UNIVERSITY OF COPENHAGEN
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PhD dissertation

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Born deaf – growing up hearing

- Outcomes of Pediatric Cochlear Implantation in Denmark

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Title / Subtitle:	Born deaf – growing up hearing. - Outcomes of Pediatric Cochlear Implantation in Denmark
Subject:	Pediatric cochlear implantation is an interdisciplinary intervention involving ENT surgeons, audiologists, engineers and speech and language pathologists. Various factors affect the outcomes of the medical-surgical intervention as regards to spoken language development and social well-being.
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“We turned a child who couldn’t hear
into a typical 2-year-old, who doesn’t listen”

New York Cochlear Implant Center

English summary

Introduction

Pediatric cochlear implantation (CI), universal neonatal hearing screening (UNHS) and bilateral cochlear implants have changed the research arena in childhood deafness markedly. The thesis studied the effect on audition, spoken language and social well-being of these medical and technological interventions for the first generation of children with CI in Denmark. The thesis points to changes in the group of children with CI over time in terms of various factors of demographic and child related characteristics.

Objective

The primary objective was an evaluation of level of audition, spoken language and social well-being for the first generation of children with CI in Denmark. Specific objectives sought to provide a greater understanding of the factors with impact on acquisition of audition, spoken language and social well-being for children with CI in Denmark. In addition the thesis investigated differences and similarities over time between two cohorts of children with CI. Furthermore, the thesis sought to identify personal and societal changes led by the introduction of pediatric CI.

Method and material

The thesis binds together four articles, all aimed at identifying factors with impact on spoken language and social well-being. All studies were countrywide survey studies and provided a status of children with CI at that particular time of testing and assessing. Two different cohorts were investigated. Study I and II included children with CI before the introduction of UNHS and bilateral CI and study III and IV included children after these introductions. A total of 250 children participated and came for testing accompanied by one or two parents. A total of nine different tests and assessments were applied to the two different cohorts. Four different factors, i.e. age at implantation, hearing age, mode of communication and educational placement, were consistently analysed in all studies. Various other factors were included in some of the studies.

Results

Parental mode of communication had a statistical significant effect on all outcome measures of audition, spoken language understanding, vocabulary, speech production and social well-being in all studies. Place of living and age at implantation were identified as influential factor for both cohorts. The factors: educational placement; diagnosis; mode of implantation; hearing age; gender; age at fitting of hearing aids prior to implantation and number of hours with support teacher affected outcome but not as consistently as parental mode of communication. The introduction of UNHS led to a decrease in age at implantation for prelingual children. Mode of implantation changed from being almost only unilateral to almost only bilateral. Parental mode of communication and educational placement changed radically between studies. The majority of children from both cohorts were assessed to have a high level of social well-being and level of social well-being was positively associated with level of spoken language.

Conclusion

The first generation of children with CI did not perform as well as described in the literature in terms of outcomes of spoken language, whereas outcomes of social well-being were consistent with findings in the international literature. Pediatric CI has led to major societal and personal changes and these changes continue to challenge both health care services and educational services.

Danish summary

Baggrund

Tilbud om pædiatrisk cochlear implantation (CI), introduktion af neonatal hørescreening (UNHS) og tilbud om bilateral CI har markant ændret forskningsområdet indenfor pædiatrisk audiologi. Nærværende afhandling undersøgte effekten af den medicinske og tekniske intervention i forhold til auditivt, talesprogligt og socialt udbytte for den første generation af børn med CI i Danmark.

Formål

Det primære mål var en evaluering af niveauet af hørelse, talesprog og social velfærd for den første generation af børn med CI i Danmark. Specifikke formål var at bidrage til en større forståelse af hvilke faktorer der havde indvirkning på udbytte af CI-behandlingen i forhold til auditivt niveau, talesprogligt niveau og niveau af social velfærd. Derudover var det et formål at undersøge forskelle og ligheder over tid mellem to forskellige grupper af børn med CI. Ydermere var det et formål at identificere i hvor høj grad introduktionen af pædiatrisk CI medførte personlige og samfundsmæssige ændringer for gruppen af børn med medfødt døvhed/svært høretab.

Metode og materiale

Afhandlingen binder fire videnskabelige artikler sammen, som alle havde til formål at identificere faktorer med indvirkning på udbytte af CI-behandlingen i forhold til talesprogligt niveau og niveau af social velfærd. Alle studier var landsdækkende undersøgelser som gav en status af børn med CI på tidspunktet for testning og vurdering af børnene. Artiklerne relaterer til to børnegrupper med CI før og efter introduktion af UNHS og tilbud om bilateral CI. Sammenlagt deltog 250 børn og familier og alle børn blev testet med deltagelse af en eller begge forældre. Ni forskellige sproglige tests og vurderingsskemaer blev anvendt på de to børnegrupper. Fire forskellige baggrunds-faktorer blev anvendt i alle undersøgelser; alder ved implantation, hørealder med CI, kommunikationsform og institutionsplacering. Derudover blev flere forskelligartede baggrunds-faktorer analyseret i de fire undersøgelser.

Resultater

Forældres valg af kommunikationsform havde statistisk signifikant effekt på alle undersøgte områder af udbytte i alle studier. Alder ved implantation og bopæl i forhold til tilknytning til

Østdansk eller Vestdansk CI center havde ligeledes statistisk signifikant effekt på udbytte af CI behandlingen for begge børnegrupper. Faktorerne institutionsplacering, diagnose, implantationsmåde, hørealder, køn, alder ved start med høreapparater inden implantation og antal støttetimer havde effekt på udbytte, men ikke så konsekvent som forældres kommunikationsform. Introduktion af UNHS har medført at prælingvale døve børn implanteres markant tidligere. Implantationsmåde ændredes over tid fra næsten udelukkende ensidig implantation til næsten udelukkende dobbeltsidig implantation. Forældres valg af kommunikationsform ændredes markant over tid. Institutionsplacering ændredes ligeledes markant og majoriteten af børn med CI fra studie III of IV var inkluderet i institutioner for normalthørende børn med varierende grader af støttetimer. Majoriteten af børn i begge grupper blev vurderet til at have et højt niveau af social velfærd og niveau af social velfærd var positivt korreleret med talesprogligt niveau.

Konklusion

Den første generation af børn med CI i Danmark havde ikke samme udbytte i forhold til niveau af talesprog, som beskrevet i litteraturen. Udbytte i forhold til niveau af social velfærd var i overensstemmelse med litteraturen. Pædiatrisk CI har medført markante ændringer i forhold til både personlige og samfundsmæssige vilkår for børn med døvhed/kraftigt høretab, og disse ændringer medfører til stadighed store udfordringer for både sundheds- og undervisningsområdet.

List of abbreviations

ABI	Auditory Brainstem Implant
AO	Auditory Oral
AVT	Auditory Verbal Therapy
CAP	Capacity of Auditory Perception
CI	Cochlear Implant
dB HL	decibel Hearing Level
dB SPL	decibel Sound Pressure Level
HA	Hearing Aid
HI	Hearing Impairment
Hz	Hertz
HL	Hearing Loss
HRQoL	Health Related Quality of Life
IF	Impact Factor
NH	Normal Hearing
NLTV	Non-Looking Vocal Turn
OR	Odds Ratio
PPVT-4	Peabody Picture Vocabulary Test 4th edition
SIR	Speech Intelligibility Rating
SL	Sign Language
TC	Total Communication
UNHS	Universal Neonatal Hearing Screening

Table of Contents

1. Introduction

This PhD thesis evaluates the first generation of children with cochlear implant (CI) in Denmark. Pediatric cochlear implantation was first introduced in Denmark in 1993 when two children with profound hearing impairment underwent the operation. A CI provides perception of speech sounds in the whole speech spectrum and also provides perception of environmental sounds. With a CI sounds can be detected, but discrimination, identification and understanding of the sounds must be learnt. The thesis presents four studies which focus on outcomes after CI in terms of levels of audition, spoken language and social well-being. The studies are presented in four articles bound together in the present thesis. The thesis provides an understanding of which factors affect the outcomes with regards to spoken language perception and production, and social well-being. The four studies involve children implanted during a period of 18 years from 1993 to 2011 in Denmark and relate to a total material of 292 children with CI and their families.

The primary purpose of offering CI to children with severe to profound hearing loss is to give the children access to listening and hence enable them to develop a spoken language (Peterson et al., 2010). An important aspect of pediatric deafness is that 95% of children born with severe to profound hearing impairment are born to parents with normal hearing (Mitchell & Karchmer, 2004). It is, therefore, only natural that the majority of parents have a strong incentive to have their child implanted in order to pursue a listening and spoken language development for their child. Ganek et al., (2012) express this as follows:

> The primary goal of implantation in children is to facilitate communication in the modality that is native to the families of the vast majority of deaf children: Spoken language.

Congenital hearing impairment is one of the most frequent functional disabilities in our society. It is estimated that severe to profound hearing impairment affects one child in every 2.000 every year (Fortnum et al., 2001). A pediatric population with hearing impairment, HI, is, however, characterised as being an in-frequent and heterogeneous group when compared to children diagnosed with, for instance, autism spectrum disorders. Deafness from birth has a profound effect on the acquisition of early communication skills which again constitute the precursors of language development (Archbold, 2010b). Therefore, children with substantial profound hearing impairment are at significant risk of serious speech and language delays that can impact on their communication, their cognitive development as well as their social development (Connor et al., 2006). Since the introduction of CI as a treatment for children with profound hearing loss there have, however, been studies documenting auditory and speech/language progress never before described for a profoundly hearing impaired pediatric population (Fulcher et al., 2012, 2014; Dettman et al., 2007; Moog, 2002; Moog & Geers, 2003; Tait et al., 2001; Waltzman et al., 2002; Wie, 2005, 2010). Various factors may have an impact on auditory and speech/language development following CI. In this thesis the results of investigations into the impact of various factors on the first generation of children with CI are described. The primary focus of the thesis will be investigations of impact factors relevant to the vast majority of children with CI. In the present thesis this is defined as prelingual deaf children born into normal hearing families, who are neurologically intact and therefore have the foundation for successful development of auditory brain pathways when cochlear implanted. Well-functioning auditory pathways enable development of listening and spoken language and the present thesis will investigate some of the factors underlying the acquisition of listening and spoken language in a child with congenital severe to profound hearing impairment, who perceives sound and speech via the CI technology.

Three major medical and technological interventions in Denmark are the introduction of pediatric CI in 1993, the introduction of universal neonatal hearing screening in 2005 and the introduction of bilateral CIs in 2006. These interventions have radically changed the life chances for children with profound hearing impairment with regards to participation in society on similar terms to children with normal hearing and typical development. The thesis studies the effect of these medical and technological interventions on children with severe HI. The thesis points to changes in the group of deaf children over time in terms of the following factors: age of implantation, types of communication, educational placement, mode of implantation. The thesis provides evidence that, since the introduction of CI, pediatric deafness must be perceived differently in order to meet the needs of the children with CI. The four studies provide a unique description of the first generation of children with CI in Denmark as the children have undergone thorough testing and assessments of levels of audition, language and social well-being.

Findings from the present thesis have both academic and societal implications. In academic terms the findings enhance understanding of the underlying mechanisms that are associated with successful listening and spoken language development following early diagnosis of pediatric hearing impairment. The documentation of possible changes in demographically related factors, such as education options for a new generation of congenital deaf children with CI, will put the societal impact of pediatric CI intervention into perspective. In terms of societal implications findings from the thesis provides insight into new opportunities for congenital deaf children with

CI in terms of inclusion in the educational system. Denmark and the other Nordic countries provide an excellent research arena on the topic of pediatric hearing impairment as universal neonatal hearing screening was introduced in the mid-2000s in Sweden, Norway and Denmark. Furthermore, Denmark and the other Nordic countries provide an excellent research arena because the influence of socioeconomic factors is minimised as CI intervention is on offer to all candidates regardless of socioeconomic status. Results will have an academic impact on the understanding of which factors can best predict outcomes of early medical, technical and educational intervention. The four studies provide the basis for an evidence based discussion of whether education of children with CI has changed in accordance with the major medical and technological changes.

2. Objectives

This thesis evaluated the first generation of children with CI in Denmark. The primary focus was to investigate impact factors relevant to the vast majority of children with CI. In the present thesis these were defined as prelingual deaf children born into normal hearing families, who were neurologically intact and therefore had the foundation for development of audition and spoken language when cochlear implanted. The objective was to evaluate the level of audition, spoken language and social well-being for the first generation of children with CI in Denmark. More specifically the objective was to provide a better understanding of the factors underlying successful acquisition of audition, spoken language and social well-being for children with CI in Denmark. The thesis focused on key factors that had been identified as predictors of outcome in the research literature. Some predictors related to demographic factors and some predictors were more related to child factors.

The demographic factors of interest were: communication mode, educational placement, parental involvement, socio-economic status, and place of living. The thesis investigated to what extent these demographic factors affected outcome and whether they changed over time in light of the introduction of UNHS and bilateral CI. Specific child factors of interest were: aetiology of the hearing impairment; age at diagnosis; age at implantation; mode of implantation; length of hearing with CI; chronological age at day of testing; additional disability and prelingual or postlingual at time of implantation. Both demographic and child factors were associated with outcome measures of social well-being and self-esteem and outcomes of audition and spoken language in terms of receptive vocabulary, expressive vocabulary, language understanding, phonology, auditory capacity and speech intelligibility.

Primary objective of thesis:

- Evaluation of the level of hearing, spoken language and social well-being for the first generation of children with CI in Denmark.

Specific objectives of thesis:

- To investigate the factors affecting the acquisition of spoken language for children with CI in Denmark.
- To investigate the factors affecting the social development of children with CI in Denmark.
- To analyse the differences and similarities between groups of children with CI implanted before and after the introduction of UNHS and bilateral CI.
- To identify the personal and societal changes which the introduction of pediatric CI has led to.

The specific research questions of study I:

- Do the factors affecting speech and language also affect the social well-being of a pediatric cochlear implant population?
- Is it possible to isolate one factor more highly associated with (positive?) outcomes than others?
- Is it possible to estimate effect-related odds ratios for cochlear implanted children’s social well-being?
- To what extent is language level post-implant associated with the level of social well-being?

The specific research questions of study II:

- Which factors affect speech and language outcomes for a pediatric cochlear implant population in Denmark?
- Is it possible to isolate one factor more highly associated with outcome than others?
- Is it possible to estimate effect-related odds ratios for cochlear implanted children’s performance with regards: level of vocabulary, phonology, speech understanding, communication, speech intelligibility and auditory capacity?

The specific research question of study III:

- To investigate whether regional differences in CI outcome exist between the eastern and western part of Denmark after the introduction of UNHS and bilateral CI.

The specific research questions of study IV:

- To identify factors associated with the level of language understanding, the level of receptive and active vocabulary,
- To estimate effect-related odds ratios for cochlear implanted children´s language level.

3. Background

Chapter 3 provides an overview of the substantial state-of-the-art literature covering various aspects of outcomes of pediatric cochlear implantation. The literary overview places the present thesis in the context of the international research within the area of pediatric cochlear implantation.

3.1 Communicative and spoken language development – implications for deaf children

Deafness from birth has a significant effect on spoken language acquisition. The auditory system begins to function in the uterus in the 3rd trimester, thus children with normal hearing, NH, are born with an auditory memory of environmental sounds and sounds of mother's voice and speech patterns (Juscyk et al., 1994). From the first days of life children with NH detect small differences in speech sounds and this ability develops further as infants are exposed to spoken language(s). During the first year of life children with NH acquire the phonology of their native language(s) and they gradually produce speech-like sounds which turn into babbling and the production of the first words (Masataka, 2006). Language is universal to a certain degree (Berwick et al., 2013) but also dependent on neuro- and socio-cultural aspects in how language is developed during childhood (Tomasello et al., 2005; Vygotsky, 1978). Spoken language development incorporates domains of phonology, grammar, pragmatics and semantics (Bishop, 1997). Audition is essential for development of all domains incorporated in spoken language acquisition and because speech primarily is a series of acoustic events, audition is therefore the only sense that can process the stream of speech in its entirety (Ling, 2002). The early auditory development facilitates the development of the early communication skills of shared attention, referencing and of communicative turn-taking, which are all crucial skills for later language learning and cognitive development (Archbold, 2010b). Early communication skills are a major predictor of later language ability and also of later interpersonal communication (Marschark, 1993).

Early childhood deafness leads to absence of exposure to auditory stimulation and spoken language, which will have a devastating effect on later language acquisition. Ninety-two percent of children with permanent hearing loss are born to two normal hearing parents and 96% of children with permanent hearing loss are born to one hearing parent and one parent with hearing loss (Mitchell & Karchmer, 2004) and the language at home is spoken language(s) and typically the mother's language is oral. A deaf child will not respond to the hearing mother in a usual way, which in most cases makes the mother change the interaction with her deaf baby. In the literature it is described how mothers change their interaction with a child with HI, because the child does not respond to the auditory communication (Calderon & Greenberg, 2003; Fagan et al., 2014; Szagun, 1997; Yoshinaga-Itano, 2014). Lack of interaction between mother and child leaves the deaf child not only with auditory deprivation but also deprived stimulation in experiencing interaction and communication. The problem of severe hearing impairment in early childhood is therefore not only a hearing problem but also an interaction and communication problem. Even though medical and technological interventions take place as early as four to six months of age, when hearing aids or CI are fitted, the deaf child has still missed out a great deal of stimulation and is therefore disadvantaged compared to a child with NH. Furthermore, Calderon & Greenberg (2003) describe how social and emotional development is based on experience and language and hence the deaf child is faced with a potential other area of delay and difficulty.

3.2 Continuum of communication methods

Before the introduction of CI to pediatric populations a bilingual educational approach in the sense of sign language and written Danish was the dominant mode of communication for children with severe to profound HI in Denmark. In the literature it is still debated which mode of communication should be recommended to congenital deaf children with CI. Knoors and Marschark (2012) sum up the bilingual history of deaf education and state:

> For over 25 years in some countries and more recently in others, bilingual education involving sign language and the written/spoken vernacular has been considered an essential educational intervention for deaf children. With the recent growth in universal new-born hearing screening and technological advances such as digital hearing aids and cochlear implants, however, more deaf children than ever before have the potential for acquiring spoken language. As a result, the question arises as to the role of sign language and bilingual education for deaf children, particularly those who are very young. On the basis of recent research and fully recognizing the historical sensitivity of this issue, we suggest that language policy should be revisited in an effort to ensure that they are appropriate for the increasingly diverse population of deaf children.

In contrast to this statement Humphries et al. (2014) argue that the language approach to pediatric populations with CI should still be bilingual. However, there is a lack convincing results of this educational approach and Knoors & Marschark (2014) suggest to further study why this approach is not as efficient as first anticipated. Furthermore, it is complex for normal hearing parents to communicate with their child in sign language, which is a foreign language for the vast majority of normal hearing parents. Use of sign language is even more complex because it is a language processed in a different sensory modality. Sundqvist et al. (2014) add to this complexity and describe how research has shown that deaf children of hearing parents have a delayed development of theory of mind, which is crucial for a total language development. In contrary, this is not always the case with deaf children of deaf parents, who presumably are immersed in a more vivid signing environment, which contribute to a better acquisition of the more subtle aspects of language. In addition, the social emotional benefits for a child of being able to have the same native language as the parents are stressed in studies of deaf children with deaf parents as opposed to deaf children of hearing parents. Deaf children of deaf parents are rated better on various social emotional scales compared to deaf children of hearing parents with a plausible explanation being that deaf children have a common native language with their deaf parents (Nicholas & Geers, 2003). Long-term language studies of deaf children in terms of reading comprehension ability show that among deaf 15-year-old students, who use American Sign Language as their primary mode of communication, reading ability is at the third-grade level, whereas the average 15-year-old student with NH is at tenth-grade-level (Ganek et al., 2012). Furthermore, the literature describes how bilingualism in the sense of two spoken languages is possible for congenital deaf children with CI (Robbins et al., 2004; Waltzman et al., 2003). Such studies indicate new ways of perceiving bilingualism for deaf children and suggest that defining bilingualism for deaf children with CI may be understood as the ability to both use and speak two spoken languages as it is for bilingual children with normal hearing.

Ganek et al. (2012) argue that if parents have chosen a CI for their child they value an auditory and spoken language development. At the same time it must be respected that not all families of children with HI share these values. However, parent choice should always be respected and parents should always have access to evidence based information about the different opportunities for their child in order for the parents to choose the language approach, which they value and which are consistent with their goals for their child's future. Figure 1 illustrates the continuum of communication options available for children with HI and the parents must choose which place on the continuum best fits their child. The two ends of the continuum represent languages expressed in two different modalities, the visual or the auditory modality, and in between there exist various other communication options (Ganek et al., 2012):

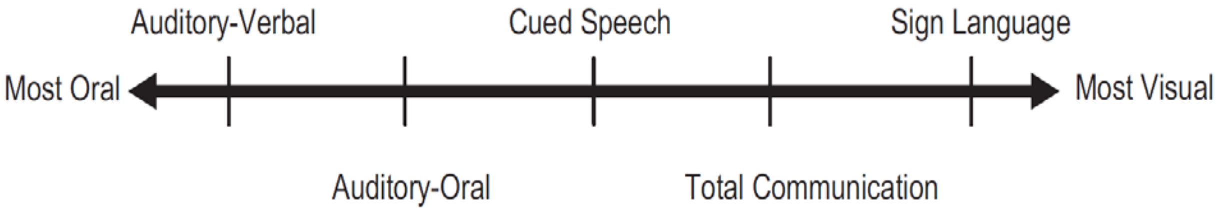


Figure 1: Continuum of communication options for children with HI (Ganek et al., 2012)

Sign language, SL, is typically associated with the deaf community. It is a visual and conceptual system and is a separate language comprised of its own unique syntax and word, but with no written form. Amplification from various hearing technology is not required for communication, as there is no focus on expressive or receptive spoken language. The entire body is used for expressive communication and to convey meaning.

In total communication, TC, the goal is to develop spoken language through speech reading with some form of manual communication. The expressive speech is developed through a combination of hearing, vision, and tactile cues. Teacher is often more fluent than parents in manual communication during early language years.

Cued Speech is a visual communication system of hand-shaped (cues) that represent different sounds of speech. The cues are used while talking to make spoken language clear through vision. This system allows the child to distinguish sounds that look the same on the lips. Cued speech is an oral option.

With the auditory-oral, AO, approach the goal is to develop spoken language primarily through hearing and speech reading. Teacher or parents are the primary language facilitator. AO often involves early social mainstreaming and school placement may also be in a mainstream setting. Use of appropriate and updated hearing technology is required for this approach.

The auditory-verbal, AV, approach aims to develop spoken language through listening. Therapy is done one-to-one, with parent or caregiver participating in each session. The parent or the primary caregiver is the primary teacher. Early mainstreaming is one goal of the AV-approach and the AV therapist must work in accordance with the 10 principles of AV Therapy. AV is an early intervention approach to teaching spoken language (AG Bell, 2015 www.listeningandspokenlanguage.org)

Yoshinaga-Itano (2014) stresses the importance of high quality of the early intervention following the detection of HI in a child. She argues that parents should have immediate early intervention from the time of detection of HI and if the parents want, they should have access to learning both a listening and spoken language method and a total communication method in order to overcome the challenges of limited interaction and communication between mother and child. Yoshinaga-Itano (2014) argues that it is a matter of quality in the early intervention and not so much a matter of combining two methods.

3.2.1 Children with CI and additional disability

Children with severe to profound HI and additional disability profit from CI and the access to auditory stimulation (Amirsalari et al., 2012; Nikolopoulos et al., 2008; Zaidman-Zaid et al., 2015). The prevalence of congenital deaf children with additional disability varies in the literature, but several studies refer to a prevalence of approximately 30-40% of children with sensorineural hearing loss >40dB to have additional disabilities (Berettini et al. 2008; Fortnum et al., 2002; Lesinski et al., 1995). Fortnum et al. (2002) found that children with cochlear implants are less likely to have disabilities concerned with learning or cognition. For children with additional disabilities it is, furthermore, important to inform parents about the various communication options available for children with HI and additional disability. A child with additional disability may have optimal auditory skills and is able to perceive language through audition. The child's own voice, however, may primarily rely on visual cues such as pictograms and signs. For instance, this could be the case for children with CI and cerebral palsy (Steven et al., 2011). Furthermore, it is important for professionals and parents to be flexible and ready to adjust to different communication methods if an auditory and spoken language approach is not possible for the individual child.

3.3 Neurological facts

The foundation of auditory development for children with CI is brain plasticity. For all children with HI, with or without additional disability, it is important to understand that in order to make use of a CI, it is necessary to work with the neurological pathways. This is only done by providing auditory stimulation and it must be kept in mind that pediatric cochlear implantation to a large extent is a matter of "use it or lose it" (Gordon et al., 2011; Kral & Eggermont, 2007). The neurological pathways have for the child with congenital deafness been out of use prior to implantation. It is therefore necessary to provide substantial amount of auditory input in order to "wake up" the neurological pathways (Borchgrevink, 2001). Parents must be informed about the neurological conditions which explain the reasons why stimulation of the auditory pathways in early childhood is crucial for success with a CI. Carol Flexer expresses this as follows:

> Hearing loss is primarily a brain issue, not an ear issue. Technology, e.g. cochlear implants and hearing aids are necessary to reach the brain of a child with hearing loss in order to create a neural structure for listening, language and literacy. The brain requires a great deal of auditory exposure and practice to develop the strong neural connections that serve as a platform for knowledge acquisition (Flexer, 2011).

Thus, the brain requires substantial amount of auditory experience in order to generate and change neural pathways in children with HI. The literature and documentation of neurological reorganisation provides evidence for the reason why early implantation followed by auditory habilitation is important in order to make use of the brain's plasticity and prevent the visual cortex to "take over" the functions of the auditory cortex (Harrison et al., 2001; Kral et al., 2002; Kral & Sharma, 2012; Sharma et al., 2002, 2005, 2009; Sharma & Campbell, 2010; Sharma et al., 2014). Studies in brain development show that sensory stimulation of the auditory centres of the brain is critically important and the organisation of the auditory pathways is indeed influenced by auditory stimulation (Boothroyd, 1997). Parents have the right to be informed about the limited timeframe for development of the auditory pathways in a child born with severe to profound HI. As with any other pediatric medical intervention, it is only ethically correct to inform parents about all dimensions of the implications of deafness and cochlear implantation. All risks, benefits and drawbacks of both surgical, communication and habilitation characters must be outlined and put into perspective for every individual child and family. Because of the neurological reorganisation, it is important to inform parents about the need for having the CI as early as possible and preferably before 3.5 years of age on the first ear. For sequentially implanted children the second implant should be as soon as possible after the first implant (Graham et al., 2009; Sharma et al. 2005, 2009; Sharma & Campbell, 2010). The auditory pathways in children with HI can be developed in the optimal developmental periods for listening from birth to 3.5 years (Dornan, 2010). This is the case for children with or without additional disabilities. If parents wish to pursue listening and spoken language outcomes for their child born with HI, an education approach that emphasises the development of auditory brain pathways through listening and spoken language is necessary. If parents choose differently for their deaf child at birth, the child will not develop similar outcome patterns, because delay in or lack of stimulation of the auditory brain results in diminished capacity for auditory brain development, thereby potentially depriving the child from the opportunity to listen and speak to their full potential. A child with HI must be regarded as a neurological emergency, because learning to listen is time-bound and has early closing windows of time (Dornan et al., 2010; Flexer, 2011).

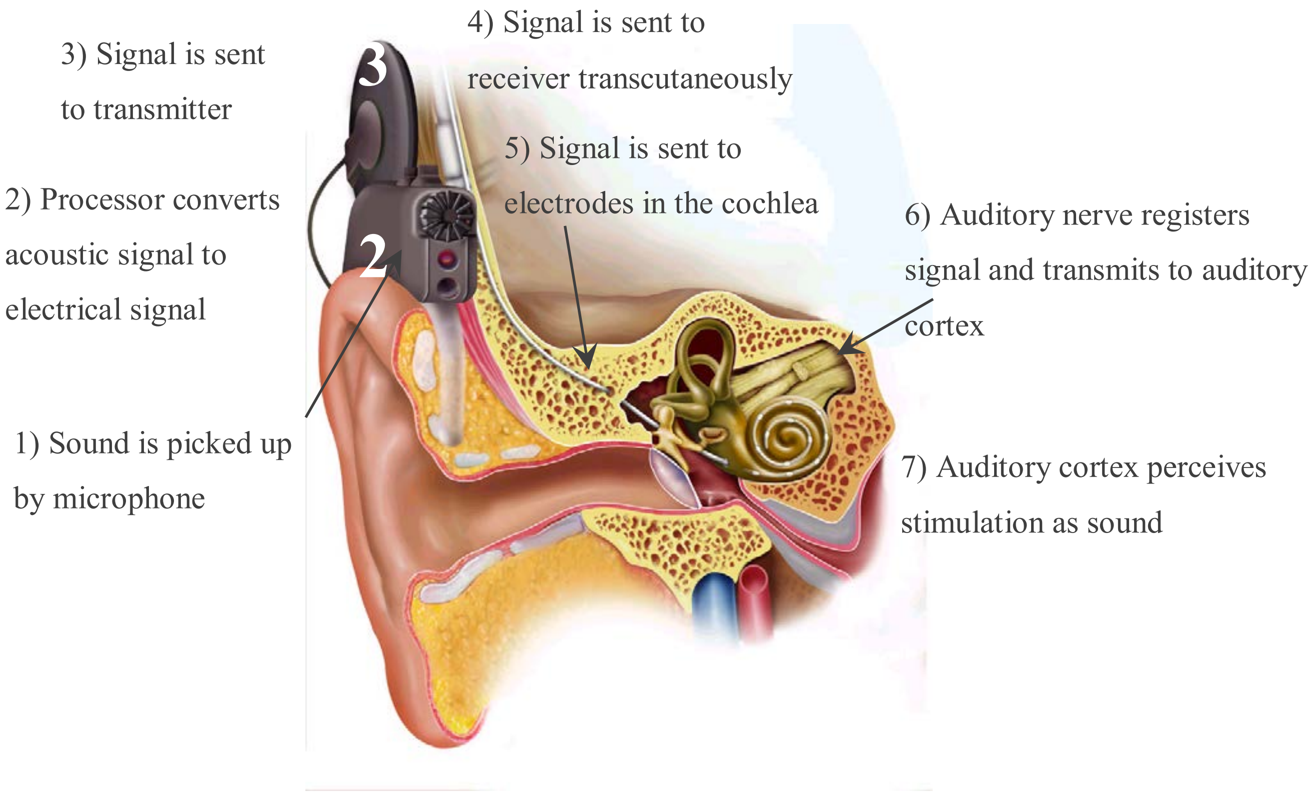
For children with additional disabilities implantation often occur later than recommended, because the children may suffer from other medical conditions, which leave the children with additional disability with a disadvantage in terms of developing the central auditory system (Amirsalari et al., 2012).

