 countries with a sample size ranging from 20 to 283 study participants. The mean age at implantation ranged from 52.9 to 74.4 years (Table 1).

Generic Instruments in Older Adults

All 6 studies reported on generic health utility metrics, which included the SF-36, GBI, and the Glasgow Health Status Inventory (GHSI) questionnaire. The SF-36 and GBI are described above. The GHSI questionnaire assesses the effect of a hearing problem on overall QoL, including physical health and social support [Orabi et al., 2006].

Disease-Specific Instruments in Older Adults

Two studies reported on disease-specific QoL instruments in adults, including the Specific Questionnaire (SQ) [Ramos et al., 2013], the NCIQ [Olze et al., 2012], and the Hearing Handicap Inventory – Adults (HHIA) [Vermeire et al., 2005]. The SQ evaluates 6 different aspects related

Table 4. Savings and cost-effectiveness associated with bilateral cochlear implantation

Study	Sample size *(n)*, age, and gender of subjects	Primary benefit	Cost analysis	Geographic origin
Studies in children				
Foteff et al., 2016	Meta-analysis of Australian children with bilateral severe-to-profound SNHL	QoL measure: HUI3 (literature derived) Gains of 0.145 and 0.208 from bilateral HAs to unilateral CI and from bilateral HAs to bilateral CI, respectively Gain of 0.063 from unilateral to bilateral CI	Cost-utility ratios‡: unilateral CI vs. bilateral HAs: USD 15,335 bilateral CI vs. bilateral HAs: USD 27,948 overall CI cohort vs. bilateral HAs: USD 22,317	Australia
Summerfield et al., 2010	Opportunity sample of 180 informants composed of clinicians, researchers, students, and parents valued the QoL of a hypothetical child born profoundly deaf	QoL gains using TTO and VAS: unilateral CI vs. no intervention: TTO: 0.11 VAS: 0.18 unilateral CI with HA vs. no intervention: TTO: 0.16 VAS: 0.25 bilateral vs. unilateral CI: TTO: 0.11 VAS: 0.13 bilateral CI vs. unilateral CI with HA: TTO: 0.05 VAS: 0.06	Cost-utility ratios‡: unilateral CI vs. no intervention: USD 30,785 bilateral vs. unilateral CI: USD 33,740	UK
Bond et al., 2009	Meta-analysis of UK children with prelingual severe-to-profound SNHL	Lifetime QALYs gained (literature-derived): gain of 4.48 QALYs for unilateral CI vs. bilateral HAs gain of 0.67 QALYs for simultaneous bilateral vs. unilateral CI gain of 0.60 QALYs for sequential bilateral vs. unilateral CI	Cost-utility ratios‡: unilateral CI vs. bilateral HAs: USD 20,924 simultaneous bilateral vs. unilateral CI: USD 63,040 sequential bilateral vs. unilateral CI: USD 84,393	UK
Studies in adults				
Foteff et al., 2016	Meta-analysis of Australian adults with postlingual severe-to-profound SNHL, average age 55–59 years	QoL measure: HUI3 (literature derived) Gains of 0.145 and 0.305 from bilateral HAs to unilateral CI and from bilateral HAs to bilateral CI, respectively Gain of 0.035 from unilateral to bilateral CI	Cost-utility ratios‡: unilateral CI vs. bilateral HAs: USD 6,877 bilateral CI vs. bilateral HAs: USD 18,785	Australia
Chen et al., 2014	$n = 90$	QoL measure: HUI3 Gains of 0.270 and 0.305 from no intervention to unilateral CI and from no intervention to bilateral CI, respectively	Bilateral implantation vs. no intervention cost of USD 111,764 yielding an ICUR of USD14,658/QALY When compared to unilateral CI, bilateral implantation yielded an ICUR of USD 55,020/QALY	Canada and USA
Kuthubutheen et al., 2015	Bilateral CI: $n = 30$ (19 female) 53 years of age at time of study The mean duration of bilateral CI use was 25 years	QoL measure HUI3, EQ5D, VAS, TTO: bilateral vs. no intervention HUI3: 0.800 vs. 0.495 EQ5D: 0.930 vs. 0.750 VAS: 0.880 vs. 0.680 TTO: 0.940 vs. 0.650 bilateral vs. unilateral HUI3: 0.800 vs. 0.795† EQ5D: 0.930 vs. 0.890† VAS: 0.880 vs. 0.810 TTO: 0.940 vs. 0.820	Bilateral vs. no intervention ICUR (per QALY): HUI3: USD 14,658 EQ5D: USD 24,837 VAS: USD 22,353 TTO: USD 15,416 bilateral vs. unilateral ICUR HUI3: USD 55,020 EQ5D: USD 48,142 VAS: USD 27,510 TTO: USD 16,047	Canada and USA

