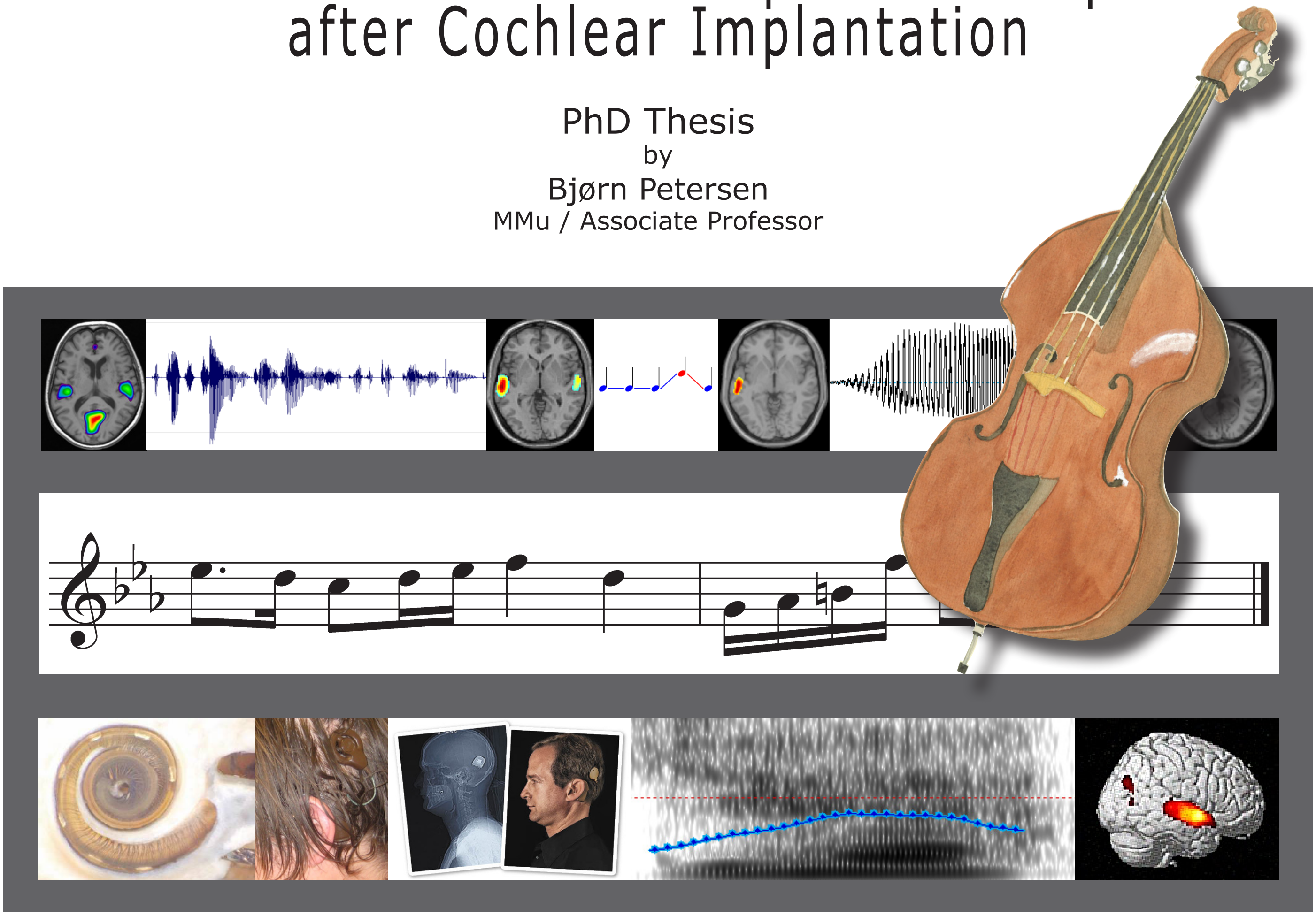
Advances in Music and Speech Perception after Cochlear Implantation

PhD Thesis
by
Bjørn Petersen
MMu / Associate Professor



Faculty of Health Sciences
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FACULTY OF HEALTH SCIENCES
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To
Jytte,
Mathias
& Louise

> As neither the enjoyment nor the capacity of producing musical notes are faculties of the least use to man in reference to his daily habits of life, they must be ranked among the most mysterious with which he is endowed.
>
> *(Charles Darwin: The Descent of Man and Selection in Relation to Sex p. 878, John Murray, London, 1871.)*

This thesis is based on the following manuscripts:

I. Petersen B., Hansen M., Mortensen M.V., Vuust P: Singing in the key of life
- musical ear training enhances musical abilities after cochlear implantation
– *submitted*

II. Petersen B., Wallentin, M., Gjedde A., Vuust P, Mortensen M.V: Cortical plasticity after cochlear implantation – *to be submitted*

III. Petersen B., Hansen R.H., Beyer K., Mortensen M.V., Vuust P: Musical Methods for Little Digital Ears –music training and testing in preschool children with cochlear implants - *in review*.

These manuscripts will be referred to as (I), (II), and (III).

This thesis was submitted to the Faculty of Health Sciences at Aarhus University at January 3rd, 2011, in partial fulfillment of the requirements for the Ph.D. degree.

PREFACE

Music is a cultural phenomenon with a tremendous impact on daily life of people around the world. A newly published survey (October 2010) performed among 2,000 Danes between 12-70 years states that 76% of Danes listen to music more than one hour daily. Seventy-eight % claim that music influences their mood, and 72% say that music means a lot to them. When asked how important music is in their everyday lives, 31% rate music as the third most important thing after family (82%) and private economy (35%). This high rate of music listening and significance supports previous studies by Rentfrow and Gosling (2003), in which college students placed music as number two on a top-ten list of leisure activities.

At a first glance, a coupling between music and deafness may appear rather paradoxical. Music has, however, historically been part of the lives of at least some deaf persons. In the 70s and 80s, Danish music and speech therapist Claus Bang used music in his work with deaf and hearing-impaired children. While unable to hear, the children could follow music through visual cues and low-frequent vibrations. In the other end of the scale, Evelyn Glennie, who is profoundly deaf, leads a career as a full-time professional classical percussion soloist, relying on vibration cues from her bare feet.

The research described in this thesis is primarily about music in the lives of deaf persons. But because of the invention of cochlear implants, the term “deaf” has been redefined. Pathologically deaf, but listening by the aid of electric hearing, means that a person functionally belongs in the hearing world. Therefore, music as a form of art, entertainment, or a means of relaxation is a realistic and plausible dream for the growing number of implant users.

The idea of developing a musical ear training program aimed at newly implanted individuals coupled with a brain imaging study of neuroplasticity was originally initiated by Peter Vuust (professor, Ph. D.) and Malene Vejby Mortensen (MD, Ph. D.), and it happened to be my privilege to materialize the plan. The study of pediatric implant users’ benefit from musical training came as a natural extension, at a time when a strike among Danish nurses had put a temporary stop for new adult candidates. The work load that I have invested in these studies has been substantial, but highly rewarding. Hopefully, the work load the reader will invest in sorting out the results of these efforts will be rewarding too. Enjoy!

Bjørn Petersen, January, 2011

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ACKNOWLEDGEMENTS

This dissertation is based on studies that were completed during my three year Ph.D.-study at Center for Functionally Integrative Neuroscience at Aarhus University. The study and the position were made possible through a united contribution from the Royal Academy of Music, Aarhus, Aarhus University Hospital, and the Faculty of Health Science, Aarhus University. I wish to express my gratitude to all three institutions for my opportunity to carry out this project.

Many people have, in different ways, contributed to the fulfillment of this work. I thank them all for their contribution, large or small. Some persons deserve special mention. Firstly, I thank my supervisor and colleague Peter Vuust, who courageously put enormous trust in my capabilities and powers of endurance, when assigning me to this project. His support, know-how, and encouragement has been decisive in finishing this project. I also wish to thank my project supervisor Malene Vejby Mortensen, for enduring support and advice. Her experience with PET and CI-patients and her knowledge about the chains of command has been essential for completing the many phases of this project. Finally, I wish to thank my supervisor Albert Gjedde for spiritual and professional support whenever needed.

I am especially grateful for all the support and good company I got from my colleagues in the Music in the Brain group: Karen Johanne Pallesen, Line Gebauer, Anders Dohn and Eduardo Garza Villareal.

Special thanks go to all the fine colleagues at CFIN and the PET centre: Kim Vang Hansen for coaching of PET-preprocessing methodology and good suggestions, Anders Bertil Rodell for extreme helpfulness and ever-ready explanations of inevitable questions, Jørgen Kold and Poul Erik Nielsen for perfect first-aid on all computer-related problems; Arne Møller for great assistance with PET-scannings and, not at least, some good humor; Helle Jung Larsen and Birthe Hedegaard Jensen for assistance in scheduling of PET-scans and tolerance and acceptance of the perfect newbie, and Søren B. Hansen for untiring help on the myriad of questions popping up at strange hours. Joel Astrup Aanerud for patience and support and some important enlightenment on brain anatomy, and Per Borghammer for his patient and selfless comments on PET-imaging and hints of possible methods of statistical analysis; Michele Gammeltoft for never-ending assistance on all issues of finance and bureaucracy – with a smile; Donald Smith for essential comments on manuscript structure and editing. Special thanks also go to my Canadian colleagues Mallar Chakravarty, Anni Landau and Ivana Konvalinka for their help on all sorts of matters and many fruitful talks. I would like to thank also all the staff at the PET center, who have taken part in the brain scannings in this project. I am truly impressed with, and respectful of, the professionalism, with which they all encompass the many aspects of their job.

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At the department of Audiology I wish to thank: Frank, Frank, Anders, Robbie, Minna, Randi, Susanne, Else, and Martin. At the hearing institute I wish to thank Lissie, Mette, and Aage.

A very special thank should of course be addressed to all the participants and parents of child participants, who committed themselves to this project.

My greatest and deepest gratitude goes to my family: my wonderful wife Jytte, without whom this work would never have seen the day of light, nor, for that matter, reached a final terminal stage. Her enduring patience and loyalty exceeds the humanly possible. Thank you! Mathias, my son, without whom this work would all have been worthless and Louise, my daughter, without whom this work would all have been meaningless.

The following funds have made financial contributions to the project, for which we wish to express our gratitude: Hørslevfonden, Augustinusfonden, Grosserer L.F. Foghts fond, Petra og Niels Nielsens Mindefond and Høreforeningen.

ABBREVIATIONS

The following is a list of some of the major abbreviations used throughout this thesis.

A/D	Analog/digital
ABI	Auditory brain stem implant
AMMA	Advanced Measures of Music Audiation
ANOVA	Analysis of Variance
BA	Brodmann area
BAB	Multitalker babble
BL	Baseline
BPM	Beats per minute
CAS	Central auditory system
CBF	Cerebral blood flow
CG	Control group
CI	Cochlear implant
dB	Decibel
e.g	For example
EEG	Electroencephalography
EP	Endpoint
F0	Fundamental frequency
Fig.	Figure
FMI	Familiar melody identification
fMRI	Functional magnetic resonance imaging
HA	Hearing aid
HAG	Hagerman
HL	Hearing loss
Hz	Hertz
i.e.	That is
ICRA	International Collegium of Rehabilitative Audiology
IFG	Inferior frontal gyrus
kHz	Kilohertz
mBq	Mega Bequerel
MCI	Melodic contour identification
mCi	Milli Curie
MD	Melodic discrimination
MEG	Magnetoencephalography
MG	Music group
MII	Musical instrument identification
MP	Midpoint
MTG	Middle temporal gyrus
NH	Normal hearing
PCD	Pitch change detection
PET	Positron Emission Tomography
PMMA	Primary Measures of Music Audiation
POST	Postlingual
PR	Pitch ranking
PRE	Prelingual
RD	Rhythmic discrimination
RS	Running speech
ROI	Region of interest
SD	Standard deviation
SPM	Statistical parametric mapping
ST	Semitone
STG	Superior temporal gyrus
TROG	Test for reception of grammar

1. INTRODUCTION

At the moment of writing electrode number 1,000 has recently been implanted into a Danish cochlea and will soon provide auditory sensation to its owner. Cochlear implants (CIs) are indeed a huge success – probably the most successful neural prosthesis ever developed with well over 120,000 users worldwide (Moore & Shannon 2009). Many of these perform remarkably well as far as speech perception in quiet is concerned, which is very good news. However, in studies with adult CI-users perception of music, especially pitch and timbre has proven extremely challenging. With only 16 to 22 electrodes, the CI-device cannot adequately code the spectrum of sound needed to perceive musical pitch. On the other hand, CI-recipients in some cases seem to overcome the technological limitations and apparently revive their long lost music enjoyment by repeated listening. Brain plasticity is one of the keys to this remarkable process, which apparently with time helps the CI-user to make musical sense of the sparse and crude signal from the implant and eventually find Elvis sounding (almost) like Elvis always sounded. Technological progress and refined mapping strategies are of great importance, but there are strong indications that to cross new boundaries and maximize implant outcome, ongoing and systematic training is a necessary supplement (Fu & Galvin III 2008). Music offers a strong and enjoyable environment for learning to discriminate key sound features like pitch, rhythm, and timbre, and could be a supplementary method of auditory training with great impact on musical as well as linguistic abilities.

The research described in this thesis examined how auditory abilities develop in CI-recipients; 1) behaviorally, by measuring the effects of individual musical ear training on the progress in musical and linguistic performance of newly implanted adult CI-users; 2) neurologically, by measuring the progress in brain activity of newly implanted adult CI-users while listening to speech over six months, and; 3) behaviorally, by measuring the effects of group-based musical training on the progress in musical and linguistic performance of early implanted pediatric CI-users.

2. AIMS AND HYPOTHESES

2.1 Study 1

Some cochlear implant users, exposed to repeated music listening, seem to enjoy music intensely despite the technological limitations of the implant. We intended to examine this phenomenon in an experiment involving the following methods:

1. To study the possible effects of musical ear training on the auditory competences of cochlear implant recipients, we created a longitudinal one-to-one musical ear training program and administered it to a group of newly implanted participants.
2. To measure the effects we adapted a battery of musical and linguistic tests, and compared the results of the intervention group with those of a group of matched controls and a group of normal hearing controls.

The results of this experiment were discussed in an article corresponding to the following hypotheses: (a) weekly one-to-one musical ear training, involving active music making and listening exercises, may substantially enhance the musical discrimination skills of CI-recipients; (b) possible enhanced discrimination skills resulting from this training could generalize to the linguistic domain, and positively affect the CI-users' recognition of speech and emotional prosody.

2.2 Study 2

Many CI-users adapt to the electric stimulation very quickly and achieve high comprehension of speech, but the outcome is very differential. We intended to examine these phenomena in an experiment involving the following method:

1. To study the cortical mechanisms underlying the initial learning process following implantation, we examined levels of brain activity during two different listening conditions with positron emission tomography (PET) at three points of time.

The results of this experiment were discussed in an article corresponding to the following hypotheses: (a) the involvement of cortical areas is different in users with a postlingual hearing loss than in users with a prelingual hearing loss; (b) Broca's area is involved in speech perception of implant users; (c) reactivation of neural pathways and engagement of cortical areas similar to those of normal-hearing controls takes place within 3-6 months after switch-on of the implant.

2.3 Study 3

To the new population of early implanted pediatric implant users with prelingual hearing loss, music training may benefit development of musical and linguistic discrimination skills. We intended to examine this phenomenon in an experiment involving the following method:

1. To study the effects of musical training on the auditory competences of pediatric CI-users, we created a music training program and administered it to a group of prelingually deaf preschool children with cochlear implants. To measure the effects we adapted a battery of musical and linguistic tests, and compared the results of the intervention group with those of a group of matched controls and a group of normal hearing controls.

The results of this experiment were discussed in an article corresponding to the following hypothesis:

1. Intensified active exposure to music and live musical sounds may expand the ability of pediatric CI-users to perform musical discrimination tasks.
2. Increased musical experience will be noticeable in these children's everyday life, expressed as increased musical interest and activity, as observed by their parents.
3. Improved musical competences may generalize to the linguistic domain.

3. BACKGROUND

3.1 The hearing system

> *"The goal is to have the patient live a normal life, not to be deprived of anything." Philip Loizou, CI-researcher*

3.1.1 Sound

Whenever an object vibrates, e. g. the back and forth movement of a guitar string or the vibration of the vocal cords when we speak, it causes variations in air pressure, i.e. sound waves. Sound waves are characterized by their wavelength, frequency, amplitude, and duration. Wavelength is the spatial period of the wave – the distance over which the wave's shape repeats. It is determined by the distance between consecutive corresponding peaks, valleys, or zero crossings (Figure. 3.1). The frequency of sound is the rate, at which the waves pass a given point. If this pattern is repeated periodically (a cycle), a pitch is produced. Frequency is measured in Hertz (Hz), expressing the number of cycles per second. Low frequencies are characterized by fewer cycles per second, higher frequency by many cycles. The physical parameter of frequency corresponds to the psychological attribute of pitch. Amplitude is the magnitude of oscillation of the wave from zero. Amplitude (or intensity) corresponds to the energy, with which the wave travels and is most often measured in decibels (dB). The physical parameter of amplitude is the psychological correlate of loudness.

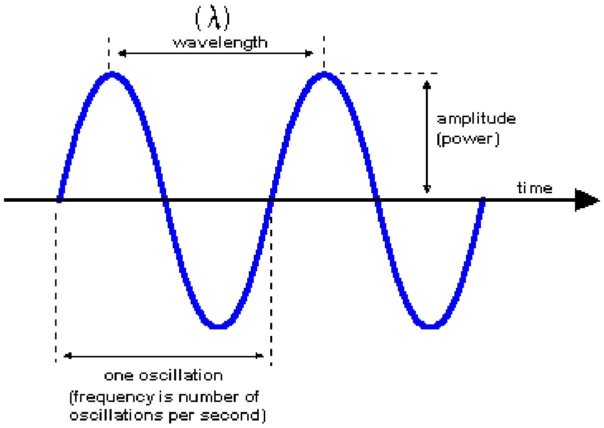

Figure 3.1 Sine wave characterized by wavelength, frequency, and amplitude illustrated on a horizontal time line.

A soundwave which encompasses just frequency and amplitude is a sine wave, sometimes named “pure tone”. Most of the sounds encountered in everyday life comprise complex tones or waveforms, whereby two or more sine waves are combined. The most basic of these have a regular repetition rate corresponding to the frequency of the fundamental - the fundamental frequency (F0). For periodic complex sounds, the frequencies of the higher components, or harmonics, are multiples of the F0 (Schnupp et al. 2011).

3.1.2 Hearing

The human peripheral auditory system (Figure 3.2) can be subdivided into three sections – the outer, middle, and inner ears. The outer and middle ears act as conductive mechanisms, detecting and transforming sound energy into mechanical vibrations for conduction to the inner ear. The inner ear houses the cochlea, a coil-shaped, fluid filled structure consisting of three chambers, the scala vestibuli, scala media, and scala tympani (Figure 3.3). There are about 3,500 inner hair cells and 12,000 outer hair cells in the (healthy) cochlea. The bony modiolus, around which the cochlear duct turns, houses the spiral ganglion. Bipolar cells of this ganglion, the spiral ganglion cells, the bases of the inner and outer hair cells, form the cochlear portion of the vestibular cochlear nerve (cranial nerve VIII). Efferent fibers to the cochlea make synaptic contacts with both inner and outer hair cells. Inner hair cells are sensitive analogue/digital (A/D) converters. They convert the mechanical force applied to the hair bundle into an electrical signal.

Tonotopy: The basilar membrane, which separates the scala media and the scala tympani, is narrow at the basal end and wide at the apex. This variation gives the cochlea its tonotopic organization (Figure 3.4). In response to an input sound wave, a travelling wave is formed in the cochlear fluid, progressing from the base to the apex of the cochlea. Different frequencies are associated with differing points of maximal displacement along the basilar membrane with low frequencies stimulating apically and higher frequencies exciting more basally. In essence, each frequency creates its own travelling wave which is spectrally analyzed by the cochlea – the location of the wave's peak becomes one of the primary bases from which pitch decisions are made by the brain (Fearn & Wolfe 2000). The range of human hearing is reported to be approximately 15 Hz to 15 kHz, although we are less sensitive to sounds at extreme frequencies (Moore & Moore 2003).

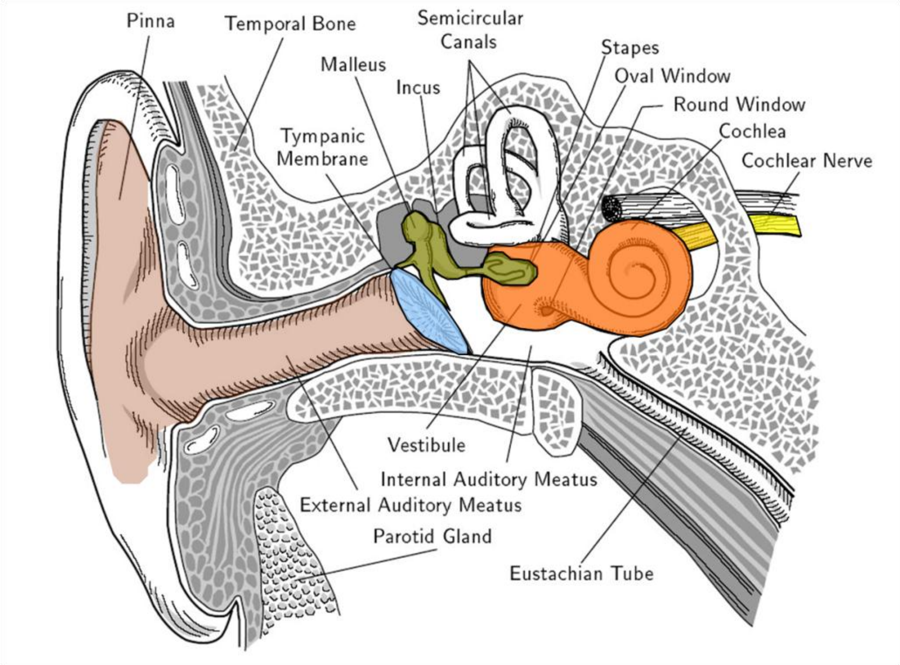

Figure 3.2. The hearing system; outer, middle, and inner ear.

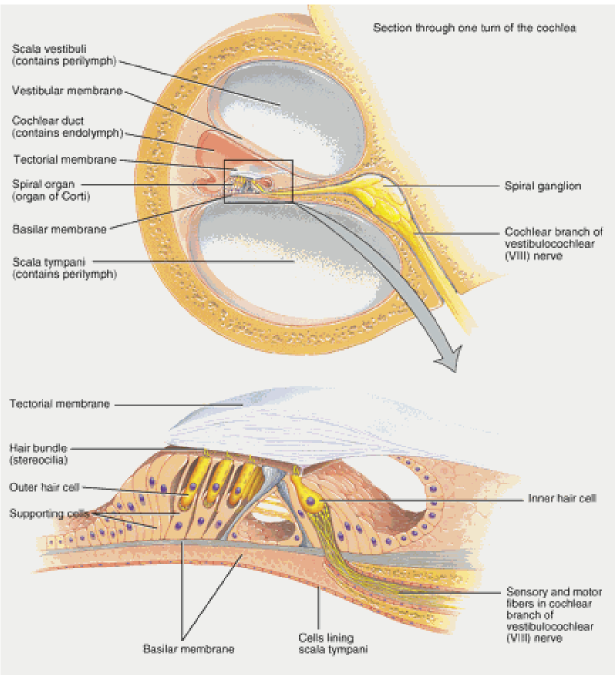

Figure 3.3 Illustration of the anatomy of the cochlea. The human cochlea has 2 ¾ turns and is about 3.5 cm long.

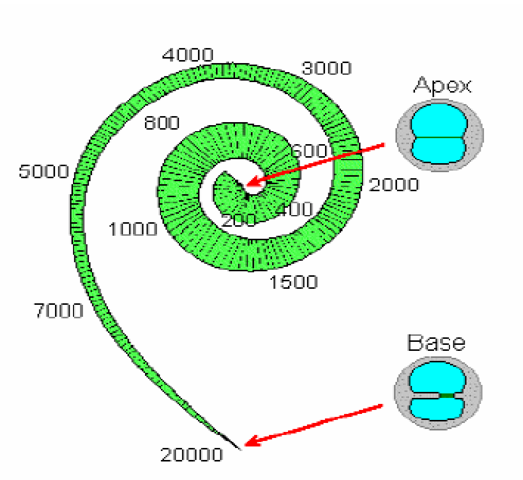

Figure 3.4 Tonotopic organization of the basilar membrane. High frequencies excite at the base, while low frequencies excite at the apex.

3.1.3 The auditory pathway

The auditory pathway is the sequence of nuclei in the brain stem leading from the cochlea up to the auditory cortex in the temporal lobe of the brain. The primary connections from the two cochlear nuclei are to the opposite hemisphere (Figure. 3.5). Each nucleus (the cochlear nucleus, superior olive, lateral leminiscus, inferior colliculus and thalamus) has a tonotopic map, which corresponds to the tonotopic organization in the cochlea. The target in cortex on each side is the superior and interior surface of the temporal lobe (Schnupp et al. 2011).

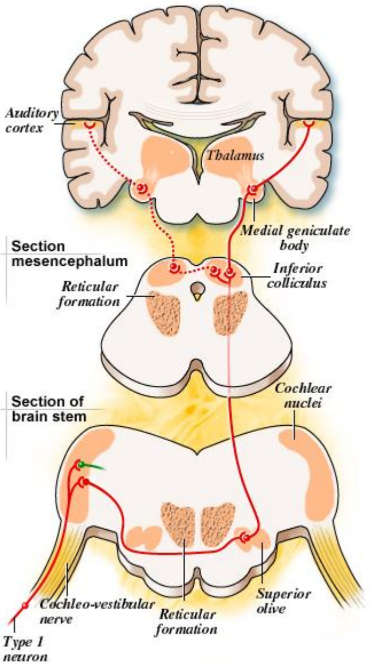

Figure 3.5 Illustration of the auditory pathway.

3.1.4 Hearing loss

Generally, hearing loss (HL) is defined as a permanent shift in thresholds of 25 dB or more at 500, 1000, 2000, and 4000Hz. The degree of a HL is defined by 5 categories of severity: (1) Mild (24-40 dB HL), (2) Moderate 40-55 dB HL), (3) Moderately-Severe (55-70dB HL), (4) Severe (70-90 dB HL, (5) Profound (>90 dB HL) (Figure 3.6) (Cole & Flexer 2007). There are no official records, but as a rule of thumb, approximately 0.1% of a population is deaf, i.e., amounting to about 4,000 individuals in Denmark. The many forms of HL can be divided into two categories: conductive and sensorineural. Conductive HL is caused by impairments in the outer or middle ear. A sensorineural HL most often arises from damage to or pathology in the hair cells in the cochlea, which hinders the transformation of mechanical vibrations into neural impulses.

Congenital or acquired: A distinction is made between a congenital or acquired HL, depending on when they occur in a person's life. Congenital HLs typically occur before, at, or shortly after birth, whereas acquired HLs happen after development of speech and language has begun. Because programming already took place in the auditory brain areas, the negative effects of an acquired HL tend to be less severe. Another distinction is made between a *prelingual* and a *postlingual* HL. A prelingual HL is usually defined as a HL, which occurred before the age of three. Thus, a *progressive* congenital HL may be postlingual, while an *acute* acquired HL may be prelingual. The earlier the on-set of the hearing loss, the more it interferes with language, learning, and development of auditory brain function. Individuals, who have an established complex linguistic system and mature neural connections in the auditory cortex, usually are those who benefit most of a cochlear implant.

Etiology of hearing loss: HL may be caused by endogenous or exogenous factors. An endogenous HL has hereditary or genetic sources (Van Camp 2002). Mutation of the gene Connexin 26 and Pendred's syndrome are examples of endogenous causes of HL. An exogenous HL is caused by external events such as bacterial or viral infection (e.g. meningitis, measles, and encephalitis), noise exposure, head injury, and medication that damage the inner ear. It is beyond the scope of this thesis to cover this field in more depth.

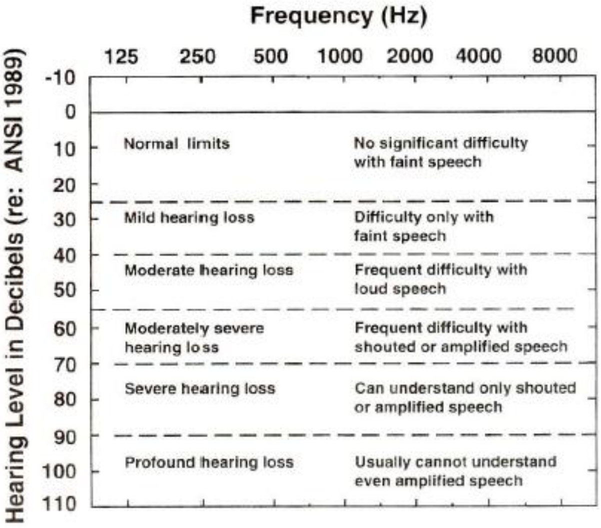

Figure 3.6 Illustration of the five categories of hearing loss according to the American National Standards Institute (ANSI).