3.4 Hearing with a cochlear implant

If parents are to rely on auditory and verbal input as a method for development of audition and spoken language for their child post implant, then it is appropriate to question what kind of hearing is provided by a CI. Firstly the process of normal and damaged hearing should be directed. In the normally hearing ear the acoustic signal is converted into electrical impulses which stimulate the auditory nerve by displacements of the hair cells in the cochlea. The hair cells in a cochlea with sensorineural severe to profound hearing impairment are damaged and even the most powerful hearing aids are unable to provide full access to the range of speech sounds through hearing. A hearing aid filters, amplifies and compresses the acoustic signal and therefore delivers a processed signal to the cochlea for transduction. A CI in contrast receives processes and transmits acoustic information by generating electrical pulses to the auditory cortex. A cochlear implant consists of two basic parts:

1. The internal part consists of a receiver and an electrical array, which is surgically inserted into the mastoid bone and the cochlea.
2. The external part consists of a microphone, processor and transmitter.

Cochlear implants bypass the damaged hair cells. Sound is picked up by the microphone and the acoustic signal is converted into electrical pulses and provides electrical stimulation to the auditory nerve, which transmits the electrical impulses to the auditory cortex of the brain (Clark, 2004). Figure 2 illustrates the insertion of the electrical array and shows how the sound processing is transmitted to the auditory cortex (Sundhedsstyrelsen, 2012).



1) Sound is picked up by microphone

2) Processor converts acoustic signal to electrical signal

3) Signal is sent to transmitter

4) Signal is sent to receiver transcutaneously

5) Signal is sent to electrodes in the cochlea

6) Auditory nerve registers signal and transmits to auditory cortex

7) Auditory cortex perceives stimulation as sound

Figure 2: Hearing with a cochlear implant (Sundhedsstyrelsen, 2012)

Both the internal electrodes and the external processor constantly improve. The electrodes become less and less traumatic, the algorithms of the sound processing become better at mapping individuals' needs and the design of the external processor become smaller and smaller. The technological advancements of the various products will continue. Graeme Clark (2004) foresees totally implantable devices in the future.

3.4.1 Speech and sound perception with CI

A CI can encode the sounds of speech with precision and, therefore, the device can provide opportunities for learning spoken language (Ganek et al., 2012). The literature on outcomes in terms of speech perception for children with CI is substantial (Fulcher et al., 2014; Geers, 2006; Geers et al.2011; Looi & Radford, 2011; Meyer et al., 1998; Moog et al. 2003; Peterson et al., 2010; Wie et al., 2007). There is today no evidence that contradicts pediatric CI in children with severe to profound HI, who are neurologically intact (Estabrooks, 2006). CI is now regarded as a standard treatment for pediatric deafness (Connor et al., 2006; Flexer, 2011; Geers et al., 2010; Papsin & Gordon, 2008). Furthermore, it is suggested that pediatric bilateral CI is a cost-effective use of health care resources (Summerfield et al., 2010). The next area of investigation in terms of perception abilities of pediatric populations is the perception of music. Results suggest that CI users can make use of temporal and spectral cues to discriminate between musical stimuli, although not yet to the extent of their NH peers (Roy et al., 2014).

3.4.2 Candidacy

In Denmark, it is estimated that number of pediatric implantations is 45-50 annually and the implantations are evenly split between the two pediatric CI centres in the Eastern and the Western part of Denmark. The number of pediatric implantations covers both congenital deafness, progressed HI and acquired severe to profound HI. Congenital deafness constitutes around half of the implantations carried out annually (Sundhedsstyrelsen, 2012). These numbers have been stable for many decades (Parving et al., 2003). In the national protocol for pediatric CI (Sundhedsstyrelsen, 2012) it is stated that it is a goal to implant children between 8 to 12 months of age. There are no specific criteria in terms of thresholds measured in dB HL for Danish children with CI. Globally the criteria change all the time and children with hearing losses of 70-75 dB HL in the range of 125 to 8000 Hz are now meeting criteria for CI candidacy (Carlson et al., 2015; Briggs, 2011). Thus, children with relatively good residual hearing are implanted in some parts of the world. The next area of investigation into pediatric CI seems to be children with single sided deafness (Boyd, 2014; van Zon et al., 2014).

There are rare cases of children born with an absence of the auditory nerve or other medical issues, where CI is not a hearing solution. The number of children born without an auditory nerve or nerve fibres not functioning well enough to transmit electrical pulses to the auditory cortex is very limited. In Denmark there is to date knowledge of less than ten children born without an auditory nerve since the introduction of pediatric CI in 1993 (East Danish CI centre, April 2016). Most of these children have been offered an auditory brainstem implant, ABI. However, a further explanation of the impact of this kind of hearing technology is not within the scope of the present thesis.

There are three major CI companies on the global market, the Australian Cochlear, the American Advanced Bionics, and the Austrian Med-El. The Danish company Oticon Medical recently bought the French CI company Neurolec. Denmark will therefore be able to keep up with long traditions of being very strong on the hearing technological market and not least being very strong in research and development of various technological hearing solutions.

3.5 History of pediatric cochlear implantation in Denmark

Pediatric cochlear implants have for more than two decades globally increased. Globally pediatric cochlear implantation took off from the late 1980s. In Denmark the first two pediatric implantations took place at the East Danish CI centre in 1993.The West Danish CI centre implanted the first two children in 1996. Prior to implantation of a pediatric population implantations on adults had been carried out with the first being done in Denmark in 1982 (Pedersen, 2007). The first two pediatric implantations were not followed by many other implantations for the next seven years, see Figure 3, which provides an overview of the first 21 years of pediatric implantations in Denmark. The number of implantations is low in 2008 due to a long lasting strike between nurses at all hospitals in Denmark, and hence more implantations were carried out in 2009.

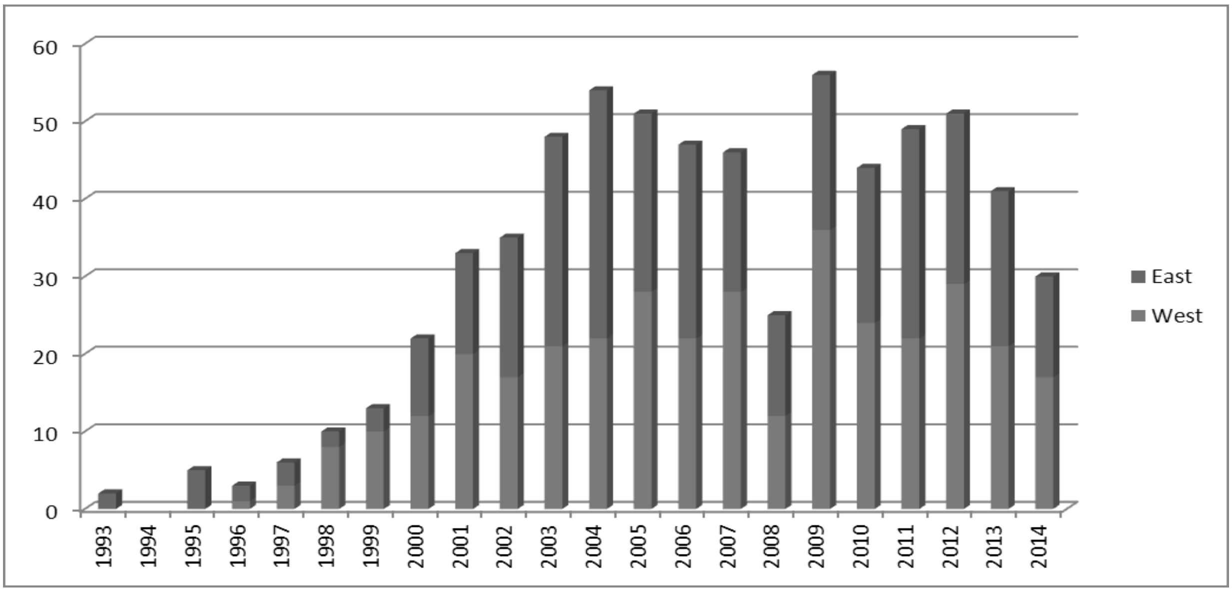


Figure 3: Number of pediatric cochlear implantations 1993-2014.

It is appropriate to question why there were so few implantations in the period from 1993 to 2000. In Denmark and the other Nordic countries the deaf community has strong and long lasting traditions of providing sign language to children with HI and their families, and in the initial area of pediatric cochlear implantation, it was regarded as a threat to this community. Pediatric CI was regarded as experimental and lacking evidence of the outcomes. Archbold (2010b) describes the challenges and controversies that was raised by the deaf community and states the following:

> The provision of implantation to children led to controversies of proportions that seem unimaginable today: Those in the deaf community argued that deafness is not a disease to be treated by medical intervention, but rather a cultural and linguistic identity, and hence children born deaf were part of that community rather than the hearing community. They claimed that a deaf child's first language is sign language, and that cochlear implantation, with its emphasis on hearing and access to spoken language was removing their right to be deaf and to their cultural identity.

The same arguments were used in Denmark and parents were advised not to pursue a CI for their child by the Danish deaf association and it was argued that cochlear implantation in children would lead them to experience severe mental health problems as adults and to reject their implants and the parents who had chosen them (Archbold, 2010b). Such statements were communicated without any scientific references and evidence.

Throughout the pediatric CI history, choice of communication mode has been and still is heavily debated (Archbold, 2010b; Graham et al., 2010; Gravel & O´Gara, 2003; Kermit, 2012) and it is recognised that the debate is of a sensitive character (Knoor & Marschark, 2012). Debates about whether children with congenital deafness are born with a sign language have continued until today and often without the explanatory neurological facts taken into account. Parents had to navigate between the fact based medical and technological guidance and guidance from people from the deaf association and former institutions of the deaf. It is understandable that this was not an easy task for parents and this perspective may contribute to explain the few implantations for the first seven years in Denmark.

3.5.1 Decibel

In 1997 five families with children with CI established the patient organisation, Decibel. The five families wanted to establish an organisation that worked towards developing new standards and updated knowledge about pediatric CI in Denmark. Decibel has grown ever since and is today the largest organisation for families with children using all kinds of hearing technology in Denmark. Decibel has become an opinion leading patient organisation on matters of choice of communication and language according to the values and wishes of the parents. Decibel has had influence on some of the milestones for children with CI in Denmark, for instance, the decision of offering bilateral CI to all pediatric candidates in 2006 and the implementation of the national protocol for pediatric CI in 2011. In 2013 Decibel established a research unit, (www.decibel.dk, 2016). Decibel is a living proof of the power of parents and shows how parents are powerful in the ability to advocate in their child's life (Estabrooks, 2012, pp 28-30).

3.5.2 Historical benchmarks

The Danish pediatric CI history is marked by the following benchmarks:

- 1993 first implantations
- 2005 introduction of UNHS (Universal Neonatal Hearing Screening)
- 2006 bilateral CIs to all relevant candidates
- 2011 implementation of a national protocol for assessment of candidacy, operation, initial rehabilitation post implant and long term monitoring of outcome.

The first implantation in 1993 is as already described the first major benchmark in the history of pediatric CI. Thirteen years later the introduction of UNHS meant that it would be possible to detect children born with all degrees of congenital HI. This introduced the opportunity of minimising the period of auditory deprivation (Morton & Nance, 2006). The aim of UNHS is to fit hearing aids before the child is 6 months of age, as this is described to be predictive of later language development (Yoshinga-Itano & Apuzzo, 1998). Introduction of UNHS meant that the need for a hospital based initial rehabilitation became evident, as children were identified at such young ages that they had not yet started attending day-cares or nurseries. Mothers/fathers on maternity/paternity leave therefore became the primary interventionists. This is in contrast to later identified children who would have started attending nursery and therefore be in contact with a professional system. To date there is still not a national protocol which describes the need for immediate and early intervention for children with all degrees of HI even though the need is well documented in the literature (Moeller et al., 2007, 2013; Yoshinaga-Itano, 2014).

The introduction of bilateral CIs for all relevant candidates is another benchmark in the history, as this provided children with severe to profound HI even better opportunities to learn spoken language as listening in background noise and the ability to localise is improved significantly (Papsin & Gordon, 2008; Summerfield et al., 2010; Wie, 2010).

When pediatric CI was first introduced Denmark, it was primarily a medical and technological introduction. In contrary to for instance England, Denmark did not introduce a clinical set-up for the initial habilitation attached to the CI centres, nor did Denmark have a standard of how to measure outcome of this new treatment of deaf children. From 1993 until 2011 the CI centres assessed the candidacy, carried out CI surgery, switched-on the processors and did fine-tunings. The former institutions for deaf children, i.e. nurseries, kindergartens and schools carried out the habilitation and in some schools did fine-tuning of the processors as well. There were regional differences in the clinical set-up between the East and West CI centres. In the West the former schools for the deaf were part of the assessment prior to implantation (Pedersen, 2007). But for the first 18 years there were no national standards of how outcome should be monitored and no standards for which methods to use in the habilitation. In 2006 the first countrywide study on outcomes of Danish children with CI was launched in a report from the Ministry of Social Affairs (Percy-Smith, 2006).

In 2011, 18 years after the first implantations, a national protocol was introduced, which describes the assessment of candidacy, the operation and the initial habilitation. The protocol is based on evidence from the neurological literature and therefore recommends to base the initial one year of habilitation on audition and spoken language. It is stressed that the parents play a vital role in this habilitation. Furthermore, it is recommended to place the initial habilitation at the CI centres in order to make use of the medical, technological and educational highly specialised professionals. In Denmark pediatric cochlear implantation is classified as a highly specialised function in the health care services, and it is a multi-professional function characterised by surgeons, audiologists, technicians/engineers and speech and language pathologists. The protocol stresses the collaboration between the hospital based CI team and teachers in the local community. The protocol, furthermore, outlines monitoring of outcome for 15 years post implant in terms of both auditory thresholds, receptive and productive language (Sundhedsstyrelsen, 2012).

3.6 Language development in children with CI

Language development in children with CI has been a well-studied area for the past 30 years and multiple factors for successful language development have been identified. Initially studies had primarily focus on speech perception in terms of phoneme/word discrimination and speech intelligibility of children with CI, but in later years studies have also focused on more subtle aspects of language development, e.g. theory of mind. Furthermore, long term language outcomes are a research area with great focus in the international literature. Ganek et al. (2012) summarise that published studies provide substantial evidence regarding the effects of CIs on language development in children and certain factors for successful language development are identified from various studies. It is important to stress the difference in language development depending on whether the child is prelingual or post-lingual in terms of spoken language. The majority of the literature has focus on congenital deaf children, i.e. prelingual children with CI. In the following the most influential factors identified in the international literature on spoken language development for prelingual children with CI is described.

3.6.1 Age at implantation

Different aspects of age are factors described to affect language development. It is important to first outline the various age definitions in the research literature. Age at implant is defined as a child's chronological age at the day of implantation. Age at switch-on or activation is defined as the child's chronological age at the day the processor was first activated post implant, which is typically a month after the implantation. Hearing age is defined as the length of device use. Some studies count hearing age from day of start with hearing aids prior to implantation and some studies count from the day of implantation or switch-on. There does not seem to be consensus of which way to count hearing age in the research literature.

Age of implantation is described as one of the most important factors with effect on language development. A substantial amount of studies provide evidence of the earlier implantation the better language outcomes and implantation before 12 months is associated with age equivalent language development (Dettman et al., 2007; Fulcher et al., 2012; Niparko et al., 2010; Tait et al., 2007; Vlastarakos et al., 2010; Wie, 2010).Various aspects of language development associated with implantation age have been described in the literature. Houston & Miyamoto (2010) found that children implanted during the first year of life had better vocabulary outcomes than children implanted during the second year of life. However, earlier implanted children did not show better speech perception outcomes than later implanted children and this indicates that vocabulary learning has a different sensitive period to perception. Sundqvist et al. (2014) found that children with early cochlear implants solved theory of mind problems to a significantly higher degree than children with late implants. Cuda et al. (2014) found that mean length of utterances and sentence complexity were significantly better for children implanted before 12 months of age compared to children implanted later. Furthermore, early implanted children performed better on both verbal and numeric tasks of auditory memory than late implanted children and it is stated that early cochlear implantation, before the age of three years, significantly improve auditory memory and contribute to better cognitive and education outcomes (Mikic et al., 2014).

Nicholas &Geers (2013) examined children implanted at 6 to 11 months compared to children implanted at 12 to 18 months. The 6-11 month group achieved higher scores on all measures of receptive vocabulary, expressive, and receptive language (including grammar) when tested at 4.5 years of age. Thus, cochlear implantation before 12 months of age seems to provide a significant advantage for spoken language achievement observed at a later age (Nicholas &Geers, 2013). It is possible to summarize from studying of the international literature that early cochlear implantation provides the best outcome in terms of listening, speech, cognition and memory due to maximal central nervous system plasticity (Mikic et al., 2014). There is consensus that early is under the age of 3.5 years but also that implantation younger than that should be pursued (Gordon et al., 2011). Cost-utility studies, furthermore, suggest that implantation as early as possible is favourable. Semenov et al. (2013) found that early implantation, before 18 months, was associated with greater and longer quality-of-life improvements, similar direct costs of implantations, and economically valuable improved educational placement, without a greater incidence of medical and surgical complications when compared to CI at older ages.

Studies of long term language outcomes indicate, however, that the effect of early implantation age diminishes with time, particularly for higher-order skills such as reading. Some children who receive cochlear implants at two years of age have the capacity to approximate the language and reading skills of their earlier-implanted peers (Dunn et al., 2014). Such findings suggest that additional factors may moderate the influence of implantation age on outcomes over time. Szagun & Stumper (2012) found that for children implanted within the sensitive period for language learning, children's home language environment contributed more crucially to their linguistic progress than age at implantation. Connor et al. described that implantation age and hearing age are interrelated and argue that there seems to be a substantial benefit for both speech and vocabulary outcomes when children receive their implant before the age of 2.5 years. However, 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 (Connor et al., 2006).

Ulrika Löfkvist states that there other factors involved in an overall language development than the age factors and points to more cognitive-related factors for learning of receptive vocabulary (Löfkvist, 2014; p. 29). Results from Quittner et al. (2013) also stress the importance of cognition on language growth as they found that cognitive stimulation was significantly associated with language growth post implant.

3.6.2 Education

Type of education and educational placement has been described as a factor to impact language development for children with CI. Archbold (2010b) describes three educational options for children with severe to profound HI:

- Institutions for the deaf, nurseries/kindergartens/schools.
- A unit for children with HI in mainstream school with varying degrees of inclusion into mainstream classes.
- Mainstream education with varying degrees of support in quantity and quality (Archbold, 2010b; p.26).

Deaf students have traditionally attended the institutions for the deaf and many schools have had residential facilities as student would come from widespread locations due to the fact that deafness is a low-frequent impairment. In the literature recent studies have described a change in the educational placement since the introduction of pediatric CI. Geers et al. (2011) found that in a longitudinal study 75% of teenagers were fully mainstreamed in high school, compared with 63% when the same teenagers were in elementary grades. Only 5% of adolescents were in full-time special education. This study indicates that educational placement change with increasing use of CI. In a study from Austria it was found that more than 80% of school-aged children attended mainstream schools. Furthermore, the educational level of pupils with CI did not differ from the Austrian population with NH (Huber, 2008). A demographic study from Finland found that 50% of the children were enrolled in kindergartens with NH peers and 43% of school-aged children with CI were enrolled in mainstream schools (Lonka et al., 2011).These data support the position that early cochlear implantation is a cost effective procedure in terms of education as it provides opportunity for children with CI to participate in a normal school environment.

Educational placement is, furthermore, associated with better language outcome. Tobey et al. (2004) found that higher speech intelligibility scores in 8- to 9-year-olds with CI were significantly associated with educational settings that emphasize oral communication development. Educational environments that incorporate exposure to peers with NH were also associated with higher speech intelligibility scores at 8 to 9 years of age. Wie et al. (2007) found that for a Norwegian population mainstream educational placement contributed to higher scores on speech recognition and speech production. The Norwegian results, furthermore, indicated that children in educational setting with increased focus on spoken language had faster growth rate of speech recognition and production. Moog & Geers (2010) found that providing intensive toddler classes with focus on developing spoken language is an optimal supplement to early parent-infant intervention for children implanted at one year of age. A study by De Raeve (2010) sums up the importance of a multidisciplinary approach to children with CI and states the following:

> The vast majority of children attend mainstream school and use spoken language as their main communication mode, but due to the evidence of heterogeneous results for children with CI it is a challenge for all educational services to meet the different individual needs. Educational services must ensure that their staff have the skills to meet these challenges: to be flexible, continually updated with the technology and changing expectations (ongoing professional training), to provide an environment which will utilise the hearing while meeting the linguistic and curricular needs of the children.

3.6.3 Parental factor

Parental involvement and communication are essential for language development in all young children. However, hearing parents of deaf children face challenges in providing language input to their children (Cruz et al., 2013).Therefore studying of parental involvement and communication is a research area of great importance. Several studies have documented that parental involvement and parental mode of communication are significantly associated with language development of children with CI (Dornan et al., 2010; Flexer, 2011; Fulcher et al., 2012; Holzinger et al., 2011). A large study of 188 children with CI, from a multicentre research group of childhood development after cochlear implantation, found that effects of maternal sensitivity on growth of language were similar to that found for age at cochlear implantation. This finding suggests that addressing parenting behaviours is a critical target for early language learning after implantation. CI teams must, therefore, include parents both in decisions on pursuing early implantation and on matters of being part of the habilitation both before and after implantation (Quittner et al., 2013).

Yoshinaga-Itano (2014) states that successful outcome after early detection of HI to a large extent depends on involvement of parents as partners in the process of diagnosis, hearing aid fitting, implantation and rehabilitation. Ganek et al. (2012) argue that parents are the most potent influence on the child’s progress. Moeller (2000) argues that two hours per week spent in therapy only make up 2% of a young child’s waking hours, whereas everyday activities, for instance feeding/changing nappy, occur at least 2000 times before the child's first birthday. Daily activities provide excellent opportunities for communication and language learning and therefore the power of families must not be neglected in the habilitation of children with HI. Lin et al. (2008) further argue that monitoring of language outcomes both before and after implantation should include parental reports. Parental reports offer a broader outcome measure and provide a potential for a fuller understanding of the true effectiveness of early implantation (Lin, 2008). Parental educational level is also found to influence various aspects of language development such as utterance length, expressive vocabulary, sentence complexity and narrative ability (Cuda et al., 2014, Murri et al., 2014). It is possible to summarise that parental involvement due to multiple factors is generally agreed to be a necessity for successful language development of children with CI.

3.6.4 Mode of communication

Mode of communication following pediatric implantation is identified as a factor with high impact on language development of children with CI. Children who are exposed to both total communication and auditory and spoken language as a whole improve their overall language performance. However, children exposed to solely audition and spoken language performed better on aspects of language development such as expressive vocabulary, morpho-syntactic use, utterance length and narrative forms children (Geers & Brenner, 2003; Geers, Brenner & Davidson, 2003; Geers, Nicholas & Sedey, 2003). In a longitudinal study Geers et al. (2011) found that use of sign to enhance spoken communication negatively influenced verbal rehearsal speed, which in turn was a strong predictor of all early outcomes, and strongly influenced later outcomes. These data suggest that early communication mode exerts a powerful influence on early outcomes with persistence into later years. In a recent study it was found that children in auditory verbal environment outperformed children in auditory oral and total communication environments. Results support consistent emphasis on oral/aural input to achieve optimum spoken communication outcomes for children using cochlear implants (Dettman et al., 2013). Children who use oral communication outperform children, who use total communication and this trend is robust in the literature (Ganek et al., 2012)

3.6.5 Other factors

Language development is also described to be influenced by gender. Gender has been documented to have some impact on scores of speech/language outcomes, as girls score higher than boys (Connor et al., 2006; Cuda et al., 2014; Meyer et al., 1998). The gender factor is, however, not described as a factor with consistent impact on overall language development. Furthermore, research has described socio-economic-status, SES, to be an influential factor on language development (Aragon &Yoshinaga-Itano, 2012).

Since the introduction of bilateral CI's to relevant candidates studies have found the method of implantation, i.e. bilateral simultaneously or sequentially or a bimodal stimulation, i.e. a combination of CI and hearing aid, to be another factor to affect language development (Graham et al., 2010; Looi & Radford, 2011; Papsin & Gordon, 2008; Sparreboom et al., 2010; Tait et al., 2010).

Several studies have investigated language development of pediatric populations with CI and additional disabilities (Amirsalari et al., 2012; Kontorinis et al., 2014; Lang, 2014). Nikolopoulos et al. (2008) found that the majority of deaf children with additional disabilities develop connected intelligible speech five years following implantation. A third of a group of children with CI and additional disabilities did not develop spoken language post implant. However, it is stressed that benefit from cochlear implantation should not be restricted to speech production alone in this specific population.

The literature study of language development of children with CI revealed that multiple factors underlie the mechanism of language development in prelingual children with CI. Furthermore, most of the factors are interrelated.

3.7 Social well-being of children with CI

It is possible to conclude that the CI technology successfully affect spoken language development for the majority of children with CI. To what extent does this positive impact of CI on spoken language acquisition also impact aspects of social and psychosocial well-being for children with CI? Are the factors identified for language outcome similar for outcomes of social well-being? Moreover, what is the status in the literature in terms of studies of quality of life for children with CI? From an ethical point of view it is important to study how children with CI interact socially in home, in school environments and in the community. It is appropriate to question the value of the listening and spoken language achievements, if the children cannot use these achievements to interact socially. A key aspect of development for every child, with NH or with CI, is the ability to interact socially. Social interactions and friendships in childhood are associated with a wide range of factors related to psychosocial well-being. Having close positive peer relationships is associated with increased self-esteem, regulation of emotion, better adjustment to school and a positive attitude to school (Batten et al., 2013).The literature describes very different rates of prevalence on psychosocial well-being of children with HI. Dammeyer (2010) studied literature on prevalence of psychosocial issues over a period of 15 years and found that rate of prevalence of psychosocial difficulties ranged from 20% to 50% among children with HI under 19 years of age. There is consensus that hearing impairment among children affects psychosocial development but there is not consensus of the rate of prevalence.

Some researchers have pointed out that pediatric cochlear implantation holds a risk of depriving the children full membership of any culture and children with CI may end up trapped between cultures. They are unable to function effectively in a hearing world but also lacking the sign language and therefore the opportunity to belong to the deaf community (Spencer et al., 2012). They risk not experiencing natural belongings to neither the deaf community nor the hearing world and, therefore, pediatric cochlear implantation risks adding on to problems of social well-being for deaf children (Kermit, 2012). It is of great importance to monitor outcomes in terms of these identity issues, which is done by investigating long-term outcomes of children who grew up with cochlear implant (Spencer et al., 2012).

In recent years children's social outcomes have begun to receive more research attention and it has become possible to investigate long-term outcomes across a wider range of domains related to children's functioning in everyday life, at home, at school and in the community (Punch & Hyde, 2011). Kumar et al. (2014) argue that cochlear implantation influences not only communication, but also psychosocial outcomes in children with severe to profound HI. They stress the importance of focusing on issues of self-reliance, social relations and education. It is argued that this knowledge will provide a more accurate and relative view of functional status of children with CI.

Connor et al. (2006) argue that children with severe to profound HI are at significant risk for serious speech and language delays and these delays impact communication, cognitive development and also social development. It is stressed that the identified influential factors on language development are interrelated with social development. The hypothesis is that higher levels of language are related to higher levels of social well-being (Connor et al., 2006). In contrast to these findings, Nicholas &Geers (2003) found that deaf children who had used a cochlear implant for 4 to 6 years coped successfully with the demands of their social and school environment, regardless of their speech and language achievements after implantation. Parents' ratings indicated that the children were emotionally and socially well-adjusted and that they benefitted from cochlear implantation. Thus, despite the language skills children with CI achieved positive outcomes in terms of social well-being. These results represent an impressive level of personal and social adjustment when compared with previous literature on adjustment problems in deaf children (Nicholas & Geers, 2003).

Moog et al. (2011) have carried out the largest study in the area of psychosocial research. They published a longitudinal study of 112 students, who had used CI up to 15 years. The study investigated psychosocial characteristics of students who had used CI since preschool and were evaluated when they were in primary school and again in secondary school. The study had four main areas of investigation: to assess to what extent the psychosocial skills which were documented in primary school maintained into secondary school; to assess whether long-term CI users identified with the deaf community or the hearing world or both; to investigate the association between group identification and the student's sense of self-esteem, preferred communication mode, and spoken language skills; and to describe aspects of social life outside school environment of the teenagers, who attended mainstream educational settings. The 112 students and their parents completed questionnaires describing their social skills, and 107 students completed questionnaires regarding issues of group identification and self-esteem. Results were compared with a control group of teenagers with NH and with norm based data. Results revealed that 70% of the students expressed either strong identification with the hearing community or mixed identification with both deaf and hearing communities. Almost all students with CI, 95%, were mainstreamed for more than half of the day, and the majority of students, 85%, were placed in classes appropriate for their age. All CI students, 98%, reported having hearing friends, and a majority reported having deaf friends. More than 75% of students with CI reported that they used primarily spoken language to communicate and that good spoken language skills enabled them to participate more fully in all aspects of their lives. Identification with the hearing world was not associated with personal or social adjustment problems but was associated with better speech perception and spoken language skills. Ninety-four percent were active participants in school activities and sports, and 50% held part-time jobs, which was a rate similar to that documented for teenagers with NH. The majority of these early-implanted adolescents reported strong social skills, high self-esteem, and at least mixed identification with the hearing world. These results indicate that mode of communication, language skills and social well-being are interrelated (Moog et al., 2011).

Bat-Chava et al. (2013) studied long-term improvements in speech and oral language skills and relationships with hearing peers of 19 children with CI. Results showed that on average, children continued to improve in both oral communication skills and quality of peer relationships up to 14 years after implantation. The largest increase in progress was especially found for the children, who initially showed poorer skills. Oral communication ability and quality of peer relationships were strongly associated at each point of assessment. Communication ability and degree of integration into the children's hearing environment were identified to be factors to impact the children's peer relationships.

Mance & Edwards (2012) studied self-perceptions and psychological well-being in the perspective of belonging to either the deaf community or the hearing world. Data revealed that the closer the implanted adolescents perceived themselves to be to their hearing peers the better their overall psychological well-being. In contrast, perceiving oneself as more similar to deaf signing peers or deaf oral peers was not significantly associated with psychological well-being.