Table 4 (continued)

Study	Sample size *(n)*, age, and gender of subjects	Primary benefit	Cost analysis	Geographic origin
Bichey and Miyamoto, 2008	*n* = 23 participants (22 female) 6–79 years of age at implantation with 6.3 (0.9–13.6) and 1.2 (0.5–3.2) years of unilateral and bilateral CI use, respectively	QoL measured using HUI3 showed statistically significant differences: before CI: 0.33 after unilateral CI: 0.69 after bilateral CI: 0.81	Bilateral vs. no intervention: USD 23,345/QALY Incremental change in cost-utility from unilateral to bilateral: USD 2,187/QALY	USA
Summerfield et al., 2002	*n* = 202	QoL measure: HUI2 More QoL is likely to be gained per unit of expenditure on unilateral implantation than on bilateral implantation	Cost-utility ratios based on volunteers' estimates: unilateral implantation versus no intervention: GBP 16,774 per QALY unilateral implantation versus management with HA: GBP 27,401 per QALY simultaneous bilateral implantation versus unilateral implantation: GBP 61,734 per QALY provision of an additional implant versus no additional intervention: GBP 68,916 per QALY	UK
Summerfield and Barton, 2003	*n* = 202	QoL measure: HUI2	Cost-utility ratio: unilateral CI: GBP 15 542 per QALY This ratio would be approximately matched if a bilateral processor cost 10% more than a unilateral processor and if 2 electrode arrays cost 10% more than 1	UK
Summerfield et al., 2006	*n* = 24	QoL measure: SSQ, GHSI, HUI3, VAS, EQ5D A significant negative change in EQ5D and a positive change in GHSI were observed before and after the second implantation Spatial hearing (SSQ) also showed a significant positive change None of the other changes were significant Overall, there was a minimal impact of health utility of 0.03	Cost-utility ratio of second CI: EUR 102,500 per QALY Increasing CI effectiveness through enhanced signal processing in binaural processors and reducing the cost of implant hardware may make the cost-utility of a second implant more favorable	UK

AQoL, Assessment of Quality of Life; CCIPP, Children with Cochlear Implants: Parental Perspectives; CHIP, Child Health and Illness Profile; CIFI, Cochlear Implant Function Index; CISQ, Cochlear Implant Satisfaction Questionnaire (a combination of Satisfaction in Daily Life Questionnaire and Client Satisfaction Questionnaire-8); EQ5D, EuroQoL-5D; GBI, Glasgow Benefit Inventory; GHSI, Glasgow Health Status Inventory questionnaire; HA, hearing aid; HHIA, Hearing Handicap Inventory – Adults; HL, hearing loss; HPS, Hearing Participation Scale; HSCL-25, Hopkins Symptom Check List, 25 items; HUI, Ontario Health Utilities Index; ICUR, incremental cost-utility ratio; IRQF, Index Relative Questionnaire Form; KINDL®, questionnaire for measuring health-related quality of life in children and adolescents including Kiddy (4–7 years), Kid (8–11 years), and Kiddo (12–16 years); NCIQ, Nijmegen Cochlear Implant Questionnaire; NH, normal hearing; PGWB, Psychological General Well-Being index; PQLF, Patient Quality of Life Form; QALY, quality-adjusted life years; QoL, quality of life; QWB, Quality of Well-Being Scale; SADL, Satisfaction in Daily Life Questionnaire; SF-36, Short Form-36 questionnaire; SNHL, sensorineural hearing loss; SSQ, Speech, Spatial, and Qualities questionnaire; TTO, time trade-off; VAS, visual analog scale; VAS-D, visual analog scale development; YQoL-DHH, Youth Quality of Life – Deaf and Hard of Hearing; SQ, Specific Questionnaire. † Not statistically significant. ‡ Converted to USD using yearly average exchange rates provided from http://www.usforex.com/forex-tools/historical-rate-tools/yearly-average-rates based on year study was published.