3.2 The cochlear implant

History: Cochlear implants began in the 1950s when Djourno and Eryies, a French surgeon and engineer, collaborated to place a coil of wire in the inner ear of two deaf people (Djourno & Eyries 1957). As a result of multidisciplinary research including bioengineering, physiology, otolaryngology, speech science, and signal processing, the technology has subsequently progressed from the early single-channel implant systems through to the current-day multi-channel implants. Signal processing science, in particular, has played an important role in developing technological solutions that would mimick the function of a normal cochlea by transforming speech signals into electrical stimuli. The evolution and expansion of cochlear implantation has been rapid, particularly in the last decade with the number of implantees exceeding 120,000 worldwide (2010).

How it works: A CI operates by bypassing the outer ear, middle ear, and the hair cells to electrically stimulate the surviving auditory neurons directly (Loizou, 1998). The CI comprises a surgically implanted internal package and externally worn components connected by a conductive link. The internal components consist of a receiver-stimulator package containing a magnetic coil connected to an electrode array. The external components consist of a speech processor and a transmitter coil (Figure. 3.7). The receiver-stimulator package decodes the radio frequency signal transmitted from the speech processor and converts it into an electrical current used to stimulate the cochlear nerve fibers via the implanted electrodes. The intracochlear electrodes (~22) are arranged in an array inserted into the scala tympani of the cochlea to an optimal depth of between 25 mm and 31 mm from the round window. This corresponds to approximately the first 1.5 turns of the cochlea. The scala tympani is a surgically accessible site in close proximity to the tonotopically arranged spiral ganglion cells. Even in cases of profound sensorineural deafness, in which few or no hair cells survive, CIs may still take advantage of the tonotopic organization of the residual auditory neurons. However, not all cases of deafness can be treated with a CI. In cases of a dysfunctional auditory nerve, auditory brain stem implants (ABI) have been developed, which provide direct electrical stimulation of the brain.

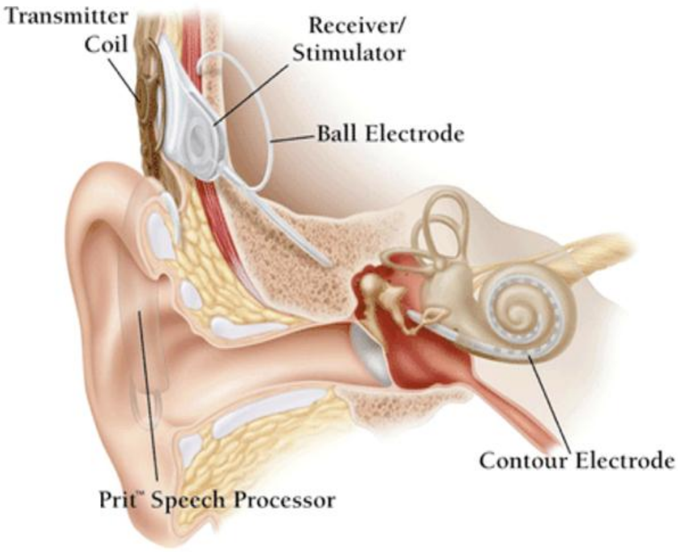

Figure 3.7: The inner and outer parts of a CI.

Sound processing: The speech processor contains a set of microphones that pick up sound, which is converted into electrical pulses, according to a preprogrammed strategy or “MAP”. The most commonly employed strategies in current sound processors are based on filterbank strategies. In all filterbank strategies, spectrum of incoming signals is split into frequency bands by means of a bank of bandpass filters with each band allocated to one channel (electrode) of the implant. At the output of each filter, the envelope of the waveform is estimated. These envelope signals are sampled at regular times, and their amplitudes are converted to appropriate stimulation current levels. In the implant, brief electric pulses are delivered by electrodes corresponding to the filters at a rate equal to the sampling rate. The channels with the greatest amplitude for each stimulation cycle are often referred to as 'maxima'.

Speech perception outcome: The clinical impact of the evolution of CIs has been nothing less than extraordinary. With current implant technology and up-to-date sound processing strategies, the average CI-listener recognizes about 90% of sentences and 55% of monosyllabic words, in quiet listening conditions, after 12 months of practice with a unilateral CI. Some users even achieve the capability of conversing over the telephone (Friesen et al. 2001; Wilson & Dorman 2007). The variability in implant outcome, however, is very large, with duration of HL and residual hearing as important predictors of the result (Summerfield & Marshall 1995; Waltzman et al. 1995). Furthermore speech perception in conditions involving background noise or competing talkers is in general described as very challenging.

3.2.1 Music perception with a cochlear implant

Because music, in many cases, has been an essential part of these patients' cultural and social life prior to deafness, CI-candidates' hope of retrieving music enjoyment is an important reason for choosing this treatment (Gfeller et al. 2000b). Furthermore, with the considerable improvements made in CI-technology with regard to speech perception, it is natural that many existing CI-users express hopes of being able to enjoy music. Many CI-users, however, are disappointed with their music experience. American CI-recipient and author Michael Chorost has described initial music listening with his implant like "walking color-blind through a Paul Klee exhibit" (Chorost 2005). In addition, several studies have concluded that recognition of melody and timbre is significantly poorer in CI-users than in normal hearing (NH) controls (Gfeller et al. 2002a; Leal et al. 2003; McDermott 2004; Kong et al. 2004; Gfeller et al. 2005; Olszewski et al. 2005; Gfeller et al. 2007).

Fine-structure: The main reason that the implant does not encode pitch well is the absence of “fine-structure” temporal encoding needed for perception of complex tones. The multiple sinusoidal components of complex sounds excite a broad range of electrodes resulting in possible frequency-to-place mismatch. Moreover, the temporal periodicity is obscured, because the processor divides the signal into a finite set of frequency channels, extract the temporal envelopes, and deliver them into the cochlea with fixed-rate pulse trains, by which process the fine-structure of the sound waves is largely lost. Finally, music depends on low frequencies for perception of the F0. The lowest-pitched string on a guitar vibrates at 83 hertz, but CI MAPs commonly has a low-cut at 250 hertz. This has been done because low-pitched sounds - air conditioners, engine rumbles - interfere with speech perception, which is the main goal of the CI.

Variable outcome: Discrimination of complex-tone pitch direction in CI-users is widely variable ranging from less than one semitone in a few implant users to as much as 12 semi-tones in others (Guerts & Wouters 2001; Gfeller et al. 2002a; Nimmons et al. 2008). This implies that other factors such as previous musical experience and low-frequency residual hearing may affect pitch perception with an implant.

Timbre: Accurate perception of timbre requires the perception of both the signal’s temporal envelope, and the energy spectrum of its harmonic components (Figure 3.8). In CIs, the coding of spectral shape is limited as a result of insufficient channels, decreased specificity in mapping frequency bands to electrodes, and a lack of precision in conveying temporal and spectral detail

(Moore & Moore 2003). These issues may in part account for CI-users' poor performances on tasks involving the identification of musical instruments or other complex sounds.

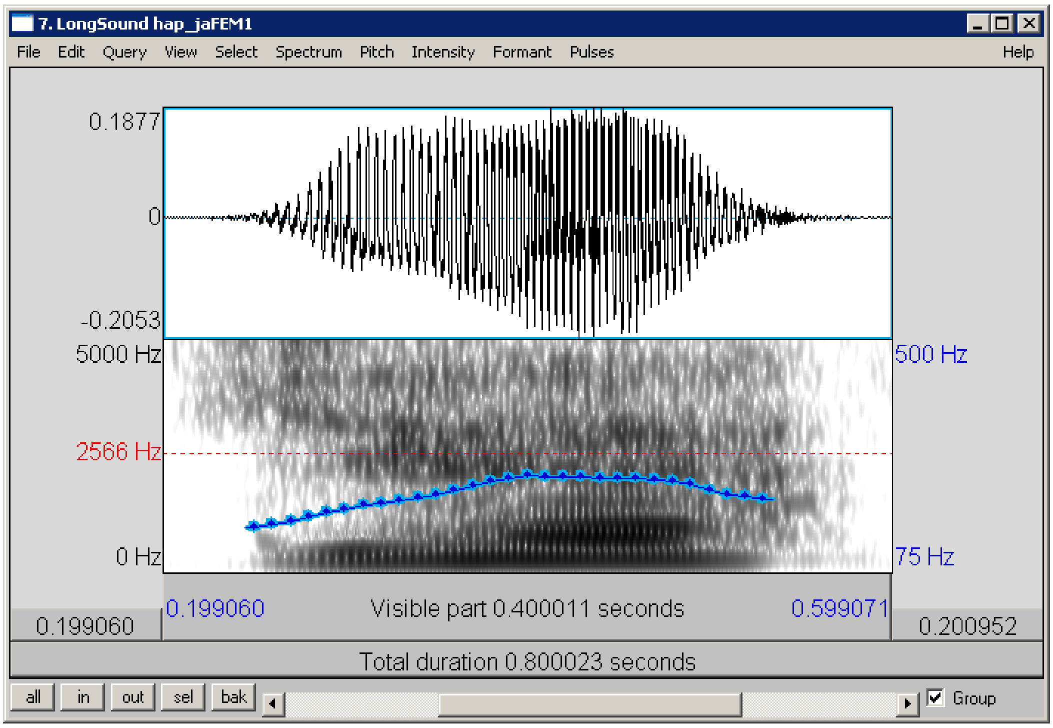

Figure 3.8 Temporal envelope (waveform) (top) and spectral envelope (bottom) of the word "yes" spoken by a female speaker. The sound is part of the EPR test in (I) (analysis window in Praat (www.praat.org)). Blue line= F0.

Rhythm: Another fundamental aspect of music perception is rhythm. A sequence of onsets defines common rhythmic patterns in music, which are well defined through the transmission of temporal envelopes in cochlear implants. Part of the reason for this is the high degree of synchrony between the electrical impulse and the nerve firing pattern. Studies on CI-users' discrimination of tempo and simple rhythm patterns have found performance close to that of NH controls (Gfeller et al. 1997; Leal et al. 2003; Limb et al. 2010). It is worth clarifying at this point, the differentiation between 'gross temporal cues' that convey a sense of rhythm, as opposed to temporal cues which provide a sense of pitch. Temporal patterns in the frequency range of 0.2 Hz to 20 Hz provide a distinctive rhythm to musical stimuli, whereas higher-frequency components of the acoustic signal provide pitch information.

3.2.2 The effects of music training

Despite the implant's technological shortcomings, some CI-users seem able to retrieve their lost musical enjoyment by repeated and persistent listening to music from their hearing past (Gfeller & Lansing 1991; Gfeller et al. 2005)[1]. A possible explanation of this apparent paradox could be that the repeated exposures may train the brain to gradually fill in missing pieces and provide additional meaning, particularly for individuals with a postlingual HL who have an internal representation of music based on pre-deafness experiences and memory. This would suggest that cortical plasticity may enable implant users to outperform the limitations of the implant and develop improved recognition of patterns in music as a result of active learning (Fu & Galvin III 2008). This notion is supported by studies involving adult CI-users, which have shown positive behavioral results from music training. Galvin et al. (2007), for instance, demonstrated significant improvement in the ability of six CI-users to identify melodic contours after a short period (~2 months) of daily computer-assisted exercises. The effect was most notable in contours with large pitch changes, but the benefit of the training interestingly generalized to the ability to identify a familiar melody. In a similar study, postlingually deaf CI-users had significantly improved recognition and appraisal of timbre of musical instruments (Gfeller et al. 2002b). A more comprehensive study investigated the effect of a computerized music training program, which trained recognition and appraisal of simple (melody-only) (Gfeller et al. 2005) and complex songs (Gfeller et al. 2000a). The program was extensive and required participants to complete 48 daily 30-minute lessons (~3 months). Pre- and posttest data indicated that particularly recognition and appraisal of complex songs improved significantly, as a result of the training program. So far, no data are at hand concerning the effects of longitudinal musical ear training based on personal tuition in CI-users.

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[1] For examples of such cases from this study see chapter 3 paragraph 3.5.1/3.5.2

3.2.3 Music and other aspects of listening

Improved perception of music may have considerable positive implications not only for music enjoyment, but also for other aspects of listening. Music training in normal listeners is beneficial for the development of specific auditory skills, such as pitch, timing, and timbre, also involved in language comprehension (Naatanen et al. 1978; Pantev et al. 1998; Koelsch et al. 1999; Naatanen et al. 2001; Vuust et al. 2005; Altenmuller 2008). Enhanced music abilities may enhance speech perception in noisy surroundings, which relies on pitch cues to separate the target from the background (Qin & Oxenham 2003), and the ability to identify voice gender and speaker, which largely depends on discrimination of timbral cues (Vongphoe & Zeng 2005). Recent brain imaging studies have shown that complex music tasks activate brain areas associated with language processing (Levitin & Menon 2003; Vuust et al. 2006). Thus, musical training of CI-users may be hypothesized to influence speech perception.

3.2.4 Music and speech perception of pediatric CI-users

Due to the introduction of newborn hearing screening programs (since 2004 in Denmark), identification of hearing loss has accelerated, dropping to 1–3 months of age, making cochlear implantation possible at around 12 months. The result is a new and growing population of prelingually deaf infants, to whom the prospects of developing spoken language, reading, and academic skills remain positive (Wie 2010). However, in contrast to adults with a postlingual HL, prelingually deaf children’s ability to use a CI is further challenged, because the brain must learn to process auditory information (Kral & Tillein 2006). Thus, a successful outcome depends on daily and ongoing auditory coaching from parents and professionals (Moog & Geers 2003; McConkey Robbins et al. 2004). In general, younger age at implantation has been associated with better language progress (Houston et al. 2003; Oh et al. 2003; Connor et al. 2006); however, the impact of early implantation on music perception and enjoyment remains unknown. Enhanced development in the central auditory system and greater cortical plasticity in early-implanted compared to late-implanted children may have favorable implications for music processing as well (Yoshinaga-Itano et al. 1998; Sharma et al. 2005).

Previous studies: Some studies have examined music perception of children with CIs. Xu et. al (2009) compared the accuracy of singing in child implant users to the performance of NH peers. While children with CIs showed a significantly poorer performance on all pitch-based assessments of singing, the rhythm based measure revealed no difference between the groups. In spite of this pitch processing deficit some children with CIs seem to be involved in a variety of musical activities like singing, dancing, listening and even instrument lessons (Gfeller et al. 1998). In a study that evaluated child implant users' ability to identify popular songs, Vongpaisal et al. found that children and teens were able to identify songs based on excerpts from original and instrumental recordings, but failed in recognizing the melody-only versions (Vongpaisal et al. 2006). The findings were replicated by Mitani et al. (2007) with TV theme songs and document that child implant users' familiarity with songs primarily stems from cues such as rhythm and lyrics.

Studies involving training: For pediatric CI, studies of the effects of musical training have been sparse. Abdi et al. (2001) studied CI-users aged 3–12.5 years following a music training program that involved either simple perceptual tasks or learning to play a musical instrument. Brief reports of the musical development of children suggested that the training may have been beneficial, although no objective evaluation, such as music tests, was described. In a case/control study including 18 pediatric CI-users, Yucel et al. (2009) investigated the outcome of a longitudinal music training program based on a take-home electronic keyboard. According to parent reports, the music group (MG) children showed significant improvement in almost all areas of music perception. However, parents may lack objectivity and accuracy in assessment of the musical skills of their children, and this may have affected the results. Recently, Chen et al. (2010) examined pitch perception in a group of children with CIs (5–14years), half of whom attended music classes.

Despite existing confounds, the authors concluded that duration of music education positively correlated with the performance of pitch perception.

Music and language: Studies which compared musically trained and untrained children have shown that music lessons may benefit non-musical domains (Schellenberg 2001). Music training, for instance, positively affects the child’s linguistic abilities such as phonological processing, early reading, and sensitivity to speech prosody (Anvari et al. 2002; Thompson et al. 2003). Furthermore, language development can be strongly facilitated by use of song, which indicates that children’s songs and lullabies may support acquisition of linguistic prerequisites (Schon et al. 2008). Because associations between music and language rely on shared resources such as melody (intonation), rhythm (timing), and dynamics (stress), music training may affect the linguistic development of pediatric CI-users too. In addition, it has been demonstrated that auditory coaching is imperative for children with CIs to develop age-appropriate communication skills, and that educational environments that emphasize the development of speech, auditory, and spoken language skills are particularly beneficial (Moog & Geers 2003; McConkey Robbins et al. 2004; Geers 2006). Since music offers a learning environment that is multisensory and enjoyable, it is plausible to believe that music can play a valuable supplementary role in auditory/oral habilitation programs.

3.3 Musical tests in CI research

3.3.1 Music test batteries targeted at adults

The lack of tests that objectively measures a broad spectrum of musical perception ability, at a level of difficulty appropriate for CI-listeners, has led to the creation of a number of test batteries such as UW-CAMP (Nimmons et al. 2008), the Zurich Music Test Battery (Büchler 2008), IMPAB (Gfeller et al. 2002a), or MTB (Looi et al. 2008). The methodologies used are often similar, but the tests have not been intended to be standardized, and it is not possible to directly compare the results across laboratories. Familiar melody identification (FMI) has been used to describe CI-users’ music perception in many studies, often using melodies assumed to be cross-culturally well-known (Happy Birthday, Jingle Bells). Because rhythm cues are readily perceived by CI-users and contribute strongly to FMI performance, FMI is often measured without rhythm cues. This manipulation is problematic, in that it relies on CI-users' memory of familiar melodies that are now distorted in terms of rhythm cues and melodic pitch (because of the CI processing). Another way to asses perception of pitch change in the context of a melody is the same/different paradigm originally suggested by Gordon (Gordon 1979) and adapted in the Musical Ear Test (MET) (Wallentin et al. 2010), as well as in the Montreal Battery of Evaluation of Amusia (MBEA) (Peretz et al. 2003). While the MET is designed to measure fine-grained melodic and rhythmic discrimination skills in music academy students, the MBEA is designed to identify individuals with the pitch processing disorder amusia. Thus, the former is at an advanced level of difficulty, while the violations contained in the latter are very obvious to a normal ear. Furthermore, most rhythm tests present rhythm patterns played with a single sine wave note, thereby restricting the complexity of rhythmic variations. In contrast the MET utilizes percussive sounds which makes it possible to present more complex rhythms found in musical styles like jazz and latin.

3.3.2 Music test batteries aimed at children

One attempt to measure musical aptitude in preschool children or toddlers, that has been made commercially available is the musical game Audie by Edwin E. Gordon (Gordon 1989) (Figure 3.9). Gordon was a pioneer in the research of music aptitude and launched the term *audiation.* Audiation, according to Gordon, is to hearing as visualization is to sight, i.e. the ability to imagine music in the mind. For the typically developing child this ability can develop in the same way language develops, by exposure and musical activity.

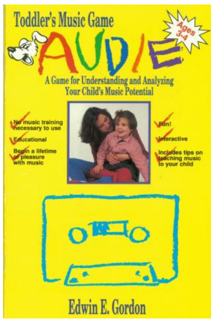

Figure 3.9: The booklet included in the Audie package.

Audie is aimed at parents, as “a tool to help monitor musical strengths and weaknesses” of their child, to assist in adapting music instruction. The game consists of a cassette tape with two games of each ten questions, one with a melodic dimension and one with a rhythmic. The design of the test is based on the same/different paradigm that constitutes Gordon’s other tests PMMA and AMMA. However, in Audie the child is not required to use the terms “same” or “different” but rather “yes” and “no”. The child is repeatedly presented to Audie’s “special song”, a 3-note falling D-major triad. The recorded speak instructs the child to say “yes” whenever it hears the special song and to say “no” whenever it hears *another* song. In the 2 games half of the questions contain a violation of either rhythm or pitch. So-called game Sheets are provided for recording of answers. The yes/no approach smartly overcomes the challenge of assessing the potential of small children. First, the test avoids introduction of terms such as melody and rhythm; second, duration is short, thereby paying due respect to the short span of concentration that most children 3-4 y of age possess.

3.4 Cortical plasticity and lateralization in the brain

Plasticity is a term used to describe the reorganization of cerebral cortex by means of synaptic changes and rewiring of neural circuits, for instance as an effect of long-term training of a specific task. To cochlear implantation, neural plasticity associated with deprivation of auditory input and adaptation to the absence of stimuli is of particular interest. Reduced input to the brain from impaired auditory pathways significantly deactivates the central auditory system (CAS). When auditory input to the brain is reintroduced, this novel auditory experience may itself induce additional plasticity. The sensory reafferentation provided by CI offers an opportunity to study the effects of preceding deafness on functional brain organization. Such studies are typically performed with modern neuroimaging such as PET and functional Magnetic Resonance Imaging (fMRI), by comparing an active brain state with a presumed inactive baseline condition. Lateralization is a term used to describe the localization of brain function in either of the two brain hemispheres, e.g. music in the right and language in the left. This traditional simple right-versus-left–hemisphere theory has changed during the past two decades. Studies examining music processing in musicians and non-musicians have demonstrated the effect of professional training on hemispheric lateralization, professionals showing left, non-musicians showing right preponderance (Altenmuller 1989; Altenmuller 2001; Vuust et al. 2005).

3.5 Preliminary research

3.5.1 Interviews

As part of the preparations for study (I) we interviewed three Danish CI-users, who were reportedly successful with music listening. They told very similar stories of how they retrieved the music of their hearing past by means of listening to the same song over and over again. Gradually, after somewhere between 20 and 50 times of listening, the sound transformed from noise to recognizable. When changing to a new song, they had to start over again. Every time, though, fewer repetitions were needed. The three cases were similar in that music had played an important part in their lives, and that they were persistent and hard working in their efforts to retrieve their music enjoyment. These reports supported the notion that repeated auditory training may facilitate CI-users in adapting to the implant signal and overcoming the technological limitations.

3.5.2 J.E. – a single case

J.E. was a 20 years old man who had a prelingual HL, but had learned to communicate by HAs and lip-reading. At the age of 19 he had received his implant. He had been fond of music since childhood and taught himself to sing and play the guitar. After his implantation he was able to continue with his musical activity. Though he did not participate in the actual study, he took part in 3-4 sessions in which he learned new songs and received ear training. His rather extraordinary abilities can be observed in three short video clips found in the file ***01_J.E. - singing playing, and imitating w. a CI.ppt*** on the accompanying CD-rom.

3.5.3 Pilot studies

To obtain a realistic impression of the level, at which musical training and testing was possible with a CI, we recruited four CI-users for five individual musical training and testing sessions. The pilot participants (3 M, 1 F) had a mean age of 34.75 years (21-48) and had an average implant experience of 1.14 years. Three of the participants were users of the Nucleus CI24 RE electrode (Cochlear ®), while one used Advanced Bionics Hi-Res 90 K ®. The pilot participants represented two cases of congenital deafness, and two cases of late onset hearing loss in early childhood. The invaluable experience drawn from these sessions formed the base, on which we created the musical training program, the musical test battery, and the musical background questionnaire.

4. MATERIALS AND METHODS

4.1 Choice of methods

Study 1: To examine the effects of musical ear training on newly implanted adult CI-users, we created a long-term musical ear training program based on music making and listening methods in a one-to-one setting. This was done for the following reasons:

1. The largest development following cochlear implantation takes place in the first six months after switch-on (Ruffin et al. 2007). We therefore expected music training to have the strongest impact, if imposed in this period.
2. Because changes as a result of training need time to internalize, an extended training period is preferable, especially considering the participants' lack of musical experience and their special hearing condition.
3. Ear training and active music-making are effective methods of training musical abilities, according to the music teaching tradition and will influence the development of auditory competences more strongly than passive listening. Guided listening practice, which links to the musical training, will appeal to the participants and create coherence. Furthermore, personal tuition is superior to computer training, in that it can be targeted, spontaneously revised, and motivating.

Study 2: To investigate the cortical mechanisms underlying restoration of the hearing sense and speech perception in the first six month period following implant switch-on, we used Positron Emission Tomography (PET). This was done for the following reasons:

1. In contrast to other imaging and recording methods, such as EEG, MEG, and fMRI, patients with (auditory) implants that are not magnet compatible can participate in PET studies.
2. PET scanning is a relatively quiet imaging modality, which is useful for both CI-users and for the study of speech.

3. Because only the head of the participants is positioned in the scanner, in contrast to fMRI, it is possible to communicate visually with the participant during scanning.

Study 3: In contrast to study 1, we decided to examine the effects of musical training on pediatric CI-users in a *group* setting. This was done for the following reasons:

1. With children, who have no previous musical experience, the elementary features of music are effectively presented and trained in a group.
2. The social aspect of group-learning motivates children to take part and sustain interest.
3. The shared experiences create a common base which may form part in activities outside the music training setting.

4.1.1 Ethics

All studies described in this thesis followed rigorously the instructions, rules and restrictions determined by *The Danish National Committee on Biomedical Research Ethics*. The studies were in accordance with the *Declaration of Helsinki pt. II*. All participants provided written, informed consent for participation in the studies. In the case of study 3, the parents of the participating children provided written, informed consent on behalf of their children.

4.2 Study 1[2]

Musical ear training with newly implanted cochlear implant users.

4.2.1 Participants

Eighteen participants in this study were involved sequentially over the course of 2 years (table 4.1).

Participant (gender)	Age at project start (y)	Etiology of deafness	Side of implant	Contra-lateral use of HA	On-set of hearing loss (y)	Duration of hearing loss (y)	[d]Degree of deafness (1-5)	Implant type	CI sound processor	CI sound processing strategy
Music group (MG)										
MG1 (F)	49.8	[a]Cong. non spec.	R		4	45.8	5	[e]Nucleus	Freedom	ACE 900
MG2 (F)	21.4	Ototoxic	R	X	0.7	20.7	5	Nucleus	Freedom	ACE 250
MG3 (M)	31.7	Meningitis	L	X	1.8	30.2	4.5	Nucleus	Freedom	ACE 900
MG4 (M)	56.0	Cong. non spec.	R	X	8	48.0	4.5	Nucleus	Freedom	ACE 1800
MG5 (F)	70.3	Cong. non spec.	R		40	30.3	4.5	Nucleus	Freedom	ACE 900
MG6 (F)	47.5	Unknown	L		30	10.5	4.5	Nucleus	Freedom	ACE 1200
MG7 (F)	56.2	[b]Hered. non spec.	R	X	19	37.6	4.5	Nucleus	Freedom	ACE 1200
MG8 (M)	58.5	Meningitis	R	X	5	53.5	5	Nucleus	Freedom	ACE 900
MG9 (F)	29.1	[c]Mon	L		10	19.1	4.5	Nucleus	Freedom	ACE 1200
Mean	**46.7**					**32.8**	**4.7**			
Control group (CG)										
CG1 (F)	44.8	Unknown	R	X	35	9.8	4.5	Nucleus	Freedom	ACE 1200
CG2 (M)	60.4	Unknown	L	X	40	16.4	4	Nucleus	Freedom	ACE 900
CG3 (F)	50.6	Cong. non spec.	R		5	47.6	5	[f]A.B.	Harmony	Fid. 120
CG4 (M)	63.5	Cong. non spec.	L	X	6	57.5	5	Nucleus	Freedom	ACE 500
CG5 (F)	63.0	Unknown	R	X	58	5.0	4	Nucleus	Freedom	ACE 720
CG6 (F)	45.8	Her. non spec.	R	X	4	41.8	5	Nucleus	Freedom	ACE 900
CG7 (M)	72.5	Unknown	R		41	21.5	4	Nucleus	Freedom	ACE 1200
CG8 (M)	53.7	Cong. non spec.	L	X	5	48.7	5	Nucleus	Freedom	ACE 500
CG9 (M)	73.3	Trauma	R		54	19.3	4	Nucleus	CP 810	ACE 720
Mean	**58.6**					**29.7**	**4.5**			

Table 4.1: Clinical and demographic data of the 18 participants included in the study. [a]Non specified congenital hearing loss, [b]non specified hereditary hearing loss, [c]Mondini dysplasia. [d]1: Mild (24-40 dB HL), 2: Moderate 40-55 dB HL), 3: Moderately-Severe (55-70dB HL), 4: Severe (70-90 dB HL, 5 Profound (>90 dB HL). [e]Cochlear ® [f]Advanced Bionics ®. Five participants in the MG and 6 participants in the CG used a HA in their contralateral ear on a daily basis.

