The effectiveness of early cochlear implantation for infants and young children with hearing loss

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This Technical Brief was authored by Dr Wasan Ali (Assistant Research Fellow), who conducted the critical appraisals, prepared the report and coordinated the project, and Dr Rebecca O’Connell (Research Fellow) who selected papers for review and contributed to the preparation of the report.

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LEVEL OF EVIDENCE CONSIDERED IN TECHNICAL BRIEFS

Technical Briefs are rapidly produced assessments of the best available evidence for a topic of highly limited scope. They are less rigorous than systematic reviews. Best evidence is indicated by research designs which are least susceptible to bias according to the National Health and Medical Research Council’s (NHMRC) criteria (see Appendix 1). Where methodologically acceptable and applicable,
appraised evidence is limited to systematic reviews, meta-analyses, evidence based clinical practice guidelines, health technology assessments and randomised controlled trials (RCTs). Where not available, poorer quality evidence may be considered.

CONFLICT OF INTEREST

None.
EXECUTIVE SUMMARY

Aim and scope

The aim of this technical brief was to assess the effectiveness of cochlear implantation at an early age when compared to implantation at a later age. Eligible studies were those that included some children less than 2 years of age at implantation, and studies where the mean or median age of the study population at cochlear implantation was less than 36 months. Studies all had sample sizes of at least 20 participants. They compared the effectiveness of cochlear implantation at a young age with implantation at an older age. The following outcomes were considered as indicators of effectiveness: audiological performance, communication outcomes, educational achievement and quality of life.

High quality secondary research (systematic reviews and meta-analyses) are considered best evidence on the topic. As there were no eligible systematic reviews, studies were included if they were cross-sectional, case control or cohort studies fitting the above selection criteria.

Methods

The search strategy considered original articles published between January 1996 and December 28, 2006 inclusive, in the English language. The search included major bibliographic and review databases and secondary sources, and mostly published and indexed literature. Databases included: Medline, Embase, Cinahl, Psychinfo, ERIC, Index New Zealand, New Zealand Bibliographic Database, Science and Social Science Citation Index, Current Contents, Cochrane Library Controlled Trials Register, Cochrane Database of Systematic Reviews, Database of Abstracts of Reviews of Effectiveness (DARE), TRIP database, NHS Economic Evaluation Database, and Health Technology Assessment Database.

Search terms and keywords included:
cochlear implants, cochlear implantation, treatment outcome, child language, speech perception, quality of life, language development, vocabulary, verbal behavior, auditory perception, voice quality, auditory threshold, postoperative complications, time factors, age factors, child-preschool

Summaries of appraisal results were presented in Evidence Tables, which detailed study design, study setting, sample, methods, results, and reported conclusions and comments based on the limitations and validity of the review. Review conclusions, implications for practice, gaps in the literature and directions for research were synthesized and overall conclusions made.

Key results and conclusions

The following conclusions are based on the current evidence available from this report’s critical appraisal of literature published on the effectiveness of cochlear implants at a young age when compared to implantation at an older age for infants and young children.

- In general, implantation at a younger age improves the effectiveness of cochlear implantation in terms of audiological performance and communication outcomes.
- This is particularly evident when cochlear implantation occurs before the age of 24 months, which is more effective than implantation after 24 months
- It is not clear whether implantation prior to the age of 12 months improves effectiveness when compared to implantation after 12 months of age.
- Because of the short length of time that implantation has been used in large numbers of infants and young children less than 2 years of age, evidence of an increase in effectiveness is only available for immediate outcomes such as communication skills, and has only been observed up to about 5-8 years after implantation.
- It is not clear what effect cochlear implantation at a younger age has on long-term outcomes such as educational achievement, and quality of life.
- It is possible that those implanted at an older age (above 24 months) develop at a slower rate but eventually reach equivalent developmental milestones.
**Implications for practice**

Whilst the current evidence base is limited, several implications for practice have been suggested by the appraised systematic studies. Implantation prior to the age of 24 months may increase the effectiveness of cochlear implantation in terms of immediate outcomes such as communication skills. This should be one of the considerations when weighing up the harms and benefits associated with cochlear implantation at this age. Another essential consideration is that a cochlear implant should not be assumed to be an appropriate intervention for each profoundly deaf child under this age. It is essential to perform prior assessment of the child and family so as to explore factors that may affect the outcomes and explain the implications to the parents.

When considering cochlear implantation before the age of 12 months, the harms and benefits of cochlear implantation at this age, other than effectiveness compared to an older age, will need to be weighed up.

**Further research/reviews required**

All of the studies included in this review were cross sectional surveys or cohort studies with relatively small sample sizes. Often the study populations were heterogeneous in terms of the degree of hearing loss and congenital versus acquired loss. Mode of communication and details on socio-economic status or educational status of parents were lacking. This could increase the possibility that confounding or bias has influenced some of the study outcomes.

While the best possible design to determine the effectiveness of cochlear implantation at a young age compared to an older age would be a randomised controlled trial this design is likely to be unacceptable to parents and clinicians as there is already evidence that younger age at implantation is more effective than at an older age, just not what the age limit of this effect is. Also, theoretically younger age is likely to be more effective as it reduces the period of sensory deprivation.

Further well-designed observational studies with large sample sizes and long-term follow-up are required, especially in the younger age groups.

Note: Readers are reminded of the limitations of technical briefs compared to more rigorous and comprehensive full systematic reviews.
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<thead>
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<tr>
<td>+ve</td>
<td>Positive</td>
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<tr>
<td>-ve</td>
<td>Negative</td>
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<tr>
<td>95% CI</td>
<td>95 percent confidence interval</td>
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<tr>
<td>95th centile.</td>
<td>95th percentile</td>
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<tr>
<td>ABR</td>
<td>auditory brainstem response</td>
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<td>CAP</td>
<td>Categories of auditory performance</td>
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<tr>
<td>CELF-P or CELF-III</td>
<td>Clinical Evaluation of Language Fundamentals P or III</td>
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<tr>
<td>CI</td>
<td>Cochlear implant</td>
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<tr>
<td>CID</td>
<td>Central Institute for the Deaf</td>
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<tr>
<td>Cinahl</td>
<td>Cumulative Index to Nursing and Allied Health Literature</td>
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<tr>
<td>DB</td>
<td>Decibels</td>
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<td>DTA</td>
<td>developmental trajectory analysis</td>
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<tr>
<td>EARS test battery</td>
<td>evaluation of auditory responses to speech test battery</td>
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<td>ELQs</td>
<td>expressive language quotients</td>
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<tr>
<td>FmHx</td>
<td>Family History</td>
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<tr>
<td>GASP</td>
<td>Glendonald Auditory Screening Procedure</td>
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<td>GP</td>
<td>General practitioner</td>
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<td>HA</td>
<td>Hearing aid</td>
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<tr>
<td>HL</td>
<td>Hearing loss</td>
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<td>HLM</td>
<td>Hierarchical linear modeling</td>
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<td>HTA</td>
<td>Health technology assessment</td>
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<tr>
<td>Hx</td>
<td>History</td>
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<tr>
<td>INAHTA</td>
<td>International Network of Agencies for Health Technology Assessment</td>
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<tr>
<td>IT-MAIS</td>
<td>Infant-Toddler Meaningful Auditory Integration Scale</td>
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<tr>
<td>LiP</td>
<td>Listening Progress Profile</td>
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<tr>
<td>MAIS</td>
<td>meaningful auditory integration</td>
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<td>MCDI</td>
<td>MacArthur Communicative Development Inventories</td>
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<tr>
<td>Abbreviation</td>
<td>Description</td>
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<tr>
<td>MeSH</td>
<td>Medical Subject Headings</td>
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<tr>
<td>MLU</td>
<td>Mean length of utterance</td>
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<tr>
<td>MoH</td>
<td>Ministry of Health (NZ)</td>
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<tr>
<td>MTP</td>
<td>Monosyllable Trochee Polysyllable Test</td>
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<tr>
<td>MUSS</td>
<td>Meaningful use of speech scale</td>
</tr>
<tr>
<td>NH</td>
<td>normal hearing</td>
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<tr>
<td>NHMRC</td>
<td>National Health and Medical Research Council</td>
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<tr>
<td>NHS</td>
<td>National Health Service (UK)</td>
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<tr>
<td>NS</td>
<td>Not stated</td>
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<tr>
<td>NZ</td>
<td>New Zealand</td>
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<tr>
<td>NZHTA</td>
<td>New Zealand Health Technology Assessment</td>
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<tr>
<td>OR</td>
<td>odds ratio</td>
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<tr>
<td>OC</td>
<td>oral communication</td>
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<td>OWLS</td>
<td>Oral and Written Language Scales</td>
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<td>Paeds</td>
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<tr>
<td>PTA</td>
<td>pure tone average/or audiometry</td>
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<td>PPV</td>
<td>positive predictive value</td>
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<tr>
<td>PPVT 3</td>
<td>Peabody Picture Vocabulary Test 3</td>
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<tr>
<td>QA</td>
<td>quality assurance</td>
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<tr>
<td>QC</td>
<td>quality control</td>
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<tr>
<td>RDLS-III</td>
<td>Renyll Developmental Language Scales</td>
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<tr>
<td>RCT</td>
<td>randomised controlled trial</td>
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<tr>
<td>RNZCGP</td>
<td>Royal New Zealand College of General Practitioners</td>
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<tr>
<td>RR</td>
<td>Relative risk</td>
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<tr>
<td>SD</td>
<td>standard deviation</td>
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<tr>
<td>SES</td>
<td>Socio-economic status</td>
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<tr>
<td>Sig diff</td>
<td>Significantly different</td>
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<tr>
<td>Acronym</td>
<td>Definition</td>
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<tr>
<td>SKI*HI</td>
<td>Skills for hearing impaired children</td>
</tr>
<tr>
<td>SNHL</td>
<td>sensorineural hearing loss</td>
</tr>
<tr>
<td>TAPS</td>
<td>Test of Auditory Perception of Speech</td>
</tr>
<tr>
<td>TC</td>
<td>total communication (simultaneous use of oral English and signs)</td>
</tr>
<tr>
<td>T-test</td>
<td>student test</td>
</tr>
<tr>
<td>UK</td>
<td>United Kingdom</td>
</tr>
<tr>
<td>UNHS</td>
<td>Universal Newborn Hearing Screening</td>
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<tr>
<td>USA</td>
<td>United States of America</td>
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<tr>
<td>Vs</td>
<td>versus</td>
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</table>
GLOSSARY

**Acquired hearing loss**  
Hearing loss which is not congenital. In practice this refers to post-lingual hearing loss.

**Age standardisation**  
A procedure for adjusting rates designed to minimise the effects of differences in age composition when comparing rates for different populations.

**Bias**  
Deviation of results or inferences from the truth, or processes leading to such deviation. Any trend in the collection, analysis, interpretation, publication, or review of data that can lead to conclusions that are systematically different from the truth.

**Blinded study**  
A study in which observers and/or subjects are kept ignorant of the group to which they are assigned. When both observers and subjects are kept ignorant, the study is referred to as double blind.

**Case-control study**  
An epidemiological study involving the observation of cases (persons with the disease, such as cervical cancer) and a suitable control (comparison, reference) group of persons without the disease. The relationship of an attribute to the disease is examined by comparing retrospectively the past history of the people in the two groups with regard to how frequently the attribute is present. See also nested case control.

**Closed-set speech recognition test**  
A test which measures the ability to recognise speech. Answers can be chosen from a limited number of choices and individuals are allowed to lip-read.

**Cochlear implant**  
An electronic device surgically implanted to stimulate the auditory nerve in the cochlea (inner ear) in order for the individual to receive and process sound and speech.

**Cohort study**  
The analytic method of epidemiologic study in which subsets of a defined population can be identified who are, have been, or in the future may be exposed or not exposed in different degrees, to a factor or factors hypothesised to influence the probability of occurrence of a given disease or other outcome. Studies usually involve the observation of a large population, for a prolonged period (years), or both.

**Confidence interval**  
The computed interval with a given probability, e.g. 95%, that the true value of a variable such as a mean, proportion, or rate is contained within the interval. The 95% CI is the range of values in which it is 95% certain that the true value lies for the whole population.

**Confounder**  
A third variable that indirectly distorts the relationship between two other variables, because it is independently associated with each of the variables.

**Critical period**  
A period during which a specific stimulus is required for the normal development of the system and during which the organism is maximally vulnerable to environmental manipulation.

**Cross-sectional study**  
A study that examines the relationship between diseases (or other health related characteristics), and other variables of interest as they exist in a defined population at one particular time.

**Decibel**  
A unit of measurement for intensity or loudness of sound. The higher the dB the louder the sound.

**Deaf**  
Deaf with an upper-case “d” refers to those identifying with the Deaf culture. Deaf with a lower-case “d” is often synonymous hearing loss.

**Degree of hearing loss**  
Degree of hearing loss is measured in dB over four audiometric frequencies 500-4000 Hz and based on assessment of the better ear. Mild is a loss from 26-40 dBL; moderate is a loss from 41-56 dBL; severe is a loss from 66-95 dBL; and profound is a loss above 95 dBL.
Effectiveness  A measure of the extent to which a specific intervention, procedure, regimen, or service, when deployed in the field in routine circumstances, does what it is intended to do for a specified population.

Evidence table  A summary display of selected characteristics (e.g., methodological design, results) of studies of a particular intervention or health problem.

Frequency  The pitch of a sound. Determined by the number of vibrations per minute of a sound and measured in Hertz (HZ).

Generalisability (applicability, external validity)  Applicability of the results to other populations.

Incidence  The number of new events (cases; e.g. of disease) occurring during a certain period, in a specified population.

Infant  A child aged less than 12 months old

Internal validity  The extent to which the design and conduct of a study are likely to have prevented bias.

Matching  The process of making a study group and a comparison group comparable with respect to extraneous factors.

Mean  Calculated by adding all the individual values in the group and dividing by the number of values in the group.

Median  Any value that divides the probability distribution of a random variable in half. For a finite population or sample the median is the middle value of an odd number of values (arranged in ascending order) or any value between the two middle values of an even number of values.

Meta-analysis  The process of using statistical methods to combine the results of different studies. The systematic and organised evaluation of a problem, using information from a number of independent studies of the problem.

Multiple regression  Any analysis of data that takes into account a number of variables simultaneously.

Neonate  A child aged less than 4 weeks of age.

Nested case control study  A case control study in which cases and controls are drawn from the population in a cohort study. That is, the case control study is “nested” within the cohort study design so that the effects of some potential confounding variables are reduced or eliminated. A case control study can also be nested into a case series study. See also case control study, cohort study, and case series study.

Observational study  A study in which the investigators do not seek to intervene, and simply observe the course of events.

Open-set speech recognition test  A test used to measure ability to recognise speech. No lip-reading is allowed and no choices given.

Population-based screening programme  A population-based screening programme is one in which screening is systematically offered by invitation to a defined, identifiable population.

Post-lingual deafness  Hearing loss occurring before the acquisition of language (i.e before about 3 years)

Pre-lingual deafness  Hearing loss occurring after the acquisition of language (i.e after about 3 years-either congenital or acquired)
Prevalence  The number of events in a given population at a designated time (point prevalence) or during a specified period (period prevalence).

Primary care  First contact, continuous, comprehensive and coordinated care provided to individuals and populations undifferentiated by age, gender, disease or organ system.

Primary research/study  ‘Original research’ in which data are collected. The term primary research/study is sometimes used to distinguish it from a secondary study (re-analysis of previously collected data), meta-analysis, and other ways of combining studies (such as economic analysis and decision analysis). (Also called original study.)