to CI: speech recognition, social interaction, telephone use, confidence, family life, and satisfaction [Faber and Grøntved, 2000; Ramos et al., 2013]. The NCIQ is described above. The HHIA is a well-validated questionnaire that assesses hearing handicap in adult populations. The HHIA is a 25-item, hearing-specific, QoL scale with 2 components that measure the emotional and situational impact of hearing loss [Vermeire et al., 2005].

Ad hoc Instruments in Older Adults

Four reports included ad hoc QoL instruments [Di Nardo et al., 2014; Olze et al., 2012; Orabi et al., 2006; Vermeire et al., 2005]. Of these, 3 studies used quantitative QoL questionnaires, and 1 [Olze et al., 2012] used a qualitative instrument.

Summary of Results for Older Adults

Overall, implanted older adults showed significant improvement in QoL after implantation [Chung et al., 2012; Di Nardo et al., 2014; Olze et al., 2012; Orabi et al., 2006; Ramos et al., 2013; Vermeire et al., 2005]. Improvements in QoL were independent of audiological performance. While all studies demonstrated significant auditory benefit of CI in all age groups, some studies [Di Nardo et al., 2014; Ramos et al., 2013; Vermeire et al., 2005] showed poorer auditory performance in the elderly group, and others showed a similar benefit of CI regardless of age [Olze et al., 2012; Orabi et al., 2006]. Regardless, nearly all studies showed significant improvement in QoL in this age group, most often reported in multiple dimensions.

Discussion

The objective of this narrative review was to comprehensively assess the literature on the QoL determination and attainment among CI recipients of all ages, and to examine cost-effectiveness measures. We were able to access a large compendium of research conducted over the past 2 decades and around the world to assess the impact of CI in deafness from a perspective that combines economic considerations with measured outcome of implantation offered by recipients or proxies. We observed a consistently high impact of implantation on perceived QoL, and a general trend towards favorable economic impact of CI that is subject to single- versus bilateral-implantation considerations.

Studies that combine clinical and economic evaluation are increasingly important in guiding comprehensive assessments and payer decision-making. Such decision-making is critical to the acceptance of new technologies; however, there is often limited, timely evidence available to payers on the effectiveness and cost-effectiveness of emerging and evolving technologies [Steiner et al., 1996]. As a result, health care stakeholders are at a disadvantage when attempting to justify interventions based not only on safety and efficacy, but also on cost-effectiveness. Economic analyses, when based on a broad range of HRQoL domains that permit comparison of outcomes across interventions, diseases, and populations, can provide guidance in programmatic initiatives designed to enhance performance and contain costs while prioritizing expenditures.

The results of this review suggest that an approach to the question "Are cochlear implants cost-effective?" is nuanced and that a meaningful answer must incorporate multiple factors that offer objective measures and reflect priorities of hearing-impaired individuals, professionals with relevant expertise, and society in general. We observed that even when priorities are recognized, the answer varies, depending on the economic measures used in the analysis.