Martin et al. (2011) published results for better outcomes for deaf children interacting in one-on-one situations compared to interactions including two other hearing children. They also found a gender effect, as girls performed better than boys. Furthermore, results of this study showed that longer use of CI and higher self-esteem were associated with better interaction with hearing peers. The results of the experimental situations were supplemented by parental reports of the children's social functioning in home, school and other social environments. The parents' reports were related to the children's performance in the experimental situation, thus revealing that parental reports of child well-being provide a valid source of information. These findings contribute to the growing literature describing the benefits of cochlear implantation in the areas of communication and socialization.

Geers et al. (2013) studied more language specific properties related to social skills. They investigated long-term outcomes of 60 children implanted within the neurological critical period for spoken language, i.e. between 12 and 38 months of age. Children were assessed at 9-12 years of age in terms of discrimination of nuances of talker identity and emotion. Results showed that well-developed social skills were more highly associated with the ability to discriminate the nuances of talker identity and emotion than with the ability to recognize words and sentences through listening. Such results indicate that early cochlear implantation enables the children to make use of some linguistic properties of speech which influenced not only their development of spoken language, but also their ability to function successfully in a hearing world. These results show new areas of research, which will positively impact both spoken language development and social well-being, as the early implanted children will have hearing and discrimination abilities beyond that seen for late implanted children.

The same relation between perception of emotional properties of conveyed sound and increased quality of life was found in a study of Schorr et al. (2009). In this study children's perceived quality of life did not significantly predict speech perception performance at the single word level, but increased quality of life predicted better performance on the emotion identification task. Furthermore, the children reported significant improvement in quality of life because of their cochlear implants, and they also reported low levels of concern about typical problems associated with wearing an implant. Implantation age was also found to predict higher perceived quality of life.

A separate study by Percy-Smith et al. (2008) evaluated social well-being and self-esteem of 164 children with CI compared to normal hearing children. Parental questionnaires, used in a national survey assessing social well-being and self-esteem parameters of 2169 children with NH, were completed by parents of children with CI in order to make direct comparisons between the two groups. Children were assessed on parameters of being bullied or bullying themselves, well-being in kindergarten/school, number of friends, management of school work and self-esteem parameters. Statistical significant differences were found for well-being in kindergarten and in school in favour of the children with CI. Boys with CI were rated significantly better at managing school work and children with CI were significantly more active and bullied less than children with NH. There was no statistical significant difference found for overall self-esteem, being bullied by other children and number of good friends. These findings are in accordance with international studies and contribute to the growing evidence that CI positively affects not only speech perception and production but also social well-being and self-esteem.

3.7.1 Quality of life

Quality of life of children and adolescents with CI is another important research area. Kumar et al. (2014) investigated issues related specifically to cochlear implantation, e.g., self-reliance, social relations, education, effects of implantation, and support. The study analysed parental perspectives of CI-specific health-related quality of life, HRQoL. The aim was to determine if parents differentially rate their child's quality of life according to psychosocial domains of communication, self-reliance and education. The study further investigated whether associations exist between quality of life domains and variables such as chronologic age, age at CI and length of CI use. Parents of 33 children with CI completed the questionnaire. Children were implanted early, mean age at switch-on was 2.47 years and children had used their CI for a mean age of 7.47 years. Parents positively rated most quality of life domains, but domains of education and effects of implantation were rated significantly less positive. The three domains, communication, self-reliance, and well-being, associated significantly with at least five other domains of HRQoL. This indicated that positivity in one domain co-occurs with positivity in other domains. Demographic variables chronologic age, age at switch-on and duration of CI use, did not associate significantly with psychosocial outcomes. Parents of children and adolescents with CI rate overall quality of life positively across psychosocial domains.

Duarte et al. (2014) also aimed at assessing HRQoL among prelingual profoundly deaf children and adolescents with cochlear implants and to compare the responses of these children to those of their parents. The sample consisted of deaf children and adolescents with CI and two control groups. One group was hearing children and adolescents and the other group was deaf children and adolescents. The ages of the participants ranged between 8 and 18 years. The hearing participants exhibited significantly higher quality of life than the deaf participants without implants in nearly all domains. In contrast, although hearing participants exhibited a slightly higher quality of life than children with CI, these differences were smaller and failed to reach statistical significance. In a study of Loy et al. (2010) the same tendency was found, as 88 children and adolescents with cochlear implant scored similarly to peers with NH in terms of quality of life. Furthermore, parents and children with CI rated quality of life similarly, which stresses the notion of parents as reliable reporters on the status of their child’s overall quality of life. In contrary to Kumar et al. (2014) this study found an effect of age at implantation as earlier implantation and longer use of CI resulted in higher quality of life scores (Loy et al., 2010).

Another study of HRQoL was conducted by Meserole et al. (2014). They investigated HRQoL of 129 children implanted between ages 6 months and 5 years in a national study. The 129 children with CI were compared to children with NH. Children with CI reported HRQoL comparable to peers with NH. The results, from both child and parent perspective, provide evidence of positive impact of CI on childhood deafness in terms of HRQoL. Results revealed that family stress was associated with worse HRQOL. Parent-child agreement in HRQoL scores was higher for CI families than NH families, which may reflect higher caregiver insight and involvement related to having a child with a CI and hence specific needs.

3.7.2 Quality of life in children with CI and additional disability

Children with CI and additional disabilities are another area of importance to investigate in terms of quality of life. Spoken language development is not always the goal for these children, but it may be an overall goal to increase quality of life by enabling the child access to sound from the CI. Palmieri et al. (2012) argue that parents often mention various benefits for children with CI and additional disabilities and by mean of a questionnaire based on observation they investigated which domains of life increased quality of life for these children. Observations of social, neuropsychological and perceptual skills were carried out on 50 children before and after implantation. The data showed significant improvements in all investigated areas. It is stressed that an observational questionnaire is often the only possible way of monitoring outcome for these children as other existing tests involve tasks too complex for these children.

A large comparative study of children with CI with or without additional needs was carried out by Mance & Edwards (2012). Reports from parents of 89 children, of whom 42% of the children were reported to have additional needs, were used to compare quality of life between the two groups. Parents' reports revealed that quality of life was poorer for children with CI and additional needs but not to a significant extent. It is concluded that cochlear implantation had a positive impact on the quality of life for both groups.

Based on the literature review above it is possible to conclude that pediatric cochlear implantation has changed life circumstances for congenital deaf children in terms of hearing and listening ability, spoken language development, education, psychosocial development, identity and quality of life. The impact of CI on both language and psychosocial well-being has proven to be positive for the vast majority of children with CI. As mentioned above the CI technology continues to improve, the age at implantation has decreased and it is common for children to receive two CIs. Spencer et al.(2012) argue that these medical and technological changes are good things and that we must integrate our knowledge into training professionals so they can apply this knowledge in the field of deaf education. With these changes and the substantial knowledge of factors with impact on outcomes in terms of language and social well-being the future looks bright for subsequent populations of users of CI.

4. Method and Material

Chapter 4 will provide an overview of the four studies. An overall description of the methodology and its limitations will be described and furthermore, the participants and the statistical analyses will be described. Finally, the ethical aspects of the thesis will be clarified.

4.1 Methodology

One major limitation in studying children with CI is that the group is small and heterogeneous in most aspects like age at diagnosis, age at implantation, aetiology and various other factors (Geers et al., 2011). This is a challenge and limits the possibilities of large randomised controlled trials (Löfkvist, 2014). Furthermore, considering the limited size of the population in a small country like Denmark there are high standards on ethical issues of not exposing the children. Even though a pediatric population with CI is small the research involves a large number of people. All pediatric research inevitably involves parents. Parents of children with CI have proven to be a highly valid source of information in assessing various aspects of the child's social and communicative development (Martin et al., 2011; Meserole et al., 2014). Pediatric cochlear implantation is characterised as a multi-disciplinary intervention involving a varied number of professionals, i.e. audiologists, surgeons, technicians/engineers, speech and language pathologists, teachers of the deaf, assistive teacher and primary teacher at the child's local institution/school. Data gathering is therefore a challenge and study design requires thorough control.

4.1.1 Study design

The thesis binds together four studies all aimed at identifying factors with impact on spoken language and social development of the first generation of children with CI in Denmark. The studies were presented in four articles published in national and international peer reviewed journals. The impact factor (IF) of each journal is presented according to Journal Citation Reports, 2014. The first article was published in Cochlear Implants International, which was a new journal and hence did not have an impact factor. Table 1 provides an overview of the four studies.

No.	Publication year	Journal	Title	Participants	Design
I.	2008	Cochlear Implants International; 9(4):199-214 IF: None	Factors that affect the social well-being of children with cochlear implants	167 pre and postlingual children and families	Countrywide survey study. Control = norm based references Score = hearing age
II.	2010	Acta Otolaryngolica; 130:708-715 IF: 1,1	Parental mode of communication is essential for speech and language outcomes in cochlear implanted children	155 prelingual children and families	Countrywide survey study. Control = norm based references Score = hearing age
Introduction of UNHS + bilateral CI					
III.	2012	Danish Medical Journal 59/5 IF:1,1	Significant regional differences in Denmark in outcome after cochlear implants in children.	83 prelingual children and families	Countrywide survey study Control = norm based references Score = chronological age
IV.	2013	International Journal of Pediatric Otorhinolaryngology;77: 184-188 IF:1,2	Language understanding and vocabulary of early cochlear implanted children	83 prelingual children and families	Countrywide survey study Control = norm based references Score = chronological age

Table 1: Summary of studies, year of publication, IF, title, number of participants and study design

The methodology used to evaluate the first generation of children with CI, was large countrywide survey studies. The studies had a cross-sectional design, i.e. “snapshots” or “one shots”, and provided a status of children with CI at that particular time of testing and assessing. The four studies did not have a control group of children with HI without CI, which precludes conclusions on causality. It was an aim to gather large data samples based on a randomised recruiting of children with CI from the whole country including Faroe Islands and Greenland. By comparing results from the four studies of the thesis it was, furthermore, possible to investigate changes in demographic and child characteristics over time. Two different cohorts of children with CI were investigated. Studies I and II included children with CI before the introduction of UNHS and bilateral CI. Studies III and IV related to another cohort of children with CI after the introduction of UNHS and bilateral CI. All children came for testing accompanied by one or two parents. All tests were carried out at the two pediatric CI centres in acoustically normal living rooms and a total of six different speech and language pathologists all experienced in testing children with HI carried out the testing. Parents completed questionnaires while the child was tested. One speech and language pathologist, i.e. the PhD applicant, scored all tests and assessments.

The variables hearing age and prelingual/postlingual are of significant importance in the thesis. Studies I and II scored children according to their hearing age with CI. Hearing age was calculated from day of switch-on of the processor, i.e. the first time the child heard with the CI.

This was decided after a pilot study, which showed very low language levels of the majority of children and hence scoring according to chronological age would have made no sense. Scoring according to hearing age allowed the child to have integrated an auditory sense and thus making development of audition and spoken language possible. Study III and IV scored speech and language tests according to each child's chronological age, as it was hypothesised that due to the introduction of UNHS and bilateral CI the children would have better chances of having closed the language gap between hearing age and chronological age. Study I included both children with prelingual and postlingual status at time of implantation. Study I focused on social well-being of the first 200 children with CI in Denmark regardless of their language status at time of implantation. This was decided because the literature describes that language level is not always significantly associated with level of social well-being (Nicolas & Geers, 2003). The three other studies included only children defined as prelingual at time of implantation as these studies focused primarily on spoken language outcomes.

4.1.2 Description of investigated factors

All responses were associated to a number of factors identified in the research literature as predictors of outcome for both spoken language and social well-being. Common factors of interest for all studies were:

1. Implantation age
2. Hearing age
3. Mode of communication
4. Educational placement

Studies I and II included gender as a factor of interest. Study II further included place of living (eastern/western part of Denmark) and implantation mode, i.e. implantation of right or left ear or both ears, i.e. bilateral. Studies III and IV added on to these factors and included start of HA prior to implantation, additional disability, diagnosis, hours of support teaching, educational level of parents and parent involvement measured as attendance at child's speech and hearing therapy sessions. A total of 13 different factors were analysed in the four studies in order to identify which factors had statistical significant impact on outcome of spoken language and social well-being for two cohorts of children with CI in Denmark.

4.1.3 Description of applied tests and assessments

A total of nine different tests and assessments were applied to the two different cohorts of children with CI. The speech and language tests were included based on the criteria that they were commonly used in the international literature and also widely used in the everyday and local practice of speech and language pathologists in Denmark. It was an aim to evaluate the population based on clinically applied tests, which were recognisable for professionals working with the children with CI. Furthermore, it was an aim to apply the tests and assessments based on standards from normal hearing pediatric populations as it is of interest for both clinicians and parents to investigate whether children with CI can close the gap between implantation age and chronological age or whether they follow language development according to their hearing age with CI. Speech and language tests most often refer to scores within/without age equivalency as it is of interest to determine a child's language level according to standards of normal development. Table 2 summarises the nine tests and assessments, their area of testing and in which study each test was included

Test/Assessment	Purpose of test/assessment	Study I	Study II	Study III	Study IV
Reynell Developmental Language Scales - III, receptive part	Language understanding	x	x	x	x
"Sproglydstesten" [Speech Sound Test]. One edition.	Phonological test	x	x	x	x
Viborgmaterialet. One edition.	Expressive vocabulary test	x	x	x	x
Tait Video analysis. One edition.	Level of auditory integration	x	x		
Peabody Picture Vocabulary Test 4th edition, PPVT-4.	Receptive vocabulary			x	x
BKS. One edition.	Discrimination test of minimal pairs			x	x
CAP. Parental assessment. One edition.	Capacity of Auditory Performance		x	x	
SIR. Parental assessment. One edition.	Speech Intelligibility Rating		x	x	
Social well-being in terms of self-esteem parameters. Parental assessment. One edition.	Degree of personal-social adjustment	x		x	

Table 2: Summary of tests/assessments, purpose and application in the four studies

The Reynell – Developmental Language Scales edition III is a widely used test in assessing language comprehension internationally for both children with NH and HI. Furthermore, the test is widely used in Denmark by local speech and language pathologists in assessing children with NH and therefore the scoring was easily understood in the everyday practice of local professionals. The receptive part was used in all four studies to investigate the children's comprehension of spoken language (Reynell et al., 1985). The standard of the Reynell test starts at two years of age and therefore the children of the four studies had either to have a minimum of

24 months of hearing with cochlear implant or to be 24 months of biological age. The child had to point to objects and move objects around in accordance to 67 different questions. An example: “Place all the pink pigs around the fence outside the field”. The manual of the Reynell test defines the score according to age equivalency in 6 months intervals, i.e. a child with a chronological age of 2½ years has a mean score of 35,6 (range = 24-48 correct answers). The scoring was based on test results from a total of 231 normal hearing Danish children from age two to seven with normal hearing and normal language development. In order to acquire a score within the norm or at age equivalent level the child with CI had to score within the range for the respective hearing age or chronological age. In accordance with the standard score for the Reynell test a discrete version was defined as performance below age or equal to/above age. In the four studies the total responses related to 165 pre- and postlingual children.

Expressive vocabulary was evaluated in all studies by use of the standardised Danish vocabulary test “Viborgmaterialet”, one edition (Pedersen et al., 2005). The standardisation is based on test results from 660 Danish children with NH. The scoring of the test is either a mean score for children between 3-7 years of age and like the Reynell test defined in 6 months intervals. The score of the Viborgtesten can also be determined according to three quartiles, i.e. a lower quartile (25th percentile), a median (50th percentile) and an upper quartile (75th percentile). An example of this: A child with a chronological age of 3:6 years scored 20 points, which placed the child in a median group. This latter ordinal scoring with three quartiles was used in all four studies of the present thesis. Responses in the thesis related to a total of 113 pre- and postlingual children with CI. During a test the child was shown photos and had to verbally state the object. Responses by use of signs only were counted as error. As the standard from children with NH starts at 36 months of age, the children with cochlear implant had to have a hearing age of at least 36 months or be 36 months of age.

Speech production in terms of production of phonemes and phoneme constellations was measured in all studies by use of the Danish “Sproglydstesten” [The speech sound test], one edition. Responses related to a total of 113 pre- and postlingual children. The test is not standardised, but it is a widely used test of phonology in Denmark. It is recommended to apply the test on children with 36 months of age in order to allow time for a phonological development to have taken place. Phonological development for spoken Danish is generally regarded as occurring for most phonemes and phoneme constellations during the first 36 months of life (Kaufmann et al., 1974). Therefore, the phonological test was applied to children with at least 36 months of hearing with CI or children who were 36 months of age. The test consists of 90 different Danish phonemes and phoneme constellations. The children had to pronounce 52 different words presented as pictures in a closed-set format. The children's productions were analysed for vowels (V) and clusters of V and consonants (C). The transcriptions consisted of the following constellations: 13 V, 15 CV, 12 VC, 16 CCV, 7 SC, 5 SCC, 9 VCC and 13 Finals. The test does not contain a standardisation as it is mainly applied in clinical settings to monitor intrapersonal assessments of speech production. In order to use the scores in the present studies the results from the phonological test were recorded in an ordinal variable with four categories: A score between 76% -100% correct, a score between 51% - 75% correct, a score between 26% - 50% correct and a score between 0% - 25% correct. Study II used a dichotomous variable, i.e. under or above 50% correct. This was decided in agreement with the statistician, because 72% (N=46) scored >50 correct and the vast majority 67% (N=31) scored >75%. A total of 18 children of study II scored < 50% correct, 11 of these children (17%) scored between 25%-50% and 7 children (11%) scored <25%. Therefore, a dichotomous variable under or above 50% correct did not introduce a bias of hiding data, as most children scored either very high or very low.

The Tait Video Analysis measures level of auditory integration and was used in studies I and II and responses related to a total of 167 pre- and postlingual children. The Tait Video Analysis is a well-established method for assessment of preverbal language skills in children with cochlear implants (Tait et al., 2007). The purpose of the Tait Video Analysis is to assess the children's auditory awareness when communicating with an adult. Children are filmed in interaction with one of the testers. Turn-takings are identified, which are the instances where the child has an opportunity to communicate. When the adult pauses, the child has the opportunity to respond. The turns are then classified as vocal, where the child uses voice to communicate with or without the addition of a sign/gesture, or as a turn with sign, gesture or facial expression without vocalisation. Vocal and gestural turn-takings are each counted as a percentage of the total number of turns, which for all subjects are 20 turns. Instances where the child does not take the opportunity to communicate are classified as no response. The turns are furthermore analysed as to whether the child communicates something which cannot be directly predicted from the adult's preceding turn. Instances where the child shows this communicative initiative are counted as autonomy. Auditory awareness of the adult's speech is measured by the non-looking-vocal-turns (NLVT). NLVT is counted when the child vocalises communicatively in a turn without being in eye contact with the adult during the adult's previous turn. No visual cue, no sign or pointing finger from the adult is given. All classifications and the scoring were made in accordance with the Tait analysis (Tait et al., 2001). In studies I and II it was the score of the NLVT which was subject for further analysis. The NLVT score was defined as a dichotomous variable, classified as a score either below or above 50% NLVT, and as the majority (70%) scored >50%, this dichotomy did not present a bias in hiding data.

Studies III and IV included measuring of receptive vocabulary by use of the Peabody Picture Vocabulary Test 4th edition, PPVT-4, (Dunn et al., 2007). The PPVT-4 is a widely used norm-referenced test of receptive vocabulary both nationally and internationally. PPVT-4 is standardized on data from approximately 3,500 subjects from the US. The sample matches the U.S. census for gender, age/ethnicity, region, socioeconomic status, and clinical diagnosis or special education placement. The Danish version of PPVT-4 was translated over a period of 1½ years by one speech and language pathologist and the PhD applicant. Two bilingual (English/Danish) persons translated the Danish translations back to English. The pictures shown to the children were not altered and as there is no Danish standardisation of the PPVT-4 the Danish children with CI were scored according to scores from American children, which involve a risk of introducing a bias of sociolinguistic character. This is, however, a common problem for a small country like Denmark, where there is only a limited sale in speech-language tests and hence not a great motivation from publishers to carry-out large trials of standardisation, which are both time and money consuming. During a test children were required to point to one of four pictures that represent the word produced by the tester. Scores according to age equivalency started at 24 months of age. An example of a scoring: A child with CI and a chronological age of 2:6 years had a raw score of 41. The raw score equalled a score of 3:0 age, i.e. the child with CI scored above age. In accordance with the standard score of the PPVT-4, the score in study III and IV was resumed in a dichotomous variable below age or equal to/above age. Responses from the PPVT-4 related to a total of 68 prelingual children with CI based on their chronological age.

Studies III and IV included a Danish discrimination test of minimal pairs, BKS, one edition, (Kjær, 1977). Children had to point to the last word heard out of two minimal pairs, i.e. hus/mus, løve/løbe, næse/læse. The score was resumed in a dichotomous variable <50% correct/>50% correct. The spread of data showed that 97% (n=32) of the children discriminated >50. Responses related to a total of 33 prelingual children.

Children’s level of audition was assessed by parents with use of CAP, capacity of auditory performance. CAP is widely used by Cochlear Implant Centres and in the present thesis CAP was used in study II and III and responses related to a total of 237 prelingual children. The purpose of the CAP rating scale is to assess the children’s auditory outcomes in everyday life. CAP is a rating scale that is rapidly applied and easily understood by parents. The CAP is based on subjective assessments, but has shown to have very high inter-user reliability and the CAP is therefore well-established as a reliable outcome measure in assessing cochlear implanted children’s auditory outcome (Archbold et al., 1998). The results from the CAP assessments are coded in an eight-point scale from 0 being “No awareness of sounds” to 7 being able to “Use of telephone with a known counterpart” (Archbold et al., 1995). In accordance with the literature the two studies resumed the results in a dichotomous variable, containing information as to whether the child was able to understand at least some sentences without lip-reading or not. Therefore, a low score was defined as CAP level 0-4 and a high score as CAP level 5-7. This division did not hide data as in both cohorts there was a quasi-complete separation, 80% and 92% respectively, scored between level 5-7.

Speech intelligibility is an important object of study for individuals with significant HI, because the ability to make oneself understood is critical to most human interaction (Flipsen & Colvard, 2006). Speech Intelligibility Rating, SIR, was investigated in study II and III as an outcome measure for conversational speech intelligibility. Responses related to a total of 237 prelingual children. The SIR has been found to be able to discriminate speech intelligibility among subjects and to be highly reliable when tested for inter-user reliability. The SIR is rapidly applied and easily understood by parents and in the present thesis the parents completed SIR (Allen et al., 2001). In accordance with the literature (Allen et al., 2001) the score was defined as a dichotomous variable, indicating whether the speech is intelligible for at least an experienced listener, when the topic is known, SIR level 3-5, or whether speech cannot be understood, SIR level 1-2. In relation to the present data this dichotomy did not hide data, as the scores for the SIR test were split into two halves and the majority of both cohorts scored at level 3-5, 69% and 72% respectively.

Study I and study III included parental reports on their child’s social well-being according to a scale used on normal hearing pediatric populations (Nielsen et al., 2001). The social well-being was defined in terms of self-esteem parameters. Parents completed the assessment of their child’s level of social well-being and data related to a total of 246 pre- and postlingual children.

The assessment consisted of a seven-point rating scale to determine the degree of the child’s personal-social adjustment, by assessing whether the child was: dependent vs. independent, passive vs. active, lonely vs. social, worried vs. not worried, sad vs. happy, insecure vs. confident. In accordance with the defined score by the National Institute of Public Health, a score below 36 was defined as a low level of social well-being and a score above 36 was defined as a high level of social well-being. Maximum score = 42 and minimum score = 7.

All of the above mentioned speech and language tests and the parental assessments were analysed for potential associations between responses in a separate study by Percy-Smith (2010). Results showed that all tests and assessments were positively associated. This substantiates the observed results. Parental assessments from CAP and SIR were positively associated with results of the four tests (Reynell, Sproglydstesten, Viborgmaterialet, and Tait Video Analysis) and this finding substantiates the parental reports on their child’s auditory and speech development.

4.2 Participants

The patient material for the first two studies related to the first 200 children with CI in Denmark and the patient material for the latter two studies related to the first 100 children implanted after the introduction of UNHS and bilateral CI. The patient material comprised a total of 292 children and their families, who were all invited to participate. A total of 250 children and their families participated.

Participation rate in the two cohorts was 85% and 88% respectively. In both of the investigated populations there were more girls than boys participating. The children were implanted during a period of 18 years from 1993-2011. Parents in both populations were normal hearing except for two mothers in population one and one mother in population two, and all of these mothers were CI recipients themselves. In the second population a total of four families from the Faroe Islands and Greenland participated. All children were implanted with Nucleus product except for one boy in population two, who was implanted abroad with a MedEl product. Inclusion criteria in all studies was at least six months of hearing with CI as this would secure that the child had integrated the auditory sense and most likely was a full-time user of CI. Children with additional disability were included in all studies if they were able to manage the speech and language tests. Three children with blindness were excluded as they would not be able to see objects, pictures and photos in the tests. Some children proved not testable on the day of testing but parental assessments of audition, speech intelligibility and social well-being were included, and therefore the number of respondents from each test varied. Furthermore, age equivalence and norm for the standardised speech and language test vary, and thus each child's hearing age or chronological age determined whether the child was eligible to perform the test, this also contributed to a variance of number of respondents in each test. Parents from all studies were informed about their child's scores on the different speech and language tests. Some families wished that the local speech and language therapist received all scores and assessments and this wish was fulfilled.

Aetiology of the HI was recorded for every child, but it is only in study IV that diagnosis of HI was applied as a covariate of investigation for possible associations between diagnosis and outcomes. In the other studies diagnosis was used as a background descriptor of the participants. All diagnoses were retrieved from the medical record of each child at the two pediatric CI centres. Table 3 summarises the different diagnoses of the participants from the two investigated populations.

Diagnosis	**Population 1 Study I + II**	**Population 2 Study III + IV**
DLA congenita hereditaria	16%	13%
DLA congenita non specificata	59%	53%
DLA congenita postinfectiosa (CMV)	2%	2%
DLA typus incertus	9%	0%
Meningitis (pneumococmeningitis/meningococmeningitis)	9%	10%
Syndroma Pendred	1%	13%
Syndroma Other (Charge, Wardenburg, Usher, Roger, Klippel-Feil)	4%	4%
Auditory Neuropathy spectrum	None	5%

Table 3: Distribution of diagnoses of participants from the two cohorts

The diagnoses were more or less equally spread between the two populations. DLA congenita non specificata was the most frequent diagnosis of both populations. DLA typus incertus was only present in the first population and auditory neuropathy was only present in the second population. Furthermore, the difference between Syndroma Pendred was noteworthy, i.e. 1% and 13% respectively for the two populations.

4.2.1 Children with CI and additional disability

In both cohorts children with CI and additional disability were invited to participate. This was decided because all studies were countrywide survey studies aimed at investigating outcomes for all children with CI in Denmark. In the first cohort three congenital deaf blind children with CI were excluded for reasons listed above. Information about the additional disability was retrieved from the child's medical record at the two CI centres. If the additional disability was not diagnosed and stated in the child's medical record it was not considered. In study IV the additional disability was used as a covariate in order to study a possible association between outcomes and the presence of an additional disability.

In population one only three children were diagnosed with additional disability. Two children were diagnosed with cerebral palsy and one child had Down's syndrome. In population two a total of 21 (22%) children were diagnosed with additional disability. This included vision problems (n=8), mental retardation (n= 8), cerebral palsy (n=2), club foot (n=1) and epilepsy (n=2).

4.2.2 Participants who declined to participate

In the first cohort 30 children and families did not participate. Eleven families (6%) actively declined and five of these families had a child diagnosed with additional disability. Nineteen families did not show up on day of testing or it was not possible to get in touch with them. Other than the five families with a child with CI and additional disability there were no common denominators in terms of gender, age at implantation, hearing age and place of living.

In the second cohort 11 families declined to participate for various reasons. There was no general picture seen for these families and also no common denominator was found in terms of gender, age at implantation, hearing age and place of living.

4.3 Statistical analysis

In all studies the statistical analysis was conducted by the Department of Biostatistics at the University of Copenhagen, hence data was analysed by statisticians who were not part of a CI team at any of the centres, thus securing a separated and objective data analysis. The statisticians had full responsibility of applying the statistical model which was considered relevant for each data sample. All tests were scored by the same speech and language pathologist, i.e. the PhD applicant.

In order to study associations between the factors of interest and the responses from the various tests/assessments data from studies I, II and IV were analysed with Fisher's exact tests and logistic regression. The estimated odds-ratios and confidence intervals were based on Wald tests, whereas p-values for covariates with more than two levels were based on likelihood-ratio tests. The odds ratio estimates, OR, were presented with a reference. An example of this was analysis of parental mode of communication; the reference was spoken language compared to spoken language combined with signs. An odds ratio estimate of 61.82 meant that children whose parents used spoken language have 61.82 times higher odds at performing at age equivalent level in the Reynell test compared with children whose parents used spoken language and signs. Another example but with different values was the analysis of age at implantation; the reference was implantation age between 5-11 months compared to implantation age between 12-17 months or 18+ months. An odds ratio estimate of 0.25 for implant age 12-17 months meant that children implanted in that period had only got 0.25 times the odds at performing at age equivalent level in the PPVT-4 test compared to implant age between 5-11 months.

Study III aimed at comparing outcomes between Eastern and Western part of Denmark. In order to study differences and similarities between place of living the data analysis comprised a comparison of categorized outcome between East and West with use of the Chi-square test or the Fishers exact test (when $N<5$ in any category). A statistical significance level of 5% is chosen.

Furthermore, studies III and IV included a sensitivity analysis between the four different speech and language pathologists, who tested the children. Results showed marked differences between regions in Denmark and it was decided to further study this difference in terms of a possible introduced bias from the four different testers. Some children were tested by a single tester and some children were tested with two testers present. By comparing logistic regressions including rater information with simple statistical models only including place of living, potential inter-rater differences were assessed. As East had more non-testable children than West, a sensitivity analysis was carried out.

4.4 Ethical aspects

All studies were of methodological character, and hence the studies did not involve any experimental design nor were any children with CI exposed to any experimenting issues, e.g. studying of function with and without CI. All personal data on children and families were kept anonymous and the child’s personal number was not used. All testing of children was only carried out when there was a signed approval from the child’s family or from the child’s legal caregiver. All results were kept anonymous and only provided to other professionals when parents had approved of that in writing. All photos and videos of children and family were only used after parents had approved of that in writing. There was no conflict of interests in any of the studies.

5. Summary of results

The following provides a summary of the results from each of the four studies. Comparisons between results are shown in order to investigate possible differences and similarities before and after introduction of UNHS and bilateral CI's.