Randomised controlled trial  An epidemiologic experiment in which subjects in a population are randomly allocated into groups to receive or not receive an experimental preventive or therapeutic procedure, manoeuvre, or intervention. Randomised controlled trials are generally regarded as the most scientifically rigorous method of hypothesis testing available in epidemiology.

Recall bias  Systematic bias due to differences in accuracy or completeness of recall or memory of past events or experiences.

Relative risk (RR)  The ratio of the risk of disease or death among the exposed to the risk among the unexposed. It is a measure of the strength or degree of association applicable to cohort studies and RCTs.

Reliability  The degree to which results obtained by a measurement procedure can be replicated. Lack of reliability can arise from divergences between observers or measurement instruments, measurement error, or instability in the attribute being measured.

Sensorineural hearing loss  Hearing loss due to damage to hair cells or damage or absence of auditory nerve.

Significant hearing loss  Refers to mild and greater hearing loss (see degree of hearing loss)

Screening  Screening is the examination of asymptomatic people in order to classify them as likely or unlikely to have the condition that is the object of screening.

Secondary care  Surgical and medical services that are generally provided in a hospital setting. In many cases, access to these services is by referral from a primary care health professional such as a general practitioner.

Secondary research/study  Re-analysis of previously collected data, meta-analysis, and other ways of combining studies (such as economic analysis and decision analysis)

Selection bias  Any error in selecting the study population such that the people who are selected to participate in a study are not representative of the reference population or, in analytic studies the comparison groups are not comparable.

Sensitivity analysis  A method to determine the robustness of an assessment by examining the extent to which results are affected by changes in methods, values of variables, or assumptions.

Statistical difference  A result that is unlikely to have happened by chance. The usual threshold for this judgement is that the results, or more extreme results, would occur by chance with a probability of less than 0.05 if the null hypothesis was true. Statistical tests produce a p-value used to assess this.

Systematic review  Literature review reporting a systematic method to search for, identify and appraise a number of independent studies.

Variance  A measure of the variation shown by a set of observations, defined by the sum of the squares of deviation from the mean, divided by the number of degrees of freedom in the set of observations.
BACKGROUND

This technical brief was requested by Vickie Rydz, Development Manager, Disability Services Directorate, Ministry of Health, New Zealand Government.

Rationale for technical brief

A universal newborn hearing screening (UNHS) programme is to roll-out in New Zealand in mid-2007. At present, between 135 and 170 infants and young children in New Zealand are identified annually as having a significant permanent congenital hearing loss (National Screening Unit 2005), with the average age of detection of moderate or greater hearing loss being about 46 months, well above the recommended 3 months of most countries (Project HIEDI and National Foundation for the Deaf 2004).

Permanent congenital hearing loss has major effects on language acquisition, cognitive development, educational achievement, social functioning, mental health and self-esteem (Conseil d’Evaluation des Technologies de la Sante du Quebec (CETS) 1997; Project HIEDI and National Foundation for the Deaf 2004). The variables known to increase the effects of hearing loss are: age of onset, severity of the loss, presence of other disabilities and intervention delay (Project HIEDI and National Foundation for the Deaf 2004).

Interventions for children with severe hearing loss include speech and language training (both oral and sign language) and use of “sensory aid” technologies such as hearing aids and cochlear implants that allow sound to either be amplified or perceived by the recipient. There is some evidence that hearing aid effectiveness is improved by usage during the first 6 months of age (Waltzman and Roland 2005) and that cochlear implantation at a young age results in more rapid and a greater improvement of speech production and perception than a later age although the optimum age for implantation is not clear (Willstedt-Svensson et al. 2004; Rizer and Burkey 1999). There seems to be evidence that implantation before 5 years is better than after 5 years of age (Kirk et al. 2002) at least after 2 years of use (Willstedt-Svenssson et al. 2004), and some studies suggest that implantation before the age of 3 years has a better outcome than after 3 years of age (Kirk et al. 2002).

These observations fit with theories of critical or sensitive periods for the auditory system (Project HIEDI and National Foundation for the Deaf 2004): those with congenital hearing loss who have early access to auditory stimuli will have an improved outcome by reducing the period of sensory deprivation and decreasing the age at which a child is first exposed to language (Connor et al. 2006; Project HIEDI and National Foundation for the Deaf 2004).

The implementation of the UNHS programme will mean earlier identification of infants with hearing loss (at birth or a few months of age) and an increase in the demand for early cochlear implantation (Houston et al. 2003). This technical brief was requested to provide evidence based information on whether the effectiveness of cochlear implantation increases with early implantation and what the optimum age is in terms of audiological performance, communication outcomes, educational achievement and quality of life.

History of cochlear implant use and age of eligibility

The first deaf adult was implanted with a rudimentary cochlear implant in Paris, in 1957 (Conseil d’Evaluation des Technologies de la Sante du Quebec (CETS) 1997). In 1961 a number of adults were implanted in the USA and in the 1970s implant programmes began in a number of countries mostly implanting post-lingually deaf adults (Conseil d’Evaluation des Technologies de la Sante du Quebec (CETS) 1997). In 1985, the USA FDA approved the marketing of two models of cochlear implants for post-lingually deaf adults (Conseil d’Evaluation des Technologies de la Sante du Quebec (CETS) 1997). Candidacy was then extended to post-lingually deaf children followed by pre-lingually deaf children. In 1990 the FDA approved the marketing of cochlear implants for children two years of age and older (Conseil d’Evaluation des Technologies de la Sante du Quebec (CETS) 1997; Geers 2006).

There were concerns at this time about implantation of children under 2 years of age including: correctly diagnosing profound hearing loss in young children; having time to judge the benefits of
amplification; needing accurate feedback to enable programming of implants; and possible surgical difficulties such as skull size and whether future growth would cause electrode extrusion in younger children (Rizer and Burkey 1999). Particular issues with children deafened by meningitis and the need for the implant to be inserted before ossification of the cochlear in these children led to a number of children being implanted before 2 years of age. This demonstrated that many of the concerns about implantation in the under twos were unwarranted or were able to be overcome (Rizer and Burkey 1999).

In 2000 the FDA approved cochlear implant for children as young as 12 months (Houston et al. 2003). Some centres are implanting children younger than 12 months, for example where there is clear lack of benefit from hearing aids (Houston et al. 2003).

**Technical description of cochlear implant device**

Cochlear implants convert sound into an electrical current that stimulates the auditory nerve and produces the sensation of sound for the recipient (Conseil d'Evaluation des Technologies de la Sante du Quebec (CETS) 1997). As the signal is an electrical current it does not make use of the ear’s mechanical function; it bypasses the external and middle ear to excite the auditory nerve directly (Conseil d'Evaluation des Technologies de la Sante du Quebec (CETS) 1997).

A cochlear implant consists of external and internal parts (Conseil d'Evaluation des Technologies de la Sante du Quebec (CETS) 1997). A microphone, voice processor and transmitting coil are located externally; the receiver/stimulator is implanted in the mastoid bone behind the ear and the electrode array is implanted in to the cochlea (Conseil d'Evaluation des Technologies de la Sante du Quebec (CETS) 1997).

**Alternative or competing technologies**

The development of the wearable electronic hearing aid more than 50 years ago had a significant effect on the acquisition of language for children with hearing loss who had some residual hearing (Geers 2006). However, hearing aids were less effective for children with profound hearing loss (Geers 2006).

The vibrotactile aid, developed in the 1980s, was a body worn vibrator “designed to present temporal and spectral aspects of speech” to profoundly deaf children (Geers 2006). It was meant to compliment the use of hearing aids and lip-reading. However, it did not improve the comprehension of speech over hearing aids alone (Geers 2006).

Cochlear implantation produces faster language acquisition than hearing aids or vibrotactile aids for children with hearing loss (Geers 2006). Even children with minimal benefit from the amplified speech of the hearing aid can have access to the perception of speech with the cochlear implant (Geers 2006).

**Other cochlear implantation considerations**

The focus of a child with a cochlear implant often becomes oral communication rather than total communication or sign language, and some deaf communities argue that this may diminish an individual’s ability to participate in the deaf community while leaving them with difficulties fully participating in the hearing world.

**SELECTION CRITERIA**

**Study inclusion criteria**

Publication type

Studies published between 1996 and 28 December 2006 inclusive in the English language, including primary (original) research (published as full original reports) and secondary research (systematic reviews and meta-analyses) appearing in the published literature.
Context

Studies reporting on the effectiveness of cochlear implantation at a young age compared with implantation at an older age where at least some of the sample was less than 2 years of age at implantation and where the mean or median of the study population was less than 36 months of age at implantation.

Outcomes

Outcomes of interest were:
- audiological performance outcomes including auditory responses to speech and sound or speech localisation, perception, and/or production
- communication (both oral and sign language) outcome and skills
- education achievements
- quality of life

Study design

Cohort studies and cross sectional studies examining the effectiveness of early cochlear implants in infants or young children.

Sample size

Studies with samples of at least 20 participants, where at least some of the sample was aged less than 2 years of age at implantation, and where the mean or median of the study population was less than 36 months at implantation.

Study exclusion criteria

Research papers were excluded if they:
- were non-systematic reviews
- were not published in English
- were letters, editorials, expert opinion, book chapters, conference proceedings, comments and articles published in abstract form, or non-published work
- were superseded by a later publication with longer follow-up data and overlap in the patient population
- reported on the effectiveness of cochlear implantation compared with another intervention and did not evaluate effectiveness when implanted early compared to a later age
- only included outcomes related to technical electrical stimulation or surgical complications.

MAIN SEARCH TERMS

Details of the search strategy are presented in Appendix 2.

MeSH headings: cochlear implants, cochlear implantation, treatment outcome, child language, speech perception, quality of life, language development, vocabulary, verbal behavior, auditory perception, voice quality, auditory threshold, postoperative complications, time factors, age factors, child-preschool

Embase subject headings (where different from Medline): cochlea prosthesis, implantation, speech discrimination, verbal communication, speech development, speech, vocalization, speech articulation, linguistics, verbalization

Additional keywords: preschool, pre-school, 12 months, 24 months, 36 months, young child$, twelve months, twenty four months, thirty six months, 1 year, 2 years, 3 years, one year, two years, three years, pediatr$, paediatr$, infant$, months.ti, ((audio-verbal or audioverbal or auditory) adj (training, or therapy)), ((neonatal or newborn) and screening and hearing)
SEARCH SOURCES

The NZHTA CORE Search was employed. Characteristics of the Core search include: essential sources only, major databases and secondary sources, and mostly published and indexed literature.

Bibliographic databases
Cinahl
Medline
Embase
Current Contents
Cochrane Library Controlled Trials Register
ERIC
Index New Zealand
New Zealand Bibliographical Database
PsychInfo
Science & Social Science Citation Index

Review databases
Cochrane Database of Systematic Reviews
Database of Abstracts of Reviews of Effectiveness (DARE)
NHS Economic Evaluation Database
Health Technology Assessment Database
TRIP Database

Other
Reference lists of retrieved papers
American Speech Language Hearing Association http://www.asha.org
Australian Association of the Deaf http://www.aad.org.au
Canadian Association of Speech Language Pathologists and Audiologists http://www.caslpa.ca/
Canadian Academy of Audiology http://www.canadianaudiology.ca/consumers/cochlear/
MedlinePlus http://www.medlineplus.gov
New Zealand Audiological Society http://www.audiology.org.nz
Royal National Institute for the Deaf (UK) http://www.rnid.org.uk/
(UK) National Cochlear Implant Users Association http://www.nciua.demon.co.uk/index.html
(US) National Institute on Deafness and Other Communication Disorders
(US) Educational Audiology Association http://www.edaud.org/
(US)National Association of the Deaf http://www.nad.org

Articles published in English language only were considered.
The search was restricted to literature published since 1996. Searching was undertaken on 5 December 2006 with follow-up searches between 15 and 18 January, 2007.

APPRaisal METHOdology

Summaries of appraisal results are shown in tabular form (known as Evidence Tables) which detail study design, study setting, sample, methods, results, reported conclusions and NZHTA reviewer conclusions/comments based on the limitations and validity of the study.

The evidence presented in the selected studies were assessed and classified according to the NHMRC’s revised hierarchy of evidence (Appendix 1).
RESULTS

From the above search strategy we identified, 946 potentially relevant articles/abstracts of which 155 were retrieved. Of these retrieved articles, 140 were excluded. Table 2 documents criteria for excluding retrieved papers and numbers excluded for each criterion.

Table 1 Reasons for exclusion of retrieved papers

<table>
<thead>
<tr>
<th>Reason for exclusion</th>
<th>Number of papers</th>
</tr>
</thead>
<tbody>
<tr>
<td>Sample size &lt;20</td>
<td>21</td>
</tr>
<tr>
<td>None in sample under 2 years at implantation</td>
<td>30</td>
</tr>
<tr>
<td>Mean age at implantation ≥ 36 months</td>
<td>35</td>
</tr>
<tr>
<td>Age at implantation not clearly defined</td>
<td>11</td>
</tr>
<tr>
<td>Outcomes ineligible or not comparison of effectiveness between young age and older age</td>
<td>38</td>
</tr>
<tr>
<td>Inappropriate study design</td>
<td>1</td>
</tr>
<tr>
<td>Data not clearly presented</td>
<td>1</td>
</tr>
<tr>
<td>Narrative review</td>
<td>1</td>
</tr>
<tr>
<td>Review not systematic</td>
<td>1</td>
</tr>
<tr>
<td>Commentary</td>
<td>1</td>
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</tbody>
</table>

Fifteen retrieved articles were appraised (listed in Appendix 4). Included papers are presented in the evidence table below. Included studies were all level III-2 or III-3 according to NHMRC’s hierarchy of evidence. All studies were observational studies including 3 cross sectional surveys and 13 cohort studies (one study used two designs).

Summary of results for the effectiveness of early cochlear implantation for infants and young children with hearing loss

Secondary studies

There were no secondary studies that assessed the effectiveness of early cochlear implantation compared to late implantation.

Primary studies

Sixteen primary studies (see Table 2, pages 11-39) fulfilled a priori eligibility criteria for the evaluation of the effectiveness of early versus late cochlear implantation for infants and young children. These studies are summarised below and in Table 2.

Connor et al. (2006) studied the existence of any added benefit to earlier cochlear implantation over and above the advantage provided by longer implant use for children compared with same age peers. Specifically they examined the latent-growth curves for 100 children, who received cochlear implants at various age groups from as early as 12 months to as old as 10 years of age using a hierarchical linear model. The model considers differences in the latent-growth curve between individual children but not within repeated measures for each child. Children were divided into four groups according to age at implantation. Two groups received their implants before 42 months of age (21 children received the implant between 12-30 months, and 15 children received their implants between 30-42 months). The latent growth curves examined speech scores and vocabulary tests. Generally, children who were younger when they received implants (particularly those implanted before 2.5 years) demonstrated stronger outcomes at any age than their same age peers who were older when they received the implant. This advantage was shown to be associated proportionally with the length of implant use. Furthermore, a burst of growth noticed immediately after implantation was primarily associated with very early implantation. This additional advantage seemed to diminish with increasing age at implantation.