4.2.2 Procedure

The study was designed as a longitudinal case/control study with three groups: (1) CI music training group (MG), (2) CI control group (CG), and normal hearing control group (NH). The MG (N=9) followed musical ear training for 6 months, whereas the CG did not. The MG and the CG were

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[2] For details see (I), methods section

tested at three points of time: 1) immediately after switch-on of the implant (baseline, BL), 2) after 3 months (midpoint, MP), and 3) after 6 months (endpoint, EP). The NH group provided reference data at a single test session.

4.2.3 Training methods

The musical ear training program included individual weekly one- hour sessions. The intention of the program was to provide active and passive experiences with pitch, rhythm, and timbre. Examples of the singing training can be heard in the file ***02_Singing with a cochlear implant.ppt*** on the accompanying CD-rom. Examples of the computer applications created for music listening exercises can be experienced in the files (a) ***03_ Recognition of melodic contours.ppt***, (b) ***04_Discrimination of musical instruments.ppt***, and (c) ***05_Recognizing rhythm patterns.ppt*** on the accompanying CD-rom.

4.2.4 Music tests

We created the music test battery with the purpose of measuring a broad range of music discrimination skills. The battery comprises five different tests: (1) musical instrument identification (MII), (2) melodic contour identification (MCI), (3) pitch ranking (PR), (4) rhythmic discrimination (RD) and (5) melodic discrimination (MD). Examples of the PR, MD and RD trials can be found in the appendices section: ***Appendix 11.1: PR test trials, Appendix 11.2 MD test trials, Appendix 11.3 RD test trials.***

4.2.5 Linguistic tests

We assessed the participants' linguistic progress by two different tests: 1) the Hagerman speech perception test (HAG), and 2) an emotional prosody recognition test (EPR). Examples of the trials in the two tests can be found in the file ***06_Linguistic tests.ppt*** on the accompanying CD-rom.

4.2.6 Data analysis

Statistical methods: All music and linguistic test scores were recorded as the percentage of correctly answered items (0-100%). Within-group results were analyzed with paired t-tests; between-group results were analyzed by t-tests. Variables with non-normal distribution were compared using the Wilcoxon/Mann-Whitney U-test. Post-hoc analysis of the responses to single trails of the PR, MD, and RD tests were performed with ANOVAs, Bonferroni corrected. In order to calculate an overall music score for each participant at each point, we converted the raw performance and gain scores from the six music tests at the three points of testing into standard scores. The conversion process of standardization is done by this formula,

$$Z = \frac{x - \mu}{\sigma}$$

where x is the raw score; μ is the mean of the population and σ is the standard deviation of the population. The standard scores (z-scores) were averaged and a group mean was calculated. We refer to these scores as *overall music z-scores* and *overall music gain z-scores*.

Pearson correlation analyses were performed with raw scores, z-scores, and background variables across all participants as well as within groups.

4.3 Study 2[3]

Neuroimaging of cortical plasticity in newly implanted adult cochlear implant users.

4.3.1 Participants

The CI-participants in Study 2 were identical to those of Study 1, except for CG 6, CG 7, and CG 8, who due to malfunction of the scanner (CG 8), claustrophobia (CG 6), and hospital fright (CG 7) did not complete the scanning procedures. The NH group involved in Study 2 is identical to the NH group involved in Study 1. Four participants (CI 2, 8, 12, and 13) had a prelingual onset of profound HL and mainly communicated via signed language, supported by lip reading. For purposes of analyses, two subgroups were identified as (1) the postlingual (POST) HL subgroup (N=11) and (2) the prelingual (PRE) HL subgroup (N=4).

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[3] For details see (II), methods section

4.3.2 Procedures

Design: The CI-participants were scanned consecutively at three points of time: 1) immediately after switch-on of the implant (baseline, BL), 2) after 3 months (midpoint, MP), and 3) after 6 months (endpoint, EP). NH participants underwent PET scanning once. The 3 milestones in study 2 were concurrent with the milestones in study 1 (± 1 day).

MRI: A high resolution T1-weighted MR scan was acquired prior to PET scanning. In the case of CI-participants, this was performed preoperatively. In three CI-participants, who were recruited after their operation, MR scans were not obtained.

PET: We measured raised or reduced cerebral activity as the change of the brain uptake of oxygen-15- labeled water, which matches the distribution of cerebral blood flow (CBF), using the ECAT EXACT HR 47 Tomograph (Siemens/CTI). Emission scans were initiated at 60,000 true counts per second after repeated intravenous bolus injections of doses of tracer with an activity of 500 MBq (13.5 mCi), which equals a radiation dose of 0.465mSv. The scan duration was 90 seconds (1 frame) with an interscan interval of 10 min. Each frame consisted of 47 3.1 mm slices. After correction for scatter and measured attenuation correction, each PET frame was reconstructed with filtered backprojection and smoothed with a post-reconstruction 10 mm Gaussian filter resulting in a resolution of 11 mm full-width-at-half-maximum (FWHM).

4.3.3 Stimuli

All participants were examined in 2 conditions: (1) multitalker babble (BAB) (ICRA 1997), and (2) ‘‘running speech” (RS) (Elberling et al. 2010). The stimuli were presented randomly and the participants were instructed to listen attentively. After each scan, participants were required to describe what they heard. Examples of the 2 stimuli are found on the accompanying CD-rom in the folder in the file ***07_Multitalker Babble and Running Speech.ppt.***

4.3.4 Preprocessing of images

Participants' MR images were co-registered to an MR template in Talairach space (Talairach & Tournoux 1988), using a combination of linear and non-linear transformations (Grabner 2006). Each summed PET emission recording was linearly co-registered to the corresponding MR image using automated algorithms. In the 3 participants for whom no MR scan was available, the PET scans were directly registered to an MR atlas brain in Talairach space.

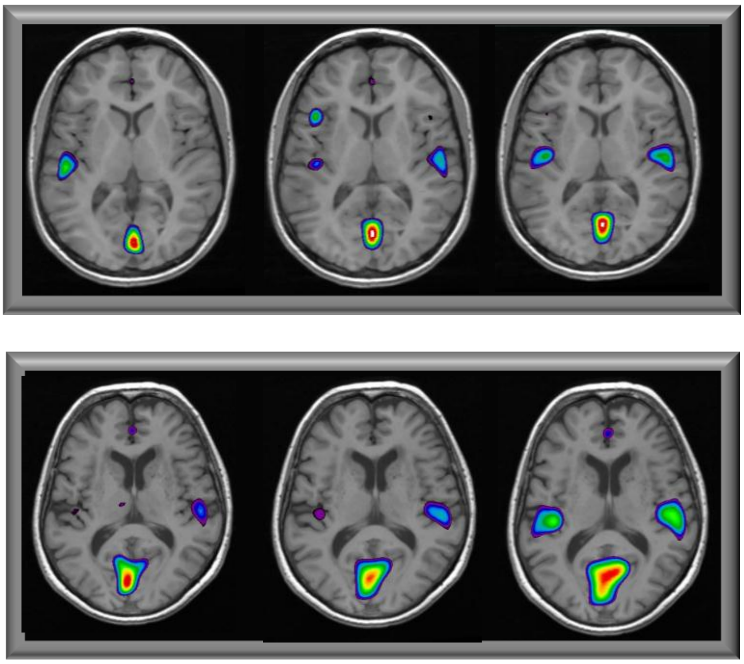

Figure 4.1: Single case studies of PET activations superimposed onto corresponding MR images, inspected by the interactive program MNI-Display (MacDonald 1995). The 3 transverse-plane images show running speech activation at BL, MP, and EP. Top: participant CI 2, 21 year old prelingually deaf woman; bottom: participant CI 5, 70 year old postlingually deaf woman. The images do not represent any statistical inference.

4.3.5 Data analysis (SPM)

SPM: All images were processed using Statistical Parametric Mapping 8 (SPM8, Wellcome Neuroimaging Department, UK; freely available at www.fil.ion.ucl.ac.uk/spm/software). SPM identifies significant changes of cortical activity by comparing brain state according to condition/stimulus. This can be performed in a factorial design where 2 or more factors are combined in the same experiment. Factorial designs enable inferences not only about effects but also about interactions, i.e. the effect of one factor, on the effect of the other. In SPM the design is represented by a design matrix (Figure 4.2).

Normalization: Differences in global activity were controlled using proportional normalization (gray matter average per volume). Significance threshold for main effects was set to $p < 0.05$, family wise error (FWE) corrected for multiple comparisons.

Analyses: We performed 3 different analyses: (1) main effects of time, condition, and hearing background (POST HL vs. PRE HL) and possible interactions between these effects. To define a Region of Interest (ROI) we created a mask based on main effect of condition. (2) Main effects of condition, time, and interactions between these factors in a ROI based on bilateral inferior frontal gyri using the WTU pick-atlas (Tzourio-Mazoyer et al. 2002). (3) Main effects of condition and group (CI vs. NH) at CI BL and possible interactions. To define a ROI we created a mask based on main effect of condition.

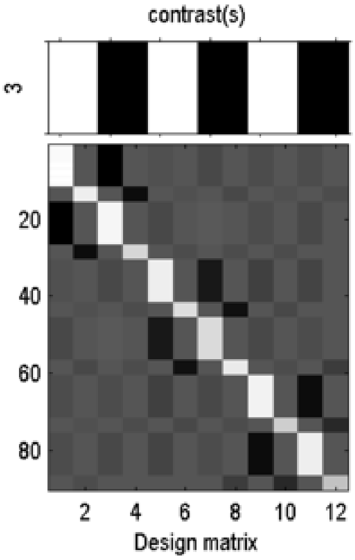

Figure 4.2: SPM design matrix of analysis 1. In this case the matrix examines the main effect of condition (F-contrast), across all 3 points of scanning. A subsequent t-contrast is needed to determine which condition (babble or speech) elicits the highest activation.

4.3.6 Behavioral measures

The Hagerman speech perception measures from study 1 were extracted for the 15 participants involved in study 2, plotted and analyzed.

4.4 Study 3[4]

Music training and testing in preschool children with cochlear implants

4.4.1 Participants

Thirty-one preschool children, 21 with CIs and 10 NHs, in the age between 3-6 years took part in this study (table 4.2).

Table 4.2 Background and clinical data for the participants in the MG and the CG

Group	Sex	Age at proj. start (months)	Number of implants	Side of implant	Hearing aid	Age at implant 1 (months)	Age at implant 2 (months)	Implant use (months)	Etiology of deafness	CI sound-processing strategy
MG										
1	M	74	2	L/R	-	19	39	55	[a]Cong. non spec.	ACE 1200 Hz
2	M	73	1	L	-	54	-	19	[b]Pen, [c]Mon	ACE 900 Hz
3	F	67	1	L	R	50	-	17	Pen, Mon, [d]AN	ACE 900 Hz
4	F	66	1	R	L	29	-	37	Cong. non spec.	ACE 1200 Hz
5	F	66	1	L	N	54	-	12	[e]Gen. (conn 26)	ACE 900 Hz
6	F	67	1	R	L	53	-	46	Gen. (conn 26)	ACE 1800 Hz
7	F	57	1	L	R	38	-	19	[f]Her. non spec.	ACE 900 Hz
8	F	49	2	L/R	-	19	40	30	Pen, Mon	ACE 1200 Hz
9	M	47	2	L/R	-	18	37	29	[g]CMV	ACE 1800 Hz
10	F	45	2	L/R	-	14	14	32	Meningitis	ACE 900 Hz
Mean		**61** *(SD 11)*				**35** *(SD 17)*	**32** *(SD 13)*	**26** *(SD 13)*		
CG										
11	F	79	1	R	L	51	-	28	Her. non spec.	ACE 2400 Hz
12	M	78	2	L/R	-	17	41	61	Cong. non spec	ACE 900 Hz
13	M	67	2	L/R	-	34	47	33	Gen. (conn 26)	ACE 1200 Hz
14	M	65	1	R	-	8	-	57	Meningitis	ACE 900 Hz
15	F	57	2	L/R	-	20	31	38	Cong. non spec.	ACE 900 Hz
16	M	56	2	L/R	-	16	41	40	CMV	ACE 2400 Hz
17	M	52	1	R	-	35	-	17	AN	ACE 900 Hz
18	F	50	2	L/R	-	13	15	35	Her. non spec.	ACE 900 Hz
19	M	50	1	L	-	34	-	16	Meningitis	ACE 1200 Hz
20	F	53	2	L/R	-	15	34	38	Her. non spec.	ACE 2400 Hz
21	M	33	2	L/R	-	13	13	20	Cong. non spec.	ACE 900 Hz
Mean		**58** *(SD 13)*				**23***(SD 13)*	**32***(SD 13)*	**35***(SD 15)*		

Table 4.2: Background and clinical data for the participants in the MG and the CG. Etiology of deafness: [a]non specified congenital hearing loss, [b]Pendred's syndrome, [c]Mondini dysplasia, [d]auditory neuropathy, [e]genetic connexin 26, [f]non specified hereditary hearing loss, [g]cytomegalovirus. All children are users of the Nucleus implant (Cochlear ltd ®, Australia).

4.4.2 Training methods

The music program was scheduled for three months in weekly 90-min modules. The training program included singing, playing, dancing, and listening activities.

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[4] For details see (III), methods section

4.4.3 Procedure

The study was designed as a case/control study with three groups: (1) CI music training group (MG; N=10), (2) CI control group (CG; N=11), and normal hearing control group (NH; N=10). The MG and the CG were tested before and at the end of the study period. The NH group provided reference data at a single test session.

4.4.4 Music tests

Measurements of music discrimination skills in preschool children are not custom in Scandinavia and thus not available. Moreover, in the context of pediatric CI-users, such tests must necessarily take into account the special circumstances of hearing through an implant. Inspired by the available literature covering music and pediatric CI, we created 3 music tests: musical instrument identification (MII), pitch change detection (PCD), and familiar melody recognition (FMI). Due to time constraints, the FMI test was performed only at the end of the 3-month period. Therefore, we were not able to measure performance gains but rather the level of FMI performance after training/no training.

Rhythm test: We were interested in examining the effects of the musical training on the rhythm discrimination skills of the MG participants. For that purpose, we created a rhythm test based on same/different judgments of 15 pairs of rhythmic phrases (Figure. 4.1). The trials were presented randomly in MACarena (see (III)). However, the paradigm was too complicated, and the test was abandoned after a first round of tests.

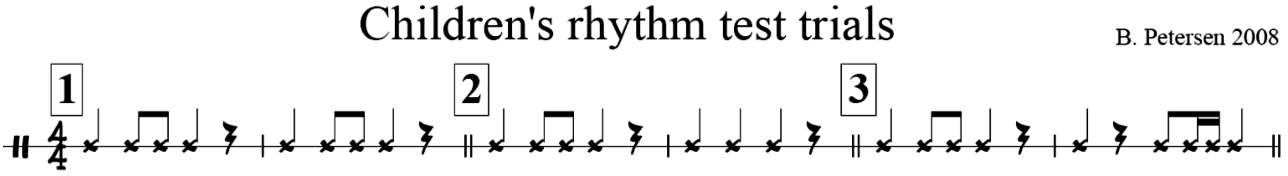

Figure 4.1: Examples of the first 3 pairs of rhythmic phrases in the abandoned children's rhythm test. The number of standards was limited to 5, with each one "same" (1) and two "different" responses (2, 3).

Melo M and Melo R: To further investigate the possibilities of making proper measurements of the children's music discrimination skills, we adapted a Danish version of the musical "game" Audie (see 3.4.2). The game (named Melo M (for melody) and R (for rhythm)) was distributed to parents to MG children with a score sheet for pilot testing (appendix 11.4). Despite follow-up requests we did, however, only receive three responses. One parent commented that she had begun playing the game several times, but her child had started to cry whenever he made an incorrect answer. Others said that they were uncertain about launching and proceeding and had therefore not tried to play the game. The two tests are found in the files ***08_Melo_M.ppt*** and ***09_Melo_R.ppt*** on the accompanying CD-rom.

4.4.5 Linguistic tests

Due to the age differences in the sample, we judged it necessary to differentiate the linguistic tests. Thus, two subgroups were formed: (1) younger children and (2) older children. Both groups performed two different tests. Younger children: a) Linguistic test 1 (expressive vocabulary test) (Bo Ege 1998) and b) Galker test (word discrimination test in background noise) (Galker & Lous 2008). Older children: a) Viborg materialet (expressive vocabulary test) (Kjøge & Pedersen 2004) and b) TROG (grammatical contrast test) (Bishop & Nguyen-Quy 2003).

4.4.6 Parental feedback

To obtain parental feedback concerning possible changes in the children's musical behavior we created a questionnaire (Questionnaire 1), which was distributed to the families in MG at the end of the program. The questions were divided into four categories: (1) general musical interest (response/attention to music), (2) interest in the actual program (talk about/refer to events/songs), (3) singing and dancing activities, and (4) linguistic development associated with musical exposure. The parents were required to state the extent to which they agreed that they had observed changes in their children following the musical program by rating a series of statements: [(1) Strongly disagree, (2) Disagree, (3) Neither agree nor disagree, (4) Agree, (5) Strongly agree]. In a follow-up questionnaire (Questionnaire 2) distributed to all 21 families, we additionally asked parents about their possible plans for their child's participation in music programs, and to what extent they expected that their child would benefit from various forms of musical activity.

4.4.7 Data analysis

All music and linguistic test scores were recorded as the percentage of correctly answered items (0-100%). Within-group results were analyzed with paired t-tests; between-group results were analyzed by t-tests. Variables with non-normal distribution were compared using the Wilcoxon/Mann-Whitney U-test. Mean, standard deviation, median, and range values were given as descriptive statistics. Significance threshold was set at 0.05. Music and linguistic test results were normalized, summed and averaged to calculate individual music and linguistic z-scores. Pearson correlation analyses were performed with raw scores, z-scores, and background variables across all participants, as well as within groups.

5. RESULTS

The most important findings are summarized in this section.

5.1 Study 1 – musical ear training with newly operated adult CI-users

(for absolute scores, p-values, and correlation coefficients, see (I))

In the following, performance gain refers to the increase in mean scores from *baseline to endpoint*. Performance scores refer to *absolute mean values*.

Overall musical performance gain: The overall music **gain** z-scores of the music group were significantly higher than those of control group (Figure. 5.1).

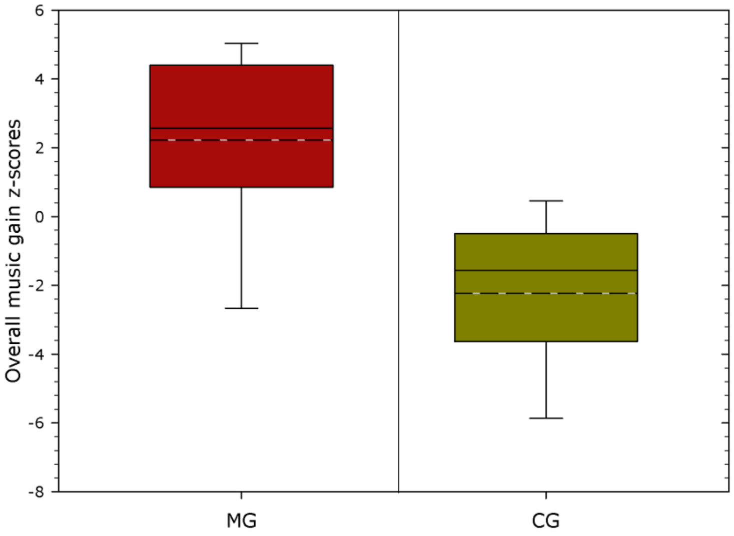

Figure 5.1: Box plot of overall music gain z-scores for MG and CG. Error bars show 10th/90th percentile. Solid box lines: median. Dotted box lines: mean.

5.1.1 Musical and linguistic skill progression in MG vs. CG

Music discrimination: The musical instrument and melodic contour identification performance gain of the music group was significant and significantly larger than that of the CG. Furthermore, the ability of the music group to rank pitches was significant, whereas that of the control group was not.

Both study groups improved their melodic discrimination performance, however non-significantly. Significant difference in performance gain was absent. The rhythmic discrimination performance gain of the music group was significant, whereas that of the control group was not. The difference in mean rhythmic discrimination performance gain between the groups approached significance (Figure. 5.2).

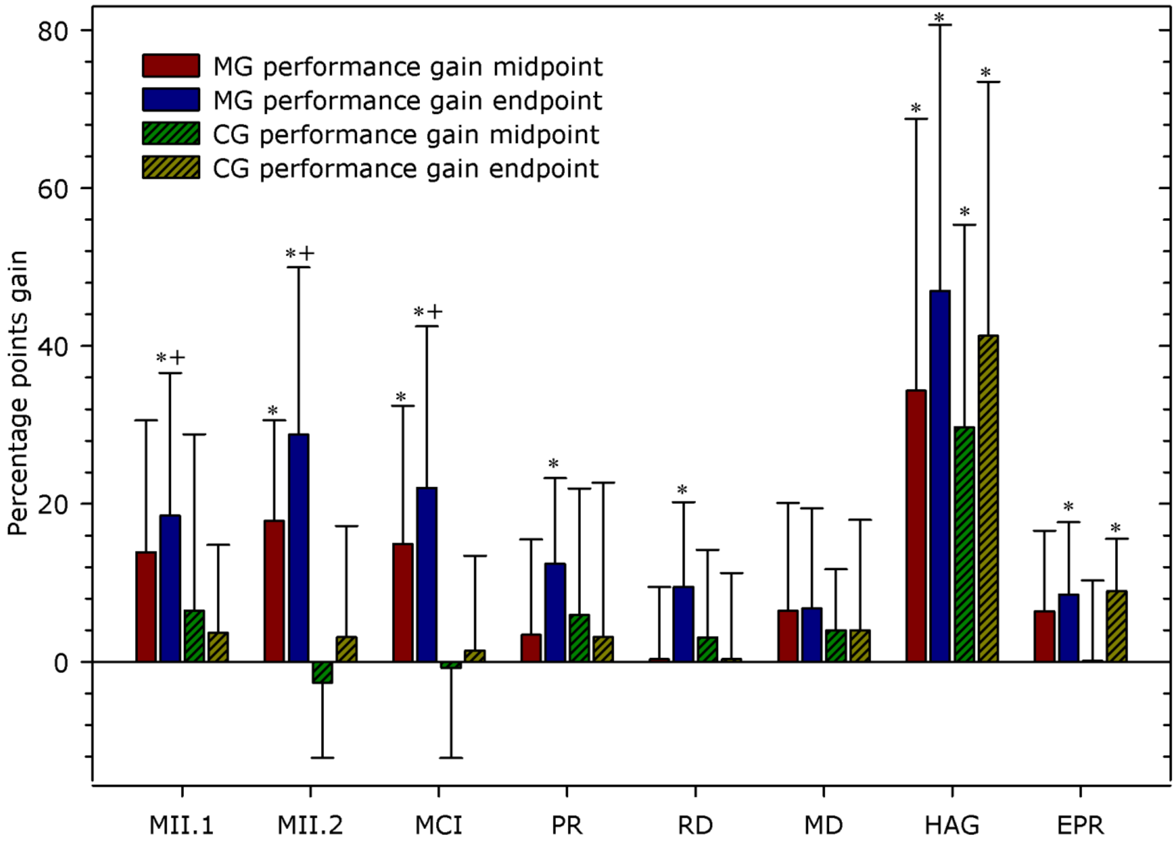

Figure 5.2: Bar graph showing gains of the music (MG) and the control group (CG) from baseline to midpoint and baseline to endpoint, in all 8 discrimination and identification tasks. Error bars show SD. * = gain is statistically significant. + = gain is significantly larger than the CG's gain.

Speech perception: Speech perception performance of the music group improved significantly, as did that of the control group. The difference in speech perception performance gain between the groups was non-significant.

Emotional prosody recognition: The emotional prosody recognition performance of the music group improved significantly, as did that of the control group. The difference in performance gain between the groups was non-significant. The main progress of the music group took place from baseline to midpoint, approaching significance, whereas the main progress of the control group took place from midpoint to endpoint (Figure. 5.2).

5.1.2 Musical skill performance in MG and CG vs. NH

Figure 5.3 shows the average EP scores of the MG, CG and NH groups. Both groups scored significantly below the NH performance level in identification of musical melodic contour, ranking of pitches, and perception of speech and emotional prosody. In the rhythm discrimination test the MG scored non-significantly higher than the NH group.

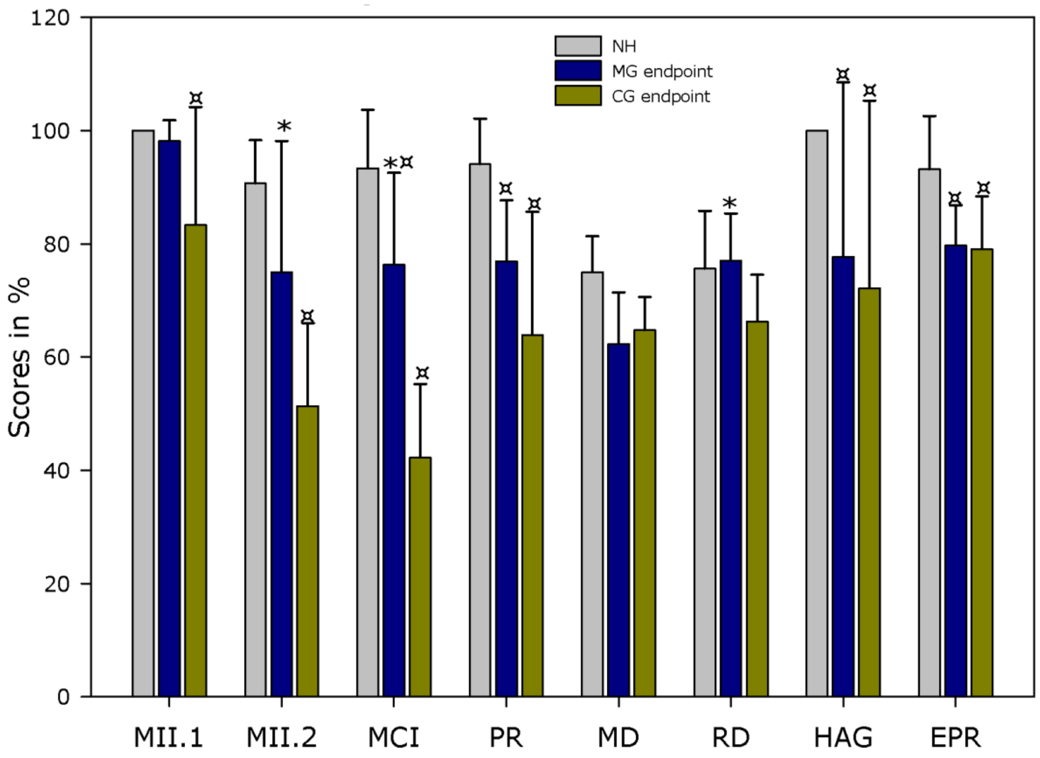

Figure 5.3: Bar chart, showing normal hearing (NH), music (MG) and control group (CG) EP scores in all 8 music and linguistic tests. Black bars = NH mean score. Error bars = standard deviation. * = the final score is significantly higher than the CG final score. ¤ = The score is significantly lower than the NH performance level.

5.1.3 Correlation analyses

Relationship between music and speech perception: Significant relationships were found across all CI-participants between overall music z-scores and speech perception scores, at all three points of measurement (Figure. 5.4). Furthermore, a significant relationship was found between overall music z-scores and emotional prosody recognition scores of the music group at mid- and endpoint.

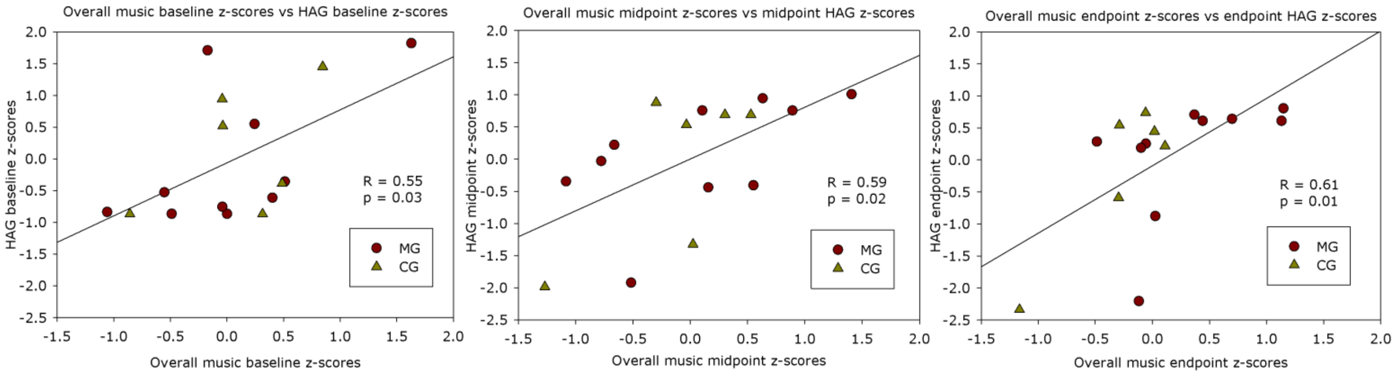

Figure 5.4: Three-panel figure showing regression plots of overall music z-scores (x-axis) and HAG scores (y-axis) of MG and CG at the three times of testing.