Economic Evaluation Measures

Changes in quality-adjusted life years that are based on HRQoL measures can vary based on several factors, including the measurement tool used (whether direct or indirect measure, disease-specific or generic measurement), and the population studied (e.g., whether those surveyed are hearing-impaired CI candidates, individuals with unilateral or bilateral implants, hearing health care professionals, parents serving as proxy respondents for their children, or members of society). We noted consistency across studies using the various instruments; however, the HUI3 is generally regarded as the most conservative and consistent measure of HRQoL for studies of CI cost utility [Kuthubutheen et al., 2015]. In most studies, the HUI3 does not show significant gain in utility from the unilateral to bilateral CI condition though it was reported clearly sensitive to unilateral CI. For this reason, the cost-utility analysis of bilateral intervention is highly driven by the increase in cost (up to double the cost of unilateral CI) with modest commensurate, enhanced utility produced by bilateral implantation.

CI analysts have struggled to understand why the impact of a technology that patients consistently value as a life-changing intervention, such as bilateral CI, is not reflected in significant increments in utility scores. It is possible that the communicative advantages produced are simply not captured in the instruments used, and even disease-specific metrics often fail to capture the nuanced ways in which access to binaural auditory inputs may confer enhanced connectivity. We noted that part of the challenge is adequately probing the benefit of a second implant when a first implant has been placed. Weighted-average QoL gains, reflected in generic QoL instruments, were observed in our literature survey to be 0.04 in children and 0.08 in adults (Ta-

Table 5. Weighted average of observed quality of life changes after cochlear implantation comparing unilateral and bilateral implantation with no intervention and bilateral implantation with unilateral implantation

QoL instrument type	Weighted-average QoL gain†					
	unilateral vs. no intervention		bilateral vs. no intervention		bilateral vs. unilateral	
	absolute gain (range)	percent gain (range)	absolute gain (range)	percent gain (range)	absolute gain (range)	percent gain (range)
Children						
Generic	0.27 (0.03–0.45)	93.4 (3.2–156)	0.26 (0.21–0.31)	90.4 (na)	0.04 (0.03–0.06)	16.9 (13–29.2)
Disease specific	0.09 (na)	18.8 (na)	na	na	0.1 (0.05–0.14)	na
Adults						
Generic	0.21 (0.05–0.4)	44.3 (0–133.3)	0.24 (0.07–0.48)	40.2 (7.5–145.5)	0.08 (0.01–0.14)	11.4 (0.6–21.2)
Disease specific	0.28 (0.2–0.5)	89.1 (41.7–312.5)	0.62 (na)	476.9 (na)	0.1 (0.06–0.15)	15.9 (9–23.1)

This represents the weighted-average QoL gain by intervention based on the data shown in the previous tables. Gains are significant regardless of the instrument used (generic vs. disease specific) whether comparing unilateral or bilateral CI with no intervention. However, gains achieved by bilateral versus unilateral CI are modest. There is a modest tendency toward disease-specific instruments being more sensitive to the benefits of bilateral implantation. † Using the quality of life gain and sample size of the studies reported in Tables 1–4.

ble 5). These same weighted-average QoL gains are, when considered in the context of bilateral implantation as compared with the baseline state, 0.26 for children and 0.24 for adults – gains in utility that are substantial in comparison to many established medical and surgical interventions.

The neurobiology of the central auditory system informs us that the system relies on bilateral stimulation to develop normal optimal function. While significant gains in localization and the ability to understand speech in the presence of background noise are realized in bilateral CI recipients [Kral and O'Donoghue, 2010], such benefits are not robust by current QoL measures. It must be appraised whether this discrepancy is a failure of CI to adequately restore an important sensory deficit, or whether current measurement tools are simply inadequate to demonstrate a meaningful gain in function. When QoL gains are modest, costs of the intervention become more important. For this reason, efforts to control incremental costs of bilateral CI, which are limited primarily to the cost of the second implant and the episode of care, are of great importance.

Sensitivity analyses are particularly useful to describe the variety of costs and other assumptions that drive the determination of cost-utility analysis. Costs can vary widely both within and between countries, and aggressive cost-cutting measures can improve financial sustainability [McKinnon, 2014]. Factors that can be examined in sensitivity analyses may include: numbers of pre- and post-CI assessments, failure rates of external devices out of warranty, discount rates (for costs as well as benefits over time), time horizon, utility gains, timing of surgeries such as bilateral sequential and simultaneous implants, and time horizon of the intervention. Cost-utility of bilateral versus unilateral CI is highly dependent upon cost of the second implant. In some health care settings, the cost of the second implant, particularly if placed simultaneously, has been discounted 50% from the first device.