5.1 Study I

Title: Factors that affect social well-being of children with cochlear implant

Objective

The aim of the study was to identify factors associated with level of social well-being for the first 200 children with cochlear implant in Denmark and to estimate effect-related odds ratios for the children's well-being. A secondary aim was to analyse associations between speech and language level and level of social well-being.

Methodology

Participants were 91 girls and 76 boys (N = 167) with either prelingual or postlingual status at time of implantation. The children were all implanted in Denmark between 1993 and 2004 at the two pediatric CI centres. Mean implantation age was 48 months and ranged from six months to 17 years. Age at day of testing ranged from 1 to 18 years with a mean of seven years. 164 children were unilaterally implanted and three children were bilaterally implanted and all children used a Nucleus product. Data were collected from August 2004 until February 2005. The inclusion criterion was implant use for a minimum of six months, in order to assure that the child had integrated the auditory sense. All parents were normal-hearing except for two mothers who used cochlear implants themselves. Parents completed the rating scale of their child's social well-being and scores were recorded as a low level, i.e. score under 36 and high level, i.e. score above 36. Five variables were studied in order to identify factors with impact on level of social well-being. The factors included: hearing age, operation/implantation age, educational placement, communication mode and gender.

In order to study possible associations between level of speech and language and level of social well-being four different speech and language tests were used. They included Tait Video analysis, Reynell test, Viborgmaterialet and Sproglydstesten [Speech Sound Test]. Both background variables and outcomes from the four speech and language tests were compared to scores from the assessment of the child's social well-being.

In order to study and describe the relationship between the response and the variables of interest, logistic regression models were used for dichotomous response. The Fisher's exact test was used to analyse possible associations between the level of social well-being and speech and language outcome data. A positive association between two variables indicated that children who performed well in one of the speech and language test in question would also be assessed to have a high level of social well-being.

Results

All children detected the six sounds of the Ling test. The mean overall social well-being score was 36.27. Sixty children (36%) had a low level of social well-being and 107 children (64%) had a high level of social well-being. The distribution of scores revealed that 82% of the children who were exposed to spoken language had a high level of social well-being, as opposed to 37% and 40% of the children who were exposed to sign support or sign language respectively. The association between level of social well-being and communication mode was highly statistically significant, p- value = 0.0006. Gender was also found to impact level of social well-being, and results showed that 70% of the girls were assessed to have a high level of social well-being as opposed to 57% of the boys. This difference was statistically significant, p-value = 0.0476. The variables educational placement, hearing age and implantation were not statistically significant associated with level of social well-being. Further quantification of the statistically significant factors in terms of estimation of odds ratios showed that children with spoken language had 7.64 higher odds of having a high level of social well-being than children with some degree of sign language. Children whose parents used sign supported communication had 3.01 higher odds of having a high level of social well-being than children who were exposed to sign language. The estimations based on the effect of gender showed that girls had 1.98 higher odds of having a high level of social well-being.

The associations between the responses from the four speech and language tests and the social well-being assessments revealed that a high level of social well-being was positively associated with level of language understanding (Reynell test), a large vocabulary (Viborgmaterialet) and level of speech production (Sproglydstesten). Data from the Tait Video analysis and the social well-being assessments were not positively associated.

Conclusion

Study I demonstrated a very strong effect of the parental communication mode on the level of social well-being. The most marked effect was found between children whose parents used spoken language as communication mode and children whose parents used sign language. Children who were exposed to their parents' native spoken language were assessed to have higher levels of social well-being. Furthermore, there was a positive association between level of spoken language and level of social well-being in terms of language understanding, vocabulary and speech production.

5.2 Study II

Title: Parental mode of communication is essential for speech and language outcomes in cochlear implanted children

Objective

The aim of the study was to identify factors associated with speech and language outcomes for prelingual children with CI and to estimate effect-related odds ratios for each factor in relation to the children's speech and language performances.

Methodology

Study II forms part of the same cohort as used in study I, but 13 postlingual children were excluded in this study. This was done in order to specifically investigate the factors underlying the mechanisms of spoken language development in children with CI. Material comprised a total of 155 children, 85 girls and 70 boys. Mean implantation age decreased to 36 months when the 13 postlingual children were removed. Mean age at day of testing decreased by one year and was for this population 6 years. All other specific characteristics of the population were the same as in study I.

In study II two more variables were included, i.e. place of living and implantation mode. Seven factors were thus included in study II: hearing age, operation/implantation age, educational placement, communication mode, gender, place of living, i.e. belonging to the East or West Danish pediatric CI centre, and mode of implantation.

The speech and language tests used were the same as in study I, i.e. Tait Video analysis, Reynell test, Viborgmaterialet and Sproglydstesten [Speech Sound Test]. Two parental assessments were included, i.e. Capacity of Auditory Performance, CAP, and Speech Intelligibility Rating, SIR.

In order to study the relationship between the responses and the factors of interest data were analysed with logistic regression models. Backwards elimination was used to choose a final model that only included the statistically significant results. Further quantification of the statistically significant factors in terms of estimation of odds ratios was carried out.

Results

Parental communication mode was the only factor with statistical significant effect on all responses from the tests and assessments. A statistical significant association was found between hearing age and the responses from Tait analysis and SIR. Gender had a statistically significant effect on Reynell test, Sproglydstesten [Speech Sound Test] and Viborgmaterialet. Implantation age was significantly associated with responses from the Viborgmaterialet. Place of living was significantly associated with responses from the Viborgmaterialet and CAP. Educational placement and mode of implantation had no statistical significant associations with any of the responses.

Forty children performed below age and 48 children equal to or above age on the Reynell test. Responses revealed extremely high odds ratios for children exposed to spoken language as compared to children with sign support and sign language, i.e. 61.82 and >100 respectively. Gender was also found to affect responses from the Reynell test in favour of girls, OR = 6.98.

Fifty children scored below 50% non-looking vocal turn, NLVT, and 104 children scored above 50% NLVT in the Tait Video analysis. The communication mode at home had the greatest effect on outcomes of the Tait Video analysis. Odds ratios were 28.00 times greater for children exposed to spoken language as opposed to children exposed to sign language from their parents.

Responses from the Sproglydstesten [Speech sound test] revealed that 18 children scored under 50% correct and 43 children scored equal to or above 50% correct production. Two variables were significant for outcome, i.e. communication mode and gender. Children with spoken language had 41.25 greater odds of scoring high in the Sproglydstesten [Speech sound test] than children exposed to spoken language combined with signs. Girls had greater odds than boys of correct phonological production, OR = 3.73.

The distribution of responses from the Viborgmaterialet showed that 23 children performed in the lower group and 38 children in the medium/upper group. Four variables were significantly associated with outcomes, i.e. communication mode at home, gender, place of living and implantation age. Again extremely high odds ratios were found for children exposed to spoken language as compared to children with sign language, OR = >100. Odds ratios for children exposed to sign support were 21.20 times higher when compared to children exposed to sign language. Children from the Eastern part of Denmark had 14.45 greater odds of a score in the medium/upper group than children from the Western part of Denmark. Girls had 7.03 greater odds than boys. Children implanted before 36 months had 0.06 greater odds of performing in the medium/upper 50% group.

The distribution of responses from the CAP revealed that 31 children scored at the low level and 124 scored at the high level. Thus 80% scored at high level and data were quasi complete and therefore the estimation of odds ratio of comparing children with spoken language and sign language did not provide any further information. However, when comparing children with sign support as opposed to children with sign language odds ratios were 6.31 times higher for children with sign supported communication mode at home.

The distribution of responses from SIR revealed that a majority of children (n=103) scored at high level, and 52 children scored at low level. Communication mode at home and hearing age were found to have significant effect on outcome of the SIR. The odds ratios for children exposed to spoken language were 10.66 times higher than for children exposed to sign language. Children with a hearing age >36 months were more intelligible than children with shorter periods of hearing/listening with CI.

Conclusion

The quantification of the effects showed that children whose parents used spoken language as communication mode had greater odds of scoring high in all tests and assessments. The greatest difference was found between children with spoken language and children with sign language. Children whose parents used spoken language combined with sign support had greater odds of scoring high on all responses in comparison with children whose parents used sign language. As for study I the parental mode of communication had a very strong effect on outcome both as regards to auditory capacities and speech/language outcomes. Furthermore, a noteworthy finding was the impact of place of living on the vocabulary test.

5.3 Study III

Title: Significant regional differences in Denmark in outcome for children with cochlear implants.

Objective

The primary aim was to investigate national effect and regional differences and similarities in outcome for a pediatric population with CI after the introduction of UNHS and bilateral cochlear implantation in Denmark.

Methodology

The patient material comprised a total of 94 children with CI, 52 girls and 42 boys. The sample included all children with CI in Denmark, who were born between January 2005 and January 2011 and who had a minimum of 6 months of hearing with their CI. Forty-nine children were implanted at the East Danish CI Centre, one child moved from abroad to the Eastern part of Denmark and was already implanted, and 44 children were implanted at the West Danish CI centre. Seventy-four per cent (N=69) of the children were diagnosed through UNHS. Twenty-five children were diagnosed later due to various reasons i.e. four children came from Greenland and the Faroe Islands where UNHS had not been implemented, eight children had hearing impairment following meningitis and thus were not born with a hearing loss, the remaining 13 children were not found through UNHS for reasons unknown. Participation rate was 88% (N=83). Twenty-two per cent (N=21) of the children were diagnosed with additional disability. All parents were normal hearing except for one mother who was a CI recipient herself. Ninety-three children had a Nucleus product and one child had a Med El product. Median age of start of hearing prior to implantation was four months. Mean age of implantation was 19.6 months (median age =14 months). Mean age at day of testing was 46.3 months (median age = 47 months). The mean age of hearing with CI was 25.9 months. Seventy-eight (83%) of the children were bilaterally implanted, 68% simultaneously and 15% sequentially.

As study III aimed at investigating similarities and differences between regions in Denmark comparisons were carried out of the distributions between East and West of the following ten factors: age at start of hearing age, age at implantation, age at day of testing, mode of implantation, number of hours of speech and hearing therapy and support teacher, parent participation in speech and hearing therapy sessions, parental mode of communication, educational placement and parents' paid compensation.

Eight different speech and language tests and assessments were applied in study III: Peabody Picture Vocabulary Test fourth edition, (PPVT- 4), the Reynell test, Sproglydstesten [Speech Sound test], Viborgmaterialet, BKS test (discrimination of minimal pairs), CAP, SIR and Self-esteem.

Results

The responses from the receptive vocabulary test showed a statistical significant difference between East and West, $p<0.001$, with better score in the east. The same was found for the test of receptive language, p-value = 0.005, speech production p-value = 0.045 and active vocabulary p-value = 0.058. Responses from CAP, SIR and BKS were similar. The parents' assessments of their child's level of self-esteem were statistically significant different from the two regions, p-value = 0.005, with higher level of self-esteem for children from the eastern part of Denmark.

Potential rater variability within either of the centres was assessed using logistic regression models. Except for SIR no significant rater variability was found. This supports that regional differences were not an artefact caused by rater variability.

Furthermore, other regional differences were investigated, i.e. number of hours of habilitation per week, parent participation in the speech and hearing therapy session, number of hours of a support teacher per week, the parental mode of communication, educational placement and paid reduction of work hours of parents. Regional differences were found for these variables except for paid compensation for the reduction of work hours. In the western part of Denmark 71% of the parents stated that they did not participate in the habilitation of their child compared to 37% in the East and this difference was statistically significant, p-value = 0.001. Ninety-five per cent of children from the West received 1-2 hours of rehabilitation per week compared to 53% from the East, p-value < 0.001. Fifty-three percent of the children from the West were provided with a support teacher for more than 15 hours per week compared to 21% from the east, p-value =

0.017. Twenty-four per cent of the parents from the West stated that they used a combination of spoken language and signing as communication mode in comparison to 7% from the East, p-value = 0.031. Ninety-six percent of the children from the East are placed in mainstream educational settings in comparison with 73% of the West children, p-value = 0.0005. There was no statistically significant difference found in terms of parents' amount of economical compensation for reduced work hours, i.e. 61% and 59% of parents from East and west respectively were paid for having reduced hours.

Conclusion

The responses from the auditory and speech intelligibility assessments were similar between regions in Denmark. On all other investigated tests/assessments there were statistically significant differences between regions with poorer results in the western part of the country. Thus, place of living significantly affected spoken language outcomes and there were statistically significant differences in demographic related factors. Furthermore, children from East were assessed to have a higher level of social well-being in terms of self-esteem parameters.

5.4 Study IV

Title: Language understanding and vocabulary of early cochlear implanted children

Objective

The aim of the study was to identify factors associated with the level of language understanding, the level of receptive and active vocabulary, and to estimate effect-related odds ratios for cochlear implanted children's language level.

Methodology

Study IV formed part of the same cohort as used in study III, therefore the characteristics of the population were similar in terms of all background variables, i.e. age at start of HA, age at implantation, age at day of testing, mode of implantation, number of hours of speech and hearing therapy and support teacher, parent participation in speech and hearing therapy sessions, parental mode of communication, educational placement and parents' paid compensation.

In order to further study the specific language levels in terms of language understanding, receptive and active vocabulary responses from three different tests were analysed, i.e. the

PPVT-4, the Reynell receptive part and Viborgmaterialet. These responses were chosen as objects for further analysis, because the literature describes them as being parameters of great importance as they are defined to be the most vulnerable parameters in language development (Salén & Nettelbladt, 1992).

Data were analysed with Fisher's exact tests and logistic regression. In the logistic models, the Viborgmaterialet had been dichotomized into below or above 75%. The estimated odds-ratios and confidence intervals were based on Wald tests, whereas p-values for covariates with more than two levels were based on likelihood-ratio tests. Four different raters, two in each region, tested the children. By comparing logistic regressions including rater information with simple models only including region of residence, potential inter-rater differences were assessed. As East had more non-testable children than West, a sensitivity analysis was carried out.

Results

Seventy-one children were tested with the Reynell receptive part and 48 (68%) of the children did not have age equivalent language understanding. Analysis of data using logistic regression revealed that children from East had 4.72 times higher odds of scoring at age equivalent level compared to children from West. The logistic regression analysis, furthermore, showed that age of HA fitting pre-implant, age at implant, amount of support teaching and educational placement were statistically significantly associated with the outcome. Parental mode of communication was also significantly associated with the outcome, when evaluated by Fisher's exact test (odds ratio estimates were infinite, as no children using sign support when communicating with their parents scored at age equivalent levels).

Sixty-eight children's receptive vocabulary was evaluated by the PPVT-4 and 45 children (66%) did not have age equivalent vocabulary. Analysis using logistic regression showed very high odds ratio estimates for children from the East for having an age equivalent receptive vocabulary compared to children from the West. Age at implantation was also statistically significant and children implanted between 5-11 months had higher odds ratios compared to children implanted between 12-17 months or older than 18 months. Mode of implantation was borderline significant with better outcomes for children with two CIs implanted either simultaneously or sequentially. Fisher's exact tests were significant for the covariates diagnosis, age at HA fitting pre-implant and communication mode of parents.

Forty-nine children were tested with Viborgmaterialet and the majority of the children (N = 22; 45%) scored in the lowest category with 0-25% correct. Logistic regression analysis and Fisher's exact test showed that age at implantation and length of hearing with CI were statistically significant. Children implanted simultaneously bilaterally had higher odds of performing at age equivalent levels than children with bimodal stimulation or sequentially implanted.

For all three tests there were no significant rater effect, p-values for PPVT-4 = 0.124, Reynell = 0.214 and Active Vocabulary = 0.397. More children in East were not testable, which may be a source of bias. In order to explore this risk, a sensitivity analysis was applied to the data sample. The superiority of the test outcomes at the Eastern CI centre was challenged, as the non-testable children were allocated to the lowest performing group for the three tests. The results of the Fisher's exact test and comparisons to the original test results showed no statistically difference between East and West. Thus, there was no reason to suspect a rater bias, i.e. to fear that the stated results as regards to regional differences were an artefact caused by different criterions for judging a child non-testable.

Conclusion
The group of children implanted after introduction of UNHS and bilateral CI were still challenged in closing the language gap between hearing age and chronological age. Place of living impacted outcome and parental mode of communication still had a significant effect on outcome.

5.5 Results between studies

In order to investigate similarities and differences between the two groups of children prior and post introduction of UNHS and bilateral CI, further analysis of distribution of variables and responses from tests and assessments were carried out. Data relate only to prelingual children when responses from speech and language tests were studied. For responses from the self-esteem assessments all children between studies were compared. All standardized tests using a score of age equivalency were for the sake of relevant and valid comparisons scored according to hearing age with CI. Table 4 summarizes the distribution of relevant comparable variables, i.e. mean hearing age, mean age at day of testing, mean age at implant, mode of implantation, educational placement and communication mode.

	Prior UNHS (n=155)	**Post UNHS (n=83)**
Mean hearing age with CI	28 months	26 months
Mean age at day of testing	72 monhts	47 months
Mean age at implantation	36 months	20 months
Mode of implantation		
bilateral	2% (n=3)	82% (n=68)
unilateral	98% (n=152)	18% (n=15)
Educational placement		
Mainstream	38% (n=59)	86% (n=71)
Special	62% (n=96)	14% (n=12)
Communication Mode		
Spoken Language	26% (n=40)	84% (n=70)
Spoken + Sign	54% (n=85)	14% (n=12)
Sign Language	20% (n=30)	0% (n=0)
		1 missing

Table 4: Distribution of direct comparable variables

Table 4 shows some marked differences between the two groups. Mean age at implantation decreased with 16 months between the populations. Mode of implantation changed from 2% to 82% of children receiving bilateral CI's. Educational placement changed from 38% to 86% of children being enrolled in mainstream educational settings. Communication mode changed from 26% to 84% of parents using spoken language. No parents used sign language as communication mode with the children in the post UNHS group.

Similarities and differences of various test responses were recorded for the Reynell test, Viborgmaterialet, Sproglydstesten [Speech Sound Test], CAP and SIR. Scores from the Viborgmaterialet were dichotomous and categorized into lower 50% and upper 50%. Table 5 summarizes the distribution of the responses.

	Prior UNHS		**Post UNHS**		
Tests	>age/low score	<age/high score	>age/low score	<age/high score	P-value
Reynell	45,5% (n=40)	54,5% (n=48)	30% (n=11)	70% (n=26)	0.1148
Vocabulary	37,7% (n=23)	62,3% (n=38)	24% (n=6)	76% (n=19)	0.3157
Phonology	29,5% (n=18)	70,5% (n=43)	18,4% (n=9)	81,6% (n=40)	0.1907
CAP	20% (n=31)	80% (n=124)	7,3% (n=6)	92,7% (n=76)	0.0135*
SIR	33,5% (n=52)	66,5% (n=103)	28% 8n=23)	72% (n=59)	0.4632

* = statistically significant
Reynell + Vocabulary; scores according to hearing age
Phonology; 0-50% correct = low score; 51-100% correct = high score

Table 5: Distribution of scores prior and post UNHS

The only scores proved to be statistically significant were the CAP assessments, P-value = 0.0135 respectively. However, children from the post UNHS group scored percentage wise higher on all tests and assessments and these findings indicated a positive development of listening and spoken language since the introduction of UNHS and bilateral CI.

Table 6 presents comparative data from the assessments of both prelingual and postlingual children's self-esteem. It must be taken into account that this comparison was between two rather different age groups in terms of the variable mean chronological age at day of testing. The population prior to UNHS and bilateral CI had a mean age of day at testing of 84 months and the post UNHS population had a mean age of day of testing of 47 months, i.e. an age difference of 37 months. As can be seen the difference between groups in terms of level of self-esteem was highly statistically significant, P-value = 0.0001.

	Prior UNHS		**Post UNHS**		
Assessment	>36/low score	<36/high score	>36/low score	<36/high score	P-value
Self-esteem	35,9% (n=60)	64,1% (n=107)	12,7% (n=10)	87,3% (n=69)	0.0001*

* = statistically significant

Table 6: Distribution of scores of self-esteem pre- and postlingual children

6. Overall discussion

The first generation of children with CI in Denmark showed high levels of auditory capacities for populations both prior and post introduction of UNHS and bilateral CI. Results from the four studies showed that the majority of children with CI were assessed by their parents to have a high level of social well-being and self-esteem with the younger group having the highest scores. However, results showed that spoken language outcomes when measured according to hearing age was a challenge for the first cohort of children and closing the language gap between hearing age and chronological age was a challenge for children of the second cohort despite early implantation and bilateral CI. In the following some of the overall results will be discussed with emphasis on clinical implications.

6.1 Impact of mode of communication

Results from all studies showed that the variable parental mode of communication was a highly significant factor with impact on all investigated aspects of levels of audition, spoken language and social well-being. Children who were exposed to their parents' native spoken language had greater odds of performing at age equivalent language level or a level equivalent to their hearing age with CI. The largest long-term study addressing this issue is carried out by Geers et al. (2011). They investigated long-term outcomes and found that children who relied on total communication to supplement their spoken communication and who continued to rely on signs demonstrated lower performance on many speech/language outcome measures relative to children, who used oral communication. Many other studies have found the same effect of mode of communication and thus the findings of the present thesis were in accordance with the international literature, where communication mode was described to significantly impact spoken language outcomes of CI (Dettman et al., 2013; Dornan, 2010; Flexer, 2011; Fulcher et al., 2012; Ganek et al., 2012; Geers & Brenner, 2003; Geers, Brenner & Davidson, 2003; Geers, Nicholas & Sedey, 2003; Niparko et al., 2010; Wie et al, 2007).

Parents' mode of communication changed markedly during the period in which data for the first generation of children with CI was collected. In the data collection in 2004-2005 the majority of children were exposed to either total communication or sign language, and in 2010-2011 only 14% of the parents used total communication with their children and no parents communicated by means of sign language. However, the majority of children in the latter group were still challenged with developing age equivalent language levels. This finding emphasised that even though parents changed communication mode there was still a challenge in providing the best language input in order to ensure age equivalent spoken language development for children with CI.

Results from study III and IV underlined that optimal outcomes of CI intervention was not only a matter of using spoken language, it was probably rather a matter of providing the right input at the right time. Niparko et al. (2010) argue that maternal engagement in early communication reflects greater scores of parent-child interactions, which is associated with increased development in spoken language skills. It was, furthermore, stressed that spoken language exposure and mentoring of caregivers provided the context for language learning. Yoshinaga-Itano (2014) argued that the intervention must be early and start from the day of detection of the HI in order to ensure intact relation and communication between mother and child. Fulcher et al. (2012) further argued that early intervention with fitting of HA by 3 months and enrolment in family centred auditory verbal intervention by 6 months allowed children to close the language gap and develop age equivalent receptive spoken language already at 3 years of age. In contrast to these arguments, Sue Archbold (2010b) argued that with very early implantation, communication mode was unlikely to be well established prior to implantation. Archbold argued that the development of communication skills after early diagnosis and early implantation was likely to bring together vision and audition and language can be learnt in a more natural way, as with hearing babies, and therefore parents did not need to make a choice of communication mode. However, the results of study IV did not indicate that spoken language developed naturally despite early diagnosis and early implantation for Danish children with CI.

The results from study IV were in line with Wie's (2010) study, which evaluated receptive and expressive speech and language levels of early bilaterally implanted Norwegian children. Sixty percent of the Norwegian children with hearing age of 24 months scored at age equivalent level for receptive language and 55% scored at age equivalent level for expressive language. These results were better than for Danish children of study III and IV but still not as convincing as the results of Fulcher et al. (2012), where 100% achieved age-appropriate language development and 96% age-appropriate speech at 36 months of biological age. Norway and Denmark are comparable in their clinical set-up and in the content of habilitation. The Norwegian children received "mainly auditory verbal, AV", but as for the Danish children, it was not certain what kind of intervention "mainly AV" really was and furthermore to what extent parents were involved in the intervention. Only 44% of Danish parents in study III and IV participated in the speech and hearing therapy sessions. If the habilitation was mainly carried out in institutions, i.e. nurseries, kindergartens, schools, without parents receiving guidance in how to communicate with their child with HI, then they risked missing out on important language learning opportunities in everyday life (Moeller, 2000).

It must be kept in mind that all of the children involved in the four studies were born before the introduction of the initial one year of auditory habilitation post implant at the two pediatric CI centres (Sundhedsstyrelsen, 2012). These guidelines emphasise parental involvement and use of audition and spoken language during the first year following implantation. However, these guidelines have not yet been evaluated in terms of documenting a possible positive effect on spoken language outcomes for children with CI in Denmark. Furthermore, the overall aim of the guidelines is only to provide an initial one year of auditory habilitation in order to ensure well-functioning auditory processing with the implants, which is done in close collaboration between multi-disciplinary staff of medical, technical and speech/language pathology background. Thus families need further and long term guidance in order to ensure learning of all aspects of a spoken language, e.g. understanding of syntax, grammatical constructions, theory of mind and vocabulary use in coherent speech. Zaidman-Zait & Young (2008) emphasised this point by stressing that the habilitation process following pediatric cochlear implantation rests heavily on parental involvement and continuing habilitation efforts are necessary after cochlear implantation in order to make the procedure effective.

In order to fully understand the impact of communication mode it is relevant to discuss the aspects of communication processed in different modalities, which is foreign to normal hearing parents. The complexity for normal hearing parents of using sign support and sign language was discussed in the literature. It was discussed that when normal hearing parents communicate with their child in a foreign communication mode, i.e. total communication, and in a foreign language, i.e. sign language, it may lead to language deprivation for the child, as a foreign language communicated in a different sensory modality risks not to be as rich and vivid as parents' native language (Archbold, 2010b; Knoors & Marschark, 2014; Sundqvist et al., 2014). This argument may contribute to explain the results of the spoken language outcomes of the participating children of the first cohort. For this cohort it was necessary to score the speech and language tests according to hearing age with CI, because the chronological spoken language level was found to be so low that it made no sense to score responses according to the children's biological age. Scoring according to hearing age allowed each child the time to acquire spoken language level according to length of use of the CI. However, results indicated that the children did not perform as well as reported in the literature in terms of spoken language understanding, as results showed that only 55% of the children scored equal to or above hearing age level. At the same time, it is relevant to stress that 80% of the children were assessed to score high on auditory capacity and all children detected the six Ling sounds, and thus had auditory access to sounds across the whole frequency range of speech. It was, however, noteworthy that despite these high auditory levels and the scoring according to hearing age the children of the first cohort showed relatively poor language levels in terms of spoken language understanding. Such results indicated that providing auditory access via the CI technology did not ensure spoken language development and indicated that listening and spoken language need to be taught.

The statistically significant effect of mode of communication on level of social well-being may also be understood by the complexity of normal hearing parents communicating in a foreign mode or a foreign language with their child with CI. Children of study I had greater odds of scoring high on self-esteem parameters when parents used their native spoken language as compared to parents who used supportive signs or sign language. In the literature it is discussed that there are obvious social emotional benefits for a child of being able to have the same native language as the parents. Deaf children of deaf parents are rated better on various social emotional scales compared to deaf children of hearing parents with a plausible explanation being that deaf children have a common native language with their deaf parents (Nicholas & Geers, 2003).

6.2 Impact of age at implantation

Age at implantation was described to significantly affect outcomes in terms of language and social well-being (Connor et al., 2006; Dettman et al., 2007; Niparko et al., 2010; Tait et al., 2007; Vlastarakos et al., 2010; Wie, 2010). Furthermore, age at implant was shown to significantly affect long term spoken language achievements (Nicholas & Geers, 2013) and long term outcomes in terms of language specific properties of social skills (Geers et al., 2013). However, the impact of age at implantation was only statistically significant for Danish children involved in study II and IV. Why was it found that Danish children of the four studies were not affected by age at implantation for neither spoken language development nor social well-being to the same extent as reported in the literature? It is noteworthy, because mean implantation age in study II was 3 years and age at implant only affected outcomes of the vocabulary test. Implantation was carried out within the 3.5-year period, where the brain was still plastic and development of intact auditory pathways was possible, which again provided opportunities for development of a spoken language (Flexer, 2011; Sharma et al., 2005, 2009). One possible explanation may be that communication by means of signs masked the effect of early implantation. Ona Bø Wie (2010) argued that implantation within a certain age may be a prerequisite for equivalent language development but that the effect of age at implantation will become less evident as other factors become important over time. The neurological literature stressed the importance of using the implant, which is done by means of auditory and spoken language stimulation; otherwise opportunities for development of the auditory pathways are lost (Borchgrevink, 2001; Gordon et al., 2011; Kral & Eggermont, 2007).

Based on these assumptions it could be argued that when total communication and sign language were used in 70% and 74% of the cases in studies I and II, then deaf born children will continue to primarily rely on visual stimuli for communication, and hence the auditory pathways did not fully develop and opportunities for development of age appropriate spoken language levels were diminished. According to Yoshinaga-Itano (2014) it is a question of the quality of the input rather than the mode of communication. It is therefore relevant to question the quality of the communication between parents and child in studies I and II. Dornan et al. (2010) emphasised that an education approach where development of the auditory brain pathways through listening and spoken language was crucial in order for the child to fully use the opportunity to learn to listen and speak. Flexer (2011) added to this by emphasizing that learning to listen is time-bound and has early closing windows of time. Doreen Pollack expressed the dilemma for children who wear hearing technology but continue to rely on vision for communication in the following statement:

> I learned that one could not simply hang a hearing aid on a child and expect them to develop hearing perceptions normally. Instead the children continued to act as if they were deaf. Sound was meaningless. When the children were encouraged to use lip-reading or signing, they continued to be visual learners and ignored sound. I came to realise that one did not have to teach deaf children to look but instead one had to teach them to listen. A hearing aid gave more hearing, but listening had to be learned. I had to make sound an important and meaningful part of everything the children were hearing (Pollack et al., 1997).

This statement summed up the dilemma of applying hearing technology to deaf children without a simultaneous educational approach with emphasis on exploiting the auditory stimulation provided from the hearing technology, e.g. hearing aids and cochlear implants.

The neurological arguments contributed to explain the relatively poor outcomes of the children in the four studies in terms of spoken language understanding and vocabulary, as the children and families were not reported to have had structured and formal educational options with emphasis on listening and spoken language. Some local speech and hearing therapists have practised in providing listening and spoken language guidance for families with children with HI, but distribution of this educational option was determined by place of residence. The auditory verbal approach is an educational intervention specifically described and specifically targeted the clinical group of children with HI and their families and auditory verbal practice is defined as a family-centred approach and an applied science with its objectively measured goals (Estabrooks, 2012). In recent guidelines from Socialstyrelsen [The National Board of Social Services] (2015) AVT was recommended as intervention for children with early detection of HI, whose parents wished to pursue listening and spoken language for their child. The AV intervention was recommended, because this intervention was based on evidence and documentation of outcomes for children with HI and families. At the same time, it was stressed that further research in types of intervention for children with HI is warranted (Socialstyrelsen, 2015).