Limitations to this study included:
- susceptibility to confounding and misclassification since the study did not use a randomised design and there was no blinding
- incomplete collection of data (SPEECH score were only available in 57% of the younger age group and 73% of the older age group).
Nicholas and Geers (2006) concluded that the amount of spoken language exhibited by profoundly deaf 3-year-old children was largely related to the length of time of cochlear implant use in infancy and very early childhood. The study also showed a reduction in the gap between a deaf child’s chronological age and the child’s spoken language level with younger age at cochlear implantation. Conclusions were based on examining data from 76 profoundly deaf children attending oral education programmes or therapy practices, who received a cochlear implant between the ages of 12 and 38 months and who have used the implant for at least 7 months. Primarily this cross-sectional study aimed to investigate possible contributions of two factors on spoken language outcomes when the child is 3.5 years of age. Audiological intervention was one factor defined by the duration of use of both a hearing aid and a cochlear implant, and the auditory perceptual experience was the other factor which was measured by aided sound field thresholds. The study hypothesis predicted better language skills at age 3 for each month of cochlear implant use. Spoken language skills of the children were tested through standardised 30-minute language (play session with own parent), parent-completed vocabulary checklist (Communicative Development Inventory-CDI), and teacher language-rating scale sample analysis (Scales of Early Communication Skills for Hearing Impaired Children). All children were tested between the ages of 40 and 44 months. However, a randomised controlled design was not used and there was no blinding so the results were susceptible to confounding and misclassification. The primary focus of the article was on duration of implantation rather than age at implantation.

In their cohort-sequential study Miyamoto et al. (2005) examined eight profoundly deaf infants who were early implanted with a cochlear implant while they were aged between 6 and 12 months and compared them to 18 infants who had their implantation performed between 12 and 24 months of age. Two behavioural methodologies were used to assess speech perception and language skills of participating infants’ after implantation. One tool (Visual Habitation –VH) assessed the ability of the infant to discriminate speech sounds (e.g. a continuous versus a discontinuous sound pattern) and the other (Preferential Looking Paradigm-PLP) assessed the ability of the infants to learn associations between speech sounds and objects (represented by looking at time differences in the target versus the non-target videos). Infants were tested at three separate interval groups and within each group the intervals ranged between one day and 18 months after cochlear implantation. Findings from this very small study were consistent with some enhanced learning ability among infants receiving very early cochlear implantation. The small sample size was a major limitation and the study was also susceptible to confounding and bias due to the study design used.

In another study of growth curves Tomblin et al. (2005) observed greater overall rates of growth in expressive language for children who were younger when they received their implants, when age at implantation was considered as a continuous variable, rather than when groups of children were compared. They used both spoken and/or sign responses to measure the growth of expressive language skills in 29 children who received their cochlear implant during infancy. They sought to assess the relationship between expressive language outcomes and cochlear implant experience.

All participating children were diagnosed within their first year of life with profound bilateral sensorineural hearing loss (SNHL); all were born to hearing parents. Participants were selected from a larger comprehensive longitudinal Cochlear Implant centre study on the basis of receiving their cochlear implantation before the age of 40 months and with at least 12 months of cochlear implant use. Both the Minnesota Child Developmental Inventory (MCDI) and Preschool Language Scale-3 (PLS-3) tools were used to measure language growth. MCDI was completed by the parent(s) and, at the same time, the investigator child administered the PLS-3 tool. Scores from both tools were converted into age-equivalent scores based on the norms provided by the tests. Data were collected at least once before cochlear implantation and then after implantation repeatedly from each child at different ages and frequencies ranging from once a month to once a year.

Available observational data from the cross-sectional design focused on outcomes prior to implantation and at 12 and 24 months after cochlear implantation. Results from both tests showed a clear negative correlation between age at initial stimulation and MCDI and PLS-3 scores indicating that children who were implanted earlier in life fared better (relative to their normal-hearing peers) than did the children who were implanted later in life. This was further highlighted when MCDI ELQ scores for 14 children implanted between 12 and 20 months of age compared to those of 13 children implanted between 21 and 48 months of age. Despite the observation of a small decline in ELQ scores following initial stimulation in children implanted early, ELQ scores were lowest for those implanted later. The difference between the two was significant at the 12-month interval after initial stimulation ($t(25) = -$}
listening skills than their peers implanted at a later age. Statistical analysis showed significant of cochlear implant use, children implanted before 2 years of age performed better in terms of auditory Monosyllable Trochee Polysyllable (MTP-tests pattern perception) tests. After one and a half to 2 years improvement over time (up to 3 years) with every test (LiP, MTP) and questionnaire (MAIS, MUSS) profoundly deaf children who received cochlear implants before the age of 12 months (mean 0.8 years) outcome in these age groups, the German versions of the MAIS and MUSS questionnaires were used. The test battery was not applied during the first 6-12 months of rehabilitation. To evaluate the material was used to evaluate speech rhythm recognition, consonant and vowel understanding, and then their progress was monitored after 3, 6, and 12 months then annually post-implantation. Closed-set implanted between August 1996 and May 2002. The children were evaluated before implantation, and 25-36 months of age and 23 between 37-48 months of age. Assessments were carried out by administering a modified open-set test of spoken and word recognition (the Mr Potato Head Task) prior to cochlear implantation and then regularly every 6 months after implantation. Measures reported for each group showed average performance on the test (represented by group mean percent correct) as a function of age. Authors stated that children implanted in their first year of life were not old enough at each testing interval to be compared to the norms for normal-hearing children. Average performance (spoken word recognition) started improving faster shortly after device activation among the later three groups but not those implanted in their first year of life. Apart from those implanted in their first year of life, speech recognition was better for children implanted early in their life than those implanted later. This was clearly shown by better performance in children implanted in their second year of life (average word recognition score = 70%) than those implanted in their third (52% score) and fourth (33%) years of life. These results were consistent for more than 3 years. The very small sample size in the first year of life is a significant limitation as the study power is insufficient to form any conclusions about the value of implantation at that age. The study was also limited by the lack of blinding and the use of a non-randomised design.

In a German speaking population Lesinski-Schiedat et al. (2004) retrospectively compared data of 27 profoundly deaf children who received cochlear implants before the age of 12 months (mean 0.8 years) to 89 children implanted between 12 and 24 months of age (mean 1.6 years). The children were implanted between August 1996 and May 2002. The children were evaluated before implantation, and then their progress was monitored after 3, 6, and 12 months then annually post-implantation. Closed-set material was used to evaluate speech rhythm recognition, consonant and vowel understanding, and discriminative ability of the children. TAPS (Test of Auditory Perception of Speech for children ‘German version’), which is an open-set method, was used to evaluate cognitive ability, and the GASP test (Glendonald Auditory Screening Procedure) in textual context was used to evaluate ability to abstract. The test battery was not applied during the first 6-12 months of rehabilitation. To evaluate the outcome in these age groups, the German versions of the MAIS and MUSS questionnaires were used. After 12 months of cochlear implant use nearly half of those implanted earlier did not complete the
tests or questionnaires for the study, and after 24 months only 8 completed the tests. Incomplete testing was also an issue in those implanted later, with 56 of 89 completing the tests after one year and 44 after 2 years. Study results showed that after 12 months of implant use children implanted later showed better speech understanding without visual assistance (open-set) scoring higher in the TAPS and monosyllable tests than those implanted earlier. However, these results reversed after 18 and 24 months of cochlear implant use and the groups of children implanted earlier achieved better lexical understanding after 24 months of implantation. Similar results were shown on the GASP and general sentences tests but only after 18 to 24 months after cochlear implantation. Earlier implanted children achieved 50% in the GASP test and 66% in the Common Phrases test compared to 30% and 53% for those implanted at a later age. Children implanted earlier also showed better speech results in terms of word understanding on the closed-set test by the age of 2.5 years, with sentence understanding in particular being more advanced in the first group (67% in general sentence) than in the second group (20%). Study limitations are detailed in Table 2. A key issue related to missing data at different time points during follow-up. As with the other studies, the lack of randomisation and blinding were also limitations.

Speech perception ability was tested and shown to be correlated with age at implantation by Manrique et al. (2004a, 2004b) in their prospective cohort study from Spain. The study performed repeated measures to evaluate and compare outcomes of 94 children with bilateral profound hearing impairment of children who were consecutively implanted with cochlear devices before their second birthday to 36 children implanted between 2-6 years of age. Children were either congenitally deaf or lost their hearing before their second birthday (pre-linguistically deaf). Children were followed up for 5-8 years with evaluations carried out before surgery and each year after implantation. Manrique et al. (2004a) reported on results from closed-set as well as open-set evaluations conducted on these children before, and annually after cochlear implantation for up to five years. Infants implanted before the age of 2 years showed better auditory perception test results than those implanted at a later age. This was specifically seen with better vowel identification results and daily words as well as better performance at bi-syllabic word test.

Children implanted before 2 years of age achieved better levels of speech perception and production than those implanted between 2-6 years. The correlation between the chronologic age and the age of vocabulary acquisition was assessed by using the Peabody Language Development test.

Children implanted before the age of 2 followed a relatively normal development of language, whereas older children showed a lag of approximately 2 years when compared with the normal baseline.

Similarly the use of the Renyll general oral expression scale showed that children implanted before the age of 2 years lagged approximately 1 year behind their normal peers, whereas children implanted at ages 2-6 years showed a lag of approximately 3 years behind their normal peers and they also tended to show slower progress.

The second paper reported on longer term auditory results of up to 8 years (Manrique et al. 2004b). The study aimed to determine the delineation between fair or poor performers versus good performers based on age at implantation. The study looked specifically at pure-tone audiometry thresholds, speech perception closed-set (Vowel Confusion test) and open-set (Bisyllabic Words test) tests. Children implanted before their third birthday showed significantly better performance over time compared with all children implanted later.

Results from both studies indicated that the earlier a child receives the cochlear implant the better his/her speech perception ability would be. This was indicated by better performances in both close-set and open-set tests of audition and speech perception irrespective of follow-up time. Furthermore, speech development of children implanted before the age of 2 years approached very closely to that of normal children.

Both studies were limited by their before and after design since this design is unable to control for other factors when assessing the role of the intervention (early CI) of interest.

In their study, McConkey-Robbins et al. (2004) looked at the effect of age at cochlear implantation on auditory development and speech perception over time in children who received cochlear implants when they were younger than 3 years of age. The study also compared the data with previously
published data from a cohort with normal hearing of the same chronological age. Participants were 45 children implanted between 12-18 months, 32 were implanted between 19 and 23 months, and 30 between 24-36 months of age. Assessments were carried out before implantation then 3, 6 and 12 months after implantation using the mean Infant-toddler Meaningful Auditory Integration Scale (IT-MAIS) scores. This tool is a structured parent interview that assesses auditory skill development in children by enquiring about spontaneous listening behaviours in everyday situations. Therefore it does not rely on the child’s compliance. All infants and toddlers showed rapid improvement in auditory skills during their first year of device use regardless of age at implantation with higher mean scores of IT-MAIS achieved by younger children. Similarly children who received cochlear implants at earlier age attained auditory skills nearer to those of their normal hearing peers at a younger age. However, regardless of age at implantation the mean rate of acquisition of auditory skills was similar to that of infants and toddlers with normal hearing.

Svirsky et al. (2004) investigated the hypothesis that implantation in the second year of life results in better speech perception and language development outcomes than later implantation. Therefore, they compared the speech perception and language skills of congenitally deaf children who received cochlear implants in the second year of life (12 children), third year of life (34 children) or fourth year of life (29 children). They used developmental trajectory analysis (DTA), which examines the curves representing changes over time, to assess the impact of early implantation throughout the child’s developmental trajectory. This was also compared to three normal-hearing reference curves of age-matched children. Analyses showed close language scores for cochlear implant users to average values from normal-hearing children as a function of age. The average estimated language age for children implanted between 12 and 24 months (measured in units of language age) was 5.7 months higher than for children implanted at 25-36 months at the same chronological age. Children implanted after 3 years of age lagged behind those implanted between 25-36 months by 5.6 months ($p<0.05$), and those implanted at 12-24 months by 10 months ($p<0.001$).

Similarly speech perception ability was correlated with age at implantation, and children who received cochlear implants before their second or third birthday showed better rates of acquisition than those implanted after their third birthday. This study had a high participation rate but was susceptible to bias and confounding due to the use of a non-randomised design.

Using both retrospective longitudinal and cross sectional study designs Govaerts et al. (2002) evaluated data from six age cohorts implanted up to 6 years of age. They assessed the auditory performance (using the indirect measure Categories of Auditory Performance CAP) preoperatively and up to 2 years after cochlear implantation. Integration into the mainstream school system decreased with increasing age at implantation. The CAP scores were rapidly (after 3 months) normalised in children implanted before the age of 2 years. Children implanted later, took longer to achieve scores similar to their normal-hearing peers. For example, 25% of children implanted after their third birthday could not achieve normal CAP scores within the first 48 months after implantation. Furthermore, this outcome was rarely achieved in children implanted after the age of 4 years. This study had a small sample size, and was susceptible to confounding due to the lack of a randomised design. Blinding was also not documented.

Hammes et al. (2002) retrospectively analysed data on 12 infants with hearing loss who received cochlear implants before the age of 18 months. These were compared to 13 infants who received their implants between 19-30 months of age, 11 children implanted between 31-40 months, and to 11 children who received their implants between 41-48 months. Comparisons according to age at implantation were performed using spoken language measures including skills for hearing impaired children, preschool language scales-3 and oral and written language scales. Spoken language data were reviewed for the children who underwent at least 6 months of cochlear implant use to assess the rate of progress and performance for the group of children who received their cochlear implants between 9 and 18 months of age (only 10 children). The study indicated that children fitted with cochlear implants at an early age showed improvement in expressive and receptive language capabilities as well as developed speech and language skills at the same rate as normal hearing children. Children who had implants inserted at or before the age of 18 months showed the best outcomes.

Regardless of age at implantation, the majority of children used total communication before receiving their implant. After receiving the implant, all of the infant subjects made a successful transition to
spoken language. This study had a small sample size, and was susceptible to confounding due to the lack of a randomised design. Blinding was also not documented.

Twenty-two young German-speaking deaf children were implanted with cochlear implants (mean age at implantation 29 months) and were prospectively followed-up by Szagun et al. (2001). Language acquisition in terms of progress in grammar and vocabulary of these children was compared to that of a group of 22 normal-hearing children over a period of 27 to 36 months. The general measure of grammatical progress was through the mean length of utterance (MLU). Vocabulary acquisition was measured by parental questionnaires. Data collection took place at two different settings for the two groups: the Cochlear Implant Centre in Hanover for children with cochlear implants and at the University of Oldenburg playroom for normal-hearing children. Correlation analyses were used to test the relationship between age at implantation and linguistic growth among children with cochlear-implants. Partial correlations were calculated between language growth measures and chronological age at implantation controlling for pre-operative hearing. Age at implantation and pre-operative hearing with hearing aids were associated with subsequent linguistic growth. Pre operative hearing had the stronger relation. The study highlighted that, among children who undergo cochlear implantation before 4 years of age, it is not until 2 to 2 and half years of cochlear implant use that one can tell whether a child develops language near normal or not. This study had similar limitations to previous studies. The sample size was small. The use of an observational design did not allow for control of confounding. There was also a probability of bias resulting from the lack of blinding and from the selection of the two different groups from different settings.