While HRQoL is traditionally used in economic analyses, there are other measures of outcome that can be considered and have not been typically measured in cost-utility analyses of CI. These measures reflect financial well-being and contributions to society by the hearing-impaired individual. Work by Emmett and Francis [2014] using the US National Health and Nutrition Examination Survey information from 1999 to 2002 demonstrated that individuals with hearing loss (World Health Organization definition of bilateral pure-tone average of 0.5, 1, 2, and 4 kHz of greater than 25 dB) were 3.21 times more likely to have low educational attainment (not complete high school), 1.58 times more likely to be of low income (family income less than USD 20,000 per annum), and 1.98 times more likely to be unemployed or underemployed (work less than 35 h per week). Other societal benefits may include lower rates of depression and dementia in elderly CI recipients. It is not yet clear if individuals with hearing loss and CI perform as well as normal-hearing people with respect to these important measures of health, well-being, and contributions to society, but several studies do suggest that income levels for adults are improved following CI and that eventual educational and employ-

Table 6. Summary of cost-utility ratios (USD) by age groupings across studies

Study	Year	TTO	VAS	HUI	EQ5D	QWB
Unilateral CI versus no intervention						
Studies in children						
Cheng et al., 2000	2000	9,029	7,500	5,197		
O'Neill et al., 2000	2000			16,546		
O'Neill et al., 2001	2001			15,000		
Barton et al., 2003	2006			34,617		
Semenov et al., 2013‡	2013			17,085		
Mean		9,029	7,500	17,689		
Studies in adults						
Wyatt et al., 1996	1996			15,928		
Palmer et al., 1999	1999			14,670		
UK Cochlear Implant Study Group, 2004	2004			35,284		
Lee et al., 2006	2006		19,223	17,387	26,064	40,474
Kuthubutheen et al., 2015	2015	14,970	19,756	9,425	18,178	
Summerfield et al., 2002†	2002			43,842		
Summerfield and Barton, 2003†	2003			24,867		
Harris and Anderson	1999					31,711
Mean		14,970	19,490	23,058	22,121	40,474
Studies in older adults						
Francis et al., 2002	2002			9,530		
Mean				9,530		
Bilateral CI versus no intervention						
Studies in children						
Bond et al., 2009	2009			29,519		
Summerfield et al., 2010	2010	34,824	23,026			
Mean		34,824	23,026	29,519		
Studies in adults						
Bichey and Miyamoto, 2008	2008			23,345		
Bond et al., 2009	2009			33,132		
Chen et al., 2014	2014			14,658		
Kuthubutheen et al., 2015	2015	22,353	15,416	14,658	24,837	
Mean		22,353	15,416	21,448	24,837	
Bilateral CI versus unilateral CI						
Studies in children						
Bond et al., 2009	2009			70,470		
Summerfield et al., 2010	2010	37,100	30,973			
Mean		37,100	30,973	70,470		
Studies in adults						
Bichey and Miyamoto, 2008‡	2008			38,652		
Bond et al., 2009	2009			86,425		
Kuthubutheen et al., 2015	2015	27,510	16,047	55,020	48,142	
Mean		27,510	16,047	60,032	48,142	

This table demonstrates that, for studies performed in most developed countries, cost-utility ratios are well below the threshold of USD 50,000. The exception to this standard is in the UK studies, which demonstrate costs substantially higher than in other parts of the developed world (e.g., Summerfield and Bond studies and NICE report). The reason for this discrepancy in cost-utility is not apparent from a reading of the literature and should be examined more deliberately in the future. TTO, time trade-off; VAS, visual analog scale; HUI, Health Utilities Index; EQ5D, EuroQoL-5D; QWB, Quality of Well-Being Scale.