5.2 Study 2 Brain maps of cortical plasticity after cochlear implantation

(for absolute scores, p-values, and correlation coefficients, see (II)

5.2.1 Neurological measures

Analysis 1: We found a main effect of speech/babble contrast across subjects, regardless of subgroup, in bilateral superior temporal gyri, driven by higher activity during running speech (Figure 5.5). There was no significant main effect of time, nor any interaction between the effects. The ROI analysis revealed significant interaction between the effects of contrast and group in BA 21/22 in the left superior temporal gyrus (Figure 5.6). A plot of contrast estimates showed a larger difference between running speech and babble in the postlingual than in the prelingual subgroup (Figure 5.7).

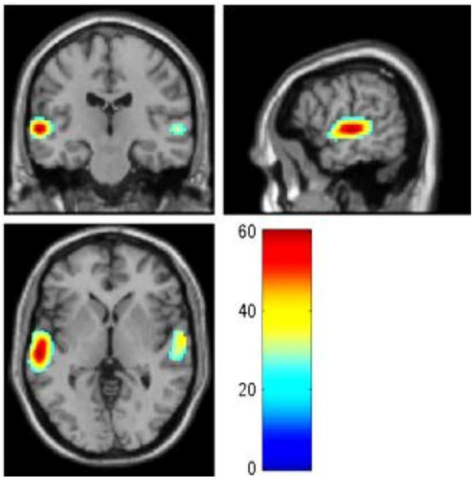

Figure 5.5: Activation map for main effect of speech/babble contrast across POST HL subgroup and PRE HL subgroups in the whole brain analysis (BA 21/22) (F: 1,78).

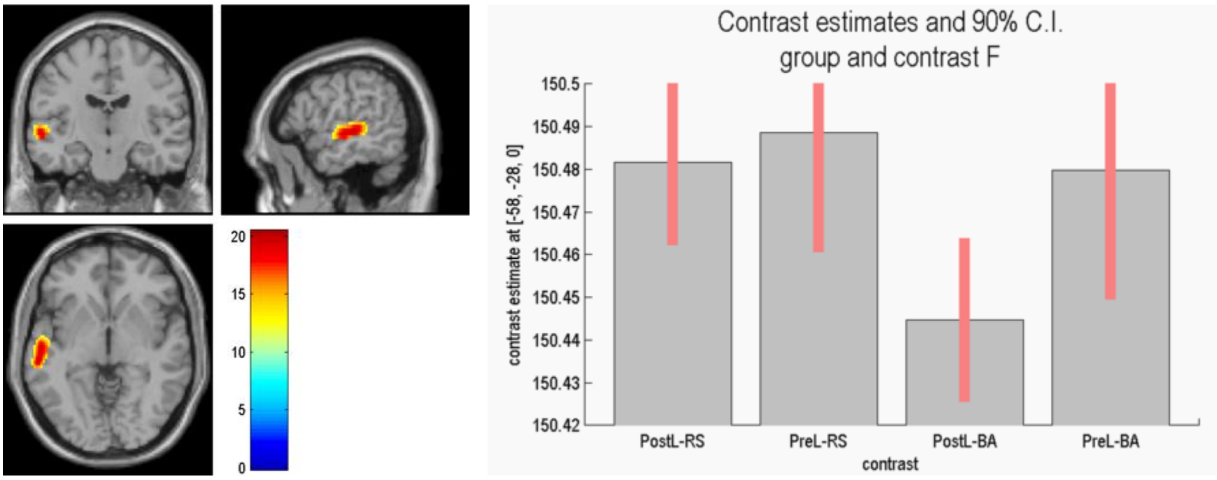

Figure 5.6: Left: Activation map for interaction between effect of contrast and effect of PRE/POST subgroup in the ROI analysis (L BA 21/22) showing greater activation during speech for the post-lingual group. Right: Bar plot showing contrast estimates of conditions in the two subgroups. PostL: post-lingual group; PreL: pre-lingual group; RS: running speech condition; BA: multitalker babble condition.

Analysis 2: In the bilateral inferior frontal gyrus ROI analysis, we found a main effect of speech/babble contrast in the postlingual subgroup, driven by higher activity during running speech. We found a main effect of time in Broca's area (BA 45) (Figure 5.8), but no significant interaction between contrast and time.

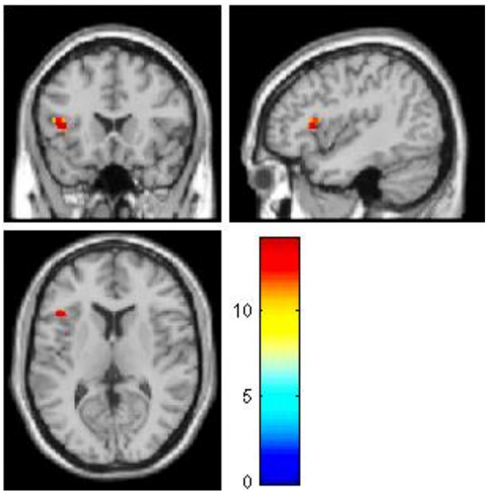

Figure 5.8: Activation map for main effect of time (Broca's area) in the separate analysis of the POST HL subgroup, with ROI based on bilateral IFG (F: 2,60).

Analysis 3: We found a main effect of speech/babble contrast across the CI and NH groups bilaterally in superior temporal gyri and in left middle temporal gyri, driven by higher activity during running speech. Furthermore, we found main effect of speech/babble contrast in the right inferior parietal lobule driven by higher activity during babble. We found a higher activation of the caudate nucleus in the NH group compared to the CI-group. The ROI analysis based on main effect of contrast yielded a main effect of CI vs. NH in secondary auditory cortex including Wernicke's area (BA 22) in the right superior temporal gyrus, due to higher activity of this area in the NH group than in the CI-group. Furthermore, we found an interaction between the effect of speech/babble contrast and the effect of group in the right inferior parietal lobule

5.2.2 Behavioral measures

The mean Hagerman speech perception performance of the entire CI-group increased 31 percentage points from baseline to midpoint and again by 10.5 percentage points from midpoint to endpoint, for a total increase of 41.5 percentage points, reflecting major differences between the two subgroups. In the postlingual subgroup, the mean Hagerman performance increased by 37.6 percentage points from baseline to midpoint and again with 13.6 percentage points from midpoint to endpoint, for a total of 51.3 percentage points. In the prelingual group, the mean Hagerman performance increased by 12.8 percentage points from baseline to midpoint, and by 1.8 percentage points from midpoint to end point for a total of 14.5 percentage points (Figure 5.9).

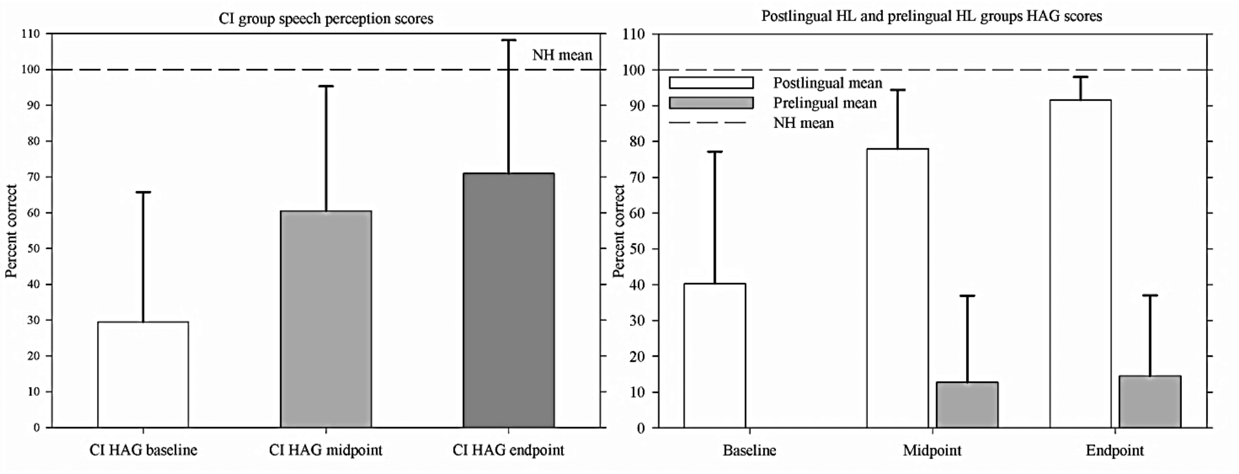

Figure 5: Left: bar plot showing mean percent correct Hagerman speech perception scores for the entire CI-group at baseline, midpoint and endpoint. Right: bar plot showing mean percent correct Hagerman speech perception scores for the POST HL subgroup and the PRE HL subgroup respectively at baseline, MP and endpoint. Dashed line shows NH mean. Error bars show standard deviation.

5.3 Study 3 – Musical training with pediatric CI-users

(for absolute scores, p-values, and correlation coefficients, see (III))

5.3.1 Musical performance

MII: The music group improved their musical instrument identification significantly as did the control group. Despite a larger gain in the performance of the music group, significant difference in gain between the two groups was absent. Both experimental groups showed final scores that were comparable to those of the normally hearing controls (Figure 5.6).

PCD: The pitch change detection performance gain of the music group was significant as was that of the control group. Despite a larger gain in the performance of the music group, significant difference in gain between the two groups was absent. The difference between the final pitch change detection scores and those of the normally hearing controls was non-significant, while the final pitch change detection scores of the control group were significantly poorer than those of the NH group (Figure 5.6).

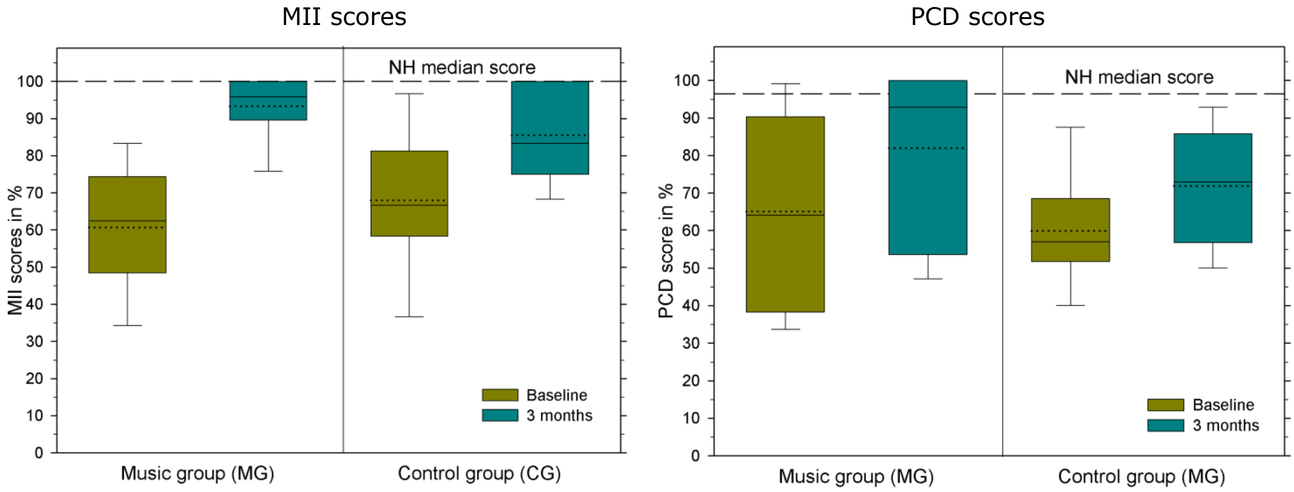

Figure 5.6: Box plots showing MII (left) and PCD scores (right) before and after the 3-month study period for MG and CG, respectively. Error bars show 10th/90th percentile. Solid box lines: median. Dotted box lines: mean. Horizontal dashed line: NH median score.

FMI: The final familiar melody identification performance was non-significantly higher than that of the control group. Both CI-groups scored significantly below the NH performance level (Figure 6.7).

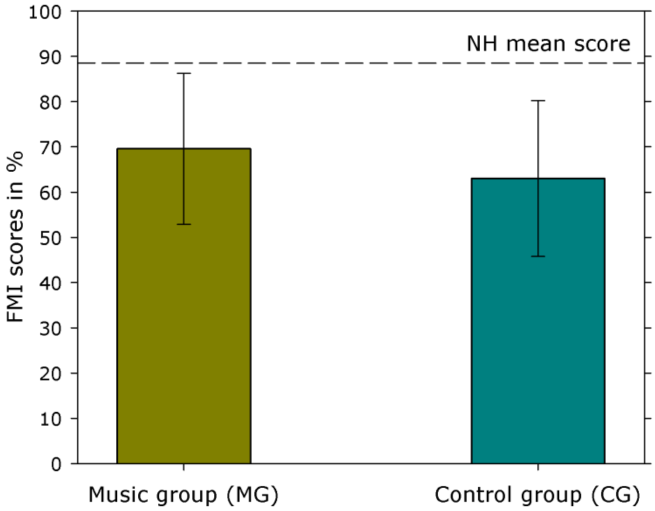

Figure 5.7: Bar chart showing FMI scores of MG, CG, and NH groups. Error bars show standard deviation. Horizontal dashed line: NH mean score.

5.3.2 Linguistic performance

Younger subgroup

In Linguistic test 1 (expressive vocabulary), performance gain of the younger music subgroup was non-significant, as was that of the control group. Significant difference in performance gains between the two subgroups was absent. The post-test scores of the music group were significantly higher than the standard Danish NH reference, which was not the case for the CG younger subgroup.

In the Galker test (phonetic discrimination), the performance gain of the younger music subgroup approached significance, whereas that of the younger control subgroup did not. Significant performance gain difference between the two subgroups was absent. The post-test scores of the younger music subgroup were significantly higher than the standard Danish NH reference, as was those of the control subgroup.

Older subgroup

The Viborg material test (vocabulary) performance gain of the older music subgroup was significant, whereas performance gain of the older control subgroup was not. Significant difference between performance gain of the two subgroups was absent. Both groups scored very close to the standard Danish NH reference.

The TROG (grammatical contrast test) performance gain of the older music subgroup was non-significant, whereas that of the older control subgroup approached significance. Significant difference in performance gains between the two groups was absent. Both groups had final TROG scores that were was significantly lower than the national Danish NH reference.

Overall linguistic performance gain:

Pooled overall linguistic performance gain (z-scores) of the music group was significant, whereas that of the control group approached significance. The difference in overall linguistic performance gain between the two groups was non-significant (Figure 5.8).

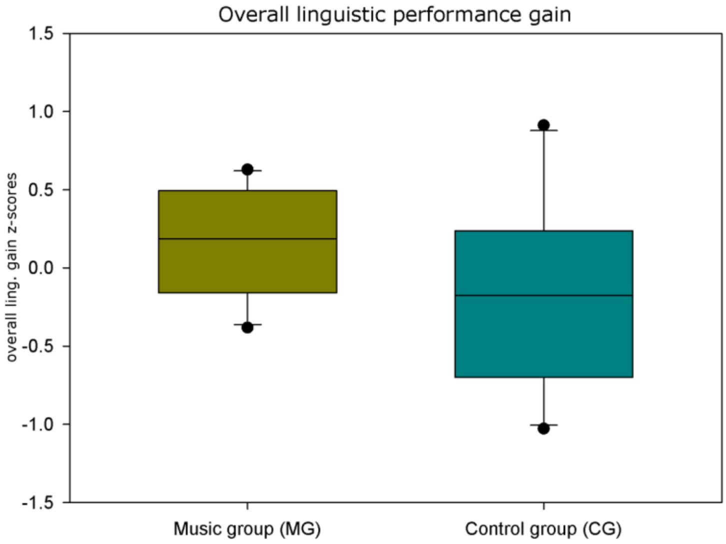

Figure 5.8: Box plot showing overall linguistic performance gain (z-scores) of MG and CG. Error bars show 10th/90th percentile. Solid box lines: median.

5.3.3 Parental feedback

Questionnaire 1: On an average, parents reported that their child expressed a generally increased interest in music during the program and frequently referred to the music program, listened to the CD, and spontaneously sung songs from the program. They observed increased and improved singing and dancing activities. Furthermore, the parents found that their children in general had improved sound discrimination, developed more prosody, and become more attentive to language nuances and rhymes.

Questionnaire 2: Thirty-eight percent of the families had plans, 10% had no plans, and 52% considered letting their child participate in a public musical teaching program. Ninety percent stated that they would be more inclined to do so, if they knew that the program was especially arranged for children with CIs. On an average, parents expected that their child would benefit to a large extent from musical activities in general and even from learning to sing and play an instrument.

5.3.4 Correlations

Music/linguistic performance vs. age at testing/implantation: Overall music z-scores correlated significantly with age at testing across all participants, while there was no correlation between combined music z-scores and age at implantation (Figure 5.9). We found no correlation between overall linguistic z-scores and neither age at testing nor age at implantation.

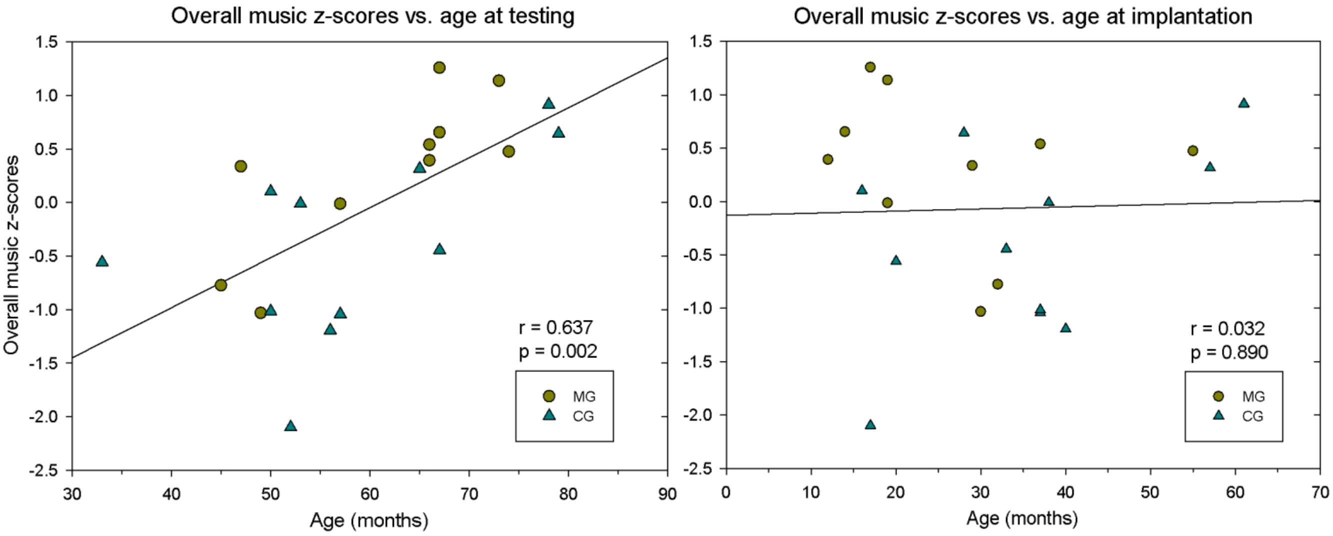

Figure 5.9: Linear regression plot of overall music z-scores vs. age at testing (left) and overall music z-scores vs. age at implantation (right).

Music performance vs. linguistic performance: A weak correlation was found between overall music and overall linguistic z-scores.

One vs. two implants: Overall music z-scores showed a weak negative relationship with number of implants across all participants, i.e. children with one implant on an average scored slightly higher than those with two. Overall linguistic z-scores and number of implants showed a weak positive correlation in the younger subgroups, but not in the older subgroups.

Music test correlations: We found a strong correlation between all music test scores, i.e. individual music discrimination was consistent across disciplines.

6. DISCUSSION

6.1 Study 1 - effects of musical ear training on newly implanted adult CI-users

Main findings: We found that six months of musical ear training significantly improved the overall music perception of the participants in the music group, compared to the control group, especially with regard to timbre, melodic contour, rhythm, and pitch. In contrast, the improvement of the control group within these areas was non-significant, and significantly lower than that of the music group in discrimination of timbre and pitch. We found a significant consistent relationship between overall music performance and speech perception. However, no significant effect of music training was found on speech perception. Recognition of emotional prosody correlated significantly with overall music performance in the music group, and showed a trend toward faster progress, relative to the control group. This suggests that musical training may be beneficial to music discrimination skills, but not necessarily to linguistic skills of CI-users.

This study and previous studies: Music training has been implemented with positive results in previous studies involving CI-listeners (Gfeller et al. 2000a; Gfeller et al. 2002b; Galvin et al. 2007). While these studies primarily were based on computer-mediated training of isolated musical tasks such as recognition of pitch, timbre, or melody, the present study employed one-to-one tuition within three different musical domains, for an extended period of time. Furthermore, in contrast to previous studies, the participants in the current study were “naïve” CI-users, i.e. without implant experience. Finally, it is important to point out that, except for the MCI, the stimuli presented in the tests were not part of the music training, i.e., there was no training for the test. Our results thus demonstrate that practical and guided musical ear training has the ability to significantly affect the general music discrimination skills, even in CI-listeners with no previous implant experience.

Musical and linguistic abilities: The remarkable progress in speech perception performance, observed across all participants, supports previous studies, which showed that most performance gains occur in the first three months of use (Spivak & Waltzman 1990; Ruffin et al. 2007). Interestingly, our correlation analyses suggested a strong relationship between speech perception performance and overall music discrimination performance. This is in contrast with Singh et al. (2009), who, in a sample of adult CI-users, found no relationship between melody recognition and phoneme discrimination, and with Gfeller et al (2008), who found association between speech perception and music perception only when lyrics were present. Our findings suggest a relation between music and speech abilities, possibly due to the necessity of low level acoustic feature extraction from sounds in both domains (Besson & Schon 2001), but no transfer effect of musical training onto the linguistic domain. These findings are consistent with the view that high level processing of music and language primarily takes place in separate brain modules (Peretz & Coltheart 2003). The fact that we found no correlation between music performance *gain* and speech perception gain indicates that the *progress* in either domain may take place independently of one another. This is illustrated by the cases of four prelingually deaf participants in our study, who, despite no or very modest endpoint speech perception gains, achieved moderate progress in their overall music perception.

Perception of timbre: The significant effect on the music group participants’ ability to identify musical instruments (timbre) is in line with other studies based on computer-assisted training (Fujita

& Ito 1999; Leal et al. 2003; Pressnitzer et al. 2005). Furthermore, the music group achieved an endpoint level that was comparable to the NH level, which is particularly encouraging, since previous studies on timbral discrimination of CI-users have found performance that was significantly poorer than that of NH participants (Gfeller et al. 2002b; McDermott & Looi 2004). The result is important, because improved perception of timbre may add positively to the aesthetic enjoyment associated with music listening.

Contour and pitch: The musical ear training program also generated a significant progress in the participants' ability to identify a melodic contour and to rank pitches. Significant improvement of melodic contour identification skills has been shown in a previous study involving computer based training (Galvin et al. 2007). Our findings suggest that this skill may be substantially improved also by training, which combines playing/singing exercises and guided listening. Since pitch ranking was not specifically trained, the significant progress of the music group's PR skills may represent a generalized effect from the active and passive musical exposure. In contrast to ranking of larger pitch differences, ranking of small pitch differences was only vaguely improved. Similar associations between size of interval and pitch ranking skills were reported by Looi et al. (2004), and illustrate the implant's restricted transmission of fine-grained pitch cues. It is, however, worth noting that while the average scores of the CI-users are significantly poorer than those of the NH group, some music group participants produced scores near the NH range, thereby correctly identifying interval changes as small as one ST. This variability indicates an effect of the training, but probably also stems from differences in the preconditions for music listening in CI-users.

Melodic discrimination: Ability to correctly identify pitch direction is a fundamental prerequisite for perception of melody, and usually strongly associated with familiar melody recognition in CI-listeners (Gfeller et al. 2002a; Looi et al. 2004; Galvin et al. 2007). Despite significantly improved pitch ranking and melodic contour identification skills, the average melodic discrimination skills of the music group participants improved non-significantly. However, in contrast to recognition of familiar melodies, as used in the former studies, the MD test used in the present study assessed comparison of unfamiliar melodies, which is substantially more challenging. The poor progress in melodic discrimination may also reflect the aforementioned poor discrimination of small intervals, and demonstrates that for a CI-listener, perception of pitch in the context of many pitches is a very challenging task. Furthermore, the same/different paradigm loads heavily on working memory, which may be restricted in some CI-users (Knutson et al. 1991).

Rhythm discrimination: Previous studies based on simple tempo or pattern discrimination tests, have concluded that perception of rhythm with CI is close to normal (Gfeller et al. 1997; Kong et al. 2004; Limb et al. 2010). In our study, the music group participants significantly improved their ability to discriminate complex rhythm patterns and reached endpoint performance levels comparable to the NH group. This progress predominantly took place in the mid- to endpoint period, in which rhythm training was introduced, and produced endpoint scores that were significantly higher than those of the control group, which is a strong indication of the effect of training. Improved perception of rhythm may positively influence the music listening outcome of CI-users.

Effect of music training on language performance: Our findings indicate that musical ear training does not affect the speech perception progress of cochlear implantees. However, a transfer effect may have been covered by other factors. First, in the initial phase of CI adaptation, the speech perception progress may be so strongly carried by the effect of daily use that other sources of training may be of lesser significance. Second, all participants followed speech therapy, which may have had substantial influence on the perceptual development. Third, the implant is specifically optimized to effectively facilitate speech perception. Finally, the Hagerman test may have been inadequate in comparing development in the two groups. A ceiling-like effect, with five of nine MG participants and two of nine CG participants scoring within the 90th percentile at the endpoint measurement, may have prevented some participants from achieving higher scores.

Emotional prosody recognition: Both groups significantly improved their abilities to recognize emotional prosody. However, in contrast to the control group, the major part of the music group's progress occurred in the initial three-month training period. Furthermore, we found a consistent significant correlation between the overall music scores and emotional prosody recognition performance in the music group, which was absent in the control group. This suggests that musical training may have affected not only the speed of the EPR progress, but also strengthened the link between music discrimination and EPR. The unexpected progress of the control group indicates that because the range of changes in pitch and timing in emotional prosody is much greater than that typical of music, these cues may be more easily identified (Ayotte et al. 2002). In line with findings of Xin et al. (2007), both groups scored significantly below the NH level, which emphasizes that these prosodic cues are particularly challenging to CI-listeners and may explain the lack of further progress in the MG.

6.2 Study 2 - cortical mechanisms in early restoring of speech in cochlear implantees

Postlingual hearing loss versus prelingual hearing loss: Our investigation of brain activation following cochlear implantation revealed that the CI-users with postlingual HL had a higher activity in BA 21 and 22 in the left superior temporal gyrus than the CI-users with a prelingual HL. This difference reflected a higher activation during speech than during babble in the postlingual listeners, versus comparable levels of activity during speech and babble in the prelingual listeners. We speculate that the former disengage attention when they are presented with the incomprehensible babble stimulus. This disengagement is then reflected in decreased temporal brain activity. In contrast, the prelingually deaf CI-listeners may be equally attentive to the two stimuli, as reflected in undifferentiated activity.

Behavioral development: The difference in cortical activity was mirrored in the behavioral measures. In contrast to the prelingual subgroup, the postlingual subgroup possessed moderate speech perception levels immediately after switch-on, as well as remarkable gains in performance, predominantly in the first three month period. This finding is consistent with expectation and implies an association between behavioral performance and brain activity related to the history of hearing loss. In prelingual deafness, the neuronal connections of the auditory pathways are not established in the appropriate time window of opportunity. The subsequent electric stimulation at some time in adulthood may produce some hearing sensation, but the discriminations of sounds and time intervals remain defective (Mortensen et al. 2005). Follow-up studies in the present population may provide interesting insight into the possible long-term speech perception progress in the prelingual subgroup.

Effect of time: We found a main effect of time exclusively in Broca's area, but only in the postlingual subgroup. This indicates that the changes following adaptation to the implant are most profoundly manifested in this specific area, which is associated with speech perception and production. Surprisingly, we found no interaction between main effect of contrast and main effect of time. This suggests that the area is increasingly activated, regardless of whether the stimulus makes semantic sense or not, or is active in the distinction between sense and nonsense.