In their longitudinal study Kirk et al. (2000) studied the rate of growth in word recognition and language skills as a function of age at cochlear implantation and whether there are sensitive periods for cochlear implantation before the age of 3 years. They prospectively followed up 106 pre-lingually deaf children who received their cochlear implants at either <2 years, between 2-4 years, or at age 5 or older. Fifty-six of the participants communicated orally and 50 children used total communication. A battery of speech and language outcome measures were administered to the children according to their model of communication before the implantation and six monthly after the implantation for at least 3 years. Data from two measures of spoken word recognition and from two measures of receptive and expressive language were analysed using analyses of variances with the length of device use, age at implantation, and communication mode as the covariates. The goal of spoken word recognition testing is to determine a child’s ability to understand and repeat speech presented through listening alone. The Grammatical Analysis of Elicited Language-Pre-Sentence Level (GAEL-P) as a closed-set spoken word recognition task. Whereas Mr Potato Head Task was used as an open-set measure of spoken word recognition task. The measures of receptive and expressive language used were the Peabody Picture Vocabulary Test-Third Edition (PPVT-III), which is a measure of receptive vocabulary, and the Renyll Developmental Language Scales (RDLS), which is a measure for receptive and expressive language developed for children with normal hearing. The study showed significant improvements in spoken word recognition and receptive and expressive language skills following cochlear implantation. The rate of improvement in both the closed-set (measured by GAEL-P) and open-set (measured by Mr Potato Head Task) word recognition skills was not affected significantly by age at implantation. On the contrary, age at implantation significantly affected the rate of growth in receptive vocabulary (measured by the PPVT-III) as well as on the development of expressive language abilities (measured by the RDLS). Children implanted before their second birthday showed higher language quotients at both initial testing and over time than those implanted later. The use of an observational design did not allow for control of confounding. There was also a probability of bias resulting from the lack of blinding and from the lack of recording some data points.
Table 2. Evidence table of appraised articles relating to effectiveness of cochlear implantation at a young age compared to an older age (population with a mean age of less than 36 months and containing some aged less than 2 years of age at implantation)

<table>
<thead>
<tr>
<th>Authors</th>
<th>Country</th>
<th>Study Design / Objectives/ Inclusion-exclusion criteria</th>
<th>Sample and Interventions</th>
<th>Methods</th>
<th>Results</th>
<th>Limitations and Conclusions</th>
</tr>
</thead>
<tbody>
<tr>
<td>Connor 2006</td>
<td>Florida USA</td>
<td>Prospective Level III-2</td>
<td>To examine the effects of developmental maturation, length of implant use and age at implantation on children's speech and vocabulary growth</td>
<td>Study setting Major cochlear implant centre Participants: children received CI between 1981 &amp; 2004 n=100 children with congenital HL who received CI between ages 12m and 10y (mean 61 months) divided in four groups. Some had used the implant for up to 13 years at the time of the study (mean 4y)</td>
<td>Clinical follow-up before-and-after cochlear implantation assessments conducted.</td>
<td>For consonant production accuracy SPEECH scores grew faster in children implanted before the age of 2.5 compared to those implanted &gt;2.5y (Table 2 in the paper) indicating sustained change in growth rate For the length of use of CI, after 2y of use, SPEECH scores significantly greater among group A1 vs. group A2, but after 3y of use scores were similar. Group A1 demonstrate early burst of SPEECH growth lasted about 2y before slowing to rates similar to those of children in group A2 A child in group A1 at age 6y expected to achieve a SPEECH score of 75 compared with a 6-y old child in group A2 with predicted score of about 55.</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Inclusion criteria: Children with congenital deafness, who received CI (1981-2004), who only used oral communication approach. Exclusion criteria: Children with history of meningitis, Mondini malformation, or additional disabilities.</td>
<td>Speech + language assessments 6 months before CI (Pre-implant). Follow-up (post-implant) each 6 months for 2 years then each 12 months thereafter. Measuring tools: PPVT-3 (Peabody Picture Vocabulary Test3) for spoken receptive vocabulary skills. Using spoken English (no sign language) in quiet room SPEECH (Consonant-production accuracy) in response to picture stimuli-transcribed by speech-pathologist &amp; entered into computer software to compute percentage of consonants produced correctly. Higher SPEECH scores = better speech intelligibility for children with normal hearing and for children with CI</td>
<td>Speech + language assessments 6 months before CI (Pre-implant). Follow-up (post-implant) each 6 months for 2 years then each 12 months thereafter. Measuring tools: PPVT-3 (Peabody Picture Vocabulary Test3) for spoken receptive vocabulary skills. Using spoken English (no sign language) in quiet room SPEECH (Consonant-production accuracy) in response to picture stimuli-transcribed by speech-pathologist &amp; entered into computer software to compute percentage of consonants produced correctly. Higher SPEECH scores = better speech intelligibility for children with normal hearing and for children with CI</td>
<td>Statistical analysis Used HLM / propensity scores to control for selection bias Regression model used to calculate propensity scores/ age at implantation as dependent variable. Independent variables: year of birth, low vs. middle SES, preimplant hearing sensitivity measures, cause of deafness, type of CI device and gender.</td>
<td>Reported conclusions (by authors). There seems to be a substantial benefit for both speech and vocabulary outcomes when children receive their implant before the age of 2.5 yr. This benefit may combine a burst of growth after implantation with the impact of increased length of use at any given age. The added advantage (i.e. burst of growth) diminishes systematically with increasing age at implantation.</td>
</tr>
</tbody>
</table>
Table 2. Evidence table of appraised articles relating to effectiveness of cochlear implantation at a young age compared to an older age (population with a mean age of less than 36 months and containing some aged less than 2 years of age at implantation) (continued)

<table>
<thead>
<tr>
<th>Authors</th>
<th>Country</th>
<th>Study Design / Objectives / Inclusion-exclusion criteria</th>
<th>Sample and Interventions</th>
<th>Methods</th>
<th>Results</th>
<th>Limitations and Conclusions</th>
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<tr>
<td>Connor 2006 Florida USA Continued</td>
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<td></td>
<td>Reviewer comments: Study highlighted these advantages with early implantation: Faster rates of vocabulary and consonant production accuracy growth immediately after implantation for those implanted &lt;2.5 years.</td>
</tr>
<tr>
<td>Authors &amp; Geers 2006 USA &amp; Canada</td>
<td>Study Design / Objectives / Inclusion-exclusion criteria</td>
<td>Sample and Interventions</td>
<td>Methods</td>
<td>Results</td>
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| **Cross-sectional** | **Level III-3** | **Objectives** To examine the contribution of pre-implant experience (duration of hearing aid and CI use) as well as auditory perceptual experience on spoken language outcomes. **Study hypothesis:** A linear relation between age at CI and language competence at 3y of age exists. **Inclusion criteria:** Presumed congenital deafness, used CI for at least 7 months, English is the primary language at home, no developmental or medical conditions that interfere with speech language development, enrolled in auditory-oral or auditory-verbal programme... | **Study setting** (Oral educational settings) 23 states across US + 1 Canadian province. Host sites (14 schools for deaf, 4 hospitals, 3 county child development centres, 4 public schools, and 7 auditory-verbal therapy practices). Participants: children received CI between 1998 & 2003 n=76 children with congenital HL who received CI between ages 12 & 38m intervention group (n=76). CI = mean age at implantation 23.16m (SD 7.73), range 12-38 m. | **Research team member travelled to child’s school or therapy location to collect data.** Measuring tools: Participant videotaped in a 30-minute play session with own parent in a quiet room. Parent: Complete a word & sentences from MacArthur Communicative Development Inventory CDI Teacher/therapist: Complete Scales of Early Communication Skills for Hearing Impaired Children **Parent report (completed vocabulary checklist), teacher language-rating scale, direct observation of child.** Outcome measures mean pure-tone average “PTA” (from language sample, parent report & teacher ratings) Spoken language (2 pre-implant variables) a. length of time from receipt of HA to CI surgery, b. sound-field threshold values obtained with use of CI to determine relation to spoken language outcomes. | **Means and standard deviations on all the language measures were presented. CLAN variables (total words, no. of different root words, bound morphemes, different morphemes, mean length utterance), CDI variables (vocabulary, irregular words, sentence complexity, longest sentence), and SECS variables (receptive scaled score, expressive scaled score).**  
• The longer the implant experience (≥1 year) the better language development  
• Correlation coefficient between language measures: All variables correlated 0.52 or higher (p<0.001)  
• Correlation between Language Factor Score and independent variables (PTA, age first aided, months of hearing aid, age at implant, post-implant PTA threshold, and duration of implant use): Language factor scores increased significantly with younger age at amplification (diagnosis & intervention; r = -0.54), also increased with younger age at cochlear implantation (r= -0.62), and increased with longer duration of implant use(r= 0.63) | **Limitations**  
• non randomised, not blind  
• no information on drop-outs  
• involved children from oral educational settings only (generalisation to other children attending other settings that use other language-teaching methods questionable)  
**Reported conclusions (by authors).** Longer use of CI in infancy & very early childhood dramatically affects amount of spoken language exhibited by 3-yr-old, profoundly deaf children. In this sample, the amount of pre-implant intervention with a hearing aid was not related to language outcome at 3.5 y of age. Rather, it was CI at a younger age that served to promote spoken language competence. The previously identified language-facilitating factors of early identification of hearing impairment and early educational intervention may not be sufficient for optimizing spoken language of profoundly deaf children unless it leads to early cochlear implantation.  
**Reviewer comments.** Although this study did not specifically evaluate the effect of age at implantation on spoken language skills it showed some linear relation between implant experience and language development. |

HA= Hearing aid, CI= cochlear implant, SD= standard deviation, CLAN= Child Language Analysis, CDI= communication Development Inventory, SECS Scales of early communication skills for hearing impaired children, PTA= pre-implant-aided
Table 2. Evidence table of appraised articles relating to effectiveness of cochlear implantation at a young age compared to an older age (population with a mean age of less than 36 months and containing some aged less than 2 years of age at implantation) (continued)

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<td>Nicholas &amp; Geers 2006</td>
<td>USA &amp; Canada... and no known problems with CI lasting more than 30 days. Exclusion criteria: Evidence or suspicion that a child had normal hearing or progressive hearing loss.</td>
<td>Functionally and behaviorally deaf children aged 1 to 3 years with a mean age of 20 months who met inclusion criteria.</td>
<td>Multiple regression linear and curvilinear regression of four independent variables used to predict language factor score. Curvilinear effects indicate that the simple slope relating duration of implant use and language test scores, with the influence of pre-implant aided threshold was estimated. Curvilinear effects indicate that the simple slope relating duration of implant use and language factor score changed as duration of implant use increased. Follow-up analyses for each level of duration of use were used to identify point at which the simple slope was significantly different from zero (in this case at 11-12 mo of use). The effect of CI use on language outcome is not apparent until after about 1 year of device use, after which a steady increase in language skill measured at age 3.5y for each additional month of use of a CI.</td>
<td>There was good agreement between measures derived from direct observation of child and measures from parent reports.</td>
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<td>Miyamoto 2005</td>
<td>USA</td>
<td>Cohort-sequential study Level III-2.</td>
<td>Study setting. Not stated Participants: n=26 infants with congenital profound hearing loss</td>
<td>Of 13 infants with profound hearing loss implanted between 6-12m, 8 infants participated in the two behavioural assessment methods. Measuring tools: VH (visual habituation) procedure assesses discrimination of sound &amp; PLP (preferential looking paradigm) assesses ability to learn association between speech sounds &amp; objects. Three interval groups testing: Group 1: tests at day 1, 1 week &amp; 1 month after implantation. Group 2: tests at 2m, 3m &amp; 6m after implantation. Group 3: at 9m, 12m &amp; 18m after implantation. Outcome measures: 'mean pure-tone average PTA' Mean looking time differences to checkerboard pattern for both VH procedure and PLP. 'Mothers' speech to infants. Mean looking time differences to the target versus the non-target video.</td>
<td>All infants showed a mean pure-tone average auditory detection of 50 dB HL or better within the first 3 months after CI (measured by visual reinforcement audiometry). VH procedure Longer looking times to the novel versus old trials for both groups but longer in infants implanted under 12 months of age. CI&lt;12m, (F(1,6) =9.20, P=0.02) CI 12-24m, (F(1,16) = 5.73, P =0.03) PLP CI &lt;12m= Significant longer looking time differences to the target versus the non-target video F(1,7) =6.45, P=0.04 CI 12-24m = did not exhibit longer looking times to the target versus the non-target video F(1,14) =0.16, P=0.7</td>
<td>Discrepancies in reporting within the article. No randomised, no blinding of intervention provider, patient and outcome assessor. Very small sample size. Assessment tools not validated. No information on drop-outs. Reported conclusions [by authors]. No surgical or anaesthetic complications occurred in this group of infants, and the pattern of listening skill development mirrors that seen in normal-hearing infants. Mothers adjust their speech to suit the listening experience of their infants. Reviewer comments. Looked at infant assessment methods to determine whether CI before the age of 12 months benefits deaf infants in terms of early speech perception and language skills.</td>
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Objectives To explore the benefits (in terms of speech perception & language skills) of early cochlear implantation in newly identified deaf infants. Inclusion/exclusion criteria: Not stated

Baseline characteristics of this group: Sex=4M, 4F, cause of hearing loss unknown in 7 and auditory neuropathy in 1. 7 received N24 implant, 1 Med-EI, mode of communication was total in 4 and oral in 4.

Comparator (n=18), CI = age at implantation between 12-24 months, mean 17.71m, range 12.39-23.24m.

Baseline characteristics of this group: Sex=11M, 7F, cause of hearing loss unknown in 13, genetic 1, auditory neuropathy1, Mondini 2, and LVA 1. 13 received N24 implant, 4 Med-EI, 1 Clarion modality of communication was total in 7 and oral in 11.
Table 2. Evidence table of appraised articles relating to effectiveness of cochlear implantation at a young age compared to an older age (population with a mean age of less than 36 months and containing some aged less than 2 years of age at implantation) (continued)