The importance of an early family-centred approach was stressed repeatedly in the literature (Ganek, 2012; Knoors & Marschark, 2014, Moeller, 2000; Niparko et al., 2010; Yoshinaga-Itano, 2014). Quittner et al. (2013) found that effects of maternal sensitivity on growth of language were similar to that found for age at cochlear implantation. Szagun & Stumper (2012) added to this finding with their results showing that properties of maternal language input, mean length of utterances and expansions were associated with faster linguistic progress independently of age at implantation. These findings suggested that addressing parenting behaviours is a critical target for early language learning both prior and post implantation. It was therefore noteworthy that the majority of Danish parents in studies III and IV did not participate in the speech and hearing therapy sessions. This raises the question of whether parents had received up-to-date evidence based guidance on how to communicate with their child pre- and post-implant. According to Ganek et al. (2012), the vast majority of parents pursue a listening and spoken language development for their deaf born child with CI, and there was no reason to believe that this should be different for Danish parents. Furthermore, the results of studies III and IV emphasised the importance of informing and involving parents both in decisions on pursuing early implantation and on matters of being part of the habilitation both before and after implantation. Health care services and educational services have the responsibility of providing up-dated and evidence based information to parents in order for the parents to make an informed choice about aspects as how to pursue listening and spoken language development in accordance with their values and wishes for their congenital deaf child with CI.

A recent report from The Danish National Centre for Social Research investigated life circumstances for deaf born children with CI (Bengtsson et al., 2014). On several issues the findings from this report complemented the findings of the present thesis. One finding of the report contributed to the above mentioned statements from the literature stressing the significance of parents receiving habilitation following cochlear implantation. An association was found between level of parental concern for their child's future and support in the habilitation following cochlear implantation. The more support in the habilitation the family had received the less concerned they were about their child's future. The support in the habilitation was more important than age at implantation, additional disability, socioeconomic status and civil status of parents. This finding emphasised that habilitation involving parents has a significant impact on a broad spectrum of parameters and not only listening, spoken language and social well-being of the children but also matters regarding parental concern.

In a further analytical note to the report two other explanatory factors were included, i.e. place of living and habilitation based on principles from AVT. The most important factor to influence identity as hearing handicapped was associated with habilitation based on principles from AVT. When habilitation was based on AVT the child's identity as hearing handicapped meant less (Bengtsson & Larsen, 2014). This finding is the first specifically targeting type of intervention in Denmark. The finding indicates that habilitation based on AVT has its eligibility in Denmark, but further investigation seems warranted.

6.3 Impact of place of living

Place of living was shown to significantly affect outcomes in terms of social well-being, audition and spoken language for both cohorts of children. Study II found that odds ratio estimates of scoring in the upper 50% in the expressive vocabulary test were significantly greater for children from the East compared to children from the West. The same advantage for children from the East was found for auditory capacity. The impact of place of living was also found in study III and IV with the same significant difference that children from East had greater odds of performing better on tests of speech and language and social well-being. However, study III and IV did not find significant differences between regions for capacity of auditory performance. Furthermore, the differences between regions were not only related to child factors but also to demographic factors, and these factors showed that children in West received significantly more hours of speech and hearing therapy, more hours of support teaching, and more children attended special educational settings. Only 30% of parents from the West participated in the speech and hearing therapy sessions and more parents used total communication. Introduction of a possible bias from the four different testers was analysed but no significant difference was found, so the difference between regions cannot be explained by a tester bias. It is noteworthy that in a small country like Denmark such significant differences were repeated in two different cohorts of children with CI.

In a further analytical note to the report from The Danish National Centre for Social Research a difference between East and West was also found. The factors of this note were related to general life circumstances and hence reflected different aspects of pediatric cochlear implantation. However, one factor to influence identity as hearing handicapped was place of living, as living in the Eastern part of Denmark meant less identity as hearing handicapped. It was noteworthy that an investigation of different types of factors also found an impact related to place of living (Bengtsson & Larsen, 2014).

It is complex to fully understand, why this difference exists in various studies. Historically there have been differences in the clinical set-up at the two pediatric CI centres. At the West Danish CI centre the special educational services for children with HI have been more involved in the assessments pre implant and in both fine-tuning and habilitation post implant (Pedersen, 2007). De Raeve (2010) stressed that educational services must ensure that their staff have the skills to meet the challenges encountered by pediatric cochlear implantation. It is stressed that educational services must be flexible, continually updated with the technology and changing expectations and provide ongoing professional training in order to provide an environment which will utilise the hearing while meeting the linguistic and curricular needs of the children. Sue Archbold (2010a) further challenged the educational services for children with HI and questioned whether these services had changed enough in accordance with the medical and technological advancements.

It is relevant to put these arguments into perspective for Danish educational services specifically when considering the results of study III and IV. The children from the West received more hours of therapy per week and more hours of support teaching, but still performed poorer than their eastern counterparts. Therefore, it is appropriate to question the content and quality of the educational services and not only for the western part of Denmark, because as a whole the children in all four studies did not show outcomes of speech and language comparable to outcomes reported in the international literature. There has been a marked decrease in number of children attending special education for children with HI, i.e. 62% of the first cohort and 14% of the second cohort. This finding was in accordance with the literature (Geers et al., 2011; Huber, 2008; Lonka et al., 2011). But as for age at implantation it was only in study IV that there was a statistically significant difference between mainstream and special educational placement in terms of outcomes of spoken language understanding. This finding indicated that it is not only a matter of educational placement, but it is rather a matter of providing the right input at the right time. It is crucial to provide up-dated and flexible educational services. This argument leads to the relevant question of how up-dated and flexible educational services are quality assured in any educational service, i.e. mainstream and special education. Quality assurance emphasises the need for evidence based intervention. Sue Archbold (2010b) questions the research tradition in the field of deaf education and its relevance in the research evaluating effect of pediatric cochlear implantation. Another complexity is that practitioners of deaf education most likely do not read the research published in various journals, which introduces a risk of not being up-dated with research (Archbold, 2010b p. 28-30).

Study III demonstrated highly significant differences between regions of the factors described in the literature to significantly affect outcome: age at implantation, mode of communication, parental involvement, bilateral implantation and educational placement. The West Danish children with CI were disadvantaged on all of these factors, and it may be argued that they all contributed to the poorer outcomes of the Western children. Therefore, it can be assumed that the impact of place of living was comprised of multivariate factors with both demographic and child related characteristics. The documented impact of place of living provides a national challenge for both health care services and educational services.

6.4 Methodological issues

As described in chapter four one major limitation in studying children with CI is that the group is small and heterogeneous in most aspects like age at diagnosis, age at implantation, aetiology and various other factors (Geers et al., 2011). This is a challenge and limits the possibilities of large randomised controlled trials. Furthermore, considering the limited size of the population in a small country like Denmark there are high standards on ethical issues of not exposing the children.

The four studies did not have a control group of children with HI without CI, which precludes conclusions on causality. The aim of the studies was to explore the influence of various factors on spoken language development and social well-being for the first generation of children with CI. For ethical reasons it was not possible to randomize or match children with similar levels of hearing who continued to use hearing aids or not use any hearing technology at all. Such trials could formally test the efficacy of cochlear implantation in children who receive implants at different ages or stages of linguistic development (Niparko et al., 2010). Instead, outcomes of the children with CI were compared to normal hearing children in standardised tests. In tests without standardisation, the children with CI were scored according to predefined categories of low versus high performance, which is a well-established method within the field, as documented in the literature. Thus, outcomes of speech and language performance are most often dichotomised, as e.g. below vs. above age equivalency. The dichotomous outcome scoring holds a risk of hiding relevant data information, as all data are categorised into only two outcome groups. However, the spread of outcome data in the various tests was remarkable, as most scores were at a distance from the cut-off value between the two categories and thus clearly fell within the low or high performance category. Thus, only a few subjects scored close to the cut-off value and most were accordingly scoring either clearly low or clearly high, which justifies the categorisation from a statistical point of view.

The four studies included children who were diagnosed with additional disability. Only three of the participating children (2%) in studies I and II were diagnosed with additional disability according to the medical record and therefore the variable was not controlled for statistically but was used to describe the population. However, in studies III and IV 22% of the children were diagnosed with additional disability according to the medical record. The factor had a statistically significant effect on outcomes of receptive vocabulary and language understanding.

Congenital deaf children had greater odds of scoring at age equivalent language level compared to children with meningitis, hereditary HI, syndroma Pendred or other types of aetiology. This finding may reflect that when a child is congenital deaf it is often more straightforward to diagnose the HI and proceed with first hearing aids and later CI, and thus reducing the period of auditory deprivation. It is a challenge for health care services to further explore this difference and make sure that distribution of audiological services is equal for children with all degrees and all types of hearing impairment in Denmark.

One limitation of the study design was that it was unknown to what extent the children may have a non-recorded additional disability that will surface with time. An additional disability may affect language development even at a time when not yet diagnosed (Wie, 2010). This could be the case for a congenital deaf child with CI and additional language impairment, e.g. dyslexia. It was an interesting finding that number of children with CI and additional disabilities increased between studies, which may reflect that more and more children with additional disability are offered cochlear implantation as argued by Amirsalari et al. (2012). Five families of the first cohort who declined to participate had a child with additional disability and three children were excluded due to blindness. Such findings suggest that future studies should address this particular clinical group with a different study design aiming at evaluating quality of life outcomes rather than spoken language outcomes, which is also stressed in the literature by Nicholopoulos et al. (2008).

The four studies were based on two “snapshots” in time and it is a challenge to compare outcomes for groups of children with CI over a period of 18 years. Cochlear implantation is technologically driven and the advancements of both internal and external parts have undergone huge changes during this period (Clarke, 2004), thus comparisons involve the risk of introducing a bias based on technological aspects. However, 249 children of the present thesis were implanted with a Nucleus Cochlear product and therefore the bias in comparing children over a long period of time was minimized in terms of not comparing technology from completely different manufacturers. The four studies of the present thesis both documented similarities and differences between groups. For that reason, the comparisons made sense and provided valuable information, which can be used in guidance of future CI candidates and their families. The documented differences in both demographic and child related factors provided knowledge of a change with societal impact. During the first 18 years of pediatric cochlear implantation in Denmark age of implantation decreased and mode of implantation changed from almost only monaural to being almost only bilateral. The vast majority of children in the latter group attended mainstream educational settings with varied amount of support teaching. Mode of communication changed from mainly total communication/sign language to almost only spoken language. These findings demonstrated huge changes and continue to challenge both health care services and educational services. It is, therefore, appropriate to question whether the educational services of the local communities were prepared to include this clinical group of children with CI. As discussed above it can also be questioned whether the special educational services for children with HI were up-to-date with the changes led by pediatric cochlear implantation and more importantly how evidence based and up-to-date knowledge was assured in special educational settings, where there are not strong traditions of documentation and publishing of research of neither quantitative nor qualitative character. Furthermore, it is appropriate to question whether the health care services were up-to-date with handling much younger children and their parents, who were faced with their child's deafness at a period of time with great vulnerability.

The four studies provided evidence that since the introduction of CI, pediatric deafness must be perceived differently in order to meet the needs of the children with CI today. The children were born deaf but they grew up hearing and thus had very different potentials for development of spoken language and for being part of a hearing family and a hearing community in comparison to congenital deaf children born before the introduction of CI. The literature describes positive personal and social adjustment levels for children with CI and it is argued that these results represent an impressive level when compared with previous literature on adjustment problems in deaf children (Nicholas & Geers, 2003). Such findings were also documented in a separate study by Percy-Smith et al. (2008) aimed at comparing children with CI and children with NH. Overall findings were that the two groups of children were comparable on parameters of being bullied and bullying, management of school work, general social well-being and self-esteem and it was therefore possible to summarise that pediatric cochlear implantation has led to personal adjustment changes also for Danish congenital deaf children. Such findings cannot be used to state that children with CI are "cured" of deafness and not challenged on various parameters of inclusion into normal hearing communities, but the results provided evidence of changed opportunities for deaf born children.

The comparison of findings of social well-being and self-esteem in studies I and III showed that the vast majority of children were assessed to have a high level of social well-being in terms of self-esteem parameters. However, the comparison between the self-esteem assessments of study I and III should be interpreted with caution as the children had very different mean ages at day of testing and the literature reports that the older the children the more problems they encounter in terms of social well-being and self-esteem (Nielsen et al., 2001). It is, therefore, relevant to question whether the highly significant difference between level of social well-being of children from before and after introduction of UNHS and bilateral CI was an artefact of the age difference of 37 months.

The comparison of the listening and spoken language outcomes between studies revealed only one statistically significant difference related to auditory capacity even though both groups had quasi complete scores in the assessments of CAP, i.e. 80% and 92.7% respectively. The difference may be interpreted as an effect of implantation mode as the vast majority in the latter group had bilateral CI. When the prelingual children from the two groups were compared on outcomes of language understanding, active vocabulary and speech production according to their hearing age with CI, there were no statistical differences between the groups. This finding was noteworthy and again questioned whether the intervention and habilitation was timely enough and how quality in the intervention was assured. The introduction of UNHS was primarily medically and technically driven, and to date (April 2015) there is no standard option of early and immediate family intervention from neither health care services nor educational services, which is stressed to be crucial and highly predictive for later language learning in the literature (Holzinger et al., 2011; Niparko et al, 2010; Quittner et al., 2013; Yoshinaga-Itano, 2014).

7. Overall conclusions and perspectives

Chapter 7 summarises the overall conclusions and answers the specific research questions of the thesis. Finally, perspectives and the needs for future research are described.

The literature regarding spoken language development in children born deaf, with CI is substantial and identifies a number of factors that affect spoken language development both pre- and post-implant, including: prelingual and postlingual status at time of implantation, communication mode, age at implantation, education, hearing age with CI, bilateral versus monaural implantation, additional disability, socioeconomic status of parents and parental involvement. Some factors were related to demographic characteristics and some related to characteristics of the child. Several studies investigated outcomes in terms of social well-being, as it is of great importance that the hearing provided by CI and the language developed by the children with CI can be used to interact socially. Findings from these studies suggest that outcomes in terms of social well-being and quality of life for children with CI are more positive than outcomes for deaf children without CI. The factors identified in the literature were used as a framework to analyse two cohorts of children with CI in Denmark. The children comprised the first generation of children who were born deaf but grew up hearing. The two cohorts related to children implanted before and after introduction of UNHS and bilateral implants. By binding together four peer-reviewed articles documenting outcome data from four different studies, the overall objective of the thesis was an evaluation of the level of hearing, spoken language and social well-being for the first generation of children with CI in Denmark.

Specific objectives of thesis:

- To investigate the factors affecting the acquisition of spoken language for children with CI in Denmark.
- To investigate the factors affecting the social development of children with CI in Denmark.
- To analyse the differences and similarities between groups of children with CI implanted before and after the introduction of UNHS and bilateral CI.
- To identify the personal and societal changes which the introduction of pediatric CI has led to.

Furthermore, each study had its own specific research questions and hence specific conclusions. In the following sections conclusions from each of the four studies in relation to the specific objectives of the thesis will be summarized. On the basis of the four studies and the comparisons of similarities and differences between studies a final summative evaluation is presented.

7.1 Conclusions from four studies

7.1.1 Conclusion study I

Study I found a positive association between parents who used spoken language and level of social well-being. The most marked difference was found between children whose parents used spoken language as their communication mode and children whose parents used sign language. Children who were exposed to their parents' native spoken language were assessed to have higher levels of social well-being. Furthermore, there was a positive association between level of spoken language in terms of language understanding, vocabulary and speech production and level of social well-being.

7.1.2. Conclusion study II

This study found an association between parental mode of communication and the level of auditory capacities and spoken language outcomes. The quantification of the effects showed that children whose parents used spoken language as their mode of communication had greater odds of scoring highly in all tests and assessments. The greatest difference was found between children with spoken language and children with sign language. Children whose parents used spoken language combined with sign support had greater odds of scoring highly on all responses in comparison with children whose parents only used sign language. Furthermore, a noteworthy finding was the impact of place of living on the vocabulary test.

7.1.3 Conclusion study III

The responses from the auditory and speech intelligibility assessments were similar between regions in Denmark. On all other investigated tests/assessments there were statistically significant differences between regions with poorer results in the western part of the country. Thus, place of living was associated with spoken language outcomes and there were statistically significant differences in demographic related factors. Furthermore, children from the eastern part of the country were assessed as having a higher level of social well-being in terms of self-esteem parameters.

7.1.4 Conclusion study IV

The group of children implanted after introduction of UNHS and bilateral CI continued to face challenges in closing the language gap between hearing age and chronological age. Place of living continued to affect outcomes and parental mode of communication also continued to have a significant effect on outcomes.

7.2 Which factors affect acquisition of spoken language for children with CI in Denmark?

In order to answer this overall research question, data from study II and IV is used. The data indicated that the parental mode of communication was the most influential factor affecting the acquisition of listening and spoken language. Parental mode of communication significantly affected outcomes in terms of auditory capacity, receptive and expressive vocabulary, language understanding and speech production, and speech intelligibility. Parental mode of communication affected outcomes both when children's responses were scored according to hearing age with CI and according to chronological age. Acquisition of age-equivalent spoken language level was more likely in cases of normal hearing parents, who used their native spoken language as mean of communication with their child with CI. These findings add weight to the international research, which stresses that parental involvement and communication are essential for language development for children with CI. The present findings are in line with several studies that documented parental involvement and parental mode of communication as significantly associated with language development of children with CI (Dornan et al., 2010; Flexer, 2011; Fulcher et al., 2012; Holzinger et al., 2011). The present data together with other findings from the literature (Quittner et al., 2013) suggest that developing parental involvement and parental communication is a critical target for early language learning after implantation. CI teams should therefore involve parents both in decisions about pursuing early implantation and in their role in preparation for implantation and rehabilitation subsequently.

In studies II, III and IV place of living was identified as a factor affecting spoken language outcomes in both cohorts, and children from the eastern part of the country scored more highly on all measures of spoken language. Factors related to demographic characteristics were statistically significantly different between eastern and western regions. It was argued that the impact of place of living was comprised of multivariate factors including both demographic and child related characteristics. The documented impact of place of living constitutes a national challenge for both health care services and educational services.

Age at implantation was positively associated with spoken language outcomes for children in study II and IV but not to the same extent as described in the literature. There was discussion about parents' mode of communication and the fact that fewer than half of the parents were involved in the speech and hearing therapy sessions which may reduce the effect of age at implantation. The factors of gender, diagnosis and educational placement were found to affect outcome but not as consistently as communication mode, place of residence and age at implantation.

7.3 Which factors affect the social development of children with CI in Denmark?

The most influential factor affecting social development was identified to be the parental mode of communication, for the first cohort of children with CI in Denmark. For the second cohort, place of living influenced the level of social well-being and self-esteem. Social well-being was, furthermore, found to be positively associated with level of spoken language understanding, vocabulary and speech production, indicating that level of social well-being and level of spoken language were interrelated. In the literature it is stressed that the factors identified as influencing language development are interrelated with social development. The hypothesis is that higher levels of language are related to higher levels of social well-being (Connor et al., 2006) and the results of the present thesis contribute to this hypothesis. At the same time the overall results showed that the majority of the children in both cohorts did not show age equivalent language level but a majority of both cohorts showed a high level of social well-being and self-esteem. These findings are in line with studies from Nicholas & Geers (2003). They found that deaf children who had used a cochlear implant for 4 to 6 years coped successfully with the demands of their social and school environment, regardless of their speech and language achievements after implantation. Like the results found in the present thesis Nicholas & Geers also found that parents' ratings indicated that the children were emotionally and socially well-adjusted and that they benefitted from cochlear implantation. Thus, despite the extent of language skills, children with CI achieved positive outcomes in terms of social well-being. These results represent an impressive level of personal and social adjustment when compared with previous literature on adjustment problems in deaf children (Nicholas &Geers, 2003). In summary: spoken language level was a factor affecting social development, but, at the same time, the majority of the first generation of children with CI demonstrated high levels of social well-being in terms of self-esteem parameters, indicating that CI in itself does not inhibit social development.

7.4 Which differences and similarities can be identified between groups of children with CI implanted before and after the introduction of UNHS and bilateral CI?

Data was gathered at two points in time and various similarities and differences were identified between groups. Parental mode of communication was identified as the most influential factor affecting outcomes in terms of spoken language for children in both cohorts. Parents' mode of communication changed markedly during the period in which data for the first generation of children with CI was collected. In the data collection in 2004-2005 the majority of children were exposed to either total communication or sign language, and in 2010-2011 a minority (14%) of the parents used total communication with their children and no parents communicated by means of sign language. Despite this change in parental communication mode the prelingual children did not perform statistically significantly better in terms of spoken language understanding, expressive vocabulary and speech production when scored according to hearing age. The majority of prelingual children from both groups were assessed by their parents as scoring highly in terms of auditory capacity, 80 % and 92% respectively, but the latter group scored significantly higher, which was explained by the fact that 82% in the latter group had bilateral implants compared to only 2% in the first group. Age at implantation decreased between groups of prelingual children from 36 months to 20 months. Educational placement changed from 38% to 86% of children with CI being enrolled in mainstream educational settings. Introduction of UNHS has led to earlier cochlear implantation and thus decreasing the period of auditory deprivation for the congenital deaf child. The two cohorts differed in the number of deaf children with CI and additional disabilities. The number of children with additional disabilities increased between the two cohorts and hence provides new challenges for both health care services and educational services. This clinical group requires different types of assessments and what constitutes a successful outcome is not necessarily spoken language.

7.5 Which personal and societal changes have the introduction of pediatric CI led to?

The results of the four studies indicate that the introduction of pediatric cochlear implantation has led to major personal changes for children with congenital deafness. The children were born deaf but they grew up hearing and therefore had the foundation for development of listening and spoken language. The introduction of UNHS led to early implantation of congenital deaf children, but there is still no standard offer of early family intervention prior to implantation, which provides a challenge for both health care services and educational services.

The results from studies III and IV showed major changes in terms of educational settings and parents' choice of communication mode. The vast majority of children in the second cohort were placed in mainstream educational settings and the data supports the position that early pediatric cochlear implantation is a cost effective procedure in terms of education, as it provides an opportunity for children with CI to participate in a local school environment. Data makes it possible to conclude that pediatric cochlear implantation in Denmark has had both a societal impact and an individual impact in terms of linguistic, educational and social well-being outcomes. Number of pediatric cochlear implantations increased markedly from 1993-2014, which indicates that pediatric cochlear implantation changed from being a controversial, to a standard, intervention for children with congenital deafness. These findings are in line with findings from the UK (Archbold, 2010b). Pediatric cochlear implantation has led to an increase in the number of parents choosing mainstream educational settings for their child with CI and thus providing the child with opportunities for interacting with normal hearing peers and being part of a local and hearing community.

7.6 Evaluation of the level of hearing, spoken language and social well-being for the first generation of children with CI in Denmark.

Based on data from all four studies it is possible to draw the following conclusions. The evaluation of the first generation of children with CI in Denmark has shown that the children had high levels of auditory capacities for populations both prior and post introduction of UNHS and bilateral CI. Evaluation on the basis of the results from the four studies showed that the majority of children with CI were assessed by their parents to have a high level of social well-being in terms of self-esteem parameters with the younger group having the highest scores. However, the evaluation also showed that spoken language outcomes, when measured according to hearing age, was a challenge for the first cohort of children and closing the language gap between hearing age and chronological age was a challenge for children in the second cohort despite early implantation and bilateral CI.

The substantial literature indicated that the changes brought about by pediatric cochlear implantation and the substantial knowledge of which factors affect outcomes in terms of language and social well-being, make the future look bright for subsequent populations of users of CI. The evaluation of the first generation of children with CI in Denmark indicated, to some extent, the same bright future in terms of access to audition and spoken language for congenital deaf children and, very importantly, that this medical-surgical and technical intervention was positively associated with high levels of social well-being.

7.7 Perspectives for further research

The findings from this thesis indicates a need for future research to include studies with a longitudinal design in order to document when and if the language gap is closed at later stages of linguistic experience.

A further critical area for further study is the educational/developmental interventions offered to children with CI and their families. Is one educational approach more effective than another? Specifically, is the auditory verbal approach, which is recommended by both Sundhedsstyrelsen (2012) and Socialstyrelsen (2015), sustainable in a Danish context? Furthermore, it is important to study how this American educational approach, which entails the intense involvement of parents, will work in a society where the vast majority of parents both work. Exploration of whether there is a need for the development of a Nordic auditory verbal approach is therefore relevant.

The impact of place of residence is another issue that warrants further investigation. Such research should be designed to include a multi-professional approach, as the documented differences between eastern and western regions provide a challenge for both health care services and educational services.

In addition, future research should include investigation of how children with CI are distributed along the continuum of communication options, when monitored over longer periods of time. Some children may need to use some degree of signs to supplement their spoken language and some parents may wish to use signs in order to feel secure in case of technical breakdowns of the internal part of the CI. The impact of such differences needs to be investigated.

Future research should include studies of a wide spectrum of language skills and also include investigation of cognitive functioning. As the children grow older the language demands will become more complex and it will be interesting to study how Danish children with CI develop language skills such as: understanding of syntax and grammatical structure, lexical-semantic abilities, understanding of language in coherent speech, understanding of more abstract language aspects such as theory of mind and understanding of subtle language used in, for instance, idioms. Furthermore, future research including analyses of various aspects of cognitive functioning and working memory would be worthwhile.

Future research should involve long-term studies of acquisition of a second spoken language, educational and vocational level for children with CI.

The group of children with CI and additional disabilities is growing. It may, therefore, be necessary to evaluate outcomes for the clinical group of children with CI and an additional disability in terms of quality of life rather than solely or predominantly in terms of spoken language outcomes. This clinical group may require different study designs based more on observation rather than formal speech and language testing.

Future research should include maternal and paternal measures of sensitivity and involvement. It is, furthermore, critical to include suitable assessments for investigation of listening and spoken language outcomes for babies identified at or shortly after birth. In addition, it is appropriate to evaluate the guidelines from Sundhedsstyrelsen (2012) in terms of effect on listening and spoken language development for children who have been part of this programme.

Pediatric cochlear implantation is now regarded as routine and future research in the overall arena of pediatric hearing impairment should involve children using all types of hearing technology, e.g. hearing aids, cochlear implants, bone anchored hearing aids and auditory brainstem implants. The focus of research interest should be on the sensory impairment and not exclusively on the hearing technology. It is important to gain knowledge of similarities and differences between children with HI wearing all types of hearing technology in order to revisit for instance criteria for CI candidacy.

8. References

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AG Bell (2015) www.listeningandspokenlanguage.org

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Factors that affect the social well-being of children with cochlear implants

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ABSTRACT *The aim of the study was to identify factors associated with the level of social well-being for cochlear implanted children and to estimate effect-related odds ratios for the children's well-being. Another aim was to analyse associations between speech and language level and the level of social well-being. Data relate to 167 children with cochlear implants. In structural interviews, parents rated their children's level of social well-being regarding the degree of their child's personal-social adjustment. Five different factors were considered. Logistic regression models and proportional odds models were used to analyse the relationship between the considered factors and the assessments. The analyses showed that the communication mode at home was the most highly associated factor. A statistically significant association was found between the level of social well-being and speech understanding, speech production and vocabulary. Children who were exposed to a spoken language had considerably better odds of having a high level of social well-being compared to children with a mixture of spoken language and sign support or sign language.*

Keywords: paediatric cochlear implantation; social well-being; odds ratio estimates; communication mode

Introduction

Children with substantial, profound hearing loss are at significant risk for serious speech and language delays that can impact their communication, their cognitive development as well as their social development (Connor et al., 2006). A substantial amount of literature has documented the difficulties of children with severe-profound hearing loss in the area of self-esteem and social-emotional adjustment (Nicholas and Geers, 2003). Since the introduction of cochlear implants as a treatment for children with profound hearing loss, many studies have documented auditory and speech/language progresses not previously described for a profoundly hearing-impaired paediatric population (Moog, 2002; Moog and Geers, 2003; Tait et al., 2001; Waltzman et al., 2002; Wie, 2005). There have, however, not been as many studies documenting matters regarding social well-being and self-esteem for children with cochlear implants. Nicholas and Geers (2003) found that children with cochlear implants cope successfully with the demands of their social and school environment and they found that parents rated their children with cochlear implants to be emotionally and socially well adjusted. Filipo et al. (1999) found that psychological well-being of both adolescents and children with cochlear implants did not cause any psychological disruption post-implant. Nicholas and Geers (2003) did not find any significant association between the level of social well-being and speech perception, speech production or language skills that the child achieved post-implant. It is interesting to study further whether some of the influential factors that have been documented for speech and language outcomes for cochlear implanted children may have an impact on the level of social well-being and to what extent the language level of cochlear implanted children is associated with the level of social well-being.

In various speech and language outcome studies it has been reported that the age of operation is a factor highly associated with auditory and speech/language outcomes (Connor et al., 2006; Harrison et al., 2001; Kral et al., 2002; Lenarz et al., 2004; Tait and Nikolopoulos, 2004; Waltzman et al., 2003). The length of device use, in the present study defined as hearing age, also affected speech/language outcomes (Beadle et al., 2005; Connor et al., 2006; Geers and Brenner, 2003; Horn et al., 2005). Another significant factor seems to be the mode of communication used for cochlear implanted children (Archbold et al., 2000; Geers et al., 2003a; Gravel and O'Gara, 2003; Meyer et al., 1998; Tobey et al., 2003; Vieu et al., 1998). In the literature, there is also evidence that educational placement can affect speech and language outcomes (Connor et al., 2000; Easterbrooks and Mordica, 2000; Tobey et al., 2004). It has also been documented that girls seem to have higher scores for speech/language outcomes than boys, thus indicating a gender-related effect (Geers et al., 2003a; Tobey et al., 2003). The five factors referred to here have been assigned various degrees of impact on the speech and language outcomes of cochlear implanted children and in the present study we aim to investigate whether these five factors also influence the level of social well-being for cochlear implanted children.

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The purpose of the study was, thus, to investigate whether the effect-related factors for speech and language outcomes also affect the social well-being of a paediatric cochlear implanted population. The study also sought to quantify data by estimating effect-related odds ratios for the level of social well-being of cochlear implanted children. Furthermore, the study sought to investigate whether the speech and language level post-implant was associated with the cochlear implanted children's level of social well-being.