Lateralization: In the entire CI-group, the bilateral middle and superior temporal gyri, more specifically the Brodmann areas 21 and 22, were significantly more active during running speech than during babble, across all points of measurement. Thus, newly implanted CI-recipients clearly distinguished between speech-like noise and speech, confirming that both hemispheres are involved in the speech perception process, also during unilateral stimulation.

CI baseline versus normally hearing: Analysis of the CI-group versus the NH group showed that right inferior parietal lobule was significantly more active during babble in the CI-group than in the NH group. Furthermore, the NH group had a significantly higher activation in the caudate nucleus than the CI-group. The latter observation may be explained by a reduction of the effort needed by the NH participants to deal with the well-known task of receiving a message. The caudate nucleus is a part of the striatum, which subserves among other tasks the learning of slowly modulated skills or habits (Gabrieli 1998). To the normally hearing listener, the reception of auditory information is an every-day experience similar to following a known route (Wallentin et al. 2006).

Conclusion: Unlike CI-listeners with postlingual hearing loss, CI-listeners with prelingual hearing loss showed undifferentiated activation of left superior temporal gyrus during speech and speech-like stimuli. This difference was reflected in behavioral data. Furthermore, Broca's area was activated as an effect of time, but only in CI-listeners with postlingual hearing loss. This confirms the key role of Broca's area in restoration of speech perception, but only in individuals in whom Broca's area has been active prior to the loss of hearing. The study clearly demonstrates that adaptation to the electrical stimulus of the cochlear implant is highly related to history of hearing loss.

6.3 Study 3 - effects of musical training on pediatric CI-users

Music discrimination outcome: The music group children significantly improved their musical discrimination performance during the training period. The improvement was larger than that of the control group, however non-significantly. The control group children significantly improved their musical discrimination performance too, but in general they scored lower than the MG children and significantly poorer than the NH controls in pitch change detection. In contrast, the MG children scored comparably to the NH controls in musical instrument identification and pitch change detection post-training. Moreover, parental feedback revealed that the music training had a strong positive impact on the children’s musical interest and musical activity. Although the study represents many challenges and some limitations associated with size and heterogeneity of the sample and shortcomings of tests, the results are very encouraging. This is emphasized by the fact that the approach of the training program was to teach and make music in the real world environment rather than within pure laboratory tasks.

Musical instrument identification (MII): The noticeable progress in the MG children’s ability to identify a musical instrument compared to the CG children is consistent with Gfeller et al (2002b), who demonstrated significant improvement in identification of musical instruments in adult CI-users after computerized training. However, in the present study, the test stimuli were exclusively presented during testing, and training did not directly aim to improve this skill. Our results show that despite poor spectral resolution, the CI delivers stimuli that are precise enough to permit recognition of timbre, and that this ability may be affected by training. Apart from an expected benefit for music appraisal, improved discrimination of timbre may have a positive impact on non-musical tasks such as recognition of voice gender and perception of speech in noise involving competing talkers. Future studies should look further into this potential association.

Pitch change detection (PCD): The MG children’s average PCD progress was non-significantly larger than that of the CG children, and their post-training scores were comparable to those of the NH group. This indicates that a majority of MG children became able to identify changes as small as a semitone, which is the smallest interval in Western music.

Familiar melody identification (FMI): The MG children's post-training FMI performance was more accurate, however non-significantly, than that of the CG children. This suggests that the increased focus on melodic contour through guided singing practice and glissando exercises may have provided some musical prerequisites for music identification. However, despite perfect recognition of the songs with lyrics, the CI-users' melody-only performance was significantly poorer than that of the NH group, even with available rhythm cues. This indicates that for pediatric CI-users, enhanced ability to detect small pitch changes does not facilitate detection of pitch changes in the context of variable tones, as required in recognition of melodies (Vongpaisal et al. 2006). It also indicates that other cues than actual pitch changes, e.g. timbral, may assist the child in the PCD task. The poor FMI results match previous research on song recognition in child implant users (Stordahl 2002; Vongpaisal et al. 2004; Nakata et al. 2005), and indicate that current implant technology induces a pitch processing deficit very similar to the phenomenon of amusia in NH individuals, as suggested by Nakata et al (2005). Interestingly, this poor perception of melody, does not seem to affect these children's music enjoyment, probably because features such as rhythm and lyrics are more effectively transmitted (Gfeller et al. 1998).

Single case observation: One congenitally and profoundly deaf girl, who had been bilaterally implanted at the age of 11 months made perfect and immediate recognition of all songs on the first occasion of testing. Her exceptional abilities were accompanied by examples of spontaneous solo singing with precise melodic and lyrical renditions of children's songs. The girl, who was 34 months old at the time of testing, had received intense family-based and professional auditory training and participated in musical activities on a regular basis. The case suggests that musical stimulation in large doses and from a very early stage may help child implant users to outperform the technological limitations of the implant.

Effect of music training on language performance: The overall linguistic performance gain of the music groups was larger and less scattered than that of the control group, but remained indicative. This lack of significant transfer from the musical to the linguistic domain may have more explanations. First, the children in both groups followed regular speech training, which may have had a high impact on the linguistic progress. Second, the duration as well as the frequency of the music training may have been insufficient to significantly influence linguistic skills. Third, other factors such as daily CI use-time and non-verbal intelligence influence linguistic skills too (Wie et al. 2007). Finally, different cognitive processes may be required for some aspects of music listening than for speech perception and production. In our study, we found internal consistency between performance on the different musical tests, but no significant correlation between music and language tests. This is in agreement with Singh et al. (2009), who found no relationship between melody phoneme recognition in child implant users. In a survey on the musical involvement of young CI-listeners Gfeller et al. found that music ratings correlated significantly with overall communication scores, indicating that older children, who successfully communicate via listening and speaking, use these skills to understand music (1998).

Prosodic speech: The children in this study spoke with genuine local dialects. This shows that while child implant users may face trouble in perceiving the smaller interval changes of music, they appear able to perceive and produce the greater range of pitch changes in speech.

Bilateral/unilateral implant: Correlation analyses showed that children with one implant performed moderately better in the music tasks than those with two, possibly due to some residual hearing on the non-implanted ear in the unilaterally implanted children. Aided residual hearing provides fine-structure cues in low-frequency ranges (<1000 Hz), crucial for pitch perception (Kong et al. 2005). One child from the music group scored 100% correct on all final music tests. The girl had some residual hearing and was bimodally aided, as well as musically inclined.

Conclusion: Music training with carefully selected material and well-considered methods is relevant to pediatric CI-users and offers a stimulating environment and substantial listening practice. The results and the parental feedback indicate that such training may support the musical, linguistic, and even psychosocial and cultural development of these children. If implemented early and regularly, the proposed program may provide a valuable supplement to other auditory habilitation initiatives.

6.4 Musical development

This research project shows that perception of music with a cochlear implant can be significantly improved. In the case of adult CI-users without previous implant experience, music discrimination skills in general, and perception of timbre, rhythm and pitch direction in particular, are significantly affected by practical and guided musical ear training. The variability is huge and some of the musically trained participants perform close to the normally hearing level. This reflects the effect of training as well as individual differences in preconditions. Discrimination and identification of melody remain inaccurate, and unaffected by training. Perception of music and speech is related, but CI-users with prelingual hearing loss can develop music discrimination abilities, despite absent or poor speech perception progress. Perception of rhythm can be trained to a normal-hearing level, which may positively influence music listening outcome.

In the case of pediatric CI-users, to whom music is a novel acoustic stimulus, music discrimination can improve significantly, even from incidental exposure. Music training and active music-making may facilitate this process. Aided low frequency residual hearing facilitates this progress. Musical stimulation at an early stage and in large doses may influence the musical development of prelingually deaf children with implants. Since the primary objective of music training is to improve the preconditions for music listening, future studies should examine music enjoyment and music involvement of adults as well as children with CIs, and the correlation between these measures and objective measures of music discrimination.

6.5 Language development

Newly implanted adult and pediatric CI-users develop their linguistic skills significantly as an effect of time, daily use, and oral/aural therapy, but not necessarily by training music. To adult CI-users, perception of emotional prosody is difficult, but may develop faster and associate with musical perceptual strategies through musical training. Pediatric CI-users have significant difficulty with discrimination of grammatical contrasts, but may have some benefit from musical training in their development of overall linguistic abilities. The melodic element of language, as expressed in dialects spoken by prelingually deaf children with CIs, seems unaffected by poor processing of musical melody. Because the linguistic skills of CI-users are far from perfect, and because music offers a stimulating environment, we believe this is an important field of research that motivates future studies.

6.6 Quality of life

This research provided no objective measures of the influence of music training on aspects of social behavior and quality of life. However, anecdotal reports and strong indications of engagement and personal benefit from the program expressed by the adult participants, as well as the children and their parents, motivate future studies on the relationship between music enjoyment and quality of life in CI-users.

6.7 Societal perspectives

As stated in the preface, the term deaf is being redefined in these years. In 20-30 years time we may be experiencing a reality, in which signed language has gradually been replaced by communication via cochlear implants. As shown in this research project, “electrical” hearing is far from normal, and implant users face many challenges in their daily auditory perception. Especially with regard to the growing number of children and young people, who are integrated into the hearing community, it is extremely important that proper and sufficient information about these children's special hearing conditions is provided to caretakers and teachers. Furthermore, improved methods of (re)habilitation are essential to help maximize implant outcome, and continued research into the field is needed. Teachers of music should learn to handle this new reality too. Special educational programs with focus on music perception and training of CI-users may be one possibility. Furthermore, edutainment software, which has proven successful for training of skills such as mathematics, reading, and foreign language, could possibly be of great help to CI-users of all ages, trying to figure out the acoustic world of music.

6.8 Neuro plasticity

The study clearly demonstrates that the cortical mechanisms underlying adaptation to the electrical stimulus of the cochlear implant is highly related to history of hearing loss. Adult patients whose hearing loss occurred after the acquisition of language involve brain areas associated with speech comprehension, which is not the case for adult patients whose hearing loss occurred before the acquisition of language.

6.7 Evaluation of methods

The employment of a one-to-one teaching setting for newly implanted adult CI-users was a good choice, because many individual differences exist in the initial phase of adaptation and because minor individual adjustments can be made to the program. It is, however, possible that adult CI-users with a longer implant experience could take part in musical training activities in a group. It is also possible that musical training at a later stage in the CI adaptation process, in which speech perception has stabilized, might bring further contribution to the challenging perception of prosody and speech in noise. In elementary musical ear training, the voice is the instrument of choice. Though a keyboard may not need refined technique to play, singing is superior because it comes natural and has direct access. Furthermore, singing involves articulation of words and phrases and thus supports the general development of vocal production. Intonation is extremely difficult with an implant. However, in this context, being in time and articulating the lyrics precisely is equally important and may provide plenty of musical enjoyment and mood elevation.

In the context of preschool children, group-based training was a proper choice of method. With older children, it may be relevant to train music individually or in small groups, in which personal direction can be provided. The choice of a “learning-by-doing” approach also proved successful, in that the children were engaged and all completed the program.

7. FUTURE STUDIES

Several studies could be proposed to expand on the studies presented in this thesis:

1) Perspectives of music with cochlear implants

Doctors and health politicians have gradually acknowledged that changes in the patient's health related quality of life (HRQOL) resulting from a medical intervention must be considered. With regard to CI, such measures are relevant for several reasons. First, because the intervention not only affects the patient's hearing and speech function but also self-esteem, daily life, capacity for work, and social life. Second, because the treatment is relatively expensive (app. 350.000dkk.), and the population of implantees (including children and adolescents) rapidly growing with estimated 900 users in Denmark (July 2010). Studies performed on small samples of CI-users show, that particularly the patient's mental well-being and social functioning may be positively influenced by the CI-treatment (Mortensen et al. 2004; Wanscher 2006). Furthermore, it has been reported that CI-users on average are on a level with or slightly above the general population within these domains (Krabbe et al. 2000). Measurements of quality of life and music listening practice among CI-users in Denmark could provide comprehensive novel knowledge about the level of QOL, music outcome and the possible correlation between these two conditions. Moreover, investigation of a number of demographic and clinical background variables from these patients may make it possible to assess if particular preconditions determine a successful outcome with CI – musically and in general. On the long term the results may have great importance for the quality of pre- and postoperative guidance and counseling provided to CI-candidates and -recipients at implantation centers. In addition, insights into the relationship between music appraisal and objective performance may help audiologists in more accurately programming and fine-tuning speech processor settings, and assist CI speech therapists in choosing and revising materials and methodologies used in auditory/oral rehabilitation. Especially regarding the growing number of pediatric CI-users, whose implant outcome is generally less easily evaluated, this may have a great positive impact.

2) Music in the implanted brain

Ethical restrictions prevented us from measuring brain activity during music listening conditions. A study, which identifies cortical areas involved in temporal and spectral listening tasks with a CI, may provide important knowledge about music perception with electrical stimulation. This could have importance for future refinements of the sound processing schemes and for creation of music specifically aimed at CI-users.

3) Bilateral and bimodal music perception

During the last couple of years, many existing adult CI-users in Denmark have been offered an implant on the contralateral ear. This provides a unique opportunity to study possible benefits from bilateral hearing on music perception. Furthermore, the impact of bimodal stimulation, i.e. a hearing aid in the contralateral ear, on music perception is not fully understood. A study that examines objective music discrimination skills and music liking, in bilateral and bimodal listening could provide some important new insight into this field.

4) Training programs

Edutainment software, which presents musical sounds and provides music understanding in a way that is relevant to children with implants, has not been developed. Such programs should be described with the intention of making them accessible on-line to CI-users world-wide.

8. SUMMARIES

8.1 English

Cochlear implantation is a treatment offered to children and adults with severe/profound hearing loss with the purpose of (re)establishing hearing and speech perception. Surgery and technology has improved dramatically during the last ten years, with the number of individuals who use cochlear implants as a means for their daily communication exceeding 120,000 worldwide. However, neither methods of maximization of implant outcome nor the processes underlying implant adaptation have been explored to an equal extent. In three different longitudinal studies, we examined how auditory abilities develop in cochlear implant (CI) recipients; (1, 3) behaviorally by studying the effects of musical ear training in adults and children with CIs and; (2) neurologically by measuring the progress in brain areas associated with perception of sound and language in newly implanted adult CI-users.

(1) Successful rehabilitation of CI-patients depends on the postoperative efforts initiated. Many CI-users achieve good speech understanding, but have poor perception of music and speech prosody. Shortly after implant switch-on, nine adult newly operated CI-users began weekly one-to-one musical ear training lessons that contained a variety of musical activities and listening exercises. To register progress in discrimination of pitch, rhythm and timbre, we created a battery of music tests. Participants in the music group significantly improved their discrimination of timbre, melodic contour, and rhythm, compared to a group of matched controls. Our results suggest that one-to-one musical ear training has a great potential as a complementary method to improve fine grained auditory skills in CI-users.

(2) Behavioral studies have demonstrated that the most dramatic progress in the restoration of hearing following cochlear implantation takes place in the first six months after the operation. With this study we aimed to study the neurologic processes that subserve this progress. We used positron emission tomography (PET) to assess regional cerebral blood flow immediately, three months, and

six months after implant switch-on. Fifteen newly operated adult implant recipients listened to either speech or speech-like noise, presented randomly in four sequential scan sessions at each milestone. Analyses of variance performed in SPM 8 showed a main effect of condition in auditory cortices bilaterally, regardless of implant side. Furthermore, main effect of time was found in Broca's area (BA45). The findings confirm the key role of Broca's area in restoration of speech perception, but only in individuals in whom Broca's area has been active prior to the loss of hearing.

(3) Prelingually deaf children who receive cochlear implants and intervention early can develop age-appropriate language skills. However, little is known about their music perception and the possible benefit of music training. Ten preschool CI-users participated in a three month music program and demonstrated higher scores than a control group in music and linguistic tests after the intervention period. Parental feedback indicated a positive impact on the children's musical interest and activity. The study's findings indicate that the proposed music program offers a stimulating environment and substantial listening practice, which may on a long term support the musical, linguistic, psychosocial, and personal development of these children.

8.2 Danish

Cochlear implantation er en behandling, der tilbydes børn og voksne med svært eller totalt høretab med henblik på at (gen)etablere hørelse og taleforståelse. Kirurgien og teknologien har oplevet en dramatisk udvikling gennem de sidste ti år, og på verdensplan bruger flere end 120.000 personer i dag CI i deres daglige kommunikation. Imidlertid har hverken rehabiliteringsmetoder der kan forbedre udbyttet af implantatet eller de neurologiske processer der betinger tilpasning til implantatet været genstand for forskning i tilsvarende omfang. I dette Ph. D.-forskningsprojekt har vi i tre forskellige langtidsstudier undersøgt udviklingen i de auditive færdigheder hos cochlear implanterede; adfærdsmæssigt (1, 3) ved at studere virkningen af musikalsk høretræning på musik-og taleopfattelsen hos voksne og børn med CI og; neurologisk (2) ved at måle udviklingen i hjerneområder associeret med lyd- og sprogopfattelse hos nyopererede voksne CI-brugere. (1)Vellykket rehabilitering af CI-patienter afhænger af den postoperative indsats. Mange CI-brugere opnår god taleforståelse, men har vanskeligt ved at opfatte musik og prosodi i tale. En gruppe på ni voksne nyopererede CI-brugere påbegyndte kort efter tilslutning et seks måneders individuelt høretræningsprogram med ugentlige lektioner, der indeholdt forskellige musikalske aktiviteter og lytteøvelser. For at kunne måle fremgangen i deltagernes musikalske skelneevne udviklede vi en række musikalske tests. Deltagerne i musikgruppen forbedrede deres evne til at skelne klangfarve, melodisk kontur og rytme signifikant i forhold til en kontrolgruppe. Resultaterne viser, at individuel musikalsk høretræning har et stort potentiale, som en metode der kan medvirke til at forbedre og forfine CI-brugeres auditive færdigheder.

(2) Undersøgelser har vist, at de mest dramatiske fremskridt i genetablering af hørelse efter cochlear implantation finder sted i de første seks måneder efter operationen. Med denne undersøgelse, ønskede vi at studere de neurologiske processer der betinger denne proces. Vi anvendte positron emission tomografi (PET) til at måle den regionale cerebrale blodgennemstrømning umiddelbart, 3 måneder og 6 måneder efter tilslutning af implantatet. Femten nyopererede voksne CI-brugere lyttede til henholdsvis tale eller talelignende støj, præsenteret i tilfældig rækkefølge ved fire sekventielle PET-målinger, på hvert af de tre tidspunkter. Analyser viste en signifikant aktivering af auditorisk korteks i såvel venstre som højre side betinget af stimulus, men uafhængigt af hvilken side operationen var udført i. Endvidere viste der sig en signifikant aktivering af Broca's område (Brodmann area 45) som en effekt af tid. Resultaterne understreger den centrale rolle, som Broca's område har i genetablering af taleforståelse, men kun hos personer hvor Broca's område har været aktivt inden de mistede hørelsen.

(3) Førsprogligt døve børn, som modtager cochlear implantater og sprogstimulering tidligt kan udvikle alderssvarende sprogfærdigheder. Meget lidt vides imidlertid om deres musikopfattelse og mulige udbytte af musikalsk træning. Ti førskolebørn med CI deltog i et tre måneders

musikprogram, der fokuserede på sangudfoldelse og melodiopfattelse og klarede sig bedre end en kontrolgruppe i musik- og sprogtests. En spørgeskemaundersøgelse blandt forældrene antydede endvidere, at programmet havde haft en positiv indvirkning på børnenes musikinteresse og –aktivitet. Undersøgelsens resultater indikerer, at musikaktiviteter rummer et stimulerende miljø og righoldig høretræning, som på lang sigt kan bidrage til disse børns musikalske, sproglige, psykosociale og personlige udvikling.

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10. CODA

The term “a jack of all trades, master of none” characterizes a person who is a generalist rather than a specialist. In Danish such a person is known as an “alt-muligmand” (all-possible man). An alt-muligmand background like mine appeared to be beneficial when launching into empirically based scientific research. In addition to my basic profession as a musician and music teacher, I found myself filling a variety of functions in carrying out this Ph. D.- project: composer, sound engineer, programmer, presenter, writer, secretary, mailman, diplomat, psychologist (lay), to name a few. Luckily, cortical plasticity in my brain helped me adapt quickly to these qualifications in demand.

“Life is understood backwards but experienced forwards”. The words of the Danish philosopher Soeren Kirkegaard (1843) certainly hold true for this author. Had I known when I launched into this project what I know now, many errors and detours could have been avoided. On the other hand; when engaging in pioneer projects like the present, mistakes will inevitably be made. Experience emerging from errors, mistakes and detours may be harshly earned, but not likely forgotten.

Engaging into this project has been incredibly satisfactory and fruitful. Playing a part, however small, in improving music perception for children and adults with a severe disability like hearing loss is a meaningful cause for me. Music is one of the core ingredients of human culture, and whether for a child getting acquainted with musical sounds or for an adult retrieving long lost musical memories, any contribution, regardless of magnitude, can be of value. I am privileged to have had the opportunity to make such a contribution.

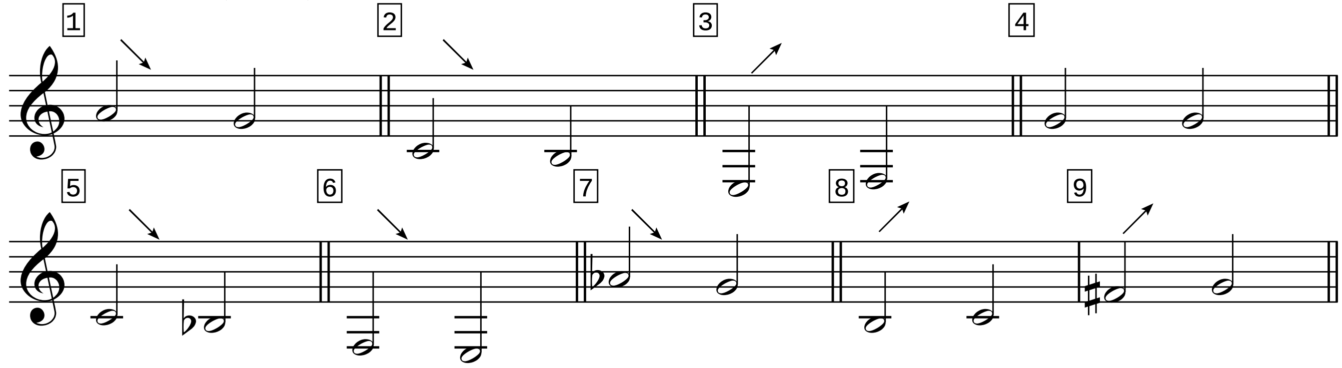
11. APPENDICES

Appendix 11.1 PR test trials.

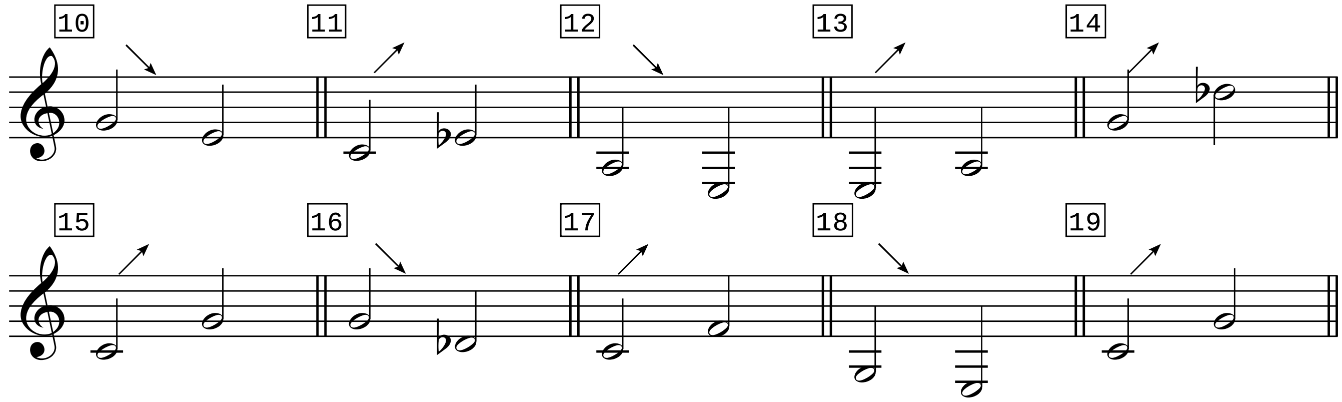
Pitch Ranking Test (PR)

B. Petersen 2008

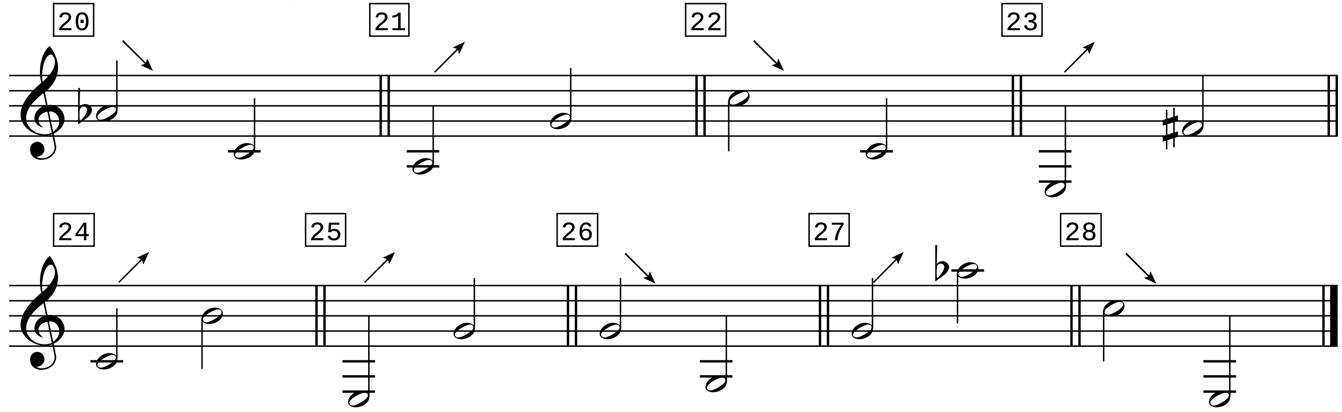
< 3 semitones (small)



3-7 semitones (medium)



> 9 semitones (large)



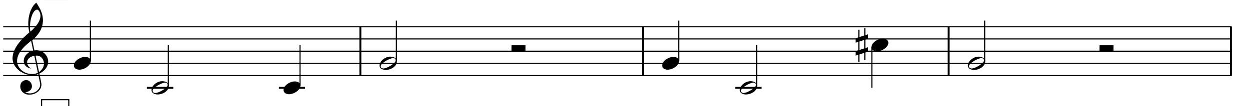
Appendix 11.2 MD test trials

Melodic discrimination trials - different trials

Organised by number of notes

B. Petersen 2008

1



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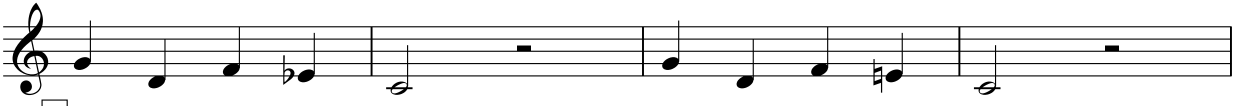

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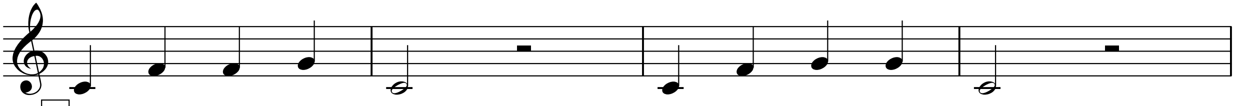

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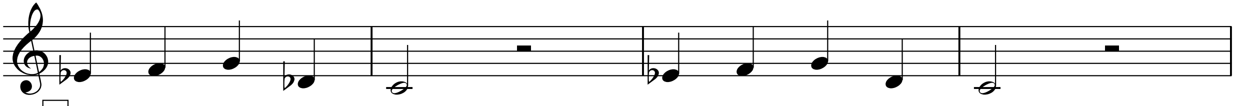

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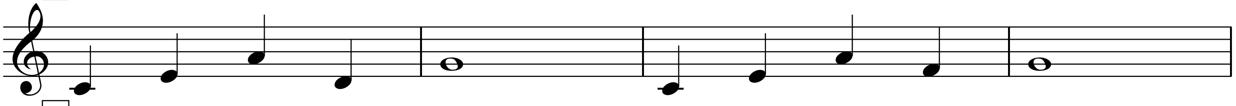