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<td>Miyamoto 2005  * USA Continued*</td>
<td>Analyses comparing groups at baseline. Baseline characteristics included sex, cause of HL, age at surgery, age at CI activation, CI type, processor, processing strategy, and communication mode. 20 used N24 implant</td>
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<td>Tomblin 2005 Iowa USA</td>
<td>Two designs (from an original longitudinal cohort) Cross-sectional and Growth curve analysis Level III-3 Objectives To evaluate the effect of receiving CI early in infancy on the expressive language development Inclusion/exclusion criteria. Although not clearly stated but referred to 2 reasons for selecting patients for this study: • Age at CI &lt;40m, • Implant experience ≥12m</td>
<td>Study setting Not stated but referred to Children participating in comprehensive CI centre study Participants: n=29 (15M, 14F) with profound bilateral sensorineural hearing loss (SNHL), normal hearing parents, no known visual abnormalities. 26 families described total communication with children (combined use of aural, manual and oral modalities in communication with and teaching hearing impaired individuals). 3 families followed oral/aural communication Spoken language was American English Within group comparison between individual participants according to age at implantation Analyses comparing groups at baseline. Age at CI placement: range 11m± SD to 40m±11d average 21m±6d SD= 7m ±2d Type of Cochlear Implant (n=29) Nucleus 24M (n=12, Nucleus 24R (n=17).</td>
<td>Systematic follow-up of children participating in comprehensive CI centre study in terms of development of communication skills. Assessment tools Two instruments: 1. MDCI (Minnesota Child Development Inventory) = parental response inventory –educational &amp; assessment tool for NH children. Parent record responses to questions on designated response forms by answering “yes” or “no” to each question regardless of the child’s modality of communication. Only the expressive language subscale (ranging from simple gestural behaviours to complex language expression) of this 8-subscale test used. 2. PLS-3 (Preschool Language Scale) = diagnostic instrument to measure language development of NH children aged 0-83m. Requires direct assessment of child At child test session an experimenter observes and records all child’s expressive communication development regardless of modality. Only expressive language subscale of this 2-subscale test used. This evaluates expressive language skills in areas of: vocal development, social communication, semantics “content”, structure “form”, and integrative thinking skills.</td>
<td>Comparison of expressive language status based on MCDI and PLS-3 of children with CI at 3 intervals relative to initial CI stimulation. In terms of the correlation of age at initial stimulation of CI with the ELQ results show: • Expressive language quotients (ELQs) showed pattern of general decline in language status relative to children with normal hearing across the three observational periods. • During peri-implant interval, no significant association between age of initial stimulation at this point in time and ELQ for both tests existed. • One year after initial stimulation a strong and significant relationship between ELQ and age at initial stimulation of CI was found for both measures (MCDI: r =-.53, p=.004; PLS-3: r =-.65, p=.0005) Comparison of the mean, MCDI ELQ scores for the 14 children with CI between 12 and 20m and 13 children with CI between 21 and 48m at 2 intervals relative to initial CI stimulation: • Children implanted at older ages fell behind the children implanted at younger ages who showed only a small decline in ELQ scores following initial stimulation.</td>
<td>Limitations • - Cross-sectional design (observational non-experimental) so does not assess outcomes over time • - Small sample size • - Discrepancy in administering 2 tests for children during peri-implant period • - Child compliance affected test administration of PLS-3 test • - Results based on data from infants with CI at or after 12m of age so results cannot be generalised to CI before this age.</td>
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<td>Tomblin 2005 Iowa USA continued</td>
<td>Outcomes: (cross-sectional design) Language measures (using each child’s preferred modality of communication ‘oral or manual’) i.e. spoken and/or signed Conversion of scores for Statistical analysis: First- creation of norm referenced scores- Raw scores from both MCDI and PLS-3 converted to age-equivalent scores and used in two ways: 1. Descriptive character of functional and expressive language levels achieved by participants: Raw scores converted to language quotient scores 2. Analysis of language growth rates of children as function of age at initial stimulation: Examined within-child growth parameters of scores derived from both MCDI and PLS-3 PLS-3 provides language quotient (standard score, 100 represents mean for each child’s chronological age, 15 represents standard deviation). MCDI provides age equivalence scores the interpretation of which considers chronological age of child. Expressive language quotients (ELQs) obtained by dividing child’s age equivalent score by child’s chronological age at time of MCDI completion.</td>
<td>Implantation during infancy ultimately “speeds up” the preimplant, decision-making process. However, it is important to acknowledge that it is unclear at this point whether early expressive language advantage will be robust later in these children’s lives. Thus, when faced with the decision to implant an infant one must consequently continue to weigh the relative, expressive language benefit against the individual needs of each family and the challenges associated with the clinical management of very young children. Reviewer conclusions This study highlighted important issues in relation to the impact of early cochlear implantation for children as young as 12 months of age, although it should be noted the sample size was small.</td>
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<td>Tomblin 2005</td>
<td>To test for the impact of earlier implantation on expressive language growth rates</td>
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<td>Second-equating of MCDI and PLS-3 scores. Both measures used jointly to avoid multiple hypothesis testing as they measure same thing. Means and variances of each test calculated (using only observation where both tests administered) resulting in observations of the sum of the two. Follow-up interval – supposed to be before CI, on day of CI, then each 2m during y1, 6m during y2&amp;y3 after CI. Actual intervals from available data: (peri-implant n=27, at 12 months n=27, and at 24 months n=21 after implantation). Growth curve analysis method using hierarchical linear modelling (HLM): Provides means of estimating linear growth rate and intercept of each individual, describing average group values of parameters, and provides for tests of association between predictors and individual differences in these predictors. Models of expressive language growth: • Unconditional model (regardless of age at initial stimulation) • Conditional model (age at initial implantation included in the model along with parameters of intercept and slope)</td>
<td>Growth curve analysis results - Modelling results - HLM modelling for a model of expressive language growth conditioned on age at initial stimulation of the CI device- Significant association of age at initial stimulation with both intercept and linear growth rate. Children who were implanted at younger ages had higher log transformed combined language raw scores than those implanted at older ages. Plot of expressive language growth based on parameters derived from combined language scores- Children implanted at younger ages initially had lower language scores than those implanted at older ages. After 3 years of implant experience, the children implanted earlier in life reached higher levels of expressive language proficiency than those implanted later in life, despite being at least 3 years younger.</td>
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<td>Anderson 2004</td>
<td>Austria + Other European countries</td>
<td>Comparative, Prospective data analysis, Level III-2</td>
<td>Study setting 28 clinics worldwide involved in the MED-EL International Children’s study (575 children implanted with MED-EL cochlear implants)</td>
<td>Assessment tool: Taken from EARS battery test (evaluation of auditory responses to speech) a multi-language to track auditory development in children aged 1-18yrs. EARS test battery include (for under 2y): LIP and MTP tests as well as the MAIS &amp; MUSS questionnaires (for teachers/ parents).</td>
<td>When the scores from children implanted &lt;2y of age entered into a one-way repeated measure ANOVA, significant improvement over time was confirmed with every test (LIP, MTP) or questionnaire (MAIS, MUSS), with all p values &lt;0.001. Data analysis shows early implanted children improve quicker than those implanted later only with the test MTP (significant interaction between age group and improvement over time, p=0.012) and with the MUSS (p=0.036), but not with the LIP and the MAIS.</td>
<td>Limitations: Non-randomised or blinded study, Small sample size, Comparator groups not similar in sample, Tests complexity (LIP &amp; MTP) in linguistic and cognition made it difficult to perform for children implanted &lt;2y despite some children demonstrating some open-skills clinically as reported by their parents, Questionnaires used (MAIS &amp; MUSS) need at least 3 years for the under two group to catch up with and improve on the older implanted children, These questionnaires may not be the appropriate tool for assessing auditory behaviour skills and monitoring the development of these skills among very young children.</td>
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<td>Objectives: To report on outcomes in children implanted under the age of two years, compared to those implanted at a later age.</td>
<td>Participants n=90 children with CI, age at CI between 10 m to 6years</td>
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<td>Inclusion/exclusion criteria: Not clearly stated, but authors stated only data from children involved in The MED-EL International Children’s study who have CI implanted before the age of 2y will be reported.</td>
<td>Intervention n=37 children with CI, median age 20.4m (range 10m-23.9m). Follow-up interval: Prior to CI, and then 2days, 1, 3, 6, and 12 months after implantation then annually (2 and 3y after implantation)</td>
<td>Baseline characteristics intervention group: 24 males, 13 females Cause of HL unknown 30, meningitis 4, hereditary 2, Waardenburg’s syndrome 1. No other medical problem Comparator groups details not shown but authors stated the groups have similar aetiologies to the under 2 group.</td>
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<td>Anderson 2004</td>
<td>Austria + Other European countries continued</td>
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<td>Compared to CI&gt;2 they develop rapidly and by 18 months after CI they match those at later CI. Results from the two questionnaires: 1. MAIS scores CI between 2-4 mean 3.1 (n=36), CI between 4-6 mean 4.11 (n=25). Parents perceived a rapid improvement of their child’s auditory integration skills within first year of CI use. One and a half years after early CI children showed same level of auditory integration as later CI children. After 1.5-2y CI use better auditory listening skills in children implanted &lt;2y than later which was constant over time (p&lt;0.001) 2. MUSS scores (assesses speech production skills, takes longer time to show improvement) CI&lt;2y n=37, CI 2-4 n=16 mean age at CI 3.1y, CI 4-6 n=13 mean age at CI 4.9y Marked improvement in progress (2nd &amp; 3rd y of CI use) in all groups. The increase in performance after 12 months was the best among those implanted &lt;2 years. This is shown as a steeper increase in mean scores from children implanted &lt;2 years compared to those implanted at other later ages. Quick rates of improvement over time in interaction between age group and improvement over time among younger MUSS p=0.036</td>
<td>Reviewer comments: The study is liable for selection bias due to lack of randomisation, blinding and small sample size. Confounding and misclassification need to be considered.</td>
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</table>

THE EFFECTIVENESS OF EARLY COCHLEAR IMPLANTATION FOR INFANTS AND YOUNG CHILDREN WITH HEARING LOSS
Table 2. Evidence table of appraised articles relating to effectiveness of cochlear implantation at a young age compared to an older age (population with a mean age of less than 36 months and containing some aged less than 2 years of age at implantation) (continued)

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<tr>
<td>Holt 2004</td>
<td>Indianapolis USA</td>
<td>Prospective Comparative Level III-2</td>
<td>Study setting Indiana University Medical Centre</td>
<td>Assessment tool: A modified open-set test of spoken word recognition involving perception and understanding (Mr. Potato Head Task). Baseline characteristics Group 1 (n=5) age at CI 7-12m, mean=10.0 (87.5% use oral mode communication*) Group 2 (n=27), age CI 13-24m, mean =19.6, (73.7% use oral mode communication*) Group 3 (n=38), age CI 25-36m, mean =30.0, (59.9% use oral mode communication*) * no supplemental signing used</td>
<td>Average performance = Group mean percent correct on the tool as a function of age at testing for each experimental group. Authors stated that children implanted in their first year of life were not yet old enough at each testing interval to be compared to the norms for normal-hearing children. Also the pronounced increase in spoken word recognition following implantation was not observed for the group of children implanted in the first year of life. Statistical comparisons of early versus later CI implantation: • Children implanted in their second year of life showed better performance than those implanted in third year. • At 4years of age children implanted in their second year had average word recognition scores of 70% compared to those implanted at their third year (with scores of 52%).</td>
<td>Limitations • Non-randomised or blinded study • Very small sample for those implanted in their first year of life • Used single measure for speech perception as a measurement tool but no information on reliability and validity Some children were not tested at every interval due to missed appointments or fatigue (no information on how many children) Reported conclusions (by authors). The results from this single measure of spoken word recognition suggest that children implanted in the second year of life quickly develop an advantage of 10-20 percentage points over children implanted in the third year of life, and this advantage persists for more than 3 years. The present results do suggest that cochlear implantation in the first 2years of life may result in an improved developmental trajectory of speech perception skills.</td>
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<tr>
<td>Lesinski-Schiedat 2004</td>
<td>Comparative retrospective analysis (pre-and-post implantation)</td>
<td>Study setting: Cochlear implant centre in Hannover, medical check-ups at Medical University of Hannover’s Department of Otolaryngology. Study period - between August 1996 and May 2002.</td>
<td>Assessment tool: Closed-set and open-set methods. Closed-set method: Pattern-perception test (12 cards-items of 1-3 syllables as visual and auditory forms). Mono- and di-syllable testing performed using 12 items for each test, minimal pairs (child shown 2 different pictures = 40 items presented with live voice, differ in one vowel or consonant). Open-set method: Mono-syllable tests, TAPS (Test of Auditory Perception of Speech for children-German version); to evaluate cognitive ability (10 items) child has to repeat auditorily presented items consisting of words 1-4 syllables in length, and GASP test (Glendale Auditory Screening Procedure) a textual context to evaluate individual ability to abstract (10 questions presented by live voice to be answered by child).</td>
<td>General post-operative (surgical &amp; anaesthetics complications): - After the implantation the youngest child (0.4 years) required 24-hour intensive medical care due to lack of blood volume. Follow-up: - After 3m = Group 1 (n=20) vs. group2 (n=56). Very good response to noise 75% vs. 69%. Ability to identify different noises 59% vs. 48%. - After 12 m of CI use children implanted later showed better scores in TAPS and monosyllable tests (indicating better speech understanding). - After 18-24m of CI use better lexical understanding in early implanted children than those implanted later. Early implanted children vs. later implanted children: - achieved 50% in GAST vs. 30%. - achieved 66% in Common Phrase test vs. 53%. - better sentence understanding (67% in general sentence) vs. 20%.</td>
<td>Limitations: Retrospective design. No statistical analysis provided for the differences in baseline characters. Questionnaires used were referenced but no information on reliability or validity. Not all children had completed 24months of cochlear implant use by the end of the study. Number of respondents to questionnaires and hearing and speech tests varied over time thus affecting the results. Open-set test not applied during the first 6-12 months of rehabilitation but used different questionnaires (referenced but not described). 7 children in group 1 (26%) and 42 children from group 2 (47%) undergone rehabilitation for up to 2 years during the analysis of data. Analyses were only from less than 50% of both groups (8 children from group 1 and 44 children from group 2). Generalisability of the study result is questioned as the sample size is small and there were baseline differences. Reported conclusions (by authors). This study revealed that children implanted before the age of 1 year were subjected to no additional risks and showed superior development of speech understanding. Cochlear implantation should therefore be performed in very young children identified as suffering from profound bilateral hearing loss.</td>
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<tr>
<td>Manrique 2004(a)</td>
<td>Prospective cohort study Level III-2</td>
<td>Study setting. Tertiary referral centre</td>
<td>Children were evaluated before, and each year after the CI for up to 5 years</td>
<td>Children implanted &lt;2y- versus children implanted between 2-6y:</td>
<td>Limitations</td>
</tr>
<tr>
<td>Spain</td>
<td>Objectives To compare the auditory abilities &amp; speech perception of children with profound pre-lingual bilateral hearing impairment when subjected to CI before or after 2y of age. Inclusion/exclusion criteria. Not stated</td>
<td>Participants: n=130 children &lt;6y with profound pre-lingual bilateral hearing impairment who received CI before the age of 2y (n=36), or between 2-6y (n=94) Baseline characteristics Group 1 (n=36) 13M, 21F, age at implantation &lt;2y, range 0-1y, mean 0.9. Reason for hearing impairment: Unknown cause= 15, Genetic n=7, Postnatal infection n=3, pre-natal infection n=1, ototoxic drugs n=1, and other causes n=6 Group 2 (companson) (n=94), 53M, 41F, age of implantation 2-6y, mean 3.3y. Reason for hearing impairment: Unknown cause= 42, Genetic n=26, pre-natal infection n=2, ototoxic drugs n=1, and other causes n=10</td>
<td>Assessment tool Auditory testing-child completes audiologic evaluation according to pure-tone audiometry and 4 tests of speech perception (vowel confusion, series of daily words, bi-syllabic words, and Central Institute for the Deaf &quot;CID&quot; sentences). Speech testing- to test speech perception and production using Peabody Picture Vocabulary and the Reynell general oral expression scale Outcome measures - PTA (in dB), performance of children in test of vowel identification (%vowels), performance levels in test of series of daily words (%daily words), performance in &quot;bi-sylabic words&quot; %, performance of CID sentence test (%CID sentences), age performance in Peabody Language Development test versus chronologic age &quot;in years&quot;, and age performance on Reynell general oral expression scale versus chronologic age &quot;in years&quot;</td>
<td>Better results for CI implant &lt;2y in: %vowel (4th y post-implant), %daily words (1st &amp; 4th y post-implant), % bi-sylabic words closed-set test (3rd &amp; 5th post-implant y), and %CID sentences (3rd &amp; 5th post-implant y) Speech test results for CI implant before the age of 2y vs. older children: Peabody Language Development test followed the normal population pattern compared to a 2-year lag for those implanted between 2 and 6y of age. Reynell general oral expression scale showed a 1-y lag with normal population compared to 3-year lag for those implanted at 2-6 years of age.</td>
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<tr>
<td></td>
<td>Level III-2</td>
<td>Tertiary referral centre</td>
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<td>Reported conclusions (by authors). When performed before 2 years of age, CI offers a quicker and better improvement of performance without augmenting the complications associated with such an intervention.</td>
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<tr>
<td>Manrique 2004(b) Spain</td>
<td>Prospective cohort study Level III-2 Objectives • Present long-term auditory results of children with pre-lingual profound deafness who received CI • Analyse role of auditory stimulation in development of communicating abilities in early implanted children • Define limits of auditory critical period.</td>
<td>Study setting. Tertiary referral centre Participants: n=182 children with congenital HL or pre-linguistic deafness (occurred before 2y of age) who consecutively received CI</td>
<td>The same as Manrique 2004(a) but with longer period of follow-up Follow-up interval 8-years</td>
<td>Regardless of the follow-up time, the auditory and speech perception tests improved significantly in all children after cochlear implant. Better performance with earlier implantation. Speech tests showed the development of children implanted before 2y of age similar to normal children.</td>
<td>As for Manrique 2004(a) Reported conclusions (by authors), When performed before 2 years of age, CI offers a quicker and better improvement of performance without augmenting the complications associated with such an intervention.</td>
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ABR= auditory brainstem response,
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<td>Manrique 2004**</td>
<td>Inclusion/exclusion criteria.</td>
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<tr>
<td>Spain</td>
<td>• Child must have prelinguistic profound bilateral hearing impairment (PTA and ABR thresholds &gt;91 dB HL in both ears)</td>
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<td>Continued</td>
<td>• No associated handicapping condition</td>
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<td></td>
<td>• Absence of medical contraindication for surgery</td>
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ABR= auditory brainstem response,
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<tr>
<td>McConkey-Robbins 2004</td>
<td>Comparative before-and-after study + retrospective comparison with previously published data Level III-3 Objectives Investigate effect of age of CI on auditory development of children who underwent CI before age 3 years.</td>
<td>Study setting Clinical centres involved in clinical trials for CLARION Multi-Strategy CI in North America Participants: Intervention group n=107 hearing-impaired children (age range 12-36m) who had received the Multi-Strategy CI during clinical trials CI = 3 groups according to age at implantation Baseline characteristics Group 1 (n=45), age at CI 12-18m Group 2 (n=32), age at CI 19-23m. Group 3 (n=30), age at CI 24-36m Comparator [n=109] (previously published data from NH cohorts), children with NH aged 5-36 months (from parents) Baseline characteristics Aged 5-36months (mean age 12.5m), 52%&lt;12m, 23% aged 12-18m, 10% aged 19-23m, 15% aged 24-36m. Analyses comparing groups at baseline. NS but included only those with the implant. Baseline characteristics included age at implant (months), mean pre-implant pure-tone average (dB HL) in both implant ear and non-implant ear.</td>
<td>Compared mean IT-MAIS scores over time for different groups (age at implantation) over time after implantation. Also compared to children with NH Assessment tools Infant-Toddler Meaningful Auditory Integration Scale (IT-MAIS) defined as a structured parent interview tool developed to yield quantitative results in children as young as newborns. For comparison with NH subjects Hebrew translation version of the IT-MAIS (n=68) or Arabic (n=41) Outcome measures (IT-MAIS) scores before and after CI Follow-up interval (12-months) before and 3 &amp; 6 months after implantation also after 12-months</td>
<td>Comparison of IT-MAIS scores between the 3 different age at CI groups (IT-MAIS) scores near zero before implantation Post-implant (showed rapid improvement over time): • All groups showed better scores post-CI. • Both groups implanted before 2 years of age showed better scores compared to those implanted between 24-36 months. • No significant difference in mean scores over time between the two youngest groups at implantation. Compared to NH children The youngest children (CI &lt;12-18m) showed 70% of the variance in data attributable to implant use, and more than two thirds of the children fell within the normal distribution of IT-MAIS scores after 6 and 12 months of CI experience. Those implanted at 19-23m showed 65% of variance attributable to CI use, and one third of the children fell within the normal distribution of the IT-MAIS scores 6 and 12 months after CI use. The later implanted age group IT-MAIS scores showed great variability in data not predicted by duration of CI use, almost all the scores in this group continued to fall below the NH range after 6 and 12 months of CI use.</td>
<td>Limitations • Data compared to that obtained from parents of NH subjects retrospectively • Tools used non-English versions (either Hebrew or Arabic) • 12-month data omitted from first analysis because of small sample size based on parent report. Reported conclusions [by authors]. Children younger than age 3 years undergoing implantation demonstrated impressive auditory skill development during the first year of device use. This data suggest that performing implantation in children with profound hearing loss at the youngest age possible allows the best opportunity for them to acquire communication skills that approximate those of their NH peers.</td>
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</table>