Materials and methods

Participants (N = 167) were children (91 girls and 76 boys) who received a cochlear implant between 1993 and 2004 at the two paediatric cochlear implant centres in Denmark. Figure 1 illustrates the diagnoses of the 167 children. Participants received their implants between six months and 17 years. Figure 2 illustrates the spread of data (mean implantation age = four years). Age ranged from one to 18 years at the day of testing with a mean of seven years. Some 164 children were unilaterally implanted and three children were bilaterally implanted. Data were collected from August 2004 until February 2005. All children used a Nucleus product (Cochlear, Lane Cove, Australia). The inclusion criterion was implant use for a minimum of six months, in order to assure that the child had integrated the auditory sense. All children detected the six Ling sounds. All parents were normal hearing except for two mothers who used cochlear implants themselves. Three children were diagnosed with another handicap (i.e. two children had cerebral palsy due to meningitis

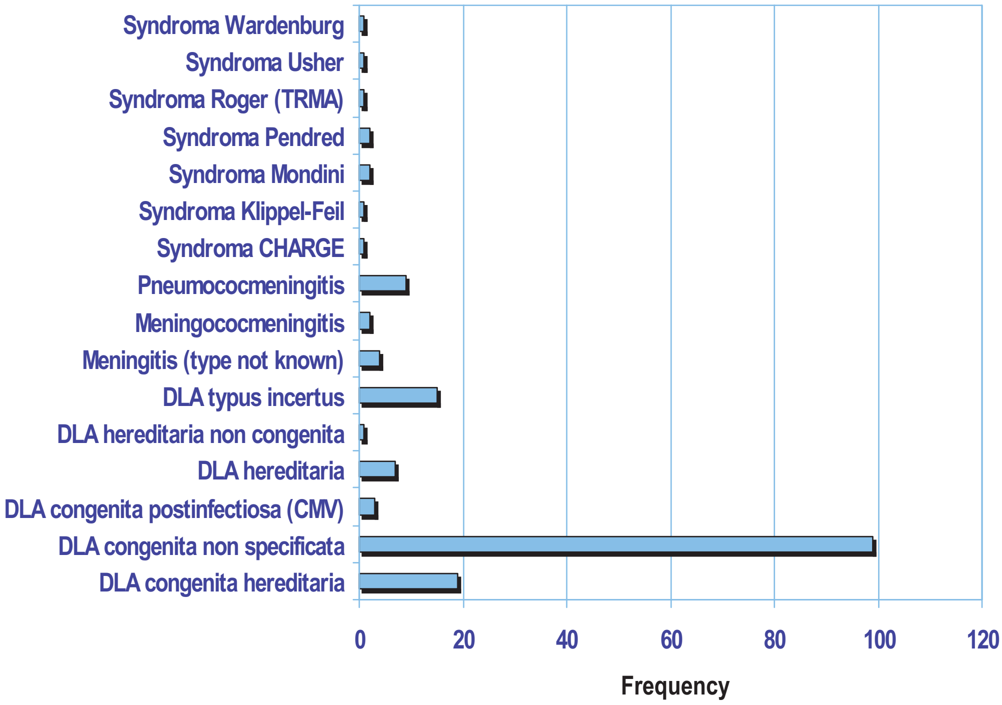

Figure 1: Diagnoses. DLA: Degeneratio Labyrinthi Acustici; CMV: Cytomegalovirus; TRMA: Thiamine responsive megaloblastic anemia syndrome; CHARGE: Coloboma, Heart defects, Atresia choanae, Retarded growth and development, Genital hypoplasia, Ear anomalies.

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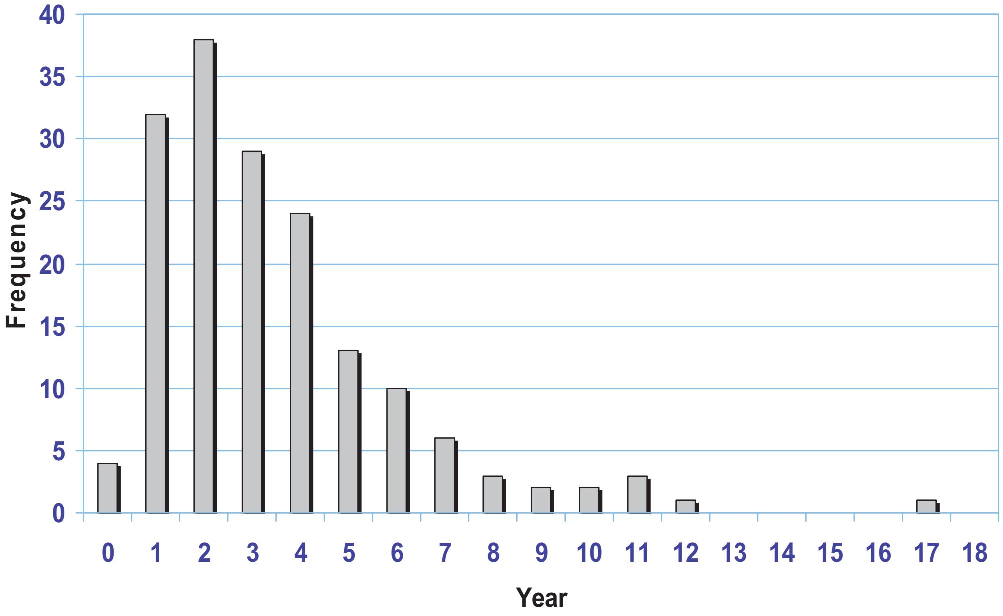


Figure 2: Age at operation. Mean: 3.95, std dev: 2.63. Max: 17.27: Q3 (75%): Q2 (50%): Q1 (25%): 2.1, min: 0.53.

and one child had Down's syndrome). The three children were included in the data sample as they had developed a spoken language and they did not encounter problems in fulfilling the test battery. The rest of the population did not have any diagnosed disability other than the hearing impairment. One child had CHARGE (Coloboma, Heart defects, Atresia choanae, Retarded growth and development, Genital hypoplasia, Ear anomalies) syndrome, but did not encounter vision problems to an extent that would exclude him from the speech and language tests. The study of children's social well-being and self-esteem post-implant was part of a large study of the first 200 children with cochlear implants in Denmark. The study sought to assess the level of communication, vocabulary, speech perception and speech production as well as the level of social well-being.

A total of 198 children and families fulfilled the inclusion criterion and were invited to participate. Some 169 families accepted, which gives a participation rate of 85 per cent. Two children were excluded due to blindness. Eleven families did not want to participate (five of these families had a child with another disability). Nineteen families did not show up on the day of testing or it was not possible to contact them. No general picture was seen with regard to the factors gender, implantation age and hearing age for the 30 children who did not participate.

Description of factors

The population was characterised by two different age variables: Implantation age and hearing age. Implantation age was defined as the chronological age on the day of operation. For further analysis of the influence of implantation age on the assessments of the children's level of social well-being, a discrete version was defined by grouping the values into three categories: <18 months, 18–36 months and >36 months. The three different groups were based on previous research by Sharma et al. (2002, 2005) and in broad terms follow the division that Connor et al. (2006) used for studying the impact of age of implantation.

Furthermore, children were divided into three groups based on their hearing age with a cochlear implant (i.e. length of device use from day of switch-on): Group one (6–23 months, N = 74), group two (24–36 months, N = 30) and group three (>36 months, N = 63). Depending on the hearing age a specific test battery was performed. The hearing age groups were based and defined on the speech and language tests' standard from a normal-hearing population. Scoring of tests according to hearing age was decided after the pilot study, run in May 2004. In the pilot study, it became evident that many children with hearing ages above the age range in the norm-based speech and language tests had great difficulties performing at their chronological age level. For some of the children it would not have been possible to do spoken language testing according to their chronological age as their general spoken language level was too poor.

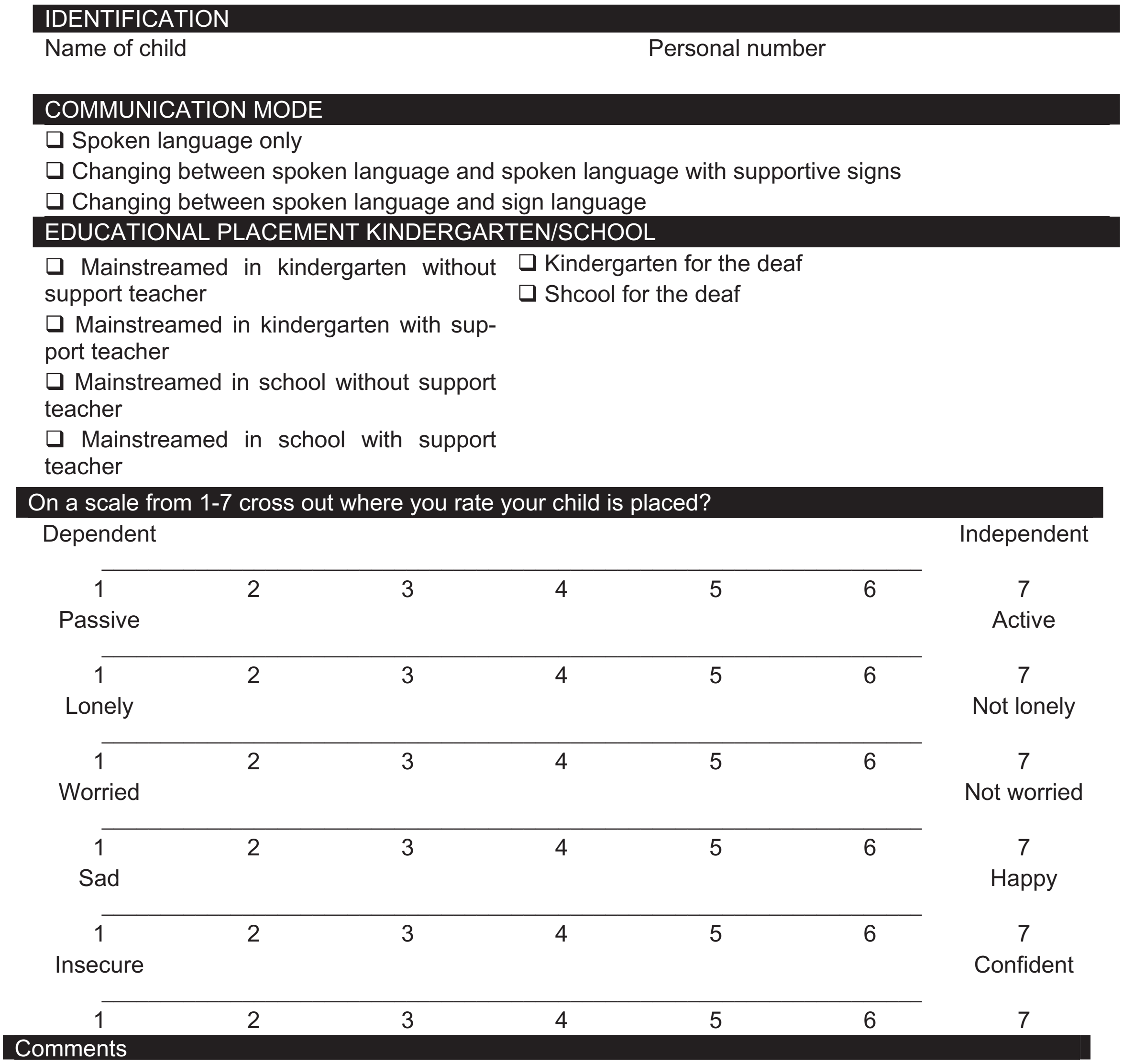
Two categories were defined depending on whether the child was placed in a school/kindergarten for the deaf or placed in a mainstream educational set-up. In a structured interview on the day of testing, the parents stated the educational placement (Figure 3). A hundred children were placed in a school/kindergarten for the deaf and 67 children were placed in a mainstream educational setting. All children placed in mainstream educational settings had a support teacher.

In order to study the impact of communication mode at home, parents were asked to choose between the following categories: use of spoken language only, use of spoken language and supportive signs or use of spoken language and sign language. The three different categories related to the conditions that children with cochlear implants encounter in Denmark. Once a child is diagnosed with a hearing impairment, the family is offered sign language courses. Some parents choose to combine signs with spoken language and some parents choose to communicate by use of spoken language and sign language. Danish sign language is without the use of the voice. The purpose of asking parents to state how they communicated with their cochlear implanted child was to get an overview of the communication status for the first 200 children with cochlear implants in Denmark and to analyse the impact of the various communication modes used. Fifty children were exposed to spoken language only, 86 children had a mixture of spoken language and supportive signs and 30 children used a mixture of spoken language and sign language. Thus, 70 per cent of normal-hearing parents use a mixture of spoken language and sign support or sign language, and 30 per cent communicate by means of spoken language only.

Gender represented the last of the five factors of interest, which are summarised in Table 1.

Description of applied tests and assessment

Assessment of the children's level of social well-being was carried out in structural interviews with the parents. The assessment scale applied was based on social



IDENTIFICATION

Name of child | Personal number

COMMUNICATION MODE

☐ Spoken language only
☐ Changing between spoken language and spoken language with supportive signs
☐ Changing between spoken language and sign language

EDUCATIONAL PLACEMENT KINDERGARTEN/SCHOOL

☐ Mainstreamed in kindergarten without support teacher
☐ Mainstreamed in kindergarten with support teacher
☐ Mainstreamed in school without support teacher
☐ Mainstreamed in school with support teacher
☐ Kindergarten for the deaf
☐ Shcool for the deaf

On a scale from 1-7 cross out where you rate your child is placed?

Dependent — Independent
1 2 3 4 5 6 7

Passive — Active
1 2 3 4 5 6 7

Lonely — Not lonely
1 2 3 4 5 6 7

Worried — Not worried
1 2 3 4 5 6 7

Sad — Happy
1 2 3 4 5 6 7

Insecure — Confident
1 2 3 4 5 6 7

Comments

Figure 3: Parental questionnaire.

Table 1: Factors of interest

Factor	Categories
Hearing age	6–23 m/24–36 m/>36 m
Operation age	<18 m/18–36 m/>36 m
Educational placement	School/kindergarten for the deaf /Mainstream
Communication mode	Only Danish/Sign support/Sign language
Gender	Girl/Boy

well-being studies of normal-hearing children performed by the National Institute of Public Health, University of Southern Denmark (Nielsen et al., 2001). Structural interviews with the parents were carried out at one of the two paediatric cochlear implant centres in Denmark. Three different speech and language pathol-

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ogists performed structural interviews with the parents and carried out speech and language tests of the children with cochlear implants. The scoring of all assessments and tests was then carried out by one speech and language pathologist, who is the first author of the present article. The parents completed a rating scale that sought to assess the degree of their child's personal, social adjustment. On a seven-point scale, parents assessed whether the child was: dependent vs. independent, passive vs. active, lonely vs. social, worried vs. not worried, sad vs. happy, insecure vs. confident (Figure 3). In accordance with the defined score by the National Institute of Public Health, University of Southern Denmark, a score below 36 defined a low level of social well-being and a score above 36 defined a high level of social well-being (maximum score = 42 and minimum score = 7).

As the study formed part of a large study that also assessed the cochlear implanted children's speech and language level, responses from four different speech and language tests were used in order to study possible associations between the level of speech and language and the level of social well-being.

Tait video analysis

The purpose of the Tait video analysis was to assess the children's auditory awareness when communicating with an adult. Children were filmed in interaction with one of the testers, and the activities being chosen had to be interesting enough to promote communication. Turn-takings were identified, which were the instances when the child had an opportunity to communicate. When the adult paused the child had the opportunity to respond. The turns were then classified as vocal, when the child used voice to communicate with or without the addition of sign/gesture, or as a turn with sign, gesture or facial expressions without vocalisation. Vocal and gestural turn-takings were each counted as a percentage of the total number of turns, which for all subjects were 20 turns in total. Auditory awareness of the adult's speech was measured by the Non-Looking-Vocal-Turns (NLVT) score. NLVT was counted when the child vocalised communicatively in a turn without being in eye contact with the adult during the adult's previous turn. No visual cue, no sign or pointing finger from the adult was given. All classifications and the scoring were made in full accordance with the Tait analysis (Tait et al., 2001, 2007). In the present study, it is the score of the NLVT that is the subject for further analysis. For statistical analysis the NLVT score was defined as a dichotomous variable, classified as a score either below or above 50 per cent NLVT. The Tait analysis was performed by all subjects.

Reynell test

In order to study the children's comprehension of spoken language the receptive part of the Reynell test was used (Reynell and Huntley, 1985). The standard of the Reynell test starts at two years of age and, therefore, the children of our study had to have a minimum of two years of hearing with a cochlear implant (N = 94).

In accordance with the standard score for the Reynell test a discrete version was defined as performance below the norm or equal to/above the norm.

Phonological test

For studying children's speech production a Danish phonological test 'Sproglydstesten [The speech sound test]' (Kaufman and Ege, 1974) was used. The phonological test was performed by children with at least 36 months of hearing (N = 64). The test consists of 90 different Danish phonemes and phoneme constellations. The children had to pronounce 52 different words presented as pictures in a closed-set format. The children's productions were analysed for vowels (V) and clusters of V and consonants (C). The transcriptions consisted of the following constellations: 13 V, 15 CV, 12 VC, 16 CCV, seven SC, five SCC, nine VCC and 13 Finals. The results from the phonological test were recorded in an ordinal variable with four categories: A score between 76 per cent and 100 per cent correct, a score between 51 per cent and 75 per cent correct, a score between 26 per cent and 50 per cent correct and a score between zero per cent and 25 per cent correct.

Vocabulary

The level of vocabulary was tested by use of a Danish vocabulary test 'Viborg materialet' (Pedersen and Kjøge, 2005). The test evaluates the children's active vocabulary. The children were shown 100 different photos and had to verbally state the object. Responses by use of signs only were counted as error. Because the normal-hearing standard is based on children with a minimum of three years of age, the children with a cochlear implant had to have a hearing age of at least 36 months (N = 64). The standard age range for normal-hearing children was three to seven years of age. The score was categorical and classified as a lower 25 per cent group, a middle 50 per cent group and an upper 75 per cent group. Table 2 summarises the applied speech and language tests and their categorisations.

Data analysis

All scoring of tests and assessments was carried out by one speech and language pathologist. Data were put into an access database created for this study. The access

Table 2: Responses of interest speech and language

Responses	Categories
Tait video analysis	Score below 50% NLT-V/Score above 50% NLT-V
Reynell test (receptive part)	Below norm/Equal to or above norm
Phonological test	<25% correct/26–50% correct/51%–75% correct/ >75% correct
Vocabulary test	Lower 25%/Middle 50%/Upper 75%

NLT-V: Non-Looking-Vocal Turns.

database was sent to The Department of Biostatistics at the University of Copenhagen, where all descriptive and statistical analyses were performed using SAS7 software. The descriptive analyses of the data were done by making cross-tabulations of the factors and responses of interest. Table 3 summarises the cross-tabulations between the factors of interest and the assessments of the children's social well-being.

In order to study and describe the relationship between the response and the factors of interest, different types of general linear models depending on the outcomes of interest were applied to the data. Backwards elimination was used to choose the final model. It consists of starting the analysis with a full model (all variables included) and eliminating variables in an iterative process. After the elimination of each variable the fit of the model was tested to ensure that the model still fitted the data adequately. When all variables in the model were significant (i.e. no more variables can be eliminated from the model), the analysis was considered completed. Data did not show any convergence problems for any of the models. Table 4 summarises the results of the backward elimination results.

In order to study further the effects found, an exploratory analysis was performed consisting of a quantification of the data by estimating odds ratios. Let π be the probability of scoring higher than 36 in the social well-being assessment. The odds of having a high level of social well-being is defined as the quotient between the probability of getting an assessment higher than 36 and the probability of getting a lower assessment, that is

$$odds = \frac{\pi}{(1-\pi)}$$

Goodness-of-fit was performed by the Hosmer-Lemeshow test.

For studying associations between social well-being and responses from the speech and language data, the Fisher exact test was used. Positive association between two variables means that children who perform well in one of the speech and language tests will also be assessed to have a high level of social well-being.

Results

Descriptive data

A score of 36 has been defined by the National Institute of Public Health as a high level of social well-being. As can be seen in Figure 4, the mean score for the whole population was 36.27. Sixty children (36%) had a low level of social well-being and 107 children (64%) had a high level of social well-being.

When looking at cross-tabulations (Table 3) between the five considered factors and the level of social well-being the following can be described: For the factor hearing age there did not seem to be any marked spread of data. For the children assessed to have a low level of social well-being they were spread equally between the three hearing age categories. Forty per cent of children having a hearing age

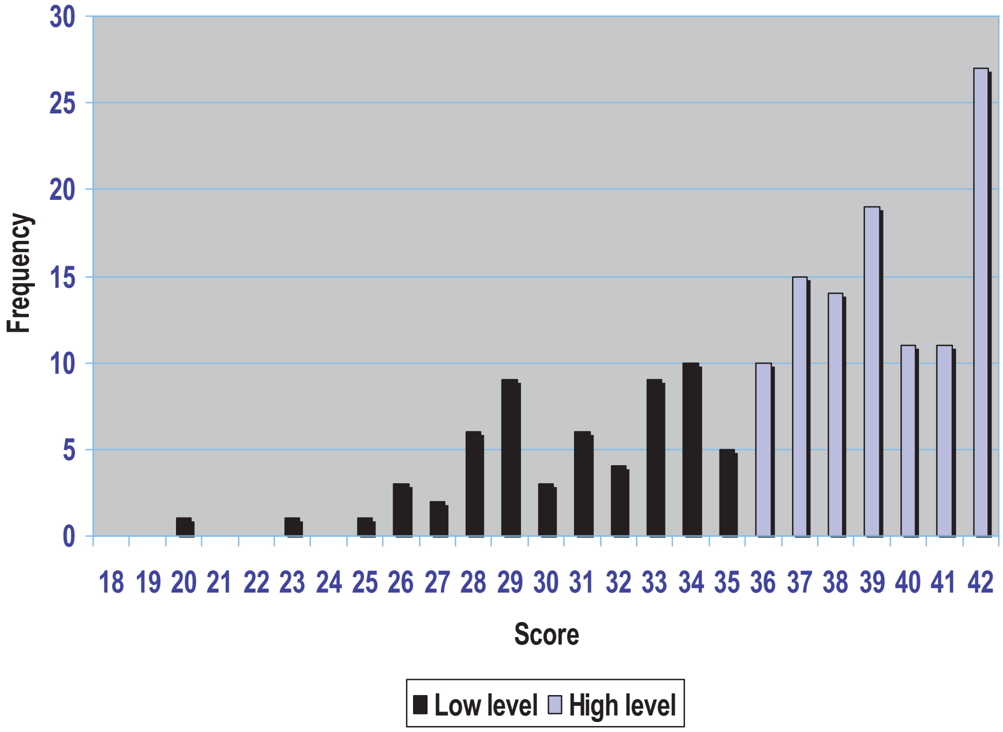

Figure 4: Social well-being scores. Mean: 36.27, std dev: 4.86. Max: 42, Q3 (75%): 40, Q2 (50%): 37, Q1 (2 %): 33, min: 20.

Table 3: Cross-tabulations factors/social well-being

Factors	Low level <36	High level >36	Total
Hearing age			
6–23 months	34%	66%	100%
24–36 months	33%	67%	100%
>36 months	40%	60%	100%
Operation age			
<18 months	20%	80%	100%
18–36 months	39%	61%	100%
>36 months	37%	63%	100%
Communication mode			
Spoken language	18%	82%	100%
Sign support	63%	37%	100%
Sign language	60%	40%	100%
Educational placement			
Institutions for the deaf	41%	59%	100%
Mainstream	28%	72%	100%
Gender			
Boys	43%	57%	100%
Girls	30%	70%	100%

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of over 36 months had a low level of social well-being as opposed to 33 per cent and 34 per cent of children having a hearing age of six to 23 months and 24–36 months, respectively. When looking at the factor implantation age, it was seen that 80 per cent of children operated under <18 months were assessed to have a high level of social well-being, whereas 61 per cent and 63 per cent of children operated at 24 months or older, respectively, were assessed to have a high level of social well-being (i.e. 61% and 63% for the implantation age categories 18–36 months and >36 months, respectively). The factor communication mode showed a marked spread of the data with 82 per cent of the children exposed to spoken language having a high level of social well-being as opposed to 37 per cent with sign support and 40 per cent with sign language. When looking at characteristics of children assessed to have a low level of social well-being, the data showed that the majority of those children were exposed to either sign support or sign language (i.e. 63% and 60%, respectively). For the factor educational placement it was found that 59 per cent of children placed in kindergartens/schools for the deaf had a high level of social well-being as opposed to 72 per cent of children placed in mainstream educational settings. For the last considered factor gender, Table 3 shows that 70 per cent of girls were assessed to have a high level of social well-being as opposed to 57 per cent of boys.

Factors with a statistically significant effect on the responses

Table 4 contains the results of the model selection for the response of the social well-being assessments, the *p*-values for the statistically significant factors are presented. As can be seen in Table 4, the two factors communication mode and gender were statistically significant associated with the level of social well-being (i.e. *p*-values = 0.0006 and 0.0476, respectively, for communication mode and gender).

Quantification of the effects: Odds ratio estimates

The observed effect of the communication mode was further analysed for the child's exposure to spoken language only, spoken language with supportive signs or spoken

Table 4: Backward elimination results – statistically significant factors (*p*-values)

Responses / Factors	Assessments of social well-being
Hearing age	X
Operation age	X
Educational placement	X
Communication mode	0.0006
Gender	0.0476

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Table 5: Odds ratio estimates and 95% confidence intervals for all responses

Odds ratio estimates – social well-being assessments

Effects		Estimate and 95% confidence intervals
Mode of communication	Danish vs. sign language	7.64 [2.72–21.45]
	Sign support vs. sign language	3.01 [1.27–7.11]
	Danish vs. sign support	2.53 [1.07–5.96]
Gender	Girl vs. Boy	1.98 [1.00–3.92]

Table 6: Positive significant associations between responses

Variables	Social well-being
Tait video analysis	–
Reynell	Positive 0.0014
Phonology	Positive 0.0010
Vocabulary	Positive <0.0001

language with sign language. The effect of gender was also analysed. A quantification, in terms of odds ratios, of both effects is summarised in Table 5, which shows that children who were exposed to spoken language had greater odds of having a high level of social well-being, than children exposed to either sign support or sign language. The greatest effect was found between children with spoken language and children with sign language. Children with spoken language had 7.64 better odds of having a high level of social well-being than children with some degree of sign language. Children exposed to sign support had 3.01 better odds of having a high level of social well-being than children exposed to sign language. The estimations based on the effect of gender showed that girls had 1.98 better odds of having a high level of social well-being than boys. Goodness-of-fit was performed by the Hosmer-Lemeshow test (Chi-square 3.0373, df 4, *p*-value 0.5516).

Associations between social well-being and the level of speech and language

In order to study whether good performance in one speech and language test was associated with a high level of social well-being, the Fisher exact test was applied to the data. A positive association between responses means that children who performed well in one of the speech and language tests will more likely also have a high level of social well-being. A negative association means that children with a high score in one specific test will more likely have a low score in the social well-being assessment. Table 6 shows the associations between responses from the

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four speech and language tests and the social well-being assessments. Table 6 illustrates that a high level of social well-being was positively associated with good speech understanding (Reynell test), a large vocabulary and good speech production, whereas there is a negative association between the score of NLVT from Tait video analysis and social well-being assessments.

Discussion and conclusion

We found a statistically significant association between communication mode and the level of social well-being. This finding is in accordance with findings of influential factors on speech and language outcomes (Tobey et al., 2004). Furthermore, it was found that children exposed to spoken language had considerably better odds of having a high level of social well-being than children who were exposed to sign support and sign language. These findings question how communication mode influences the level of social well-being. A possible explanation may be that the parents are normal hearing and when they use sign support or sign language they communicate in a foreign communication mode and a foreign language. The literature of bilingualism for normal-hearing populations is substantial and the importance of parents using their native language as the mode of communication with their children is stressed repeatedly. In addition, the social and emotional benefits for a child to be able to have the same native language as its parents are stressed in studies of deaf children with deaf parents as opposed to deaf children of hearing parents. Deaf children of deaf parents are rated better on various social and emotional scales compared to deaf children of hearing parents with a plausible explanation being that deaf children have a common native language with their parents (Nicholas and Geers, 2003). Cochlear implantation has facilitated new possibilities for deaf children, as it provides sufficient hearing to develop age-appropriate speech and language and, thus, the possibility of having the same native language as their parents.

Like the study of Nicholas and Geers (2003) we found that parents' ratings were higher for girls than for boys. The gender effect has also been documented in speech and language outcome studies (Tobey et al., 2004; Wie, 2005). Implanted girls have a better receptive language and a better production of phonemes than boys, which is equivalent to normal-hearing children acquiring language. Future research seems warranted in order to study whether the gender effect regarding social well-being is found for normal-hearing populations as well.

We found no statistically significant effect of educational placement, which is a factor that has been shown to be of influence as regards to speech and language outcome. When looking at cross-tabulations between educational placement and social well-being it was, however, evident that 41 per cent of the children assessed to have a low level of social well-being were placed in kindergartens/schools for the deaf. In contrast, 28 per cent of the children in mainstream educational set-ups were assessed by parents to have a low level of social well-being and 72 per cent were assessed to have a high level of social well-being. Thus, our data did not indicate that cochlear implanted children in mainstream settings, where the children rely on speech exclusively for communication, have greater social and emotional difficulties. This is in accordance with findings of Filipo et al.'s (1999) study showing that children in mainstream settings are positive and well adjusted. Nicholas and Geers (2003) found the same tendency. These findings are in contrast with several studies of deaf children without cochlear implants placed in mainstream settings. We can thus conclude that cochlear implants have given deaf children the possibility of being well integrated and adjusted in mainstream schools.

In many studies of speech and language outcomes the age of implantation has been shown to have great influence, as younger age is associated with better outcomes as regards to both speech perception and speech production (Connor et al., 2006; Harrison et al., 2001; Kral et al., 2002; Lenarz et al., 2004; Tait and Nikolopoulos, 2004; Waltzman et al., 2003). We found no statistical significant association between age at implantation and the level of social well-being. It is, nevertheless, noteworthy that 80 per cent of children implanted younger than 18 months of age were assessed to have a high level of social well-being. In contrast, 63 per cent and 61 per cent of children implanted at 24 months or older than 36 months, respectively, were assessed to have a high level of social well-being. This may indicate that early implantation is of importance also for outcome data regarding social well-being. The wide range of implantation age, six months to 17 years, may have introduced a bias in the present study, as the population was not homogenous regarding the factor implantation age. It could be argued that the study should have included only children born deaf, implanted before three years of age and with a hearing age of at least two years. The purpose of the present study was, however, to get an overview of how the first 200 children with cochlear implants in Denmark managed social well-being parameters such as loneliness, independence, happiness, confidence, activity and worry. Future research with inclusion of a more homogenous population seems warranted.