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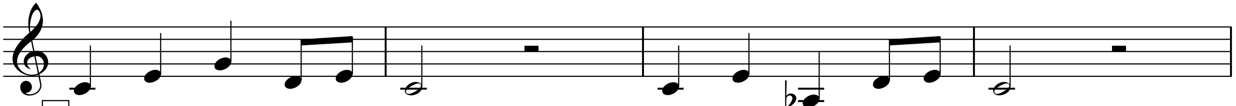

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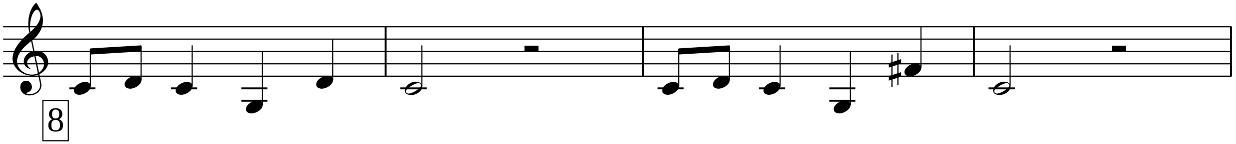

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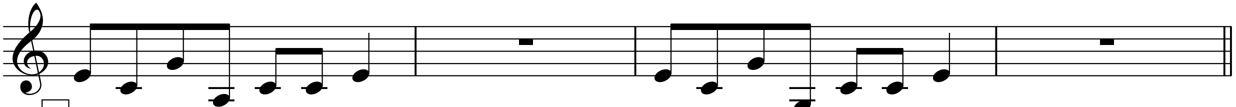

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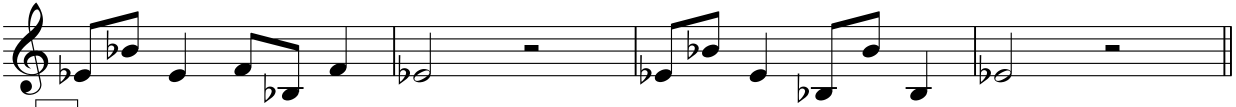

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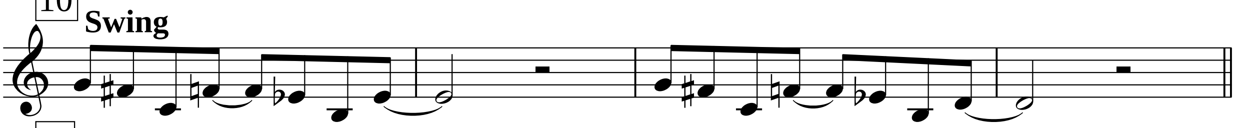
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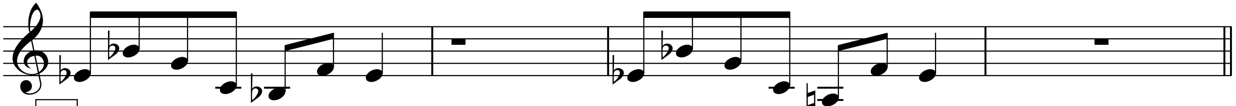

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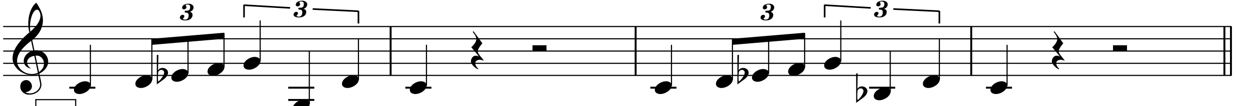

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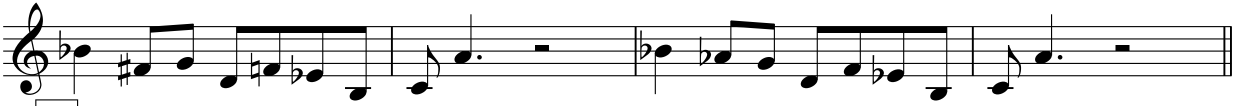

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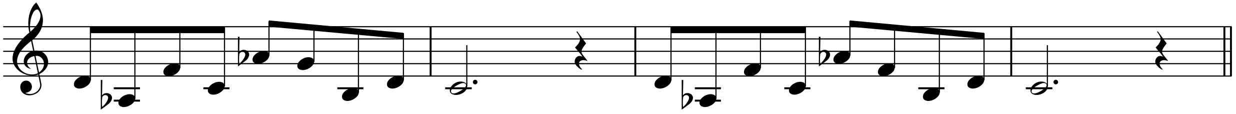
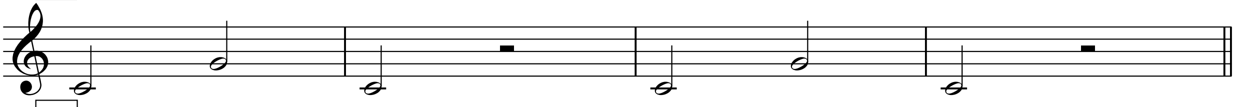


Appendix 11.2 MD test trials - 2

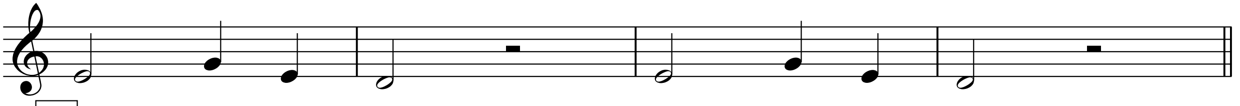
Melodic discrimination trials - same trials

Organised by number of notes

15



16



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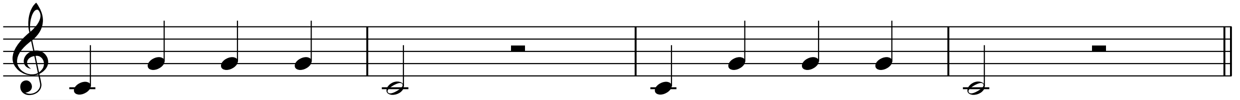

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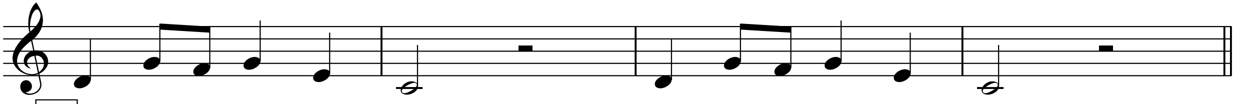

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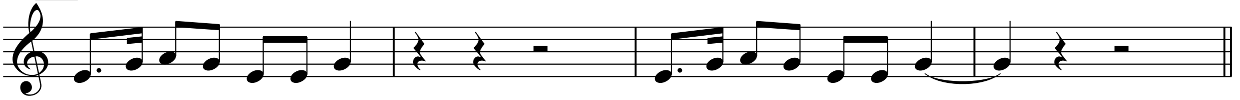

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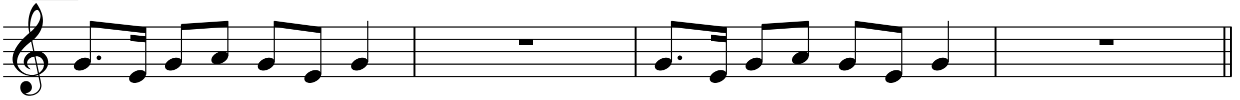

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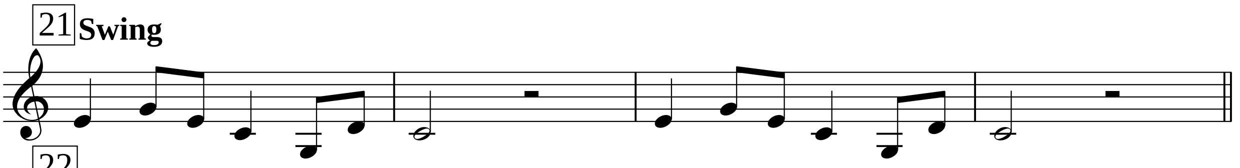

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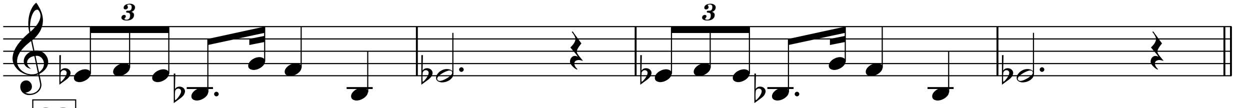

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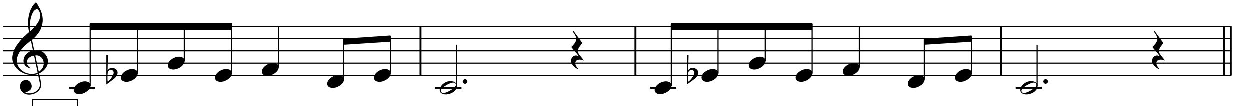

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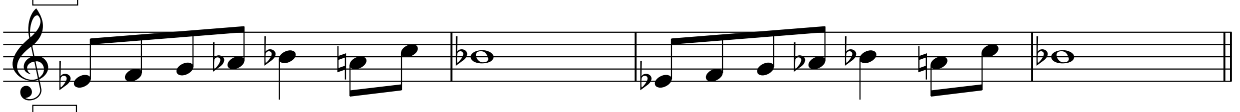

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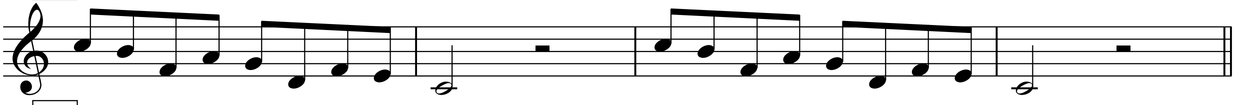
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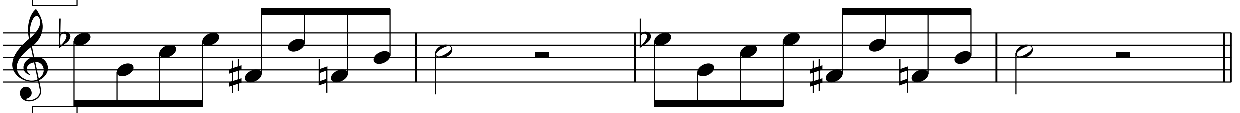
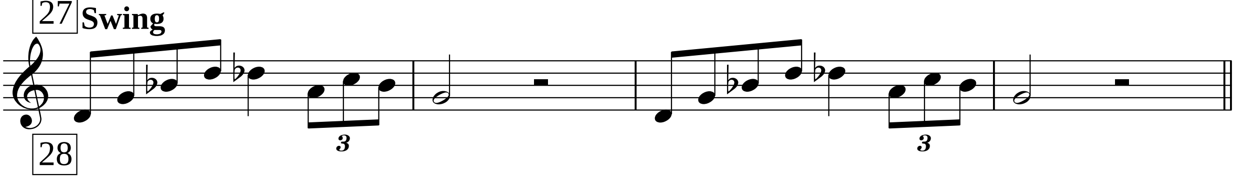


27



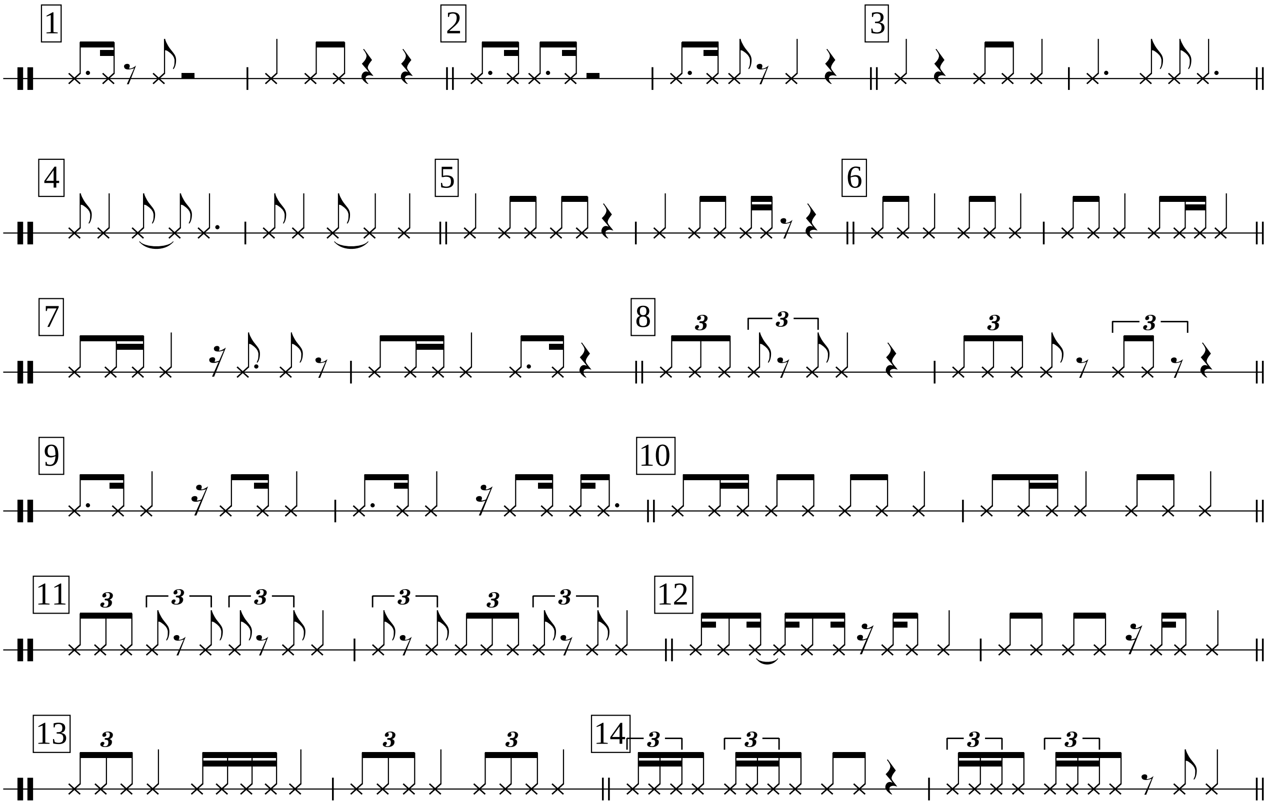
28

Appendix 11.3 RD test trials.

Rhythmic discrimination - different trials

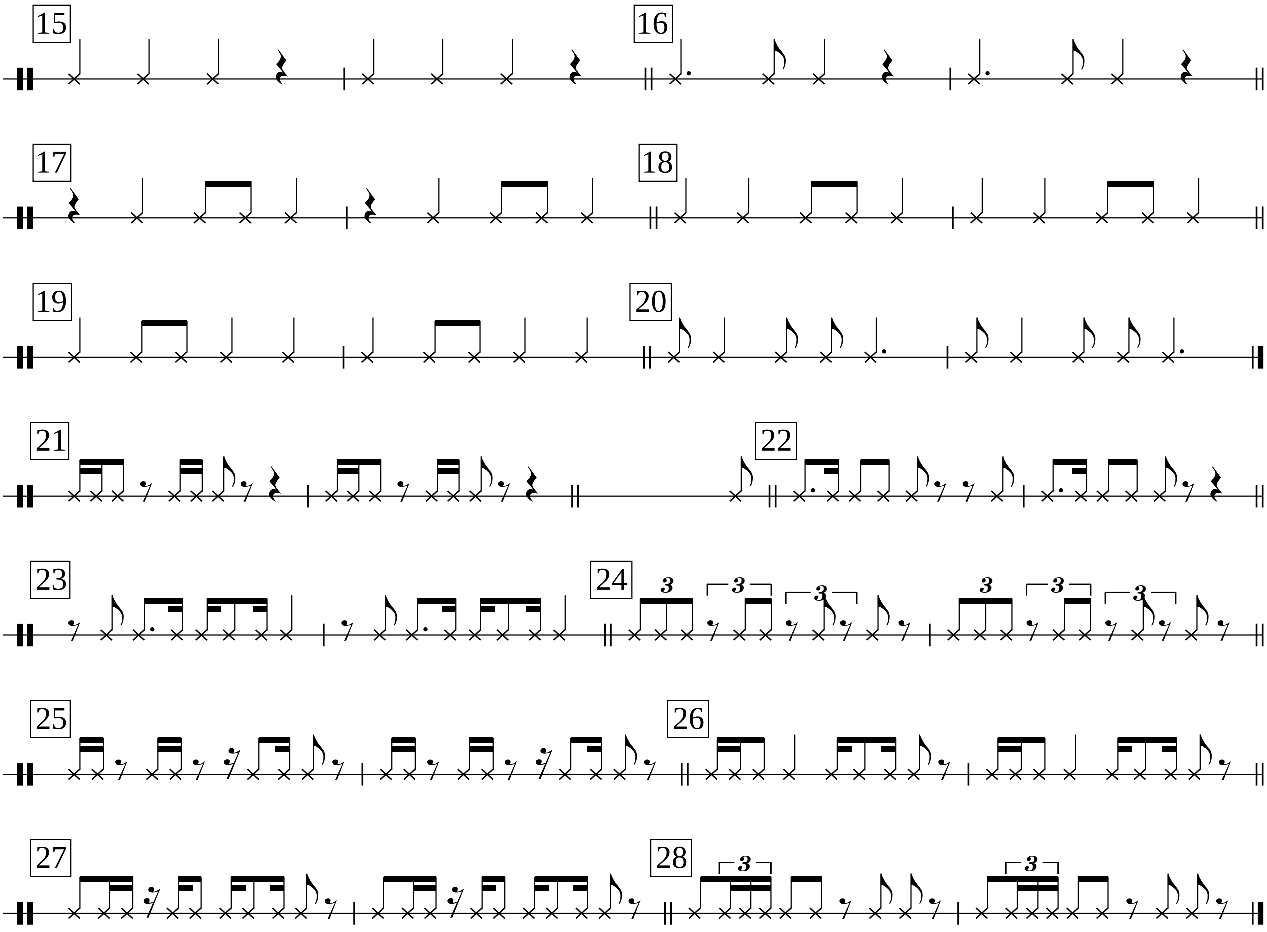
B. Petersen 2008

Organised by the number of beats



Rhythmic discrimination - same trials

Organised by the number of beats



Appendix 11.4. Melo M and Melo R score sheet

SVARARK MELO-M

MUSIKTEST FOR 3-5-ÅRIGE

MELO_M: **Melodisk** skelneevne

____

Testpersonens navn

____

Dato

____

Fødselsdag

	JA	NEJ	?
1	☐	☐	☐
2	☐	☐	☐
3	☐	☐	☐
4	☐	☐	☐
5	☐	☐	☐
6	☐	☐	☐
7	☐	☐	☐
8	☐	☐	☐
9	☐	☐	☐
10	☐	☐	☐

SVARARK MELO-R

MUSIKTEST FOR 3-5-ÅRIGE

MELO_R: **Rytmisk** skelneevne

____

Testpersonens navn

____

Dato

____

Fødselsdag

	JA	NEJ	?
1	☐	☐	☐
2	☐	☐	☐
3	☐	☐	☐
4	☐	☐	☐
5	☐	☐	☐
6	☐	☐	☐
7	☐	☐	☐
8	☐	☐	☐
9	☐	☐	☐
10	☐	☐	☐

MELO MUSIKTEST – SVARARK FOR MELODISK OG RYTMISK SKELNEEVNE

Singing in the key of life - musical ear training enhances musical abilities after cochlear implantation

B. Petersen[1,2], M.V. Mortensen[3], M. Hansen[1], P. Vuust[1,2]

1 Center of Functionally Integrative Neuroscience, Aarhus University Hospital, Aarhus, Denmark
2 Royal Academy of Music, Aarhus/Aalborg, Denmark
3 Department of Otolaryngology, Aarhus University Hospital, Aarhus, Denmark

ABSTRACT

Objectives: Cochlear implantation helps deaf people regain hearing abilities and many cochlear implant users achieve good speech understanding. While a few implant recipients enjoy music intensely despite the limitations in the sound signal of the implant, most users are disappointed with the music experience. We hypothesized that significant improvement of musical skills can be achieved, if active learning processes are initiated, and that these skills could enhance other auditory abilities such as perception of prosody. The objective of this study was to investigate the effect of one-to-one musical ear training on the perception of music, speech, and emotional prosody of newly implanted CI-users.
Design: Eighteen adult newly operated CI-users were assigned to a music group and a control group. Shortly after switch-on, participants in the music group began weekly one-to-one musical training sessions, involving active music making and listening exercises. For home practice, we provided computer assisted audio-visual training material. The control group received no musical training, but followed standard aural therapy. To measure the progress in musical discrimination skills, we created a battery of music tests. To measure linguistic progress we used the Hagerman test and an emotional prosody recognition test. Testing took place at three different points: immediately after switch-on of the implant, after 3 months, and after 6 months. A group of age matched normal-hearing participants provided reference performance levels.
Results: The music group significantly improved their overall music perception compared to the control group. Discrimination of timbre, pitch, and melodic contour improved in particular whereas rhythm discrimination equaled normal hearing levels. Both groups showed remarkable improvement in average speech perception performance, but no significant difference was found between groups post-training. In contrast to the control group, the music group showed an earlier on-set of progress in recognition of emotional prosody, while endpoint performances were comparable. A significant relationship was found between overall music performance and speech perception across all participants at all milestones. In general, average implant user performance was significantly poorer than the normal hearing reference, even after training. Finally, correlation analyses showed that duration of deafness had significant negative effect on speech perception performance, while age was predictive of the overall music discrimination progress.
Conclusion: Our results indicate that one-to-one musical ear training has a great potential as an effective method to improve overall music perception in CI-users. In particular perception of timbre, pitch and rhythm may be positively affected. Perception of speech may not necessarily benefit from musical ear training, while development of emotional prosody recognition may develop faster by musical ear training. All participants completed the program and showed great enthusiasm for the musical ear training, especially singing-related activities, indicating that such a program may also serve a motivational purpose in rehabilitation of cochlear implantees. If implemented in aural/oral rehabilitation therapy, the proposed musical ear training program could form a valuable complementary method of auditory rehabilitation, and may, on the long term, contribute to an improved general quality of life in CI-users.

Introduction

Cochlear implantation is a surgical treatment, which effectively provides hearing sensation and speech recognition in individuals with pre- or postlingual hearing loss (Friesen et al. 2001; Wilson & Dorman 2007). The number of CI-listeners is rapidly growing, with over 120,000 users worldwide. The variability in implant outcome is very large, and important predictors for a good result are short duration of deafness prior to implantation and some residual hearing (Summerfield & Marshall 1995; Waltzman et al. 1995). Because music, in many cases, has been an essential part of these patients' cultural and social life, CI-candidates' hope of retrieving music enjoyment is an important reason for choosing this treatment (Gfeller et al. 2000b). Despite the fact that the implant is designed primarily to promote speech perception, and therefore does not transmit the complex sound of music satisfactorily, some users seem to enjoy music intensely (Gfeller & Lansing 1991; Gfeller et al. 2005).

However, for many CI-users, the music experience is disappointing (Mirza et al. 2003). Moreover, several studies have concluded that recognition of melody and timbre is significantly poorer in CI- users than in normally hearing (NH) controls (Gfeller et al. 2002a; Leal et al. 2003; McDermott 2004; Kong et al. 2004; Gfeller et al. 2005; Olszewski et al. 2005; Gfeller et al. 2007). Some reports suggest a possible correlation between successful music outcome and factors such as musical background, musical exposure and residual hearing (Mirza et al. 2003; Gfeller et al. 2006; Gfeller et al. 2008). Furthermore, studies involving computer-assisted music training have demonstrated significant improvement of discrimination of melodic contour, musical timbre, and recognition and appraisal of songs (Gfeller et al. 2000a; Gfeller et al. 2002c; Galvin et al. 2007). This indicates that the possibility of overcoming the limitations of the implant and develop improved musical pattern recognition, relies on "active" learning efforts (Fu & Galvin III 2008). So far, no data are at hand concerning the effects of longitudinal musical ear training based on personal tuition in CI users.

Improved perception of music may have considerable positive implications not only for music enjoyment, but also for other aspects of listening. Music training in normal listeners is beneficial for the development of specific auditory skills, such as pitch, timing, and timbre, also involved in language comprehension (Naatanen et al. 1978; Pantev et al. 1998; Koelsch et al. 1999; Naatanen et al. 2001; Vuust et al. 2005; Altenmuller 2008). Enhanced music abilities may enhance speech perception in noisy surroundings, which relies on pitch cues to separate the target from the background (Qin & Oxenham 2003), and the ability to identify voice gender and speaker, which largely depends on discrimination of timbral cues (Vongphoe & Zeng 2005). Recent brain imaging studies have shown that complex music tasks activate brain areas associated with language processing (Levitin & Menon 2003; Vuust et al. 2006,Vuust and Roepstorff, 2008). Thus, musical training of CI-users may be hypothesized to influence speech perception.

Since an important function of music is to convey emotion (Juslin & Laukka 2003), musical training has especially been suggested as a way to enhance processing of the emotional aspects of language, which are mediated by loudness, speech rate, and pitch contour. Thompson et al. (2003) found that musically trained participants outperformed untrained participants in extracting prosodic cues in speech, and Besson et al. (2007) found that musical training of 9-11 year olds enhanced their ability to perceive prosodic cues compared to a control group receiving drama lessons. Therefore we also expected musical training to enhance emotional processing of speech in CI-users.

The aim of the present study was to develop a one-to-one musical ear training program, targeted at newly operated adult CI-users. We hypothesized that weekly training, involving active music making and listening exercises, would substantially enhance the musical discrimination skills of the participants, compared to a group of controls. We also hypothesized that the possible enhanced discrimination skills could generalize to the linguistic domain, and positively affect the CI-users' recognition of speech and emotional prosody. To measure progress of the participants, we developed a music test battery targeted at CI-users, which objectively and effectively would measure a broad range of music-related perceptual skills.

Materials and Methods

Ethical approval: The study was conducted in accordance with the Helsinki declaration and approved by the Research Ethics Committee of the Midlands Province of Denmark by Sept. 2008. Informed consent was obtained from all participants.

Participants: Over the course of 2 years, patients who were approved for transplantation were contacted by mail and invited to take part in the research project. Of a total of 41 patients 18 replied positively and were assigned to either a music group (MG: 6F 3M, 21.4-70.3 years of age M=46.7 SD 16) or a control group (CG: 4F 5M, 44.9-73.3 years of age M 58.6 SD 10.6), matched according to duration of deafness, musical background and, equally important, availability for the weekly music training sessions. The difference in the mean age of the two groups was not statistically significant ($p > 0.05$). According to Danish practice, all participants were unilaterally implanted. Five MG participants and 6 CG participants were bimodally aided with a hearing aid (HA) in the non-implanted ear. Table 1 lists the demographic and clinical data for the 18 participants.

Musical background: To account for past training and experience, all participants filled out a questionnaire concerning their musical background. None of the participants had a professional musical background, or any formal music instruction beyond grammar school.

Normal hearing reference. To obtain a reference for the complete test battery, we recruited a group of NH adults (4F, 2M; 47-64 years of age, M=54.29; SD 7.55) for a one-time test session. All NH participants met the criteria for normal hearing, by passing a full audiometric test.

Design: Immediately after switch-on, the participants were tested for speech and music perception (baseline). Subsequently, the participants received either musical ear training (MG), or no musical ear training (CG). Test procedures were repeated after three months (midpoint) and six months (endpoint), respectively. After three months of musical ear training, which focused on pitch-related tasks, rhythm training was introduced and added to the existing training program. This strategy was applied to make the training more manageable for the MG, while at the same time allowing for a comparison between training and no training, and possible effects of rhythmic training. All participants followed standard auditory/oral therapy for six months, in parallel with the music training study.

Pilot training and testing.

Four CI-users (3 M 1 F, 21-48 years of age M 34.75 mean implant experience 1.14 y) acted as pilot participants in five individual musical training and testing sessions but did not take part in the actual training program. The experience drawn from these sessions formed the framework, on which we created the musical training program, the musical test battery, and the musical background questionnaire.