NH= Normal Hearing, NA= North America, NS = not stated, HL= Hearing Loss, CI= cochlear implant, F=female, M=male, oral mode= use exclusively spoken language, total mode= combination of spoken language with Signed Exact English, IT-MAIS = Infant-Toddler Meaningful Auditory Integration Scale
Table 2. Evidence table of appraised articles relating to effectiveness of cochlear implantation at a young age compared to an older age (population with a mean age of less than 36 months and containing some aged less than 2 years of age at implantation) (continued)

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<td>Svirsky 2004</td>
<td>Indianapolis, USA</td>
<td>Prospective comparative, Level III-2</td>
<td>Study setting</td>
<td>Subjects tested 2-8 times as follows:</td>
<td>In general results indicated implantation in second year of life results in better speech perception &amp; language development outcomes than later implantation.</td>
<td>Limitations: More than 90% of children who qualified for research protocol participated. Few children changed the mode of communication during course of study. Referred to using both scores as being normed on children with NH and also been applied to users of either OC or TC.</td>
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<td>Participants: N=75 congenitally deaf children divided into 3 groups based on age at CI</td>
<td>1st session 1-3months before initial CI activation.</td>
<td>On average, Language proficiency levels for those implanted at 12-24m close to 1 SD below NH means versus 2 SD below NH means for those implanted at 25-36m.</td>
<td>* RDLS-III = Reynell Developmental Language Scales, MCDI = MacArthur Communicative Development Inventories</td>
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<td>To investigate the effect of age at CI on speech perception and language skills of congenitally deaf children using developmental trajectory analysis (DTA). To compare language skills shown by profoundly deaf children with CI to those of age-matched children</td>
<td>Study setting</td>
<td>After 2nd test, testing interval becomes 6months</td>
<td>DTA results indicated an average estimated language age for children in the first group 5.7m higher than for children in second group at same chronological age (P&lt;0.01).</td>
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<td>Speech-language pathologist sees children 2-3 times per week.</td>
<td>Keep contact with schools and recommendations made to ensure appropriate rehabilitation for each child.</td>
<td>CI at 25-36m had advantage of 15.8% over CI at 37-48m (P&lt;0.001)</td>
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<td>Outcome measures</td>
<td>In general results indicated implantation in second year of life results in better speech perception &amp; language development outcomes than later implantation.</td>
<td>For speech perception DTA shows average advantage for CI 12-24m over CI at 25-36m by 12.2% and at 26.5% over CI at 37-48m (P&lt;0.001).</td>
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<td>Underlying language abilities independently of ability of child to understand spoken language or to produce intelligible speech. Measures used:</td>
<td>CI at 37-48m showed children lagged behind CI at 25-36m by 5.6m (P&lt;0.05), by 10m behind CI at 12-24m (P&lt;0.001)</td>
<td>CI at 25-36m had advantage of 15.8% over CI at 37-48m (P&lt;0.001).</td>
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<td>Expressive &amp; receptive language assessed using the RDLS-III (expressive language scores only). Scores expressed as age-equivalent scores.</td>
<td>Spoken word recognition was assessed using a modified open set test (Mr. Potato Head task).</td>
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<td>Early language development assessed using the MCDI (based on parent report). Provides 2 levels of complexity and administered according to age of child</td>
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<td>* All children attended schools that promoted development of auditory/oral skills with or without using signs. Some children used oral communication (OC), others used total communication (TC) = oral English + signs.</td>
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<td>Inclusion/exclusion criteria. Monolingual English-speaking children, implanted &lt;5y, used SPEAK/ACE or CIS strategies since initial device fitting, and who had no other handicapping conditions (mental retardation or speech motor problems)</td>
<td>RDLS-III =Reynell Developmental Language Scales, MCDI=MacArthur Communicative Development Inventories</td>
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RDLS-III =Reynell Developmental Language Scales, MCDI=MacArthur Communicative Development Inventories
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| Govaerts 2002 | Belgium | 1. Retrospective longitudinal (evidence Level III-3)  
Objectives To assess the relationship between cochlear implantation in young children and age at implantation.  
Inclusion/exclusion criteria.  
Inclusions were all congenitally deaf children who:  
• Underwent CI between Jan 94 & Aug 99.  
• Aged 1-6y at time of CI, with follow-up of 2 years.  
• Aged <1y at CI irrespective of follow-up time.  
Exclusions were all children with:  
• Severe mental retardation or cochlear malformations | Study setting: St Augustinus Hospital  
Participants: Intervention group n=48 congenitally deaf children received CI, (15 child received CI before 24m). Participants received CI:  
• At median age 8m (n=6) [age range 5-10m],  
• At median age 19m (n=9) [age range 13-23m]  
• At median age 30m (n=7) [age range 25-35m]  
• At median age 40m (n=13) [age range 37-47m]  
• At median age 56m (n=7) [age range 50-60m]  
• At median age 70m (n=6) [age range 63-71m]  
CI = multichannel bipolar LAURA CI Comparator group (n=113) children with NH aged 11-32 months, four groups.  
• Age group 12m (n=26), mean CAP score 2  
• Age group 18m (n=28), mean CAP score 5  
• Age group 24m (n=36), mean CAP score 6  
• Age group 30m (n=23), mean CAP score 7 | Questionnaire administered to parents and professional therapist monitoring the child.  
CAP score determined before CI and 3, 6, 12 and 24 months after CI.  
Scores calculated according to responses by parents and professional therapist to questionnaires.  
Outcome measures  
• Auditory performance in the form of Categories of auditory performance scores (CAP)  
• Integration into mainstream school system  
Follow-up: 2 years  
CAP scores:  
• 0 = no awareness of environmental sound  
• 1 = awareness of environmental sounds  
• 2 = responds to speech sounds  
• 3 = recognises environmental sounds  
• 4 = discriminates at least 2 speech sounds  
• 5 = understands common phrases without lip-reading  
• 6 = understand conversation without lip-reading with a familiar talker  
• 7 = can use telephone with familiar talker | Results from the retrospective design:  
• All children achieved higher CAP scores after CI  
• Implantation between the age of 2 and 4y of age always resulted in normal CAP score after 3 years.  
• 67% of children implanted before the age of 2y integrated in the mainstream school system. | Limitations  
• Retrospective design  
• Small sample size  
• Non randomised, not blind  
Reported conclusions (by authors), All children with congenital deafness who underwent implantation before the age of 6 years appeared to benefit from the implant. However, these data add evidence to the importance of early implantation (before the age of 2 years). Intervention before the age of 4 years seemed to be critical to achieve optimal results. |
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<td>Govaerts 2002</td>
<td>2, cross-sectional (evidence Level III-3)</td>
<td>Baseline characteristics included median age at implantation and CAP scores</td>
<td></td>
<td>Outcome measures:</td>
<td>Limitations: Cross-sectional design, so change overtime is not assessed</td>
</tr>
<tr>
<td>Belgium</td>
<td>Objectives: To assess the relationship between cochlear implantation in young children and age at implantation</td>
<td>Study setting: St Augustinus Hospital</td>
<td></td>
<td>Percentage of children from each age group that reaches normal CAP scores when compared with NH children of the same age</td>
<td>Small sample size, non randomised, not blind</td>
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<td></td>
<td>Inclusion/exclusion criteria.</td>
<td>Participants: Intervention group n=70 congenitally deaf children received CI (aged 12-71 mths at CI), (21 children received CI before 24m). Participants grouped into 7 groups according to age at CI:</td>
<td></td>
<td>Time taken for 50%, 75% and 90% of children with CI to reach normal CAP scores.</td>
<td>Reported conclusions (by authors). All children with congenital deafness who underwent implantation before the age of 6 years appeared to benefit from the implant. However, these data add evidence to the importance of early implantation (before the age of 2 years). Intervention before the age of 4 years seemed to be critical to achieve optimal results.</td>
</tr>
<tr>
<td></td>
<td>Inclusions were all congenitally deaf children:</td>
<td></td>
<td></td>
<td>Results from evaluating auditory performance using Categories of Auditory Performance (CAP) score:</td>
<td>Reviewer comments. Study is susceptible to confounding due to the lack of a randomised design.</td>
</tr>
<tr>
<td></td>
<td>• Underwent CI between Jan 94 &amp; Aug 99,</td>
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<td></td>
<td>• 3 months after CI all children implanted &lt;2y of age obtained normal CAP scores</td>
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<tr>
<td></td>
<td>• Aged 9m-6y at time of CI</td>
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<td></td>
<td>• More than 25% of those implanted after 3y of age failed to reach normal CAP scores within 48m after CI</td>
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<tr>
<td></td>
<td>Exclusions were all children with:</td>
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<td>• Children implanted after the age of 2y took longer than 3m to achieve CAP scores similar to their normal peer</td>
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<tr>
<td></td>
<td>• Severe mental retardation or</td>
<td></td>
<td></td>
<td>• It takes a median time of 3 months before 90% of children implanted before the age of 2y to reach normal CAP scores</td>
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<tr>
<td></td>
<td>• with cochlear malformations</td>
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Table 2. Evidence table of appraised articles relating to effectiveness of cochlear implantation at a young age compared to an older age (population with a mean age of less than 36 months and containing some aged less than 2 years of age at implantation) (continued)