Hearing age was not found to be a statistically significant factor for the level of social well-being and the cochlear implanted children's score is spread almost equally between the three hearing age variables defined for this study (Table 3). It is, nevertheless, interesting to study further whether longer use of cochlear implants and hence greater speech and language development will make more parents perceive their child as better socially and emotionally adjusted.

In conclusion, the present study demonstrates a very strong effect of the parental communication mode on the level of social well-being. Estimation of odds ratios for scoring high on parental social well-being assessments has been documented in the study. The children exposed to spoken language had considerably better odds (7.64) of scoring high on the social well-being assessment as compared to children exposed to a mixture of spoken language with sign support or sign language. Children exposed to sign support had better odds than children exposed to sign language. The most marked effect was found between children exposed to spoken language only and children exposed to sign language. Thus, our findings suggest a very clear benefit of spoken language communication with a cochlear implanted child, not only for obtaining and securing a high level of speech and language, but also and very importantly to obtain and secure a high level of social well-being.

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ORIGINAL ARTICLE

Parental mode of communication is essential for speech and language outcomes in cochlear implanted children

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Abstract

Conclusion: The present study demonstrates a very strong effect of the parental communication mode on the auditory capabilities and speech/language outcome for cochlear implanted children. The children exposed to spoken language had higher odds of scoring high in all tests applied and the findings suggest a very clear benefit of spoken language communication with a cochlear implanted child. *Objectives:* The aim of the study was to identify factors associated with speech and language outcomes for cochlear implanted children and also to estimate the effect-related odds ratio for each factor in relation to the children's speech and language performances. *Methods:* Data relate to 155 prelingually deafened children with cochlear implant (CI). A test battery consisting of six different speech and language tests/assessments was used. Seven different factors were considered, i.e. hearing age, implantation age, gender, educational placement, ear of implantation, CI center, and communication mode. Logistic regression models and proportional odds models were used to analyze the relationship between the considered factors and test responses. *Results:* The communication mode at home proved essential to speech and language outcome, as children exposed to spoken language had markedly better odds of performing well in all tests, compared with children exposed to a mixture of spoken language and sign support, or sign language.

Keywords: *Pediatric cochlear implantation, communication mode, odds ratio estimates*

Introduction

Children with profound hearing loss are at significant risk of serious speech and language delays that can impact their communication skills and their cognitive development, as well as their social development [1]. However, since the introduction of cochlear implantation as a treatment for children with profound hearing loss there have been studies documenting auditory and speech/language progress never before described for a profoundly hearing-impaired pediatric population [2–4]. Various factors may have an impact on the auditory and speech/language development following cochlear implantation. In the present study, we aimed to investigate the impact of a number of these factors for the first 155 prelingually cochlear implanted children in Denmark.

Several studies have described that age at implantation is a factor highly associated with auditory and speech/language outcomes [1,5–7]. The length of device use, in the present study defined as hearing age, also affects the speech/language outcomes [1,8,9]. Implantation age and hearing age are furthermore described to be highly inter-related [1]. Another significant factor seems to be the mode of communication used towards the cochlear implanted children [10,11]. In the literature there is also evidence that the educational placement can affect the speech and language outcomes [12–14]. It has also been documented that girls seem to have higher scores

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for speech/language outcomes than boys, thus revealing an effect of gender [9]. The five factors referred to have been assigned various degrees of impact on the speech and language outcome of cochlear implanted children across countries, languages, and cultures.

On the basis of the literature above, the five factors referred to were chosen for further analyses in relation to a Danish cochlear implant (CI) population. A sixth factor of interest was included to analyze whether there was a significant difference between the two pediatric CI centers in Denmark and a seventh factor was included regarding whether ear of implantation and/or monaural vs bilateral implantation affected the speech and language outcome. Thus, the purpose of the present investigation was to identify the effect of the seven factors on auditory communication and spoken language outcomes for prelingually implanted children. In addition, we sought to isolate one factor that was more highly associated with outcome than others. Effect-related odds ratios for the children's performances as regards level of vocabulary, phonology, speech understanding, communication, speech intelligibility, and auditory capacity were estimated.

The international literature is massive and goes back a long time as regards documentation as to which kind of rehabilitation should be offered after pediatric cochlear implantation. At the beginning of 2000 many studies, as listed above, documented the effect of oral communication for children with CI, so why is it relevant to futher study this in the Nordic countries? In Scandinavia there is a historically strong and long tradition of using sign language and sign-supported communication with hearing-impaired children. Furthermore, there is a well established deaf community and sign language courses are offered to family and relatives shortly after diagnosing a child with a moderate to severe hearing impairment. Sign language in the Scandinavian way is signing without the use of voice. These conditions make it relevant to particularly study speech and language outcome for children in Scandinavia, as the conditions and content of rehabilitation are different from those in studies referred to in the international literature.

Material and methods

The study formed part of a large study that also assessed the self-esteem and social well-being of children with CIs in Denmark [15].

Of the first 198 cochlear implanted children in Denmark, 2 were excluded due to blindness and 13 were excluded as they were implanted post-lingually. Of the remaining 183 children and families, 28 did not want to participate or did not turn up on the day of testing, or it was not possible to contact them by telephone to arrange a day for testing and interviewing (5 of these families had a child with another disability).

Thus 155 children (85%) and their families participated (85 girls and 70 boys). The children were implanted between 1993 and 2004 at one of the two pediatric CI centers in Denmark.

Mean implantation age was 3 years. All children had used their implant for a minimum of 6 months, assuring integrated auditory sense.

A few children implanted after 3 years of age had only used hearing aids sporadically and were communicating by means of sign language with some spoken words or verbal utterances prior to implantation. These children were classified as prelingual and thus included in the study.

Three children were diagnosed with another handicap, i.e. two children had cerebral palsy due to meningitis occurring before 3 years of age and one child had Down's syndrome. These children were included, as they had developed a spoken language and were able to complete the test battery.

Data were collected from August 2004 until February 2005. The mean age on the day of testing was 6 years. All children used a Nucleus product. All children detected the six Ling sounds. All parents were normal-hearing except for two mothers who used CIs themselves.

Description of factors

Implantation age. The population was characterized by two different age variables: implantation age and hearing age. Implantation age was defined as the chronological age on the day of implantation. For further analysis of the influence of implantation age on test responses, a discrete version was defined by grouping the values in two categories: <36 months ($n = 74$) and >36 months ($n = 81$). This categorization was based on previous research by Sharma et al. [16].

Hearing age. The children were further divided into three groups based on their hearing age with CI (i.e. length of device use from day of switch-on): group one (6–23 months; n = 66), group two (24–35 months; n = 27), and group three (≥ 36 months; n = 62) (Table I).

The mean age of hearing with CI was 2.4 years. Depending on the hearing age, a specific test battery was performed. The hearing age groups were based and defined on the speech and language test standards from a normal-hearing population. Scoring in the tests according to hearing age was defined after a

Table I. Distribution of factors of interest.

Factors	*n*	%
Hearing age		
6–23 months	66	43%
24–35 months	27	17%
≥ 36 months	62	40%
Implantation age		
< 36 months	74	48%
≥ 36 months	81	52%
Educational placement		
Mainstream	59	38%
Kindergartens/schools for the deaf	96	62%
Communication mode		
Danish	40	26%
Sign support	85	54%
Sign language	30	20%
Center		
East	83	54%
West	72	46%
Gender		
Girls	85	55%
Boys	70	45%
Ear implanted		
Right	106	68%
Left	46	30%
Bilateral	3	2%

pilot study performed in May 2004. In the pilot study, it became evident that a number of children had difficulties performing at their chronological age level and some were simply not able to complete language testing accordingly, as their level of general spoken language was too poor.

Educational placement. Categories were defined as placement in a school/kindergarten for the deaf or in a mainstream educational set-up. The educational placement provided information of the kind of intervention the child was receiving. Characteristically, the children placed in kindergartens/schools for the deaf communicated mainly by means of sign-supported Danish or sign language. Thus, 77% of the children who were exposed to sign support or sign language were placed in preschools/schools for the deaf. In a structured interview on the day of testing, the parents stated the educational placement. Ninety-six children were placed in a school/kindergarten for the deaf and 59 children were placed in a mainstream set-up (Table I). All children placed in mainstream educational set-ups had a support teacher.

Communication mode. To study the impact of communication mode at home, parents were asked to choose between the following categories: use of spoken language only, use of spoken language and supportive signs, or use of spoken language and sign language. Forty children used spoken language only, 85 children had a mixture of spoken language and supportive signs, and 30 children used a mixture of spoken language and sign language (Table I). Thus, 74% of the normal-hearing parents used a mixture of spoken language and sign support or sign language, and 26% communicated by means of spoken language only.

CI center. Geographical placement was a factor of interest in order to analyze whether there was a significant difference between the two pediatric CI centers in Denmark. In all, 83 children attended the East Danish CI Center and 72 children attended the West Danish CI Center (Table I).

Implantation ear. To study whether implantation on the right vs left side and whether monaural vs bilateral implantation affected the speech and language outcome, implantation ear was included as a factor of interest.

Gender. Gender represented the last of the seven factors of interest. The categorical distribution of the children is summarized in Table I.

Description of applied tests

All children came to one of the two pediatric CI centers in Denmark for testing accompanied by one or two parents. Three different speech and language pathologists tested the children and performed structural interviews of the parents. All tests were carried out using spoken language. If necessary, some children were given explanation about the test by use of supportive signs. All tests and assessments were scored by one speech and language pathologist. As described above, the test battery performed by each child depended on the child's hearing age.

Tait video analysis. In all, 154 children performed the Tait video analysis (1 missing value). The purpose of the Tait video analysis was to assess the children's auditory awareness when communicating with an

Table II. Distribution of scores from all tests.

Tests	Scores for all children (n)	%
Tait video analysis		
$<50\%$ NLVT	50	32%
$>50\%$ NLVT	104	68%
Reynell test		
Below norm	40	45%
Equal to or above norm	48	55%
Phonological test		
$<50\%$ correct	18	30%
$>50\%$ correct	43	70%
Vocabulary		
Lower 50%	23	38%
Upper 50%	38	62%
Capacity of auditory performance, CAP		
Low score, CAP 0–4	31	20%
High score, CAP 5–7	124	80%
Speech intelligibility rating, SIR		
Low score, SIR 1–2	52	34%
High score, SIR 3–5	103	66%

adult. Children were filmed in interaction with one of the testers, and turn-takings were identified, which were the instances where the child had an opportunity to communicate. The auditory awareness of the adult's speech was measured by the number of non-looking-vocal-turns (NLVTs). An NLVT was counted when the child vocalized communicatively in a turn without being in eye contact with the adult during the adult's previous turn. No visual cue, no sign or pointing finger from the adult was given. For the statistical analysis, the NLVT score was defined as a dichotomous variable, either below or above 50% NLVT (Table II).

Reynell test. The Reynell receptive part was used to study the children's comprehension of spoken language. The standard of the Reynell test starts at 2 years of age and therefore the children in our study had to have a minimum of 2 years hearing age (n = 88). In accordance with the standard score for the Reynell test, a discrete version was defined as performance below the norm or equal to/above the norm (Table II).

Phonological test. To study the children's speech production, a Danish phonological test 'Sproglydstesten' (Speech Sound Test) was used. The phonological test was performed by children with at least 36 months of hearing (n = 61). The test consists of 90 different Danish phonemes and phoneme constellations. The children had to pronounce 52 different words presented as pictures in a closed-set format. The children's productions were analyzed for vowels (V) and clusters of V and consonants (C). The transcriptions consisted of the following constellations: 13 V, 15 CV, 12 VC, 16 CCV, 7 SC, 5 SCC, 9 VCC, and 13 finals. The results from the phonological test were recorded as a dichotomous variable: a score $<50\%$ or a score $>50\%$ correct production (Table II).

Vocabulary. The children's vocabulary was tested by use of a Danish vocabulary test 'Viborg materialet'. The test evaluates the active vocabulary and was performed by children with a hearing age of at least 36 months (n = 61). The score was categorical and classified as a lower 50% score and an upper 50% score (Table II). The children's scores were based on a standard from normal-hearing children with an age range from 3 to 7 years.

Capacity of auditory performance (CAP). The parents assessed the children's performance of auditory capacity (CAP). The results from the CAP assessments were coded with an eight-point scale from 0 being 'No awareness of sounds' to 7 being 'Use of telephone with known listener'. The test was performed for all hearing ages (n = 155). The results were resumed in a dichotomous variable, containing information as to whether the child was able to understand at least some sentences without lip-reading or not. Therefore, a low score was defined as CAP level 0–4 and a high score as CAP level 5–7 (Table II).

Speech intelligibility rating (SIR). The SIR was chosen as an outcome measure for conversational speech intelligibility. The test was performed for all hearing ages (n = 155). The score was defined as a dichotomous variable, indicating whether the speech was intelligible for at least an experienced listener, when the topic is known (SIR level 3–5), or whether the speech could not be understood (SIR level 1–2) (Table II).

As described, the responses were categorized in all tests and it should be noted that responses were not available for all children in all tests, since the child's performance depended on the hearing age. Table II summarizes the distribution of children in all test response categories.

Data analysis

The Department of Biostatistics, University of Copenhagen performed all statistical analyses. The categorization of the factors of interest and the test responses was carried out after guidance from the

Table III. Logistic regression results – statistically significant factors.

	Responses					
Factors	Tait video analysis	Reynell test	Phonology	Vocabulary	CAP	SIR
Hearing age	$p = 0.0002$ $\chi^2 = 17.43$	X	X	X	X	$p < 0.0001$ $\chi^2 = 22.70$
Implantation age	X	X	X	$p = 0.0154$ $\chi^2 = 5.87$	X	X
Communication mode	$p < 0.0001$ $\chi^2 = 20.62$	$p < 0.0001$ $\chi^2 = 18.50$	$p = 0.0148$ $\chi^2 = 8.43$	$p = 0.0025$ $\chi^2 = 11.99$	$p = 0.0010$ $\chi^2 = 13.76$	$p = 0.0017$ $\chi^2 = 12.78$
Center	X	X	X	$p = 0.0147$ $\chi^2 = 5.95$	$p = 0.0294$ $\chi^2 = 4.74$	X
Gender	X	$p = 0.0053$ $\chi^2 = 7.78$	$p = 0.0498$ $\chi^2 = 3.85$	$p = 0.0271$ $\chi^2 = 4.88$	X	X

statistician and was made in order to be able to use exact statistical tests and also due to the relatively small sample size. In order to study and describe the relationship between the responses and the factors of interest listed above, data were analyzed with logistic regression models. Backwards elimination was used to choose a final model, including only the statistically significant factors. Table III summarizes the significant results. Goodness of fit was performed by use of the Hosmer-Lemeshow test. Odds ratio estimates for the final model fitted for each of the responses are presented in Table IV.

Results

Factors with a significant effect on test responses

As shown in Table III, the communication mode at home was the only factor that had a statistically significant effect on all test responses. A statistically significant effect was also found for hearing age (Tait video analysis score and SIR score), for gender (Reynell test score, phonological and vocabulary test score), for implantation age (vocabulary test score), and for center (vocabulary and CAP test scores). The two factors

Table IV. Odds ratio estimates and 95% Wald confidence intervals for all responses.

Effects		Tait video analysis	Reynell test	Phonology	Vocabulary	CAP	SIR
Mode of communication	Danish vs sign language	28.00 [6.21, 126.31]	> 100*	41.25 [3.34, 508.5]	> 100*	> 100*	10.66 [2.85, 39.85]
	Sign support vs sign language	5.94 [2.13, 16.56]	3.53 [0.64, 19.49]	3.4 [0.66, 18.25]	21.20 [1.67, 267.83]	6.13 [2.35, 15.98]	3.66 [1.34, 9.98]
	Danish vs sign support	4.71 [1.27, 17.49]	61.82 [6.75, 566.10]	11.8 [1.33, 104.5]	39.05 [2.56, 594.60]	> 100*	2.91 [0.96, 8.86]
Hearing age	Hearing age > 36 months vs 24–35 months	2.38 [0.67, 8.41]					5.12 [1.49, 17.57]
	Hearing age > 36 months vs 6–23 months	7.90 [2.91, 21.43]					12.25 [4.35, 34.50]
	Hearing age 24–35 months vs 6–23 months	3.32 [1.09, 10.11]					2.39 [0.88, 6.52]
Gender	Girl vs boy		6.98 [1.95, 24.97]	3.73 [1.00, 13.93]	7.03 [1.25, 39.63]		
Implantation age	> 36 months vs < 36 months				17.43 [1.73, 175.82]		
Center	East vs West				14.45 [1.69, 123.60]	2.84 [1.11, 7.27]	

*The effect is highly significant but an exact quantification is not meaningful.

educational placement and ear of implantation did not have any statistically significant effects on the speech and language outcome for the present population.

Quantification of the effects (odds ratio estimates)

Table IV shows that children exposed to spoken language only had greater odds of scoring high in all tests, compared with children exposed to some degree of either sign support or sign language. The greatest difference was found between children with spoken language only and children with sign language. Children exposed to sign support had higher odds of scoring high than children exposed to sign language, except for the results from the Reynell test. A more detailed account of the findings for each of the tests is presented below.

Tait video analysis

Of 154 children, 50 children had a score below 50% NLVT and 104 children had a score above 50% NLVT (Table II). The interpretation of the odds ratio for the Tait video analysis is as follows: the odds of a child exposed to spoken language having a score higher than 50% NLVT is 28.00 times greater than that of a child exposed to sign language (Table IV). Thus, the communication mode at home had the largest effect on the Tait video analysis score. Goodness-of-fit: $\chi^2 = 2.8207$, df = 7, p value = 0.9011.

Reynell test

Forty children performed below the norm and 48 children performed equal to or above the norm (Table II). As seen in Table IV, extremely high odds ratios (61.82 and > 100) for performing equal to or above the norm were found for children exposed to spoken language only, compared with children exposed to sign support or sign language. No significant difference existed between children exposed to sign support and sign language. The gender effect showed that girls perform better than boys with an odds ratio of 6.98. Goodness-of-fit: $\chi^2 = 0.6152$, df = 4, p value = 0.9614.

Phonological test

The scores for the phonological test were recorded in a dichotomous variable with a score < 50% or a score > 50% correct production (Table II). In all, 18 children (30%) scored < 50% correct and 43 children (70%) scored > 50% correct and thus showed good capabilities of learning to produce Danish phonemes and phoneme constellations correctly.

As seen in Table III, two variables were statistically significant for the outcome, i.e. gender and communication mode. Table IV shows that communication mode had the largest effect. Children given spoken language only had 41.25 times higher odds of scoring high in the phonological test, compared with children communicating by a mixture of spoken language and sign language. Girls had 3.73 times higher odds of performing better than boys. Goodness-of-fit test: $\chi^2 = 6.0932$, df = 4, p value = 0.1923.

Vocabulary

A total of 23 children performed in the lower group and 38 children performed in the medium/upper group (Table II). As seen in Table III, four variables with significance for the outcome were found, i.e. communication mode at home, gender, center, and implantation age. Table IV shows that the children exposed to spoken language had > 100 times higher odds of performing in the upper 50% group than children exposed to sign language, whereas children exposed to sign support had 21.20 times higher odds than children exposed to sign language. Girls had 7.03 times higher odds of performing in the upper group. Compared with the West Danish Cochlear Implant Center, children from the East Danish Cochlear Implant Center had 14.45 times higher odds and children implanted before 36 months of age had 0.06 times higher odds of performing in the upper 50% group. Goodness-of-fit: $\chi^2 = 1.4551$, df = 8, p value = 0.9934.

Capacity of auditory performance, CAP

In all, 31 children scored at level 0–4 and 124 scored at level 5–7 (Table II). Thus, 80% of the children displayed high auditory potentials, with a minimum of being able to understand common phrases without lip-reading in everyday situations and a maximum of being able to understand a well-known speaker on the phone. The communication mode at home had a significant effect on the children's CAP (Table III). Due to the pattern of the data (quasi-complete separation), the algorithm converge, but the estimation of the odds ratio comparing children exposed to spoken language with children exposed to sign language or sign support did not give any further information. However, children exposed to sign support had 6.13 times higher odds of performing at CAP level 5–7 than children exposed to sign language (Table IV). Goodness-of-fit: $\chi^2 = 0.3958$, df = 3, p value = 0.9411.

Speech intelligibility rating, SIR

A total of 52 children were assessed by the parents to perform at the low level 1–2 and 103 children performed at the high level 3–5 (Table II). Two factors with significance for outcome were found: hearing age and communication mode (Table III). The odds ratio for performing well in the SIR was 10.66 times higher for children exposed to spoken language compared with children exposed to sign language (Table IV). The significance of the hearing age variable showed that children with a hearing age > 36 months were more intelligible than children with hearing ages between 6 and 23 months and between 24 and 36 months. Goodness-of-fit: $\chi^2 = 4.4897$, df = 6, p value = 0.6107.

Discussion

As noted, a very influential effect of the communication mode used at home was documented in our study. The communication mode was highly associated with all test responses and it was more highly associated with speech and language outcomes than any of the other investigated factors. This finding leads to the question as to why sign support and sign language was used as the communication mode by 74% of the parents in our study, although they were normal-hearing. Is it because the parents valued signs/sign language and found that the communication worked better when signs were used? This assumption was stressed in a study by Archbold et al. [17]. On the other hand it could be argued that the normal-hearing parents chose to communicate by means of sign language or sign support because they were guided to do so by the professionals that take care of hearing-impaired children in Denmark. The complexity of parents communicating in either a foreign language, i.e sign language, or foreign communication mode, i.e. sign support, is stressed in our study, as the results reveal a significant effect on the children's speech and language outcome.

In Denmark, there is a well-developed sign language program available for all families with a profoundly hearing-impaired child. Families can be referred to sign language courses following diagnosis. Furthermore, the guidance and teaching of children with CIs and their families occur at the schools for the deaf. This system has been/is optimal for deaf children without CIs, but may not be ideal for children with CIs. The significant difference found between the two CI centers may reflect the fact that at the West Danish CI Center, the schools for the deaf constitute a part of the CI center and thus provide the initial and primary guidance of the parents as a standard. The findings in our study stress the need for a revision of the aftercare of Danish children with CIs and their families. It is furthermore important to stress the importance of a continuous follow-up of the cochlear implanted children, to document the timeframe for full integration of the auditory sense and development of a verbal language. Setting a standard for how long it takes to integrate an auditory sense will enable us to inform future implant candidates and troubleshoot for complications regarding the technique or specific speech/language problems for a given child.

We found no effect of educational placement, which is unlike studies from Connor et al., Easterbrooks et al., and Tobey et al. [12–14]. In our population, 62% of the children were placed in schools/kindergartens for the deaf and 38% in mainstream educational placements with a support teacher. When looking at cross-tabulations between communication mode and educational placement it appears that 97% of children exposed to sign language were placed in schools/kindergartens for the deaf, 69% of the children with sign support were placed in schools/kindergartens for the deaf, and 20% of the children exposed to spoken language were placed in schools/kindergartens for the deaf. This indicates that educational placement and communication mode are factors that are somehow linked together.

Ear of implantation and bilateral vs monaural implantation did not show any statistically significant effect on the speech and language outcome for the present population. At the time of testing, only three children were bilaterally implanted and hence the data sample was too small to perform a statistically meaningful comparison with the unilaterally implanted children.

Unlike many other studies we have not found any effect of implantation age except for the results of the vocabulary tests [1,5–7]. This could possibly be explained by the fact that our study examined the children's outcomes at one point of time, rather than in a longitudinal set-up. The studies by Geers et al. [9,18,19] did not find an effect of implantation age either. These studies also documented outcome at one point in time rather than in a longitudinal set-up. Further investigation of the role of implantation age seems warranted. However, an effect of the hearing age was found, as longer use of the device leads to better performances as regards speech intelligibility and integration of the auditory sense in communication. This effect of hearing age is in agreement with previous investigations [1,8].

It could be argued that the scoring in the speech and language tests according to hearing age instead of chronological age may have introduced a bias in the present investigation. However, as described above, the spoken language level for many of the children was too poor to perform speech and language tests according to their chronological age. As an example, 69% of the children who scored below the norm in the Reynell test had hearing ages above 36 months. This is contrary to the finding described by Beadle et al. [8] and Connor et al. [1], that the length of implant use has a positive effect on language performance.

The extremely high odds ratio estimates for the significant effects of the communication mode for the Reynell test and the vocabulary test, i.e. > 100, may be due to the fact that there were relatively few observations. It should be noted that only 88 of the 155 children were eligible to perform the Reynell test and only 61 the vocabulary test. Therefore, it would be of great interest to further study these statistically significant effects on a larger sample size.

In conclusion, the present study demonstrates a very strong effect of the parental communication mode on the auditory capabilities and speech/language outcome for cochlear implanted children. The children exposed to spoken language had higher odds of scoring high in all tests applied, compared with children exposed to a mixture of spoken language with sign support or sign language. Children exposed to sign support had higher odds than children exposed to sign language. The most marked effect was found between children exposed to spoken language only and children exposed to sign language. Thus, our findings suggest a very clear benefit of spoken language communication with a cochlear implanted child.

Declaration of interest: The authors report no conflicts of interest. The authors alone are responsible for the content and writing of the paper.

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Significant regional differences in Denmark in outcome after cochlear implants in children

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ABSTRACT

INTRODUCTION: The objectives of the present study were to study regional differences in outcome for a paediatric cochlear implant (CI) population after the introduction of universal neonatal hearing screening (UNHS) and bilateral implantation in Denmark.

MATERIAL AND METHODS: Data relate to 94 subjects. A test battery consisting of eight different tests/assessments was performed in order to report the level of audition, speech, language and self-esteem. For data analyses of any associations between the regions, Fisher's exact test was used. Potential rater variability within either of the centres was assessed using logistic regression models.

RESULTS: The levels of audition were comparable between the group from West Denmark (West) and the group from East Denmark (East). In contrast, all tests of speech and language revealed a statistically significant difference between East and West. In all tests, West subjects scored significantly lower than East subjects. West children received more hours of speech therapy, more learning support assistance, and more parents used signing. Furthermore, the parents from West were significantly less involved in the auditory rehabilitation of their children than parents from East.

CONCLUSION: The results were remarkable and call for a thorough evaluation of both the quality and organization of the paediatric CI population with particular concern for the paediatric CI population of West Denmark.

FUNDING: The Oticon foundation financially supported this study. Trial registration was not relevant as the study is designed as a prospective case series.

TRIAL REGISTRATION: not relevant.

The implementation of universal neonatal hearing screening (UNHS) and the increased use of bilateral cochlear implants (CI) have provided more and more children with profound hearing loss access to bilateral auditory signals during their first year of life [1, 2]. Recent reports indicate that unilateral cochlear implantation in prelingually deaf children within the first year of life may result in speech and language skills comparable to those of children with normal hearing [3-6]. In Denmark, UNHS was implemented in January 2005, and children have been offered bilateral implantation either simultaneously or sequentially as from September 2006. It has previously been reported that outcomes of CI were associated with the Danish Region from which the children originated [7], and it is therefore of great interest to study whether such regional differences still exist after the introduction of UNHS and bilateral implantation. This new group of children with CI has now reached an age where testing of outcomes of audition, speech and language is possible. The aim of this study was to investigate whether regional differences in CI outcome still exist between East and West Denmark.

METHODS AND MATERIAL

The patient material comprised a total of 94 children with CI, 52 girls and 42 boys. The sample included all children in Denmark who were born between January 2005 and January 2011, and who received CI and had a minimum of six months of hearing with their CI. Fifty children were implanted at the East Danish CI Centre, Copenhagen University Hospital (Rigshospitalet), and 44 children were implanted at the West Danish CI Centre, Aarhus University Hospital. 74% (n = 69) of the children were diagnosed through UNHS. A total of 25 children were diagnosed later for various reasons, i.e. four children came from Greenland and the Faroe Islands where UNHS has not been implemented; eight children had hearing impairment following meningitis and thus were not born with a hearing loss; the remaining 13 children were not found through UNHS for reasons unknown. The distribution of diagnoses was: congenital non-specificata = 53%, congenital hereditary = 13%, post-infectious cytomegalovirus (CMV) = 2%, meningitis = 10%, Pendred syndrome = 13%, auditory neuropathy spectrum = 5%, CHARGE association, Waardenburg and Usher syndromes = 4%. 22% (n = 21) of the children were also diagnosed with an additional handicap, including vision problems (n = 8), mental retardation (n = 8), cerebral palsy (n = 2), club foot (n = 1) and epilepsy (n = 2). 52% of the children with additional handicaps were implanted at the East Danish CI Centre and 48% were implanted at the West Danish CI Centre. In general, the two centres followed the same procedures after implantation as regards to time interval from operation to first switch-on and the frequency of tuning sessions, which varied depending on the individual child's and its family's needs. The tunings at the two centres were carried

TABLE 1

Characteristics of all 94 cochlear implant recipients

	East	West	Total
Median age for start of hearing aid pre-implantation	4 months (n = 44)	6 months (n = 38)	4 months (n = 82)
Median age of implantation	12 months (n = 50)	19 months (n = 44)	13.5 months (n = 94)
Median age for day of testing	47 months (n = 46)	46 months (n = 37)	47 months (n = 83)
Simultaneous bilateral implantation	82% (n = 41)	52% (n = 23)	68% (n = 64)
Sequential bilateral implantation	14% (n = 7)	16% (n = 7)	15% (n = 14)
Unilateral implantation	4% (n = 2)	32% (n = 14)	17% (n = 16)

TABLE 2

Applied tests/assessments, age criteria for testing, number of recipients tested and test result categories

Tests/assessments	Minimum age criteria for testing	Number of CI recipients tested	Test result categories
Receptive vocabulary (PPVT-4)	24 months	68	Below age/equal to or above age
Receptive language (Reynell)	24 months	71	Below norm/equal to or above norm
Phonological test	35 months	49	< 25% correct/26-50% correct/ 51%-75% correct/> 75% correct
Active Vocabulary test	35 months	49	Lower 25%/middle 50%/upper 75%
CAP	6 months	82	Low score (CAP 0-4)/ High score (CAP 5-7)
SIR	6 months	82	Low score (SIR 1-2)/high score (SIR 3-5)
Discrimination of minimal pairs	48 months	33	< 50% correct/>50% correct
Self-esteem	17 months	79	Low score (< 36)/high score (> 36)

CAP = capacity of auditory performance
CI = cochlear implant
PPVT-4 = Peabody Picture Vocabulary Test-4
SIR = speech intelligibility rating

out by technicians at the East Danish Centre and by audiologists/engineers/technicians at the West Danish Centre. 11% (n = 10) of the children had a non-Danish ethnical background. All parents were normally hearing except for one mother who had a CI herself. Ninety-three children had a Nucleus product and one child had a Med El product. Further characteristics of the population regarding age of implantation, bilateral simultaneously versus sequential implantation age are summarised in **Table 1**. Parents received a letter with a description of the study and were then contacted by telephone in order to find a date for testing and interviewing. The participation rate was 88% (n = 83). Eleven families chose not to take part in the study for various reasons. No common denominator was observed among the children who did not participate regarding age, additional handicap or origin (East or West Denmark).