Table 1

Participant (gender)	Age at project start (y)	Etiology of deafness	Side of implant	Contra-lateral use of HA	On-set of hearing loss (y)	Duration of hearing loss (y)	[d]Degree of deafness (1-5)	Implant type	CI sound processor	CI sound processing strategy
Music group (MG)										
MG1 (F)	49.8	[a]Cong. non spec.	R		4	45.8	5	[e]Nucleus	Freedom	ACE 900
MG2 (F)	21.4	Ototoxic	R	X	0.7	20.7	5	Nucleus	Freedom	ACE 250
MG3 (M)	31.7	Meningitis	L	X	1.8	30.2	4.5	Nucleus	Freedom	ACE 900
MG4 (M)	56.0	Cong. non spec.	R	X	8	48.0	4.5	Nucleus	Freedom	ACE 1800
MG5 (F)	70.3	Cong. non spec.	R		40	30.3	4.5	Nucleus	Freedom	ACE 900
MG6 (F)	47.5	Unknown	L		30	10.5	4.5	Nucleus	Freedom	ACE 1200
MG7 (F)	56.2	[b]Hered. non spec.	R	X	19	37.6	4.5	Nucleus	Freedom	ACE 1200
MG8 (M)	58.5	Meningitis	R	X	5	53.5	5	Nucleus	Freedom	ACE 900
MG9 (F)	29.1	[c]Mon	L		10	19.1	4.5	Nucleus	Freedom	ACE 1200
Mean	**46.7**					**32.8**	**4.7**			
Control group (CG)										
CG1 (F)	44.8	Unknown	R	X	35	9.8	4.5	Nucleus	Freedom	ACE 1200
CG2 (M)	60.4	Unknown	L	X	40	16.4	4	Nucleus	Freedom	ACE 900
CG3 (F)	50.6	Cong. non spec.	R		5	47.6	5	[f]A.B.	Harmony	Fid. 120
CG4 (M)	63.5	Cong. non spec.	L	X	6	57.5	5	Nucleus	Freedom	ACE 500
CG5 (F)	63.0	Unknown	R	X	58	5.0	4	Nucleus	Freedom	ACE 720
CG6 (F)	45.8	Her. non spec.	R	X	4	41.8	5	Nucleus	Freedom	ACE 900
CG7 (M)	72.5	Unknown	R		41	21.5	4	Nucleus	Freedom	ACE 1200
CG8 (M)	53.7	Cong. non spec.	L	X	5	48.7	5	Nucleus	Freedom	ACE 500
CG9 (M)	73.3	Trauma	R		54	19.3	4	Nucleus	CP 810	ACE 720
Mean	**58.6**					**29.7**	**4.5**			

Table 1: Clinical and demographic data of the 18 participants included in the study. [a]Non specified congenital hearing loss, [b]non specified hereditary hearing loss, [c]Mondini dysplasia. [d]1: Mild (24-40 dB HL), 2: Moderate 40-55 dB HL), 3: Moderately-Severe (55-70dB HL), 4: Severe (70-90 dB HL, 5 Profound (>90 dB HL). [e]Cochlear ® [f]Advanced Bionics ®. Five participants in the MG and 6 participants in the CG used a HA in their contralateral ear on a daily basis.

The musical ear training program.

Participants who were assigned to the MG committed themselves to weekly 1-hour music training sessions and home-practice for a period of six months. Training took place in a rehearsal room at the Royal Academy of Music in Aarhus. The room was acoustically dampened and well isolated from external noise. The intention of the program was to provide active and passive experiences with (1) pitch, (2) rhythm, and (3) timbre.

(1) Pitch-related training aimed at developing a sense of basic musical attributes such as high/low, up/down, and far/close. This was facilitated by:

- *Singing:* The participants were required to vocalize and imitate short phrases with a range of vowels such as *ooh, ah,* and *eeh*, and to sing well-known Danish songs of their own choice. Emphasis was put on rhythmic precision, articulation of the lyrics, and intonation.
- *Playing:* The participants were required to imitate short phrases and to play well-known folk and children's songs with a limited range of notes [c4 (261.6 Hz) to g4 (392 Hz)] on the piano. For home practice the participants were provided with an electronic keyboard (Yamaha PSR-E313, Yamaha Corporation, Japan).
- *Listening:* For home training two different audio-visual applications were installed on the participants' personal computers. a) Piano renditions of 7 simple songs in the key of C major, identical to those used for the playing exercises. The sound was accompanied by the songs in standard music notation. b) Five-note melodic sequences with different contours and pitch distances in different frequency ranges. In both applications, sound clips were activated by clicking corresponding pictorial representations on the screen. On a final "quiz"-page, the participant had the opportunity to test his or her identification of the melodies/contours.

(2) Rhythm-related training aimed at strengthening perception of basic features of rhythm such as pulse/meter, beat/subdivision, fast/slow, and weak/strong. This was facilitated by:

- *Drumming:* The participants were required to replicate rhythmic patterns either by clapping, tapping, or drumming, putting specific focus on accentuation of beats and dynamic expression.
- *Energizing:* The participants practiced the rhythm of specific melodies by lyrics and rhythm only, sometimes accompanied by drumming.
- *Listening:* Computer applications similar to those described above were provided. a) a quiz game, which required matching of a rhythm pattern with a pictorial representation of a rhythm. b) two training applications, presenting series of patterns with increasing difficulty for imitation exercises.

(3) Timbre-related training aimed at improving the distinction between light/dark, attack/decay, and hard/soft in the quality of the tone of different instruments. This was facilitated by the means of a computer application, which presented flute, clarinet, trumpet, trombone, double bass, guitar, violin, cello, and accordion playing two different melodies. The participants were able to measure their progress by recording their answers on a sheet and obtain the score at the sessions.

Musical test battery.

To assess the development of the participants' musical discrimination skills, we created a battery consisting of five music tests: (1) musical instrument identification (MII), (2) melodic contour identification (MCI), (3) pitch ranking (PR), (4) rhythmic discrimination (RD) and (5) melodic discrimination (MD).

(1) The MII test required the participant to identify the instrument playing randomly presented parts (A, B, C, or D) of a well-known Danish children's song ("Mariehoenen Evigglad"/ Ladybug Ever Happy, Fig. 1). Instruments were presented in random order. To take into account the presumed differences in baseline preconditions, the test was divided into two subtests: MII.1 (easy) and MII.2 (difficult).

MII.1 was a 3-alternative forced-choice test including three instruments from different instrument families: flute (woodwind), piano (pitched percussion), and double bass (string). Prior to MII.1 the participant was presented with each instrument playing melody part A (Fig. 1), while corresponding icons were shown on the computer screen. Familiarity with the musical instruments was not required.

Figure 1

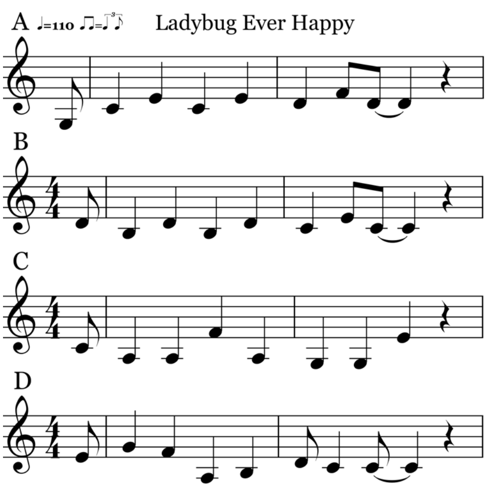
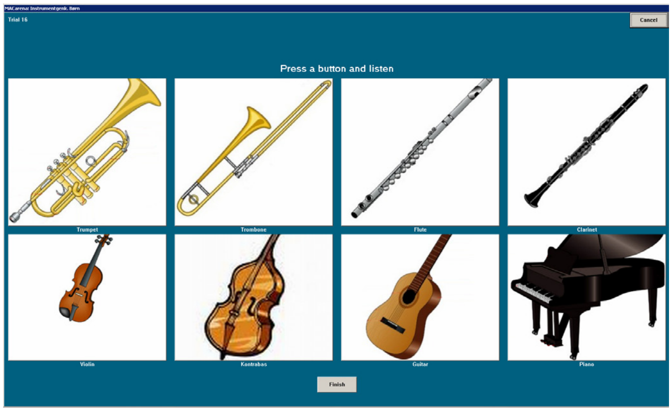



Figure 1: "Ladybug Ever Happy" in musical notation. The melody comprises an octave from G3 (196 Hz) to G4 (392 Hz). Letters A–D indicate the four parts presented randomly in the test.

Figure 2


MACarena: Instrumentgenk. Børn

Trial 16

Press a button and listen

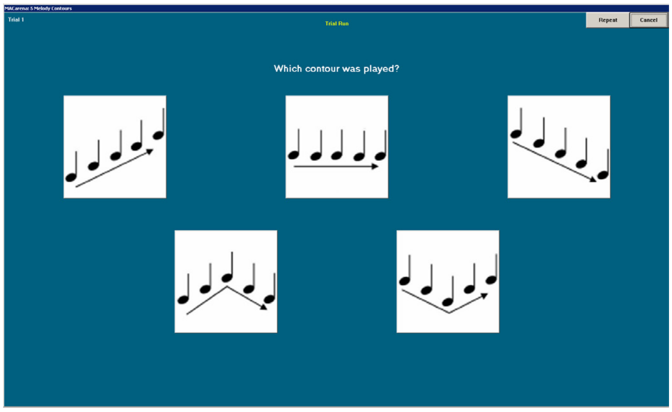
Figure 2: The MII 2 computer screen illustrating the 8 alternative choices.

MII.2 was an 8-alternative forced-choice test including the instruments from MII.1, together with clarinet (woodwind), violin (string), trumpet (brass), trombone (brass), and guitar (string). Prior to this test the five new instruments were presented.

The division of the melody into four parts used in the MII test was inspired by The Zurich Music Test Battery (Büchler 2008). The random presentation of phrases ensures that the instrument identification is not associated with a single melodic feature.

(2) The MCI test required the participant to identify the melodic contour of a 5-note sequence: 1) rising, 2) falling, 3) flat, 4) rising-falling or 5) falling-rising (Fig. 3). The sequences were played with the timbre of a modified digital sampling of a clarinet (3-tone complex) diatonically in the key of A major, ranging from A3 (220Hz) to E4 (329,6Hz). Contours were presented in random order with each variant appearing twice, thus adding up to a total of 10 trials. No feedback was given during the test. The MCI test is an existing part of the Zurich Music Test Battery (Büchler, M. 2008) originally adapted from Galvin et al. (2007).

Figure 3


MACarena: 5 Melody Contours

Trial 1

Trial Run

Which contour was played?

Figure 3: The MCI computer screen with pictorial representations of the 5 contours.

(3) The PR test consisted of 28 trials, which required the participant to judge which of two tones was the higher. The tones were presented in the range between E3 (164.8 Hz) and G#5 (830.6Hz). Note distance was divided in 3 categories: small

1-2 semitones (ST); N=9, medium 3-7 STs; N=10, and large >7 STs; N=9. Half of the trials were descending, half ascending. The tones had a rhythmic value of a half note and played back at 85 beats per minute (BPM), equaling tone duration of 1400 ms. To create a relation to the training practice and a real-life listening reference, we used a grand piano sampling for the timbre.

(4) The **MD** test was a same/different test presenting 28 pairs of melodic 1-bar phrases in 4/4 time, played at 100 BMP. Half of the trials were *same* trials and half were *different*. In an attempt to avoid floor/ceiling effects the phrases were of varying complexity (3-10 tones) and difficulty. The MD phrases were played by pure tones in the pitch range from G3 (196 Hz) to D#5 (622.3 Hz). The *different* trials contained a violation of pitch. In five of these, the violation also constituted a violation of contour. In nine trials, the deviant note was diatonic (within the scale), and in five trials the deviant note was non-diatonic (outside the scale). The deviant note was either 1-2 STs (N=9), 3-7 STs (N=4), or > 7 STs (N=3) away from the standard. Thirteen of the 28 melodies were in the major key, 11 in the minor key, and 4 were neither (i.e. contained no determining major or minor third). To make the test as varied as possible, the melodies represented a variety of musical styles (fig. 4).

(5) The **RD** test was a same/different test presenting 28 pairs of rhythmic 1-bar phrases in 4/4 time played at 100 BMP, half of which were *same*, half *different*. The RD phrases used the sampled sound of a cowbell for the first part (call) and the sound of a woodblock for the second (response). Twenty-five trials began on the downbeat, 2 began later and 1 began earlier (upbeat). Eleven trials contained syncopated beats and 6 trials contained triplets. The different patterns contained a violation of the rhythm. Five patterns were violated by a delayed beat, four by an anticipated beat, and five were violated by addition or omission of a beat. Beats were delayed or anticipated by 1/8- or 1/16-notes, which equals 300 and 150 ms respectively. A 4-beat metronome count-in preceded all RD trials (Fig. 4).

Test procedure

At all occasions of the MCI, PR, MD and RD tests the participants were given two example trials with feedback prior to the actual test, while at the same time looking at the corresponding pictorial representations on the computer screen. Preceding the MD test, the participants were informed that they were required to detect possible changes in the melodic content and that the rhythmic content was identical in all melodic pairs. Similarly, in the RD test, participants were carefully prepared for the three different sounds of the count-in, the "call", and the "response", and informed that they were required to distinguish between the 2nd and the 3rd pattern.

All tests in the music battery were presented in the computerized test environment MACarena (Lai 2000), played back on a laptop computer through a small active loudspeaker (Fostex 6301B, Fostex Company, Japan) placed in front of the participant. The stimuli were presented at 65dB sound pressure level (SPL), and CI users were instructed to adjust their processors to a comfortable loudness level. The stimuli were presented in random order and the test examiner registered the participants' responses by clicking corresponding pictorial representations on the computer screen. One person administered all musical tests. Participants used their preferred processor settings during the entire test session. Participants who continued to use a contralateral hearing aid (HA) were allowed to use it during training, whereas all tests were performed by CI only.

Linguistic tests

We assessed the participants' linguistic progress by two different tests: (1) the Hagerman speech perception test (HAG) and (2) an emotional prosody recognition test (EPR).

(1) **HAG** is an open-set test, which presents sentences organized in lists of ten in background noise. The sentences have identical name-verb-number-adjective-noun structures such as "Peter buys five red flowers", which the participant is required to repeat. Scores are registered as number of correctly repeated words. Since all participants were included immediately after switch-on, and therefore had no implant experience, we chose to perform the test without the standard background noise. The participants were given one example list with feedback and two trial lists without feedback (max. score=100 pts.). To reduce the risk of learning effect, we used different lists at the three times of testing. Sound was played back at the most comfortable hearing level.

Figure 4

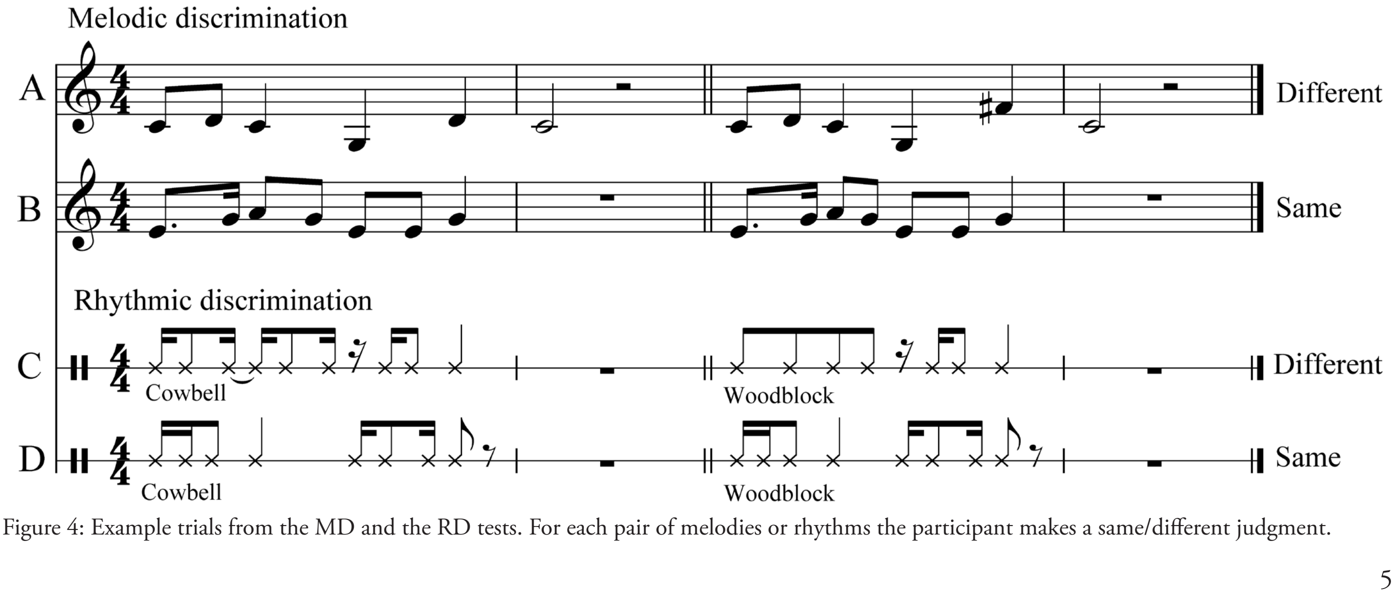


Figure 4: Example trials from the MD and the RD tests. For each pair of melodies or rhythms the participant makes a same/different judgment.

(2) The **EPR** test required the participant to judge from the prosodic content of 44 different spoken words and sentences, whether they expressed a sad or happy emotion. The EPR trials were taken from the Danish Emotional Speech Database (DES) (Engberg & Hansen 1996), which holds words and sentences in Danish, spoken by two female (32y; 52 y) and two male (32y; 52y) actors in five different emotions. The 44 EPR trials were compiled from the total of 88 happy and sad samples and balanced on emotion (22 happy, 22 sad), speaker gender, and age. Single words were "yes" and "no", while sentences were every day utterances. The trials were presented in random order, using the same method as in the music tests. Prior to the test, participants were given two example trials with feedback.

Software used: Training applications were programmed as slideshows in MS Powerpoint ®, Microsoft Corp. USA. PR tone pairs and MII, MD, and RD phrases were programmed in Cubase 4.1 (Steinberg Media Technologies GmbH, Hamburg, Germany), using musical instrument digital interface (MIDI). MD pure tones were produced in Audacity (http://audacity.sourceforge.net), and played back by the software sampler Halion 2.0 (Steinberg). The PR piano sound and the MII musical instrument sounds were high quality samplings taken from the library found in Halion 1.

Statistical methods: All music and linguistic test scores were recorded as the percentage of correctly answered items (0-100%). Data were analyzed and plotted with Sigmaplot for Windows 11.0 (Systat Software Inc) and NCSS (Hintze 2009). Paired t-tests were performed to compare within-group results, and t-tests were performed to compare results between groups. Variables with non-normal distribution were compared using the Wilcoxon/Mann-Whitney U-test. To calculate an overall music score for each participant at each point, we standardized, summed, and averaged the raw performance and gain scores from the six music tests at the three points of testing. The mean of these scores is in the following referred to as overall music z-scores and overall music gain z-scores. Pearson correlation analyses were performed with raw scores, z-scores, and background variables across all participants as well as within groups.

Figure 5

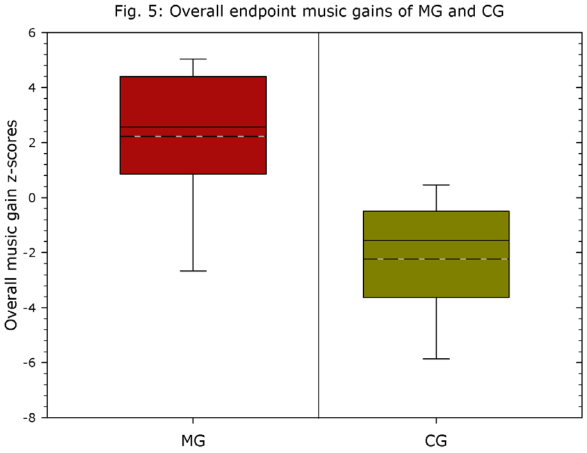


Figure 5: Box plot of overall music gain z-scores for MG and CG. Error bars show 10th/90th percentile. Solid box lines: median. Dotted box lines: mean.

Figure 6

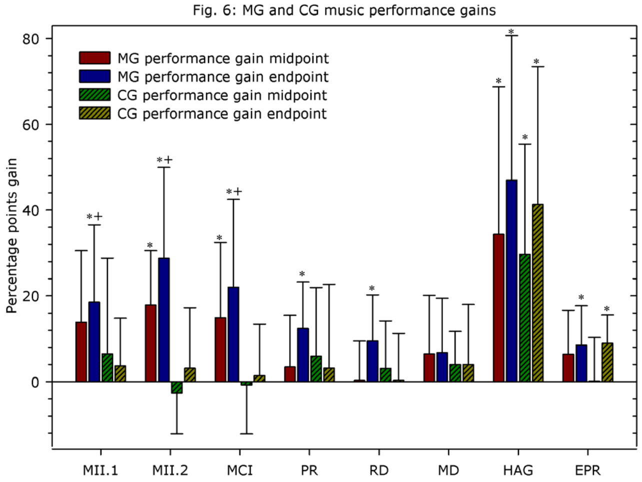


Figure 6: Bar graph showing gains of the music (MG) and the control group (CG) from baseline to midpoint and baseline to endpoint, in all 8 discrimination and identification tasks. Error bars show SD. * = gain is statistically significant (p < 0.05). + = gain is significantly larger than the CG's gain (p < 0.05).

Results

In the following, performance gain refers to the increase in mean scores from baseline to endpoint. Performance scores refer to absolute mean values.

Musical skills: The overall music gain z-scores of MG were significantly higher than those of CG ($p < 0.001$) (Fig. 5).

The MG MII.1 and MII.2 performance gain was significant (MII.1: +18.5 percentage points (pp); $p = 0.015$; MII.2: +28.8 pp; $p = 0.01$). Furthermore, MG MII.1 and MII.2 performance gain was significantly larger than that of CG (MII.1: $p = 0.05$; MII.2: $p = 0.02$) (Fig. 6). The CG scored significantly below the mean NH level in both subtests ($p < 0.05$). Ceiling performance (100% correct) was observed in one NH participant in the MII.2 subtest, and in the endpoint scores of seven MG, four CG, and all NH participants in the MII.1 subtest. Due to very low MII.1 baseline scores, two participants in the MG and two participants in the CG did not perform the MII.2.

MG MCI performance gain was significant (+22 pp; $p = 0.012$). Furthermore, MG MCI performance gain was significantly larger than that of the CG ($p = 0.019$) (Fig. 6). MG scores were significantly higher than those of the CG (Mann-Whitney: $p = 0.002$). Both groups scored significantly below the NH level ($p < 0.05$). Ceiling performance (100% correct) was observed in one of nine MG participants and four of six NH participants (Fig. 7).

MG PR performance gain was significant (+12.4 pp; $p = 0.009$), whereas CG PR performance was not (+ 3.18 pp; $p = 0.64$). Although mean performance gain was larger for the MG than for the CG, the difference was non-significant ($p = 0.231$) (Fig. 6). Both groups scored significantly below the NH level ($p < 0.05$) (Fig. 7). An analysis of variance showed that MG endpoint performance was significantly better on pitch pairs with large note distance than pairs with small pitch distance (p = 0.016, Bonferroni), whereas no effect of note distance was found in the CG endpoint PR performance.

MG MD performance gain (+6.8 pp) was non-significant, as was that of the CG (+4 pp). The difference in gain between the MG and the CG was non-significant (Fig. 6). Both groups scored significantly below the NH level ($p < 0.05$) (Fig. 7).

MG RD performance gain was significant (+9.5 pp; p = 0.009), whereas that of the CG was not (+ 0.328 pp; p = 0.93). The difference in mean performance gain between the MG and the CG approached significance (p = 0.09) (Fig. 6). MG RD performance scores were significantly higher than those of the CG (p = 0.015) and non-significantly higher than the NH scores. (Fig. 7).

Linguistic skills: MG HAG performance improved significantly (+47 pp; p = 0.003), as did that of the CG (+41.6 pp; p = 0.015). The difference in HAG performance gain between MG and CG was non-significant ($p > 0.05$) (Fig. 6). Both groups scored significantly below the NH level. Ceiling performance (100% correct) was observed in one MG participant and in all NH participants. Four MG participants and two CG participants scored within the 90th percentile at endpoint. There was considerable variability in performance in both groups (MG range: 93 (7-100), CG range: 95 (3-98) (Fig. 6). To maintain a true comparison between the groups, two CG participants were excluded from the HAG data analysis, due to 0 point scores at all three points of testing.

MG EPR performance improved significantly (+8.5 pp; $p < 0.05$), as did CG performance (+8.9 pp; $p < 0.05$). The difference in endpoint performance gain between MG and CG was non-significant (Fig. 6). The main progress of MG took place from baseline to midpoint (+6.4 pp; p = 0.09), whereas the main progress of CG took place from midpoint to endpoint (+ 8.8 pp; p = 0.06). Both groups scored well above chance levels, but significantly below the NH level ($p < 0.05$) (Fig. 7). Ceiling performance (100% correct) was observed in one NH participant.

Correlations

Relationship between music and speech perception

Significant relationships were found between overall music z-scores and HAG scores at all 3 points of measurement, across all participants (Fig. 8). Furthermore, a significant relationship was found between overall music z-scores and EPR scores of the MG at mid- and endpoint (Table 2). Other correlations between overall music z-scores and HAG and EPR across participants and within groups vary in strength (Table 2).

Figure 7

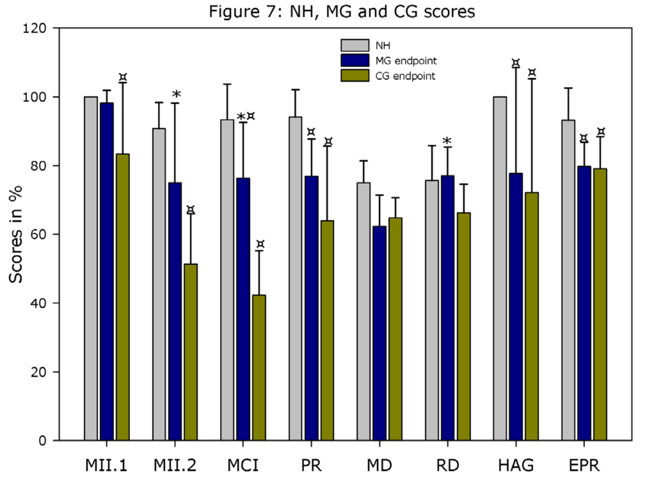


Figure 7: Bar chart, showing normal hearing (NH), music (MG) and control group (CG) endpoint scores in all 8 music and linguistic tests. Black bars = NH mean score. Error bars = standard deviation. * = the final score is significantly higher than the CG final score. ¤ = The score is significantly lower than the NH performance level ($p < 0.05$).

Table 2

Table 2. Correlations between overall music z-scores and HAG/EPR

	Baseline				**Midpoint**				**Endpoint**			
	HAG		EPR		HAG		EPR		HAG		EPR	
	r	p	r	p	r	p	r	p	r	P	r	p
Music group (MG)												
EPR	0.35	0.36	-	-	**0.72**	**0.03**	-	-	**0.76**	**0.02**	-	-
Overall music z-scores	0.59	0.10	0.13	0.74	0.59	0.09	**0.66**	**0.05**	0.62	0.08	**0.66**	**0.05**
Control group (CG)												
EPR	0.48	0.28	-	-	-0.15	0.74	-	-	-0.24	0.60	-	-
Overall music z-scores	0.48	0.28	0.01	0.97	0.57	0.19	-0.04	0.92	0.86	0.01	0.04	0.92
MG and CG pooled												
EPR	0.34	0.19	-	-	0.19	0.49	-	-	0.22	0.42	-	-
Overall music z-scores	**0.55**	**0.03**	0.09	0.72	**0.59**	**0.02**	0.34	0.17	**0.61**	**0.01**	0.26	0.30

Table 2. Correlations between overall music z-scores and Hagerman speech perception and emotional prosody recognition scores respectively in the MG, CG, and across all participants. Statistically significant values are indicated in bold.

Figure 8

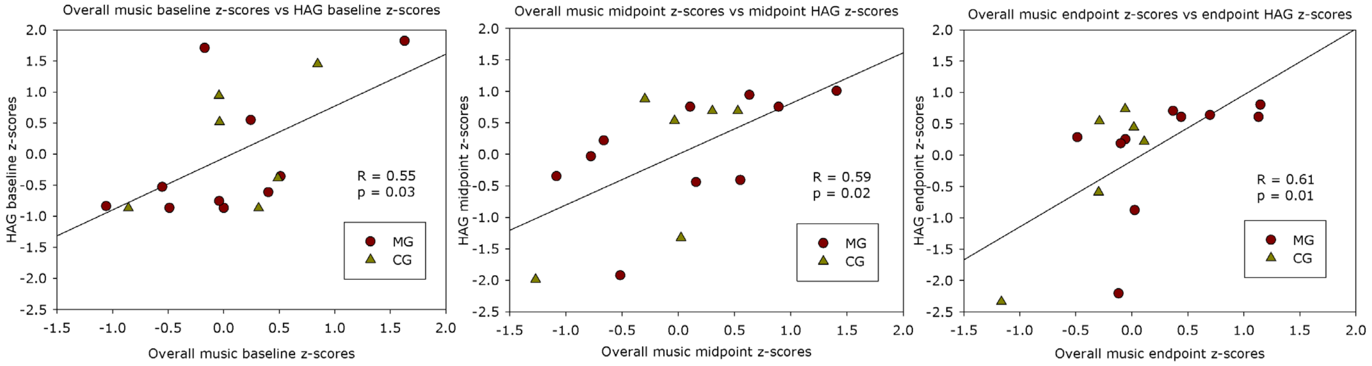


Figure 8: Three-panel figure showing regression plots of overall music z-scores (x-axis) and HAG scores (y-axis) at the three times of testing.