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</tr>
</thead>
</table>
| Govaerts 2002 | Belgium | Comparator group (n=113) children with NH aged 11-32 months, four groups. | - Age group 12m (n=26), mean CAP score 2  
- Age group 18m (n=28), mean CAP score 5  
- Age group 24m (n=36), mean CAP score 6  
- Age group 30m (n=23), mean CAP score 7 | | | |
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<tr>
<td>Hammes 2002 USA</td>
<td>Retrospective longitudinal data analysis and additional comparative data Level III-3 Objectives To present spoken language data for infants who underwent implantation at 18 months of age or less. To compare performance of children with at least 6 months of CI use to children who underwent CI at older ages.</td>
<td>Study setting Carle Clinic and Foundation Hospital Participants (n=47): Children underwent CI at the above settings, age range 9-48m at CI, assigned to four age groups according to time at CI.</td>
<td>Spoken language measures: • Skills for Hearing Impaired Children (SK*HI) (designed for children with HL based on parent report) • Preschool Language Scales-3 (PLS-3) (designed, normalised on NH children) • Clinical Evaluation of Language Fundamentals P or III (CELF-P or CELF-III) (designed, normalised on NH children) • Oral and Written Language Scales (OWLS) (designed, normalised on NH children) For open set word recognition abilities a comparison measure used the Phonetically Balanced-Kindergarten (PBK) word lists Outcome measures For comparative data: • Open-set word recognition (abilities), • Communication mode and • Spoken language achievement levels. Speech perception scores for the best performance demonstrated by each child reviewed. Spoken language abilities analysed in relation to outcomes at the most recent test interval. Follow-up Retrospective for 84months</td>
<td>Longitudinal data for spoken language outcomes (SK*HI): • Similar language-learning rates from both NH children and the majority of infants with HL Comparative data • (PBK): Children who underwent CI at ≤18m showed that infant group achieved a word score average of close to 80% correct compared to 60% for those implanted between 19-30m or 31-40m, and to 40% of those implanted between 41-48m • Communication: Spoken language-Children implanted before the age of 30m were more likely to have spoken language skills within 12m of their chronological age than children implanted after that age • Children with CI ≤30m showed better open-set word recognition abilities compared to CI &gt;30m • Children implanted at or before the age of 18m were most likely to rely solely on spoken language for communication</td>
<td>Limitations • Retrospective • Very small sample size • non randomised, not blind Reported conclusions (by authors), Early implantation is desirable. Children who undergo implantation as infants may develop language skills commensurate with those of their hearing peers. Reviewer comments Study susceptible to confounding due to lack of a randomised design and blinding.</td>
</tr>
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<tr>
<td>Hammes 2002 USA</td>
<td>Significant cognitive delays with hearing loss, progressive hearing loss, or stopped using device</td>
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<tr>
<td>Szagun 2001</td>
<td>Germany</td>
<td>Prospective Longitudinal comparative study Level III-2</td>
<td>Objectives To assess the relationship between age at cochlear implantation, pre-operative hearing and subsequent linguistic development (particularly progress in grammar and vocabulary acquisition). Inclusion/exclusion criteria. Intervention group: Only children with CI: ( &lt;4 \text{y} ) at CI ( &lt;24 \text{m} ) of age, 14 children at ( &lt;36 \text{m} ), and 3 children implanted at ( \geq 36 \text{m} ) of age. Baseline characteristics of intervention group: • 12 girls, 10 boys, age at CI=14-46m, mean 29m ±SD 9m • All attend CI Centre for aural rehabilitation program (hearing + speech + language education). Comparator n=22 NH children (12 girls, 10 boys) aged 16m at time of data collection, age-appropriate levels of cognitive development as measured by object permanence tests of Infant Psychological Developmental Scales</td>
<td>Study setting Intervention group Hannover Medical School (20 children) and Freiburg University Teaching Hospital (2 children) Control group-3 paediatricians’ practices + 2 day care centres (Oldenburg, northern Germany) Participants: n= 44 German speaking children (24 girls, 20 boys) Intervention: n= 22 deaf children with CI, (5 children had their CI implanted ( &lt;24 \text{m} ) of age, 14 children at ( &lt;36 \text{m} ), and 3 children implanted at ( \geq 36 \text{m} ) of age) Baseline characteristics of intervention group: • 12 girls, 10 boys, age at CI=14-46m, mean 29m ±SD 9m • All attend CI Centre for aural rehabilitation program (hearing + speech + language education). Comparator n=22 NH children (12 girls, 10 boys) aged 16m at time of data collection, age-appropriate levels of cognitive development as measured by object permanence tests of Infant Psychological Developmental Scales</td>
<td>Data collection took place in playroom at the Cochlear Implant Centre for CI children, and in playroom at the University of Oldenburg for NH children. Pre-operative audiometric assessment First 2y post-implantation • At the centre, 5-day rehabilitation every 10wks (1-2 language training sessions of 45-min long) • At home one session per week NH children &amp; CI children matched for initial language level (measured by MLU) &amp; number of words. Everything spoken by child transcribed using CHILDES system for transcribing &amp; analysing child speech. Outcome measures Mean length of utterance (MLU) for grammatical progress. Number of words (vocabulary) Follow-up interval 27-36 months Used correlation analyses (Pearson product moment correlation coefficients). Linguistic progress measured by MLU in terms of total number of words a child produced (as indicated by parent questionnaire).</td>
<td>Grammatical development: CI children-MLU ranged 1-1.23 (mean 1.04 SD 0.06), NH children-MLU ranged 1-1.2 SD 0.08 CI children-vocabulary assessed by parent report 0-72 words mean 20.9 SD 20.8, NH children- vocabulary parent report 0-88 words mean 17.5 SD 19.4 Relation between implantation age, pre-operative hearing and linguistic growth: Partial correlation calculated between language growth measures and chronological age at implantation. Age at implantation correlated significantly with linear growth in MLU at three data points, but less strongly, accounting for 25% of the variability.</td>
</tr>
<tr>
<td>Limitations and Conclusions</td>
<td>Limitations</td>
<td>Non randomised study</td>
<td>Very small sample size</td>
<td>Acquired deafness in 3/5 children implanted ( &lt;2 \text{y} ) (meningitis), lost hearing at 1, 8, and 18m (all considered pre-lingually deaf)</td>
<td>Child who lost hearing at 18m had limited verbal skills</td>
<td>Children are German speaking and the outcomes were mainly about language acquisition including progress in grammar and vocabulary acquisition (which may not be applied to English language)</td>
</tr>
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### Table 2. Evidence table of appraised articles relating to effectiveness of cochlear implantation at a young age compared to an older age (population with a mean age of less than 36 months and containing some aged less than 2 years of age at implantation) (continued)

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<tr>
<td>Szagun 2001</td>
<td>Germany</td>
<td>Inclusion/exclusion criteria. Control group were NH children: • With no diagnosed developmental delays or hearing problems • Monolingual environment</td>
<td></td>
<td></td>
<td></td>
<td>The use of an observational design did not allow for control of confounding. The lack of blinding and selection of the two different groups from different settings result in the study being more liable for bias.</td>
</tr>
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<tr>
<td>Kirk 2000</td>
<td>USA</td>
<td>Longitudinal (prospective) Level III-2</td>
<td>Study setting Indiana University School of Medicine Participants: pre-lingually deaf children with CI, n=106, divided into 2 groups according to method of communication and 3 subgroups according to age at CI • Oral communication (OC) n=56, age at CI &lt;2y n=8, CI 2-4y n=37, and CIB5y n=11 • Total communication (TC) n=50, age at CI &lt;2y n=6, CI 2-4y n=33, and CIB5y n=11 Analyses comparing groups at baseline Variables within each group included: • Mean age at onset of deafness, • Mean age at fit with CI, • Mean unaided pure-tone average Also included number of participants for each device type used for the 3 groups of age at CI irrespective of type of communication.</td>
<td>Battery of speech and language tests performed on children before and every 6 months after CI Measuring tools: Spoken word recognition tests 2 measures • Closed-set using the Grammatical Analysis of Elicited Language-Pre-Sentence Level (GAEL-P), test fist familiarise child with 30 objects in auditory-plus-visual modality, then child must identify target word through listening only • Open-set using Mr Potato Head Task, child asked to assemble toy in response to 10 verbal commands presented in auditory-only modality Test instructions administered according to child’s preferred modality, child with CC = audition + speech reading used &amp; child with TC= simultaneous signed + spoken English used Outcome measures Average rate of improvement in GAEL-P scores as function of age at implantation indicated - No interaction between age at implantation and length of device use No significant relationship between age at implantation and rate of improvement in close-set word recognition skills Performance on open-set word recognition test (Mr Potato Head) indicated – There was no interaction between age at implantation and length of device use Rate of improvement in open-set word recognition abilities did not differ among the three age-at-implantation groups There was a significant effect of communication mode on open-set spoken word recognition performance (p &lt;.01) Children communicating orally showed significantly higher open-set recognition scores than those who use total communication modality</td>
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<td></td>
</tr>
<tr>
<td>Limitations and Conclusions</td>
<td>Limitations</td>
<td>• non randomised, not blind • length of CI use varied across children • incomplete tests conducted due to time constraints on family, child’s attention span • incomplete testing at every interval of interest • number of data points varied across tests and children • small numbers in &lt; 2 year group</td>
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</table>

Reported conclusions (by authors). Age effects on the development of receptive and expressive language differed from those on spoken word recognition. Earlier implanted children demonstrated superior language abilities. Children implanted prior to 2 years of age had significantly faster rates of receptive vocabulary and language development than later-implanted children. Furthermore, children implanted prior to age 2 had superior expressive language abilities compared with those implanted after that age. There were no significant differences in rates of language development between the oral and total communication groups. |
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<td>Kirk 2000</td>
<td>USA</td>
<td>Excluded children were • Using previous-generation speech processors or • Previously used earlier versions and then upgraded to current CI speech processors/strategies.</td>
<td>Measuring tools: Receptive and expressive language measures: • Measure of receptive vocabulary using the Peabody Picture Vocabulary Test-Third Edition (PPVT-III) • Measure of receptive + expressive language using the Reynell Developmental Language Scales (RDSL). Instructions &amp; tests administered in child’s preferred modality. Outcome measures All language measures include test battery with raw scores converted to age equivalency using normative data from children with NH. Age-equivalency scores converted to language quotients (&lt;1.0 indicates language age less than chronological age, &gt;1.0 = language age &gt; chronological age) Follow-up interval (6 monthly for at least 3 years). Data analysis Subjects as random effect with the following as covariates: • Length of device use, • age at CI, • communication mode</td>
<td>Growth in receptive vocabulary knowledge over time as a function of age at implantation for the two communication groups – • Language quotient scores increased significantly with increased length of device use (p&lt;.0001) • Significant effect of age at CI on rate of growth in receptive vocabulary; • Significantly faster growth rates in PPVT-III language quotient scores in children implanted before the age of 2y compared to those implanted between 2-4y (p&lt;.0001) • Growth in RDSL receptive language quotient scores over time as a function of age at implantation for the two communication groups – • No significant effect of age at implantation or communication mode on receptive language skills • A significant interaction between age at CI and length of CI use (p&lt;.0001) Rate of improvement in RDSL receptive language quotients over time increased significantly faster in children implanted before the age of 2y compared to those implanted between 2-4y (p&lt;.0001)</td>
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</tr>
</thead>
</table>
| Kirk 2000 USA continued | Rate of improvement in RDLS expressive language quotients as function of age at CI (2 communication groups)  
- Significant improvement of expressive language abilities with increased use of CI ($p<$0.0001)  
- Age at CI significantly affected development of expressive language abilities ($p<$0.01)  
- Children implanted at an age <2y had significantly higher RDLS expressive language quotients than children implanted between 2-4y ($p$<0.01). | | | | |
SUMMARY AND CONCLUSIONS

Summary of designs and levels of evidence

The search yielded a total of 946 articles. From 155 articles identified from abstracts and titles as being potentially eligible for inclusion, a final group of 16 were selected for appraisal, all of which were primary research.

All 15 papers were cohort (13) or cross sectional studies (3) as one study used two designs Govaerts et al. (2002) and all studies were graded III-2 or III-3 as per NHMRC. The best possible design to determine the effectiveness of cochlear implantation at a young age compared to an older age would be a randomised controlled trial which would assign children to have the cochlear implantation at either a young age or an older age. However, this design is likely to be unacceptable to parents and clinicians as there is already evidence that younger age at implantation is more effective than at an older age, just not what the age limit of this effect is.

Key results

While the tests used to measure outcomes were mixed there was consensus that, in general, implantation at a younger age improves the effectiveness of cochlear implantation in terms of communication outcomes. Also those children achieved stronger skills after sustained implant use than did children who received the implants more recently. Studies by Manrique et al. (2004a; 2004b), Svirsky et al. (2004) and Tomblin et al. (2005) showed stronger receptive and expressive vocabulary scores and stronger language development when children were implanted early in life.

These children develop auditory skills at an earlier chronological age (Anderson et al. 2004). This allows for quicker language development, which would then affect reading, writing and other educational skills. Auditory perception was better in children implanted before the age of 2 years than those implanted later (Govaerts et al. 2002; Manrique et al. 2004a). The latter study also showed better integration into the mainstream system with the decrease of age at implantation. Level of speech perception and production was also better in children implanted before the age of 2 years (Manrique et al. 2004a; Manrique et al. 2004b; Svirsky et al. 2004). Speech recognition was better in children implanted in their second year of life than children implanted later (Holt et al. 2004). The rate of growth in word recognition and language skills was affected by the age at which a child received cochlear implantation (Kirk et al. 2000). Significant improvement in spoken word recognition and receptive and expressive language skills were shown with children implanted before the age of 2 years. However all studies were level III2/3 as per the NHMRC.

Three studies compared implantation prior to the age of 18 months to implantation at an older age and showed similar results (Hammes et al. 2002; McConkey Robbins et al. 2004; Miyamoto et al. 2005).

A number of studies compared implantation prior to the age of 12 months to implantation at an older age. For example Holt et al. (2004) found no advantage for five children implanted at 6 to 12 months of age compared with those implanted at 12 to 24 months in terms of speech perception results. Three other studies were documented as including children under 12 months of age. Lesinski-Schiedat (2004) included 27 children aged under 12 months at implantation. In general, these children had poorer short-term outcomes but improved performance more than 24 months after implantation when compared with children implanted at a later age. Miyamoto et al. (2005) included eight patients who were implanted at under 12 months of age. There was some suggestion of improved outcome with early implantation. Finally, Govaerts et al. (2002) included six patients aged between five and 10 months at implantation. However, the results presented focused on two years as the cut off of interest. Based on the above findings, there was no clear evidence that implantation of a cochlear implant prior to the age of 12 months improves effectiveness when compared to implantation after 12 months of age.

Because of the short length of time that implantation has been used in large numbers of infants and young children, evidence of an increase in effectiveness is only available for immediate outcomes such as communication skills and has only been observed up to about 5-8 years after implantation.
Differences in long-term effects such as educational achievement and quality of life will take many years to become evident, if they exist, or if the effects are large enough to be detected.

Children implanted at an early age showed improved expressive and receptive language capabilities and also similar rates of speech development and language skills as their normal hearing peers (Govaerts et al. 2002; Hammes et al. 2002).

Other important outcomes are the rate of language acquisition as it is possible that those implanted at an older age develop at a slower rate but eventually reach equivalent developmental milestones.

**Implications for practice**

Whilst the current evidence base is limited, several implications for practice have been suggested by the appraised systematic studies. Implantation prior to the age of 18 months may increase the effectiveness of cochlear implantation in terms of immediate outcomes such as communication skills. This should be one of the considerations when weighing up the harms and benefits associated with cochlear implantation in this age group.

When considering cochlear implantation before the age of 12 months, the harms and benefits of cochlear implantation at this age, other than effectiveness compared to an older age, will need to be weighed up. Outcomes such as surgical outcomes/complications were not included in the protocol for this technical brief but would be one of the considerations when weighing up harms and benefits of implanting children younger than 12 months of age.

Other issues to be considered are understanding that while a cochlear implant can dramatically aid speech and language development for deaf children, using the device alone does not guarantee spoken communication. In considering how cochlear implants affect communicative development, it is helpful to distinguish between speech perception (understanding sounds), speech production (using sounds to convey messages), and receptive and expressive language (understanding and using words for communication).

**Methodological issues and recommendation for future research**

All of the studies included in this review were cross sectional surveys or cohort studies with relatively small sample sizes (range between 26-216 participants). This is because of the low incidence of severe to profound hearing loss and the small number of children in any centre at any given time being fitted with a cochlear implant (Geers 2006). Because of this, often the study populations were heterogeneous (or details were lacking) in terms of the degree of hearing loss, congenital versus acquired loss, mode of communication, socio-economic status or educational status of parents. Where studies were conducted over a long time period children were also fitted with different models of cochlear implant which may vary in effectiveness. These factors will increase the possibility that confounding or bias has influenced some of the study outcomes. For instance children from higher socio-economic groups are likely to be detected earlier, be implanted earlier, and have greater access to rehabilitation programmes. They will also have parents of higher educational levels. In an ideal situation it would be important for study groups to be comparable in terms of confounding factors such as socio-economic status but this may not be possible in many countries/centres.