The participating families came to one of the two paediatric CI centres for testing accompanied by one or both parents. Four speech- and language pathologists, two from each centre, carried out the tests and the parental interviews. All testers used spoken language and all test results were scored according to standards from normally hearing children. **Table 2** summarizes all tests and assessments used, the age criteria for each test, number of responses and standardised test categories for scoring.

The Peabody Picture Vocabulary Test-4 (PPVT-4) is a widely used norm-referenced test of receptive vocabulary. During a test, children were required to point to one of four pictures that represented the word produced by the tester. For the study of the children's comprehension of spoken language, the Reynell receptive part was used. The study of the children's speech production was carried out with the "Sproglydstesten". A test consisting of 90 different Danish phonemes and phoneme constellations in a closed-set format. The children's active vocabulary was tested by use of the Danish "Viborgmaterialet". In this test, the children had to actively name an object shown on a picture. For test of auditory discrimination, the Bent Kjærs (BKS) test was used; children had to point to the last word heard out of two minimal pairs. The parents assessed the children's capacity of auditory performance (CAP), and parents also assessed the children's speech intelligibility (SIR). For a high score on the CAP, the child must be able to understand at least some sentences without lip-reading; and for a high score of the SIR, the child's speech must be intelligible at least for an experienced listener. The parents, furthermore, assessed the child's self-esteem. The assessment scale applied was based on social well-being studies from normally hearing children performed by the National Institute of Public Health [8]. Parents completed a seven-point rating scale to determine the degree of their child's personal-social adjustment by assessing whether the child was: dependent versus independent, passive versus active, lonely versus social, worried versus not worried, sad versus happy, and insecure versus confident. In the interview, respondents stated their educational background, the number of hours their child had a support teacher per week, the number of hours of speech and language rehabilitation and the degree of parental involvement in the rehabilitation.

As referred, the responses were categorized according to standard in all tests, and it should be noted that responses were not available for all children in all tests, since the child's performance depended on the child's age. In addition, not all children were willing to cooperate in all tests. Table 2 summarizes the categorization and numbers of all test responses.

Data analysis

The data distribution for all tests and the regional differences are presented. For comparison of categorized outcome between East and West, the χ^2 test or Fisher's exact test (when n > 5 in any category) was used. A statistical significance level of 5% was chosen. Potential rater variability within either of the centres was assessed using logistic regression models.

Trial registration: not relevant.

RESULTS

Table 3 summarizes the distribution of all responses for the East versus the West population. The responses from the receptive vocabulary test showed a statistically significant difference between East and West, p < 0.001, with better scores in the East. The same was found for the test of receptive language, p = 0.005, speech production p = 0.045, and active vocabulary p = 0.058. Responses from CAP, SIR and BKS did not show any statistically significant differences between East and West. The parents' assessments of their children's levels of self-esteem were significantly different between the two regions, p = 0.005, with a higher level of self-esteem among children from East than from West Denmark. Potential rater variability within either of the centres was assessed using logistic regression models. Except for SIR, no significant rater variability was found. This indicates that regional differences are not an artifact caused by rater variability. **Table 4** summarizes other regional differences, i.e. number of hours of rehabilitation per week, parental participation in the rehabilitation, number of hours with a learning support assistant per week, the parental mode of communication, educational placement and paid reduction of work hours for parents. As for the test responses, regional differences were also found for these variables except for the reduction of work hours. In West Denmark, 71% of the parents stated that they did not participate in the rehabilitation of their child compared with 37% in East. This difference was statistically significant, p = 0.001. 95% of the children from West received 1-2 hours of rehabilitation per week compared with 53% from East, p < 0.001. 53% of the children from West were provided with a support teacher > 15 hours per week compared with 21% in East, p = 0.017. 24% of the West parents stated that they used a combination of spoken language and signing as communication mode in comparisons with 7% in East, p = 0.031. 96% of the East children were placed in mainstream educational settings in comparison with 73% of the West children, p = 0.0005. No significant difference was found as regards to parents' amount of financial compensation for reduced working hours, i.e. 61% and 59% of parents from East and West, respectively, were paid for reducing their number of working hours.

DISCUSSION

Children with CI in Denmark can hear and discriminate just as well as children reported in international studies [4, 9-11]. It is remarkable, however, that on speech and language parameters such as receptive vocabulary, active vocabulary, receptive language and speech production, the results are significantly poorer in West than in East Denmark. Receptive vocabulary and receptive language are, furthermore, parameters of great importance as they are defined to be the most vulnerable parameters for language development [12]. These findings call for explanations why such differences exist in Denmark.

Although speculative, the fact that children were implanted somewhat later and that only 67% received bilateral implants in West Denmark (compared with 96%

TABLE 3

Distribution of test results in relation to geographical region.

		East, % (n)	West, % (n)	p values
Receptive vocabulary PPVT-4 (n = 68)	< age	41 (14)	91 (31)	0.00002
	> age	59 (20)	9 (3)	
Receptive language Reynell (n = 71)	< age	51 (18)	83 (30)	0.005
	> age	49 (17)	17 (6)	
Phonology Sproglydstesten (n = 49)	< 25%	4 (1)	17 (4)	0.045
	26-50%	12 (3)	4 (1)	
	51-75%	16 4)	42 (10)	
	> 75%	68 (17)	38 (9)	
Active vocabulary Viborgmaterialet (n = 49)	Lower 25%	28 (7)	63 (15)	0.058
	Middle 50%	36 (9)	17 (4)	
	Upper 75%	36 (9)	21 (5)	
Capacity of auditory performance (n = 82)	Low level	7 (3)	8 (3)	1.00
	High level	93 (42)	92 (34)	
Speech intelligibility rating (n = 82)	Low level	31 (14)	24 (9)	0.62
	High level	69 (31)	76 (28)	
Discrimination minimal pairs (n = 33)	< 50%	6 (1)	0 (0)	1.00
	> 50%	94 (16)	100 (16)	
Self-esteem (n = 79)	< 36	2 (1)	24 (9)	0.005
	> 36	98 (41)	76 (28)	

TABLE 4

Regional differences of rehabilitation of the 83 tested recipients.

		East, % (n)	West, % (n)	p values
Rehabilitation hours per week (n = 80)	None	37 (16)	5 (2)	0.00008
	1-2	53 (23)	95 (35)	
	3-5	9 (4)	0 (0)	
Parent participation (n = 62)	No	37 (10)	71 (25)	0.001
	Yes	63 (17)	29 (10)	
Support teacher hours per week (n = 78)	None	31 (13)	17 (6)	0.017
	5-15	48 (20)	31 (11)	
	>15	21 (9)	53 (19)	
Parental mode of communication (n = 82)	Spoken + sign	7 (3)	24 (9)	0.031
	Spoken only	93 (42)	76 (28)	
Educational placement (n = 83)	Special	4 (2)	27 (10)	0.005
	Mainstream	96 (44)	73 (27)	
Paid compensation (n = 80)	No reduction	37 (17)	41 (14)	0.82
	Reduction	63 (29)	59 (20)	

Test person Caroline in communication with normally hearing brother (photo reproduced with permission from the parents).

in East) may partly explain their poorer outcome [13, 14]. The documented differences in parental communication mode, parental participation, educational placement, number of rehabilitation and learning support assistant hours, however, suggest that the most likely explanation for the significant differences in CI outcome in Denmark lies in the different organisations of the Danish paediatric CI population. In East Denmark, the primary guiding comes from the CI centre in close collaboration with the local speech and language therapist. In West Denmark, the former schools for the deaf play an important role in the guiding of the parents. In the West, children are referred to the paediatric advisory services at the former schools for the deaf once the diagnosis of hearing impairment has been established at the audiological hospital clinic. These advisory services at the former schools of the deaf with a core tradition of teaching sign language to deaf children are thus the parents' first-hand rehabilitation contact; their contact is not with the team at the CI centre, which focuses on and stresses the evidence-based importance of the use of spoken language and parental involvement in the rehabilitation of the children with CI. This fact probably explains why 24% of the parents in West Denmark as opposed to 7% in East still were using a combination of spoken language and signs for their child, even though the detrimental impact of this on the outcomes of speech/language and social well-being is well documented [7, 15]. Some may argue that West children do not get enough speech and language therapy after implantation, and that they do not have enough learning support assistance. This study does, however, question the value of the contents and the absolute amount of provided hours of learning support assistance, as well as speech and language therapy provided by advisory services at the former schools for the deaf. The West population received markedly more hours of rehabilitation and support teaching, even though they do not have more frequent or more severe additional handicaps and although they hear and discriminate as well as their East counterparts. However, as presently indicated, this has no positive effect on their outcome. This questions the contents, quality and socio-economic value of these efforts.

The significant difference in parental participation is noteworthy, as it is reported that parents play the most important roles in infants' language development and that rehabilitation of babies and toddlers must involve parents in order to secure positive outcomes [16-19]. This is, however, not the case for a stunning 71% of the families in West Denmark and a far too high 37% in East. This finding is, furthermore, in contrast to our finding that 59-61% of the parents received financial compensation for reduced hours of work and were thus provided with the possibility of being intimately involved in the rehabilitation of their child. In January 2011, the National board of Health launched a protocol on paediatric cochlear implantation which stresses the importance of evidence-based auditory stimulation and parental involvement after implantation. The protocol recommends that the initial auditory rehabilitation is placed at the CI centres in close contact with local speech and hearing therapists [20]. This protocol is, indeed, a great step forward for the total paediatric CI population in Denmark, but it cannot be overheard that 91% of the West children and 41% from the East did not perform at an age-equivalent level in the receptive vocabulary test. These children are at serious risk of never closing the gap between chronological and hearing age, which is the core purpose of paediatric cochlear implantation. This is again very likely to have a lifelong negative impact on the future educational level and subsequent vocational status for the implantees, thus effecting the socio-economic investment and outcome negatively.

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Language understanding and vocabulary of early cochlear implanted children

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ABSTRACT

Objectives: The aim of the study was to identify factors associated with the level of language understanding, the level of receptive and active vocabulary, and to estimate effect-related odds ratios for cochlear implanted children's language level.

Methods: The patient material included all children born in Denmark between January 2005 and January 2011, having received a cochlear implant (CI) and with a minimum of 6 months of hearing with their CI ($N = 94$). The participation rate was 88% ($N = 83$). Sixty-eight (82%) of the participating children were implanted bilaterally. Mean age at implantation was 19.6 months. The mean age at test was 46.3 months and the mean age of hearing with CI was 25.9 months. The children were tested with three different tests, the PPVT-4, the Reynell receptive part and a Danish test "Viborgmaterialet" for active vocabulary. Logistic regression models were used for analysis of the potential influence of eighteen different factors upon the test outcomes.

Results: The majority of children did not have age equivalent language understanding and vocabulary. There was significant effect of the following factors upon the test outcomes: age at hearing aid start before implantation, age at implantation, length of hearing, communication mode, mode of implantation, amount of support teaching, residence and educational placement. Children who started HA treatment before 6 months of age, were implanted before 12 months or did not use total communication had the highest odds of having age equivalent language understanding and vocabulary.

Conclusions: The majority of hearing impaired children in Denmark received hearing aids before six months of hearing and the majority was implanted before 18 months of age. Despite these medical and technical advances the vast majority did not have age equivalent language understanding and vocabulary. Data suggest that the language gap is not closed in two years after implantation.

1. Introduction

Cochlear implantation as a treatment for children with profound hearing loss is nowadays regarded as standard [1–4] and various studies document auditory and speech/language progresses never before described for a profoundly hearing impaired pediatric population [5–7].

The implementation of universal neonatal hearing screening (UNHS) and the increased use of bilateral cochlear implants (CIs) have provided more and more children with profound hearing loss access to bilateral auditory signals during their first year of life [7–10]. Recent reports indicate that cochlear implantation in prelingually deaf children within the first year of life may result in speech and language skills comparable to those of children with normal hearing [7,8,11–14]. In addition, a study of the cost-effectiveness of pediatric bilateral cochlear implantation, furthermore, suggests that it is a cost-effective use of health care resources [15].

Various factors may have an impact on the auditory and speech/language development following cochlear implant. Thus, several studies have described that age at first hearing aid (HA) fitting is a factor predicting later language levels for hearing impaired children [16–18]. Another important factor described in the literature is age of implantation and recent studies [2,5–7] report that implantation before 12 months is associated with age equivalent language development. The length of device use affects the language outcomes [2,7], which is also the case for the rehabilitation and the educational placement following cochlear

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implantation [4,19,20]. Gender has been documented to impact scores on speech/language outcomes, as girls score higher than boys [1,13]. The educational level of the parents also affects the speech/language outcomes [18]. In addition, the method of implantation, i.e. bilateral simultaneously or sequentially or a bimodal (CI in one ear and HA in the other) stimulation, may be an important factor to take into consideration when investigating the children's language outcomes [3,21–24]. Children with syndromes and additional handicaps such as cerebral palsy, mental retardation, e.g. global developmental delay or autistic spectrum disorders, are now not routinely precluded from being offered a cochlear implant [25,26] and thus it is of interest to investigate outcomes of children with CI and additional handicaps.

For a Danish pediatric CI population, it has previously been described that parental mode of communication is essential for language outcomes and furthermore that differences exist between region of residence [27]. In the present study, we investigate the level of receptive language, active vocabulary and language understanding for a Danish pediatric cochlear implant population after the introduction of the nationwide UNHS. Furthermore, the impact of various factors upon language outcome is analyzed. In Denmark, UNHS was implemented in January 2005 and since September 2006 children have, when indicated, been offered bilateral implantation, either simultaneously or sequentially.

2. Materials and methods

The patient material consisted of all children born in Denmark between January 2005 and January 2011, having received a CI and with a minimum of 6 months of hearing with their CI ($N = 94$).

The parents were mailed a letter with a description of the study and then contacted by telephone in order to find a date for testing and interviewing. The participation rate was 88% ($N = 83$). Eleven families chose not to take part in the study for various reasons. There were no characteristic differences seen for the children that did not participate regarding gender, age, coming from the eastern or western part of Denmark, implanted sequentially or simultaneously or having a bimodal stimulation, diagnosis, educational placement. The only noteworthy fact was that three of the eleven children that did not participate were diagnosed with mental retardation. The distribution of gender was 36 boys (43%) and 47 girls (57%). Fourty-six (55%) children came from the East Danish CI center and 37 (45%) came from the West Danish CI center. Fourty-nine (59%) of the parents were paid compensation from government to have time off during the week in order to handle their hearing impaired child. Seventy-one children (86%) were placed in a mainstream educational setting, which referred to a situation where the child with CI was included into a normal hearing educational setting. Twelve children (14%) were placed in special education which referred to kindergartens specifically for hearing impaired children. Table 1 summarizes the distribution and further characteristics of the population regarding diagnosis, pre-implant treatment, age of implantation, bilateral simultaneously/sequentially or bimodal stimulation, implantation age, age at test, additional handicap, communication mode, parents' educational level and number of support teaching per week. The mode of implantation referred to whether a child was implanted on both ears simultaneously or sequentially, i.e. one ear at a time. Data were not gathered for the interval between the sequential implantations. Bimodal referred to children who wore a cochlear implant on one ear and a hearing aid on the other ear. The variable rehabilitation included only the number of hours provided to each family and not what kind of rehabilitation the child received. For the variable start age of hearing age preimplant data was not collected for whether that also included rehabilitation. Support teacher referred to whether the child had a special teacher allocated specifically to him and if so the number of hours of support teaching. Seventy-four per cent ($N = 69$) of the children were diagnosed through UNHS. Twenty-five children were diagnosed later due to various reasons i.e. four children came from Greenland and the Faroe Islands where UNHS has not been implemented, eight children had hearing impairment following meningitis and thus were not born with a hearing loss, the remaining 13 children were not found through UNHS for reasons unknown.

Eleven per cent ($N = 10$) of the children had a non-Danish ethnical background. All parents were normal hearing except for one mother who had a CI herself. Ninety-three children had a Nucleus product and one child had a Med El product. Fifty children were implanted and tested at the East Danish CI center and 44 children were implanted and tested at the West Danish CI center.

Sixty-eight (82%) of the children were implanted bilaterally. Mean age at implantation was 19.6 months (median; 14, range 5;

Table 1
Characteristics of tested children ($N = 83$) and distribution of explanatory variables.

Covariate	Category	N (%)	Covariate	Category	N (%)
Diagnosis	Auditory neuropathy	5 (6%)	Support teacher hours per week	None	19 (23%)
	CMV	2 (2%)		5–10 h	16 (19%)
	Cong nonspecified	45 (54%)		10–15 h	15 (18%)
	Hereditary	9 (11%)		>20 h	16 (19%)
	Meningitis	7 (8%)		Missing	5 (6%)
Additional handicap	Cerebral palsy	2 (2%)	Communication mode	Spoken	70 (84%)
	Epilepsy	2 (2%)		Spoken + sign	12 (14%)
	Mental retardation	5 (6%)		Missing	1 (1%)
HA pre-implant start age	No pretreat	9 (11%)	Education mother	<10 years	10 (12%)
	0–5 months	45 (54%)		10–11 years	6 (7%)
	6–11 months	9 (11%)		12–13 years	11 (13%)
	12+ months	20 (24%)		>13 years	54 (65%)
Age at implantation	5–11 months	28 (34%)		Student	2 (2%)
	12–17 months	19 (23%)	Education father	<10 years	9 (11%)
	18+ months	36 (43%)		10–11 years	4(5%)
Age at test	6–23 months	8 (10%)		12–13 years	15 (18%)
	24–35 months	20 (24%)		>13 years	52 (63%)
	36+ months	55(66%)		Student	2 (2%)
Hearing age with CI	6–23 months	46 (55%)		Missing	1 (1%)
	24–35 months	13 (16%)	Method of implantation	Sequential bilateral	12 (14%)
	36+ months	24 (29%)		Simultaneous bilateral	56 (67%)
				Bimodal (HA + CI)	15 (18%)

Table 2
Distribution of test outcomes.

Test/assessment	Categories	Test age	Number of children tested	Not testable	Not relevant for age	Scores
PPVT-4	<Age >Age	≥24 months	68	7	8	<Age = 45 (66%) >Age = 23 (34%)
Reynell	<Age >Age	≥24 months	71	4	8	<Age = 48 (68%) >Age = 23 (32%)
Vocabulary	0–25% 26–75% 76–100%	≥35 months	49	8	26	0–25% = 22 (45%) 26–50% = 13 (27%) 76–100% = 14 (29%)

55). Mean age at test was 46.3 months (median 47, range 17–74). Mean age of hearing with CI was 25.9 months (median 21, range 6–59). Nine children did not wear HAs before implantation and hence the mean and median age for this parameter is based on 74 subjects. Mean age of first HA-fitting was 8.0 months (median 4, range 3–36).

2.1. Description of applied tests

The Peabody Picture Vocabulary Test-4, PPVT-4, is a widely used norm-referenced test of receptive vocabulary. During the test, children were required to point to one of four pictures that represented the word produced by the tester. For studying of the children's comprehension of spoken language, the Reynell receptive part was used. Receptive vocabulary and receptive language are, furthermore, parameters of great importance as they are defined to be the most vulnerable parameters for language development [28]. The children's active vocabulary was tested by use of the Danish "Viborgmaterialet". In this test, the children had to actively name an object shown on a picture. In an interview, the parents stated their educational background, the number of hours their child had a support teacher per week, the number of hours of speech and language rehabilitation and degree of parent involvement in the rehabilitation.

As referred, the responses were categorized according to standard in all tests and it should be noted that responses were not available for all children in all tests, since the child's performance depended on the child's age. In addition, not all children were willing to cooperate in all tests. Table 2 summarizes the categorization, numbers of all test responses, number of non-testable children, number of children for whom the test was not relevant for age and the distribution of the scores in number and percentage.

2.2. Data analysis

Statistical analysis was conducted by the Department of Biostatistics at the University of Copenhagen. Data were analyzed with Fisher's exact tests and logistic regression. In the logistic models, the vocabulary test (Viborgmaterialet) has been dichotomized into below or above 75%. The estimated odds-ratios and confidence intervals were based on Wald tests, whereas *p*-values for covariates with more than two levels were based on likelihood-ratio tests. The odds ratio estimates, OR, are presented with a reference, e.g. the reference was implantation age between 5 and 11 months compared to implantation age between 12 and 17 months or 18+ months. An odds ratio estimate of 0.25 for implant age 12–17 months means that children implanted in that period have only got 0.25 times the odds at performing at age equivalent level in the PPVT-4 test compared to implant age between 5 and 11 months. Four different raters, two in each region, tested the children. Some children were tested by a single tester and some children were tested with two testers present. By comparing logistic regressions including rater information with simple models only including region of residence, potential inter-rater differences were assessed. As East had more non-testable children than West, a sensitivity analysis was carried out.

3. Results

Seventy-one children were tested with the Reynell receptive part and 48 (68%) of the children did not have age equivalent language understanding (Table 2). Table 3 shows the results of this outcome when it was evaluated by logistic regression. Table 3 only shows the significant variables derived from the logistic regression analysis. It can be seen that when the regions were compared to one another the children from East had 4.72 times higher odds of scoring at age equivalent level compared to children from West (OR = 4.7). The logistic regression analysis, furthermore, showed that age of HA fitting pre-implant (OR = 0.06 for 12+ months vs. 0–5 months), age at implant (OR = 0.08 for 18+ months vs. 5–11 months), amount of support teaching (OR = 0.15 for more than 15 h vs. none), and educational placement (OR = 0.15) were found to be statistically significantly associated with the outcome (Table 3). Parental mode of communication was also significantly associated with the outcome, when evaluated by Fisher's exact test (odds ratio estimates were infinite, as no children using sign support when communicating with their parents scored at age equivalent levels).

Sixty-eight children's receptive vocabulary was evaluated by the PPVT-4 and 45 children (66%) did not have age equivalent vocabulary (Table 2). When the results of the PPVT-4 were evaluated by logistic regression analysis, the covariate region of residence again showed very high odds ratio estimates for children from the East for having an age equivalent language level compared to children from the West (OR = 14.76). Age at implantation was also found to be statistically significant and revealed that children implanted between 5 and 11 months had higher odds ratios compared to children implanted between 12 and 17 months or older than 18 months (OR = 0.25 for 12–17 months vs. 5–11 months and OR = 0.06 for 18+ vs. 5–11 months). Mode of implantation was borderline significant (OR = 8.57 for sequential vs. bimodal and 7.84 for simultaneous vs. bimodal; *p*-value = 0.051). Fisher's exact tests were significant for the covariates diagnosis, age at HA fitting pre-implant and communication mode of parents.

The children's level of active vocabulary was evaluated by use of the Danish vocabulary test "Viborgmaterialet". Forty-nine children were tested and the majority of the children (*N* = 22; 45%) scored in the lowest category with 0–25% correct (Table 2). When the scores from the vocabulary test was evaluated by logistic regression analysis and Fisher's exact test, the covariates age at implantation (OR = 0.69 for 12–17 months vs. 5–11 months and OR = 0.10 for 18+ months vs. 5–11 months) and length of hearing with CI (OR = 1.87 for 24–36 months vs. 6–23 months and OR = 6.25 for 36+ months vs. 6–23 months) were found to be statistically significant (Table 3). Children implanted simultaneously bilaterally had higher odds of performing at age equivalent levels than children with bimodal stimulation or sequentially implanted.

Table 3
Effect of significant covariates on test outcomes.

Covariates		Test		
		PPVT-4	Reynell	Vocabulary
		Odds ratio estimates (95% CI)	Odds ratio estimates (95% CI)	Odds ratio estimates (95% CI)
Region	West	Reference	Reference	No significant effect
	East	14.76 (4.277;70.361)	4.72 (1.641;15.17)	
Diagnosis	Cong. nonspec.	<0.001[*]	Reference	No significant effect
	Hereditary		0.20 (0.01;1.295)	
	Other		0.60 (0.115;2.553)	
	Meningitis		0.60 (0.115;2.553)	
	Pendred		0.311 (0.043;1.428)	
Age HA preimplant	No pretreat	<0.001[*]	0.55 (0.071;3.247)	No significant effect
	0–5 months		Reference	
	6–11 months		0.55 (0.105;2.471)	
	12+ months		0.059 (0.003;0.332)	
Age at implant	5–11 months	Reference	Reference	Reference
	12–17 months	0.25 (0.059;0.97)	0.34 (0.087;1.239)	0.69 (0.124;3.732)
	18+ months	0.06 (0.014;0.253)	0.08 (0.018;0.303)	0.10 (0.017;0.525)
Mode of implantation	Bimodal	Reference	No significant effect	No significant effect
	Sequential	8.57 (1.091;182.876)		
	Simultaneously	7.84 (1.353;149.558)		
Age hearing with CI	6–23 months	No significant effect	Reference	Reference
	24–36 months		0.67 (0.091;3.294)	
	36+ months		3.98 (1.325;12.814)	6.25 (1.329;45.9959)
Support	None	No significant effect	Reference	No significant effect
	5–15 h		0.81 (0.245;2.698)	
	>15 h		0.15 (0.029;0.628)	
Placement	Mainstream	No significant effect	Reference	No significant effect
	Special		0.15 (0.008;0.868)	
Communication	Spoken	0.012[**]	0.013[**]	No significant effect
	Spoken + S			

[*] *p*-Value from Fisher's exact test (not accounting for region) as the OR was estimated to be infinity or zero due to no observations in a cell in the underlying contingency table.

[**] *p*-Value from Fisher's exact test (not accounting for region) as the OR was estimated to be infinite since there are no children who use sign that have the best outcome for the tests.

The number of children tested by each tester was evaluated by a likelihood ratio test in order to find out whether the strong effect of region could be explained by a rater bias. This potential inter-rater difference (bias) was assessed by testing whether a model including precise information on who rated the child could be reduced to a simple model only including test center (East or West Danish CI center). For all three tests, the models could be reduced and all three tests showed no significant rater effect, *p*-values for PPVT-4 = 0.124, Reynell = 0.214 and active vocabulary = 0.397. More children in East could not be tested at all, which may be a source of bias (Table 4). In order to explore this risk, a sensitivity analysis was applied to the data sample. The superiority of the test outcomes at the eastern CI center was challenged, as the non-testable children were allocated to the lowest performing group for the three tests. Table 4 shows results of the Fisher's exact test and comparisons to the original test results. The two approaches reached the same qualitative conclusions. Thus, there is no reason to suspect a rater bias, i.e. to fear that the stated results as regards to regional differences are an artifact caused by different criterions for judging a child non-testable.

4. Discussion

The educational level of the parents was very high, as the majority of both fathers and mothers had more than 13 years of education. Other studies have shown a correlation between parental educational level and language outcomes [1,12], but we did not find any significant effect of this covariate. Despite the high educational level of the parents the vast majority of children did not perform at age equivalent level when tested for language understanding and for both receptive and active vocabulary. This again is in great contrast to similar studies of language understanding and vocabulary [29]. We found that the region of residence (eastern or western Denmark), age at HA start pre-implant, age at implantation, length of hearing with CI and parental communication mode have significant influence on the

Table 4
Regional distribution of actually tested and non-testable children and Fisher's exact test for regional differences for actually tested children vs. all children.

Test	East actually tested children (*N*)	West actually tested children (*N*)	East non-testable children (*N*)	West non-testable children (*N*)	Actually tested	All[a] children
Reynell	35	36	4	0	0.005	0.014
PPVT-4	34	34	5	2	<0.001	<0.001
Vocabulary[**]	25	24	7	1	0.058	0.314

[a] Non-testable children artificially allocated to the lowest category.

[**] The *p*-values for the dichotomized versions are 0.345 and 0.548 respectively.

language outcome. Except for the regional effect, all the other effects have been reported to affect outcome for pediatric populations with CI in international studies [7,8,11,12]. Thus, this study raises the question, why such poor outcome was found for the present population. A possible explanation could be that the specific guiding of the parents and the education of the hearing impaired children has failed development to match the technical and medical advances made. This issue has also been subject for a study by Archbold, who addressed the problem by raising the question "has deaf education changed enough?" [30]. The question seems extremely relevant in Denmark, where children and families did have optimal prerequisites for acquiring age equivalent language as the start age of HA was early (mean = eight months) and mean age of implantation was 19.6 months. Furthermore, 82% of the children were bilaterally implanted, which is a large number compared to other studies [3]. In line with other studies [5,6,9], we also found a linear effect of age at implantation, such that the lower the age at implantation, the better results. The results, furthermore, stress the importance of bilateral implantation, as children implanted either simultaneously or sequentially for all test responses showed higher odds of having age equivalent language and vocabulary than children with bimodal implantation.

The highly significant effect of diagnosis for the response of the PPVT-4 is an interesting finding. Children with congenital deafness performed better than children with hereditary deafness, Pendred syndromes, meningitis and other diagnoses. This can probably be explained by the fact that children with congenital deafness were diagnosed earlier than children with other diagnoses and hence also started both HA and CI treatment earlier. It must be noted that children diagnosed with hereditary deafness, Pendred syndromes, CMW and other diagnoses are congenital but the hearing may fluctuate and this may result in later age at implantation.

The effect of the amount of support teaching was remarkable, as children who received more than 15 h per week had very poor odds of performing at age equivalent level. It could be argued, that the children with extensive support need it, because their language is not age equivalent. On the other hand, the surprising finding also raises the question of the content and quality of the support teaching, and should initiate an examination and discussion of the current content and quality of the pre- and post-implant rehabilitation of children with CI in Denmark.

It is relevant to discuss, whether the study was carried out at a stage too early for the children to have acquired age equivalent language understanding and vocabulary. The mean age of the children at the time of the testing was 46.3 months, but the length of hearing with cochlear implant was 25.9 months and thus it can be argued that the children's length of hearing with CI was too short. Future studies of early implanted children must have a longitudinal character in order to monitor when and if the language gap is closed. This again has prognostic importance when organizing the postimplant rehabilitation of the children, as this study brought evidence that the gap is not closed within a time frame of the first two years postimplant. In Denmark, a national protocol for pediatric cochlear implantation was implemented in January 2011 [31]. The protocol recommends that the auditory verbal rehabilitation at the two pediatric CI centers is offered to families one year postimplant. Our data suggest that the rehabilitation lasts for a longer period of time in order to close the language gap of children with CI.

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