†Using USD exchange rates for the year the study was published. ‡Median value calculated by authors.

ment attainment for children with CI may be on par with those with normal hearing [McKinnon, 2014]. Hence, as difficult as it is to define all possible costs of an intervention, it is equally challenging to fully reflect all possible associated benefits to society. These benefits can be, but have not yet been, measured in financial terms.

The final step in the economic analysis requires that incremental cost-utility be determined to reflect the incremental cost per HRQoL benefit from CI. Thresholds are set based on the incremental cost-utility ratio: if it falls below this set level, an intervention is determined to be "cost-effective." This varies by country in the developed world. This threshold is approximately USD 50,000 in the USA, and an equivalent-currency cost in the UK. Here, we observed that despite variance in utility associated with different methodologies, the calculated cost-utility ratios for the USA and Canada consistently met this threshold, indicating high value for expenditures on CI (Table 6). Some of the studies, particularly those performed in the UK, revealed higher cost-utility ratios; the reason for this is not known, but should be explored further. Such data can also be assessed in developing countries, wherein the threshold for cost-effectiveness suggested by the World Health Organization as a factor of gross domestic product can be developed using a methodology that drives CI feasibility in lower income countries [Emmett et al., 2015].

Range of Outcomes Observed (Unilateral)

Results observed in this survey demonstrated mean health-utility gains in children receiving unilateral implants ranging on average from 0.27 with generic measures to 0.09 with disease-specific measures (single study). In adults, utility gains in generic QoL measures ranged on average from 0.21 with generic measures to 0.28 with disease-specific measures. While there is variability in these pooled results, mean gains in utility that were observed to consistently exceed 0.20 are impressive and rarely observed in health utility studies of interventions commonly covered, for example, within the US health care system.

Range of Outcomes (Bilateral)

Health utility gains following bilateral CI may be compared either to no intervention, in which case gains are robust (average 0.26 and 0.24 using generic measures for children and adults, respectively, and 0.62 using disease-specific measures for adults), or to unilateral implantation, in which case benefits, as mentioned previously, are more nuanced. Absolute gains in health utility for 2 versus 1 CI average from 0.04 to 0.1 in children for generic versus disease-specific measures, and from 0.08 to 0.1 in adults for the same. In each case, there is a small tendency for the disease-specific measures to be more reflective of the benefits of bilateral CI. This makes sense, as bilateral CI is less likely to show incremental benefits in overall QoL than to benefit the individual in listening in complex auditory environments, particularly with adequate time for follow-up and refinement of auditory skills.

Overall Valuation and Related Ethical Considerations

The overall economic value of CI is generally regarded as greater in children who are implanted early in life than in adults, both because of the greater benefit over longer mean time horizons, and because of the fact that young children with significant hearing loss do not develop normal speech and language skills. This raises an ethical consideration: What if lack of spoken language is considered acceptable, "normal" and even desirable for a segment of the population? For example, this is the often discussed case with a deaf community stance. How should their valuation of utility of intervention for deafness, which they do not regard as a disability, be considered? We note that this situation is quite different from the stroke victim for whom health care providers are considering a costly intervention, or for a potential transplant recipient. One argument is that, while a dialogue around issues related to health valuation can include a choice of perspective (patients or able-bodied individuals) as well as the perimeter of assessment (medical or nonmedical care), the community finance of treatment such as bilateral CI for children is justified once it is shown that the disability has a significant impact on the fundamental social achievements of the individual and that such treatment can reduce inequalities effectively [Thebaut, 2013]. Evidence of the first point has been advanced [Emmett and Francis, 2014], and evidence of the second point continues to accumulate.

Conclusions

Considerable work has been done on the QoL attainment and health economic implications of CI. Unilateral CI across all age groups leads to reported sustained benefits in the recipients' overall and disease-specific QoL. With increased cost associated with bilateral CI, further study is needed to characterize its costs and benefits with respect to the recipients' health, well-being, and contributions to society.

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