While the best possible design to determine the effectiveness of cochlear implantation at a young age compared to an older age would be a randomised controlled trial this design is likely to be unacceptable to parents and clinicians as there is already evidence that younger age at implantation is more effective than at an older age, just not what the age limit of this effect is. Also, theories around sensory deprivation, and sensitive and critical periods indicate that younger age is likely to be more effective as it reduces the period of sensory deprivation.

Further well-designed observational studies with large sample sizes and long-term follow-up are required, especially in younger age groups. It is important to disentangle the effects of age at implant and length of use of implant.
**Issues raised by the included studies**

In general, results of studies on the effect of the duration of implant use or corresponding age-of-implantation effects may be confounded by the fact that those children who are getting a CI at the youngest age may also have the advantage of earlier diagnosis, earlier hearing aid intervention, and earlier educational intervention (Nicholas and Geers 2006).

Nicholas and Geers (2006) argued that the time interval between the onset of deafness and receipt of a cochlear implant may play a critical role in achieving the optimum success of cochlear implantation.

Advantages of early implantation include improved auditory experience and spoken language during a critical developmental period (Svirsky et al. 2004). Early implantation would provide an earlier start in language learning as well as capitalising on brain systems that are better suited for language learning during infancy (Tomblin et al. 2005).

Tomblin et al. (2005) highlighted the need to characterise the growth trajectory or rate. Previous studies typically have not distinguished between the benefits of early implantation that come from simply “starting early,” versus those benefits that come from faster growth rates. Only one study (Svirsky et al. 2000) to date has formally examined both the paediatric CI users’ mean differences in language test scores and their language growth over time, but this particular study did not address age at initial stimulation as a factor in these children’s growth; thus, cochlear implantation’s long-term benefit on language development in infants implanted before the age of 36 months remains unclear.

For children implanted before the age of 5 years language growth rates were similar to growth rates in hearing children once the deaf child received an implant. Differences between children with implants and their hearing-age counterparts in terms of their language performance may be due primarily to the existing delay in performance at the time of implantation (Nicholas and Geers 2006). They concluded that implantation should occur before delays are present and not only early enough for normal language progress to be achieved (Nicholas and Geers 2006).

**Other issues not specifically raised by included studies**

Candidacy issues were raised by Edwards et al. (2006) as they may play a role in predicting the degree of benefit from a cochlear implant in childhood. The paper referred to the wide spectrum of outcomes in terms of speech and language development cited in the literature, and on the difficulty in accurately predicting the degree of benefit in any individual case. They referred to the criteria for selecting appropriate paediatric cochlear implant candidates, which seems to be broadened to include audiological criteria, age of the child and the presence of additional disabilities. They also highlighted that outcomes of cochlear implants may be affected by type of school placement, signed versus oral language, duration of implant use, family size, and gender. However, Edwards et al. stated that age at implantation was the most robust recent finding in the literature to be of critical importance in determining outcome in congenitally and pre-lingually deaf children.

**Conclusions**

The purpose of this review was to determine whether early cochlear implantation in young children and infants is better than later implantation. This technical brief identified 16 eligible papers investigating the effectiveness of cochlear implantation in young children and infants compared to an older age group. These papers were all cross-sectional surveys or cohort studies. There was a consistent finding that implantation in a younger age group (before 18 months of age) was more effective in terms of than implantation in an older age group. However, the evidence was less certain among children receiving implants before 12 months of age.

Studies to date have examined only a small number of children in this age group which challenges generalising the findings across the infant cochlear implant population. Other studies have used retrospective designs, which have resulted in inconsistent and incomplete language measures.
REFERENCES


APPENDIX 1: LEVELS OF EVIDENCE*

Level I  Evidence obtained from a systematic review (or meta-analysis) of relevant randomised controlled trials.

Level II  Evidence obtained from at least one randomised controlled trial.

Level III.  1 Evidence obtained from pseudorandomised controlled trials (alternate allocation or some other method).

2 Evidence obtained from comparative studies (including systematic reviews of such studies) with concurrent controls and allocation not randomised, cohort studies, case control studies or interrupted time series with a control group).

3 Evidence obtained from comparative studies with historical control, two or more single-arm studies or interrupted time series without a parallel control group.

Level IV  Evidence obtained from case series, either post-test or pretest/post-test.

* From NMHRC 2000
APPENDIX 2: SEARCH STRATEGY

SEARCH STRATEGIES

**Medline**

1. cochlear implants/ (4049)
2. cochlear implantation/ (1495)
3. 1 or 2 (5142)
4. treatment outcome/ (292366)
5. child language/ (2109)
6. speech perception/ (11208)
7. quality of life/ (59499)
8. language development/ (5745)
9. vocabulary/ (4375)
10. verbal behavior/ (11697)
11. auditory perception/ (15555)
12. postoperative complications/ (221123)
13. voice quality/ (2687)
14. auditory threshold/ (11346)
15. or/4-14 (596436)
16. (preschool or pre-school).tw. (11432)
17. months.ti. (9219)
18. infant$.tw. (220072)
19. (12 months or 24 months or 36 months).tw. (82208)
20. (1 year or 2 years or 3 years).tw. (162455)
21. (one year or two years or three years).tw. (85937)
22. (twelve months or twenty four months or thirty six months).tw. (2836)
23. time factors/ (757519)
24. age factors/ (288167)
25. *child, preschool/ (570)
26. young child$.tw. (21374)
27. (pediatr$ or paediatr$).tw. (121742)
28. or/16-27 (1572467)
29. 3 and 15 and 28 (745)
30. clinical trial.pt. (467833)
31. randomized controlled trial.pt. (242391)
32. trial.ti. (69131)
33. random$.tw. (380181)
34. animals/ (4163880)
35. humans/ (9958560)
36. 34 not (34 and 35) (3147770)
37. or/30-33 (700210)
38. 37 not 36 (650487)
39. 3 and 28 and 38 (61)
40. meta-analysis.pt. (14993)
41. (systematic$ adj3 (review$ or overview$)).tw. (12318)
42. (meta-analy$ or metaanaly$).tw. (17787)
43. exp epidemiologic studies/ (972389)
44. exp case control studies/ (350868)
45. exp cohort studies/ (625301)
46. cross-sectional studies/ (74304)
47. (case control or cohort analy$ or cross sectional).tw. (103115)
48. (longitudinal or retrospective).tw. (201674)
49. (cohort adj (study or studies)).tw. (28976)
50. ((follow up or observational) adj (study or studies)).tw. (38748)
51. or/40-50 (1107069)
3 and 28 and 51 (450)
29 or 39 or 52 (887)
limit 53 to english (808)
(letter or news).pt. (694633)
54 not 55 (806)
aged/ or middle aged/ or "aged, 80 and over"/ (2772110)
56 not 57 (597)
"Case Reports [Publication Type]/ (1306615)
58 not 59 (549)

**Embase**

1. cochlea prosthesis/ (2895)
2. implantation/ (11854)
3. 1 and 2 (472)
4. cochlear implant$.mp. (2897)
5. 3 or 4 (2959)
6. age/ (66044)
7. (preschool or pre-school).tw. (3705)
8. Preschool Child/ (40738)
9. months.ti. (3775)
10. infan$.tw. (76834)
11. (12 months or 24 months or 36 months).tw. (46117)
12. (twelve months or twenty four months or thirty six months).tw. (1322)
13. (one year or two years or three years).tw. (37023)
14. (1 year or 2 years or 3 years).tw. (87439)
15. young child$.tw. (9295)
16. (pediatr$ or paediatr$).tw. (66154)
17. *Time/ (559)
18. infant/ (80964)
19. infant disease/ (1983)
20. age distribution/ (26303)
21. or/6-20 (421285)
22. 5 and 21 (871)
23. (age adj implant$).mp. (210)
24. 5 and 23 (38)
25. 22 or 24 (871)
26. clinical trial/ (338713)
27. randomized controlled trial/ (98926)
28. randomization/ (19175)
29. single blind procedure/ or double blind procedure/ (48311)
30. crossover procedure/ (14182)
31. placebo/ (54760)
32. (randomized controlled trial$ or randomised controlled trial$).tw. (19890)
33. rct.tw. (1472)
34. (random$ adj2 allocat$).tw. (6634)
35. (singl$ or doubl$ or tripl$ or trebl$) adj (blind$ or mask$ or dummy)).tw. (43881)
36. prospective study/ (53621)
37. case study/ (2456)
38. case report.tw. (58880)
39. abstract report/ or letter/ (223934)
40. or/37-39 (283989)
41. or/26-36 (412623)
42. 41 not 40 (398939)
43. exp meta-analysis/ (25321)
44. (meta-analy$ or metaanaly$).tw. (13552)
45. (systematic$ adj3 (review$ or overview)).tw. (10775)
46. (reference list$ or manual search$ or hand search$ or relevant journals or bibliograph$).tw. (6176)
47. (data extraction or selection criteria or medline or embase or cinahl or psychlit or psychinfo).ab. (19129)
THE EFFECTIVENESS OF EARLY COCHLEAR IMPLANTATION FOR INFANTS AND YOUNG CHILDREN WITH HEARING LOSS

Cinahl

1  Cochlear Implant/ (1301)
2    limit 1 to (newborn infant <birth to 1 month> or infant <1 to 23 months> or preschool child <2 to 5 years>) (416)
3    ((elder$ or geriatric or adult$) not (child$ or infan$)).ti. (78933)
4    94 not (95 or 96) (543)
clinical trial.pt. (19179)
clinic$ adj trial$.tw. (9631)
((single$ or doubl$ or trebl$ or tripl$) adj (blind$ or mask$)).tw. (5806)
randomized control$.tw. (8332)
random assignment/. (14138)
random$.af. (86080)
quantitative studies/ (2975)
meta-analysis/ (4981)
(meta analy$ or metaanaly$).tw. (3374)
exp literature review/. (5092)
(systematic$ adj (review$ or overview)).tw. (6212)
or/7-18 (114498)
prospective studies/ (51653)
exp case control studies/ (11697)
correlational studies/ (7184)
nonconcurrent prospective studies/ (22)
cross sectional studies/ (18170)
(cohort adj (study or studies)).tw. (5289)
(observational adj (study or studies)).tw. (2313)
or/20-26 (85209)
letter.pt. (38286)
Animals/ (798)
28 or 29 (39053)
19 not 30 (112876)
27 not 30 (84631)
6 and 19 (13)
6 and 32 (125)
33 or 34 (135)
treatment outcome/ (31278)
Language Development/ (1587)
Speech Perception/ (1507)
"Quality of Life"/ (14055)
VOCABULARY/ (290)
Verbal Behavior/ (423)
Auditory Perception/ (696)
Auditory Threshold/ (759)
Postoperative Complications/ (7166)
Voice Quality/ (623)
Time Factors/ (20848)
Age Factors/ (23419)
or/36-47 (92595)
6 and 48 (225)
limit 49 to abstracts (149)
limit 49 to review (10)
50 or 51 or 35 (214)

PsychInfo
1 exp Cochlear Implants/ (534)
2 limit 1 to english (527)
3 limit 2 to yr=1996-2006 (477)
4 limit 3 to (120 neonatal <birth to age 1 mo> or 140 infancy <age 2 to 23 mo> or 160 preschool age <age 2 to 5 yrs>) (103)
5 limit 4 to all journals (91)

Cochrane Central Register of Controlled Trials
1 cochlear implants/ (42)
2 cochlear implantation/ (15)
3 1 or 2 (53)
4 treatment outcome/ (34737)
5 child language/ (31)
6 speech perception/ (254)
7 quality of life/ (4952)
8 language development/ (97)
9 vocabulary/ (121)
10 verbal behavior/ (417)
11 auditory perception/ (272)
12 postoperative complications/ (8685)
13 voice quality/ (57)
14 auditory threshold/ (150)
15 or/4-14 (46351)
16 (preschool or pre-school).tw. (823)
17 months.ti. (1606)
18 infan$.tw. (10453)
19 (12 months or 24 months or 36 months).tw. (11830)
20 (1 year or 2 years or 3 years).tw. (15725)
21 (one year or two years or three years).tw. (7811)
22 (twelve months or twenty four months or thirty six months).tw. (387)
23 time factors/ (30149)
24 age factors/ (4221)
25 *child, preschool/ (0)
26 young child$.tw. (1257)
27 (pediatr$ or paediatr$).tw. (5406)
28 or/16-27 (75527)
29 3 and 15 and 28 (9)

Current Contents
1. TS=Cochlear implant*
2. TS=Age SAME implant*
3. #1 AND #2
4. TS=infan* OR child*
5. #3 AND #4
6. TS=12 months  OR 24 months  OR 36 months
7. TS=Twelve months  OR 24 months  OR 36 months
8. TS=One year  OR 2 years  OR 3 years
9. TS=1 year  OR 2 years  OR 3 years
10. TI=months
11. TS=(preschool OR pre-school)
12. #6 OR 37 OR #8 OR #9 OR #10 OR #11
13. #5 OR #13
14. TI=((elder* OR geriatric) NOT (child* OR infan*))
15. #14 NOT #15

Cross database additional searches – Medline, Embase, Cinahl, Psychinfo
1 ((neonatal or newborn) and screening and hearing).mp. (2337)
2 cochlear implant$.mp. (8157)
3 1 and 2 (94)
4 limit 3 to english (75)
5 remove duplicates from 4 (55)
6 (audioverbal therapy or audio-verbal therapy or audioverbal training or audio-verbal training or auditory verbal therapy or auditory verbal training).mp. (27)
7 limit 6 to english (21)
8 remove duplicates from 7 (16)
9 8 and 2 (6)
10 5 or 9 (61)

Current Contents additional search
1. TS=(audioverbal therapy OR audio-verbal therapy OR auditory verbal therapy)
2. TS=(audioverbal training OR audio-verbal training OR auditory verbal training)
3. TS=((neonatal OR newborn) SAME hearing screening)
4. \textit{TS=cochlear implant*}
APPENDIX 3: EXCLUDED RETRIEVED PAPERS


National Health and Medical Research Council (2000). *How to use the evidence: assessment and application of scientific evidence*. Canberra: NHMRC.


perception functioning in children after cochlear implant surgery. *Archives of Pediatrics &
Adolescent Medicine, 157*, 552-558.

questionnaire to evaluate children’s Auditory Behavior in Everyday Life (ABEL). *American
Journal of Audiology, 11*, 72-82.

versus language outcome after cochlear implantation in children. *International Journal of
Pediatric Otorhinolaryngology, 67*, 497-504.

Richter, B., Eissele, S., Laszig, R., & Lohle, E. (2002). Receptive and expressive language skills of 106
children with a minimum of 2 years’ experience in hearing with a cochlear implant.

Clinics of North America, 32*, 1117-1125.

of Clarion implant use. *Annals of Otolaryngology, Rhinology, & Laryngology - Supplement, 185*, 94-
95.


implantation between 5 and 20 months of age: the onset of babbling and the audiologic


speech perception in children with cochlear implants. *Proceedings of the National Academy of
Sciences of the United States of America, 102*, 18748-18750.

benefit analysis of pediatric cochlear implantation: German experience. *Otology &
Neurotology, 23*, 674-681.

and articulation in children with cochlear implants. *International Journal of Pediatric
Otorhinolaryngology, 66*, 115-123.

Sharma, A., Dorman, M. F., & Spahr, A. J. (2002). A sensitive period for the development of the
central auditory system in children with cochlear implants: implications for age of

up with access to cochlear implants. *Laryngoscope, 114*, 1576-1581.

one to three years of age: child, family, and linguistic factors. *Journal of Deaf Studies & Deaf
Education, 9*, 395-412.


APPENDIX 4: APPRAISED RETRIEVED PAPERS