No significant relationship was found between overall music *gain* z-scores and HAG gain scores or EPR gain scores, neither across all participants nor within groups at any point of time.

Relationship between tests: We found significant internal correlations between baseline, midpoint, and endpoint measures in all music tests and in the 2 linguistic tests ($p < 0.05$). Furthermore, MII.1 performance predicted MII.2 performance.

Background variables vs. music and speech perception/gain.

Use of contralateral HA showed no significant relationship with any single speech or music performance. However, across all participants, use of contralateral HA showed borderline significant negative correlation with overall music endpoint gain z-scores ($r = -0.451$; $p = 0.06$), and significant negative correlation with EPR endpoint gain scores ($r = -0.560$; $p = 0.01$). Age showed a significant negative correlation with overall music gain z-scores at midpoint ($r = -0.650$; $p = 0.003$), as well as endpoint ($r = -0.543$; $p = 0.02$), across all participants. Duration of deafness correlated negatively with midpoint HAG scores ($r = -0.668$; $p = 0.004$) and endpoint HAG scores ($r = -0.512$; $p = 0.04$) across all participants.

Discussion

We found that six months of musical ear training significantly improved the overall music perception of the participants in the music group, compared to the control group. With regard to timbre, melodic contour, rhythm, and pitch, the MG improved their average musical discrimination abilities significantly during the training period. In contrast, the improvement of the control group, within these areas, was non-significant, and significantly lower than that of the music group in discrimination of timbre and pitch. We found a significant consistent relationship between overall music performance and speech perception. However, no significant effect of music training was found on speech perception. Overall music performance correlated significantly with recognition of emotional prosody exclusively in the music group, and the music group's emotional prosody recognition skills showed a trend toward faster progress relative to the control group. This suggests that musical training may be beneficial to music discrimination skills, but not necessarily to linguistic skills of CI-users.

Music training has been implemented with positive results in previous studies involving CI-listeners (Gfeller et al. 2000a; Gfeller et al. 2002c; Galvin et al. 2007). While these studies were primarily based on computer-mediated training of isolated musical tasks such as pitch, timbre, or melody recognition, the present study employed one-to-one tuition within three different musical domains, for an extended period of time. Furthermore, in contrast to previous studies, the participants in the current study were "naïve" CI-users, i.e. without implant experience. Finally, it is important to point out that, except for the MCI, the stimuli presented in the tests were not part of the music training, i. e. there was no training for the test. Our results thus demonstrate that active music training has the ability to significantly affect the general music discrimination skills, even in CI-listeners with no previous implant experience.

The remarkable progress in the average speech perception observed across all participants is in line with findings of previous studies, which showed that most performance gains occur in the first three months of use (Spivak & Waltzman 1990; Ruffin et al. 2007). Such drastic development following implantation not only shows the efficiency of the CI technology, but also the potential of cortical plasticity to reactivate inactive areas in the brain. Interestingly, our correlation analyses suggested a strong relationship between speech perception performance and overall music discrimination performance. This is in contrast with Singh et al. (2009), who, in a sample of adult CI-users, found no relationship between melody recognition and phoneme discrimination, and with Gfeller et al (2008), who found association between speech perception and music perception only when lyrics were present. Our findings suggest that there may be a relation between music and speech abilities, possibly due to the necessity of low level acoustic feature extraction from sounds in both domains (Besson & Schon 2001), but no transfer effect of musical training onto the linguistic domain. These findings are consistent with the view that high level processing of music and language primarily takes place in separate brain modules (Peretz & Coltheart 2003). The fact that we found no correlation between music performance gain and speech perception gain indicates that the progress in either domain may take place independently of one another. This is illustrated by the cases of four prelingually deaf participants in our study, who, despite HAG baseline scores of 0% correct and no or very modest endpoint gains, achieved moderate progress in their overall music perception.

Musical instrument identification

The largest impact of the musical training was found on the MG participants' ability to identify musical instruments (timbre), which improved significantly compared to the control group, even after three months of training. This finding is in line with other studies, which showed enhanced MII skills after short- and long-term computer-assisted training (Fujita & Ito 1999; Leal et al. 2003; Pressnitzer et al. 2005). Furthermore, the music group achieved an average endpoint level in the difficult subtest of the MII that was comparable to the NH level. This indicates that the implant transmits sufficient spectral information to allow CI-users to learn to identify musical instruments by their timbre, and is particularly encouraging, since most studies, which examined discrimination of timbre in CI-users have found performance significantly poorer than that of NH participants (Gfeller et al. 2002b; McDermott & Looi 2004). Even though our study doesn't allow for direct comparison between one-to-one musical training and computer-mediated training, we may speculate that the better performance observed in our study regarding timbre is a consequence of a more ambitious training program. The result is important, because improved perception of timbre may add positively to the aesthetic enjoyment associated with music listening. Furthermore, CI-listeners' ability to distinguish several instruments, playing at the same time, could be positively affected.

Pitch-related tasks

Melodic contour identification.

The MG participants significantly improved their ability to identify a melodic contour compared to baseline measures, as well as compared to the CG. The result is in line with Galvin et al. (2007), who found significant MCI progress in CI-users, following daily computer based training. This suggests that MCI may be substantially improved by training, which combines playing/singing exercises and computer based listening. The fact that the NH group performed significantly better than the CI-listeners indicates that discrimination of melodic direction remains challenging with a CI even after musical training.

Pitch ranking.

We found that the MG participants improved their ability to rank two pitches significantly, while the CG did not. Because it requires detection of direction of pitch pairs of changing interval sizes on a floating frequency base, the PR task is considerably more challenging than the MCI, which uses a fixed frequency base. Since pitch ranking was not specifically trained, this indicates that the significant MG progress represents a generalized effect from the active and passive musical exposure. Further scrutiny of the scores showed that the MG participants performed significantly better on large intervals after training, compared to the CG, while the progress only affected small intervals vaguely. Similar associations between size of interval and PR skills were reported by Looi et al. (2004), and illustrate that the implant signal does not facilitate fine-grained pitch perception. It is, however, worth noting that while the average scores of the CI-users are significantly poorer than those of the NH group, some MG participants produced scores near the NH range, thereby correctly identifying interval changes as small as one ST. This variability indicates an effect of the training but probably also stems from differences in the preconditions for music listening in CI-users.

Melodic discrimination.

Ability to correctly identify pitch direction, as measured in MCI and PR, is a fundamental prerequisite for perception (and production) of melody, and usually strongly associated with familiar melody recognition in CI-listeners (Gfeller et al. 2002a; Looi et al. 2004; Galvin et al. 2007). Despite significantly improved PR and MCI skills, the average melodic discrimination skills of the music group participants improved non-significantly. However, in contrast to recognition of familiar melodies, as used in the former studies, the MD test used in the present study assessed the comparisone of unfamiliar melodies, which is substantially more challenging. This indicates that CI-listeners may require exposure to melodies learned prior to their deafness in order to extract sufficient information from the stimulus for melody recognition. The poor progress in melodic discrimination possibly reflects the aforementioned poor discrimination of small intervals, and demonstrates that for a CI-listener, perception of pitch in the context of many pitches is a very challenging task. Furthermore, the same/different paradigm loads heavily on working memory, which may be restricted in some CI-users (Knutson et al. 1991).

Rhythm discrimination.

Several studies have concluded that perception of rhythm with CI is close to normal (Gfeller et al. 1997; Kong et al. 2004; Limb et al. 2010). Many of these studies, however, used simple tempo or pattern discrimination tests exhibiting ceiling effects in both CI and NH groups. In our study, the music group participants significantly improved their ability to discriminate complex rhythm patterns, and reached endpoint performance levels comparable to the NH group. This progress predominantly took place in the mid- to endpoint period, in which rhythm training was introduced, and produced endpoint scores that were significantly higher than those of the control group, which is a strong indication of the effect of training. The finding is quite encouraging, and evidence of the high accuracy with which current implant technology transmits temporal information. It is assumed that those CI-users, who successfully listen to music, primarily depend on lyrics and rhythm (Gfeller et al. 2008). This implies that enhanced discrimination of rhythm, as a result of training, may assist CI-users in general when listening to music.

Effect of music training on language performance.

We found no significant effect of the musical training on speech perception performance, as tested by the Hagerman test. This indicates that there is no transfer effect of musical training on speech perception of cochlear implantees. However, a transfer effect may have been covered by the magnitude of progress found in both study groups, as well as by other confounding variables. First, in the initial phase of CI adaptation, the speech perception progress may be so strongly carried by the effect of daily use that other sources of training may be of lesser significance. Second, all participants followed speech therapy, which may have had substantial influence on the perceptual development. Third, the implant is specifically optimized to effectively facilitate speech perception. Finally, the Hagerman test may have been inadequate in comparing development in the two groups. A ceiling-like effect with five of nine MG participants and two of nine CG participants scoring within the 90th percentile at the endpoint measurement, may have prevented some participants from achieving higher scores.

Emotional prosody recognition.

Both groups significantly improved their abilities to recognize emotional prosody. However, in contrast to the control group, the major part of the music group's progress occurred in the initial three month training period. Furthermore, we found a significant correlation between the overall music scores and emotional prosody recognition performance in the music group, which was absent in the control group. This suggests that musical training may have affected not only the speed of the EPR progress but also strengthened the link between music discrimination and EPR. The unexpected progress of the control group indicates that because the range of changes in pitch and timing in emotional prosody is much greater than that typical of music, these cues may be more easily identified (Ayotte et al. 2002). However, the earlier development in the music group and the significant relationship between HAG and EPR performance in the music group only, indicates that this group may employ a different strategy in extracting emotional prosodic information. In line with findings of Xin et al. (2007), both groups scored significantly below the NH level, which emphasizes that these prosodic cues are particularly challenging to CI-listeners and may explain the lack of further progress in the music group.

The effect of contralateral hearing.

The strong negative relationship between use of contralateral HA and overall music and EPR endpoint gain implies that combined acoustic and electric hearing did not facilitate progress in music discrimination, or ability to decode emotional prosody. It should be emphasized, however, that some studies have demonstrated significantly improved music perception performance in participants who combined their CI with a HA in their non-implanted ear (Kong et al. 2005; Gfeller et al. 2007; Looi et al. 2008). Here, a majority of participants kept using their HAs, but were tested CI-only. Some of these participants may have exhibited better music performance, had they been allowed to use their HA. Future studies on music listening with bimodal and bilateral hearing should examine these possible advantages more thoroughly.

The influence of age on the benefits of musical training.

Age showed a significant negative relationship with overall music gain across all participants. This implies that younger participants had a relatively larger improvement of their general music discrimination skills than older participants. Since age showed no relationship with overall music performance or HAG and EPR gain, a possible explanation of this finding is that greater cortical plasticity facilitates learning of the more complex discrimination tasks associated with music listening. The finding may also reflect a higher music listening frequency in younger CI-recipients than in older, as observed by Mirza et al. (2003), or suggest that older persons may require more extensive training, to achieve similar benefit compared to younger adults, as proposed by Driscoll et al. (2009).

The influence of duration of deafness on the benefits of musical training.

As expected, duration of profound deafness was predictive of speech perception, meaning that a short period of deafness pre-implantation was associated with better speech perception. However, duration of profound deafness was not related to speech perception gain, which suggests that duration of deafness may not necessarily predict speech progress. Quite surprisingly, no correlation was observed between duration of deafness and overall music or EPR performance. As discussed earlier, this suggests that long-time deafness does not preclude acquisition of aspects of music perception.

Limitations of the study.

While the two groups in this study were relatively uniform with regard to CI experience, type of implant, implant settings and use of HA, there was some interparticipant variability in hearing background. Five participants were prelingually deaf with baseline speech perception scores of 0%. Three of these showed varying degrees of progress, whereas two participants in the CG, for unknown reasons, remained unable to perceive speech throughout the study period. To maintain a true comparison between groups, we chose to disregard the results of those two participants in our speech perception analysis. Our sample size was relatively small. However, considering that the study involved individual training, more participants could not realistically have been included. Furthermore, all participants completed the program. The music tests in general proved valid in measuring music discrimination skills, had acceptable time consumption, were sufficiently motivating, and demonstrated high test/retest reliability. Additional tests of music appraisal could have added some important information about to which degree the improved perception of musical components influenced liking of music (Gfeller et al. 2008). However, the broad-spectred nature of this music training study demanded a wide range of tests, and more tests would probably have exhausted the participants, resulting in less reliable results. Despite shortcomings related to their provisional nature, the audio-visual material, in general, proved valid for supplementary home training. However, according to verbal reports, the amount of time MG participants spent on home-practice was quite varied. While it would have been preferable to hold this parameter constant, the variability reflects the differential job and family background found among the MG participants.

The music training program.

The music training program was not formally evaluated, but the participants in general gave positive feedback. MG 5 and MG 9 were interviewed about their experiences with the program. MG 5 commented: "The activities have been exciting and amusing, and the recurrent measurements of my progress have been tremendously motivating. The opportunity to sing out loud and be guided in my performance has been wonderful and learning to accompany myself with chords on the piano was beyond my wildest imagination." MG 9, who was interviewed on national radio, commented: "The music listening exercises have helped me a lot. It has supported my ability to segregate sounds and focus my listening– but it has taken some time." About her experience with singing she added: "I have always been incredibly shy to sing at birthdays and Christmas. Even with my family I never felt like singing along. This year, maybe I'll sing a bit louder."

Surprisingly, the majority of MG participants found singing particularly fruitful and profitable, in spite of the obvious challenges of intonation. Of course, singing comprises all the important elements of ear training; it is expressive and impressive at the same time, it features pitch, timing, and timbre, and, evenly important, has a linguistic-lyrical dimension. Furthermore, we observed that several participants spoke and sang in strikingly soft voices, probably due to long-time insecurity about the loudness of their own voice. Having received their new electrical "ear", these CI-listeners were getting acquainted with their voice anew, and the different vocal exercises in many cases were beneficial in gaining more volume and improved voice quality, also in the context of talking. Playing the keyboard, in contrast, appeared less motivating, and most participants gradually stopped practicing at home after some time. However, for ear training exercises, a keyboard (or a piano) is the most appropriate choice of instrument, in that it has a steady pitch and does not require advanced technical skills.

Perspectives.

Music enjoyment through an implant is not only a function of perceptual accuracy. Many factors such as the quality of sound, acoustic environment, familiarity with the music, and, in particular, the structural features and style of music, influence music enjoyment. However, for the majority of implant users, who find music "hard to follow" or unpleasant, introduction to the key features of music and training of the ability to discriminate different musical sounds, as examined in the present study, may be very helpful in the struggle for a higher music satisfaction. Even sparse improvements of music enjoyment may have considerable positive influence on the quality of life of CI-users (Lassaletta et al. 2007). Future research should elucidate the association between these factors.

The positive impact of training on specific musical attributes may be beneficial in the non-musical domain too. Improved discrimination of timbre may be beneficial in aspects of listening such as recognition of gender or speaker in auditory-only acoustic communication, which are notoriously challenging with CIs (Vongphoe & Zeng 2005). Moreover, poor perception of rhythm has been associated with poor perception of syllable stress and dyslexia (Huss et al.; Leong et al. 2010), and it is possible that training of rhythm, on the long term, could form a valuable and beneficial part in auditory-oral therapy, directed not only at adult, but also pediatric CI-users.

The findings of this study underlines that perception of music, in contrast to speech, does not improve as a result of incidental exposure to music, but can be improved significantly by training. This important information could profitably be part of the counseling provided to the many CI-recipients who hope for retrieving music enjoyment. Furthermore, the indication of an independent progress of music and speech perception suggests that formalized music discrimination measures could provide valuable additional information about CI outcome, if implemented in audiological follow-up routines.

Effect of music training on speech perception was absent. However, musical ear training at a later stage in the CI adaptation process, in which speech perception has stabilized, could provide further improvement, especially of recognition of emotional prosody. Future research should examine this potential benefit.

Conclusion

This study measures the progress of musical and linguistic skills in newly operated adults with CIs, following training or no training. We conclude that musical ear training, based on one-to-one tuition and active music making methods, may have a great potential as a motivating and efficient method to improve the overall perception of music in CI-users. Especially, discrimination of timbre, pitch and melodic contour may be enhanced, thereby providing improved prerequisites for fundamental aspects of music listening. Furthermore, perception of rhythm can be positively affected by training and reach normal hearing levels, which may benefit music understanding in general and some aspects of speech perception. Perception of speech may not necessarily benefit from musical ear training in this initial phase of CI adaptation, whereas perception of emotional prosody may progress more quickly and associate with musical skills. Despite its obviously challenging nature, singing may be fruitful and profitable to some CI-listeners, as a means of training pitch perception, articulation, and phrasing. Thus, the proposed musical ear training program could form a valuable complementary method of auditory rehabilitation, and, on the long term, contribute to an improved general quality of life in CI-users. The great compliance of the participants indicates that such measures could be relatively easily implemented.

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II

Cortical Plasticity after Cochlear Implantation

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Abstract

The most dramatic progress in the restoration of hearing takes place in the first six months after cochlear implantation. To map the brain activity underlying this progress, we used positron emission tomography (PET) to assess regional cerebral blood flow at three different timepoints post-implantation: immediately, at three months, and at six months. Fifteen newly operated adult implant recipients listened to running speech or speech-like noise, presented randomly in four sequential PET sessions at each milestone. A group of normally hearing control participants was tested once as reference. Speech perception was measured at the PET sessions. Analyses of variance across all cochlear implant (CI) users showed a significantly increased activation of auditory cortices bilaterally during speech. Participants with postlingual hearing loss had a significantly higher activation of Wernicke’s area relative to participants with prelingual hearing loss, when they listened to speech. This group difference was also reflected in a behavioral advantage for patients with post-lingual hearing loss. Broca’s area had significant activation as a function of time in CI-users with postlingual hearing loss. Comparison of the cochlear implant group and the normally hearing group revealed significantly higher activity in the caudate nucleus of the normally hearing group. The findings confirm the key role of Broca’s area in restoration of speech perception, but only in individuals in whom Broca’s area has been active prior to the loss of hearing.

Introduction

The cochlear implant (CI) transforms acoustic signals from the environment into electric impulses, which are then used to stimulate intact fibers of the auditory nerve. With this treatment, individuals with profound hearing loss (HL) are given the opportunity to gain or regain the sense of hearing. Current technology and speech processing strategies allow many CI recipients to achieve impressive accuracy in open-set speech recognition, and CI arguably is the most effective neural prosthesis ever developed (Friesen et al. 2001; Wilson & Dorman 2007; Moore & Shannon 2009). However, the success of the outcome depends both on duration of deafness prior to implantation (Gantz et al. 1994; Tyler et al. 1997), and on the onset of deafness before (prelingually) (Gantz et al. 1994; Tyler et al. 1997; Manrique et al. 1999), or after (postlingually) (Green et al. 2005) the acquisition of language. In many cases, the greatest gains of performance occur in the first three months of use (Spivak & Waltzman 1990; Ruffin et al. 2007). The dramatic improvements following implantation not only demonstrate the efficiency of the CI technology, but also points to the role of cortical plasticity as a means to reactivate brain function.

Plasticity is a term used to describe the reorganization of the cerebral cortex by means of synaptic changes and rewiring of neural circuits. In cases of cochlear implantation, neural plasticity associated with deprivation of auditory input and adaptation to the absence of stimuli is of particular interest. Reduced input to the brain from impaired auditory pathways imposes significant changes to the central auditory system (CAS). When auditory input to the brain is reintroduced, this novel auditory experience may itself induce additional plasticity. The sensory reafferentation provided by the CI thus offers a unique opportunity to study the effects of preceding deafness on functional brain organization.

Neuroimaging experiments comparing auditory responses of CI users and normally hearing control participants, while listening to speech or complex non-speech, generally reveal activity in the bilateral primary and secondary auditory cortices, including both superior and middle temporal gyri (Naito et al. 1997; Wong et al. 1999; Naito et al. 2000; Giraud et al. 2000; Giraud et al. 2001a; Giraud et al. 2001c). One consistent outcome of these studies is the more dominant right temporal activity of CI-users listening to speech, i.e., the observation of more bilateral activity than rigidly held by the classical presumption of left-lateralized activity of language processing in normal hearing (Gjedde 1999). However, in these studies, activation of other classic language regions such as Broca's area was not a consistent finding. Naito and colleagues found Broca's area to be activated only when the CI-participants silently repeated sentences (Naito et al. 1995; Naito et al. 2000). Mortensen et al. (2006) compared brain activity in experienced CI-users according to their level of speech comprehension performance. They found that, unlike CI-users with low speech comprehension, single words and speech yielded raised activity in the left inferior prefrontal cortex (LIPC) in CI-users with excellent speech perception.

Some observed activations outside the classic language areas, including anterior cingulate, parietal regions and left hippocampus, have been attributed to non-specific attentional mechanisms and memory in CI-users (Naito et al. 2000; Giraud et al. 2000). Giraud and colleagues consistently demonstrated visual activity in CI-users in response to speech stimuli (Giraud et al. 2001b). They found that the visual cortex responded increasingly to sounds after implantation, and that the process was associated with improvement of lip-reading proficiency, but this cross-modal interaction has not been replicated. The reasons for these mixed results include differences in experimental paradigms, small sample sizes, a heterogeneous population, liberal statistical thresholds and a lack of control of post-operational plasticity.

With the present study, we tested the cortical mechanisms underlying the restoration of hearing and speech perception in the first six month period following implant switch-on. We expected to see inactive neuronal pathways reactivated in CI-recipients within three to six months of switch-on, and engagement of cortical areas resembling those of the NH control participants. Furthermore, previous findings notwithstanding, we expected to see Broca's area involved in speech perception. Finally, we expected to see a difference in the progress of adaptation and the involvement of cortical areas between CI-users with postlingual hearing loss and CI-users with prelingual hearing loss.

Materials and Methods

Participants

Six normally hearing (NH) participants and 15 newly implanted CI-users were included in the study. All participants were right-handed Danish-speakers. The NH participants were two men and four women (47-64 years of age, *M* 54.29, *SD* 7.55). NH participants met the criteria for normal hearing by passing a full audiometric test. The CI-users were nine men and six women (21-73 years of age, *M* 51.8, SD 15). The duration of deafness ranged from 5 to 57.5 years (M: 29.7). The implant participants were included through a longitudinal recruitment procedure, by which patients, who were approved for transplantation, were contacted by mail and invited to volunteer for the research project, commencing immediately after switch-on of the implant. Of a total of 41 patients 13 replied positively prior to their operation, and three agreed to participate while hospitalized post-operation. In this report one of the 16 participants was excluded due to a possible malfunction in the implant. Ten CI-participants had right-sided implants and five had left-sided implants. Fourteen participants chose an implant device manufactured by Cochlear ©, while one chose a device by Advanced Bionics$^{©}$. Four participants (CI 2, 8, 12, and 13) had a prelingual onset of HL, indicated by their estimated age at onset of deafness (0.7-6 y) and main use of signed language as communicative strategy. Of the remaining 11 participants, 10 had a postlingual or progressive onset of HL (4-58 y), as indicated by their main use of residual hearing, supported by lip-reading. One participant (CI 3) had a prelingual onset of severe hearing loss, but with sufficient residual hearing to communicate with a hearing aid. For purposes of analyses, two subgroups were identified as (1) the postlingual (POST) HL subgroup (N=11) and (2) the prelingual (PRE) HL subgroup (N=4). The clinical and demographic details of the CI-participants are listed in Table 1.

The study was conducted at the PET center, Aarhus University Hospital, Denmark, in accordance with the Declaration of Helsinki. The research protocol was approved by the Research Ethics Committee of the Midlands Province of Denmark by Sept. 2008. Informed written consent was obtained for all participants prior to participation in the study.

Design

NH subjects underwent PET once, while CI subjects were tested consecutively at three points of time; 1) immediately after switch-on of the implant (baseline, BL), 2) after three months (midpoint, MP), and 3) after six months (endpoint, EP). In parallel with the PET study, the participants took part in a behavioral study, in which we examined the possible effects of musical ear training on music and linguistic discrimination performance. The number of musically trained and untrained participants was balanced in the two subgroups (POST 6/5; PRE 2/2). The music intervention study is to be separately reported.

Table 1. Clinical and demographic data for the 15 CI-users included in the study

Participant (gender)	Age at project start (y)	Etiology of deafness	Side of implant	Age at on-set	Duration of HL	[d]Degree of deafness	[e]Pre/ post	CI type	CI sound processing strategy
CI 1 (F)	49.8	[a]Cong. non spec. prog.	R	4	17.8	5	Post	[f]Nuc. Free	ACE 900
CI 2 (F)	21.4	Ototoxic	R	0.7	20.7	5	Pre	Nuc. Free	ACE 250
CI 3 (M)	31.7	Meningitis	L	1.8	30.2	4.5	Pre	Nuc. Free	ACE 900
CI 4 (M)	56.0	Cong. non spec. prog.	R	8	48.0	4.5	Post	Nuc. Free	ACE 1800
CI 5 (F)	70.3	Cong. non spec. prog.	R	40	30.3	4.5	Post	Nuc. Free	ACE 900
CI 6 (F)	47.5	Unknown, prog.	L	30	10.5	4.5	Post	Nuc. Free	ACE 1200
CI 7 (F)	56.2	[b]Hered. non spec.	R	19	37.2	4.5	Post	Nuc. Free	ACE 1200
CI 8 (M)	58.5	Meningitis	R	5	53.5	5	Pre	Nuc. Free	ACE 900
CI 9 (F)	29.1	[c]Mon, prog.	L	10	19.1	4.5	Post	Nuc. Free	ACE 1200
CI 10 (F)	44.8	Unknown, prog.	R	35	9.8	4.5	Post	Nuc. Free	ACE 1200
CI 11 (M)	60.4	Unknown, prog.	L	40	16.4	4	Post	Nuc. Free	ACE 900
CI 12 (F)	50.6	Cong. non spec.	R	5	47.6	5	Pre	[g]A. B. Harm.	FID 120
CI 13 (M)	63.5	Cong. non spec.	L	6	57.5	5	Pre	Nuc. Free	ACE 500
CI 14 (F)	63.0	Unknown, prog.	R	58	5.0	4	Post	Nuc. Free	ACE 720
CI 15 (M)	73.3	Trauma, prog.	R	54	19.3	4	Post	Nuc. CP 810	ACE 720
Mean	**51.8 (SD 15)**				**29.7**	**4.5**			

Table 1: Table showing clinical and demographic data of the 15 participants included in the study. [a]Non specified congenital HL, [b]non specified hereditary HL, [c]Mondini dysplasia; prog: progressive deafness; [d]1: Mild (24-40 dB HL), 2: Moderate 40-55 dB HL), 3: Moderately-Severe (55-70dB HL), 4: Severe (70-90 dB HL, 5 Profound (>90 dB HL); [e] Pre- or postlingual HL; [f]Nucleus Freedom (Cochlear ®), [g]Advanced Bionics Harmony®.

Apparatus and stimuli

MRI

A high resolution T1-weighted MR scan was acquired prior to PET scanning. In the case of CI-participants, this was performed preoperatively. In three CI-participants, who were recruited after their operation, MR scans were not obtained.

Stimuli

All subjects were examined in 2 conditions: (1) multitalker babble (BAB) from multiple simultaneous speakers with a complexity close to that of speech and perceived by the listeners as speech-like but devoid of meaning (ICRA 1997), and (2) ‘‘running’’ speech (RS), narrating the history of a familiar geographical locality at the rate of 142 words per minute, generated in Danish by a standard female voice (Elberling et al. 2010). The stimuli were played back on a laptop computer in the freeware sound editor software Audacity (http://audacity.sourceforge.net), and delivered directly from the computer's headphone jack to the external input port of the implant speech processor. Bimodally aided subjects removed their hearing aid and were fitted with an earplug in the contralateral ear during the tomography. The NH subjects listened to the stimuli binaurally through a pair of headphones (Sennheiser). All stimuli were presented at the most comfortable level. To define this level, subjects were exposed to the two stimuli once before the tomography. In the tomograph, prior to bolus injection, participants had no information about the nature of the next stimulus, but were instructed to listen attentively in all cases. After each of the four scans, subjects described what they had heard, and, if possible, reviewed the content of the narration.

PET

Positron emission tomography (PET) is a molecular imaging method that yields brain activity, by means of detecting changes in regional cerebral blood flow (rCBF). This is done by computing and comparing the spatial distributions of the uptake of a blood flow tracer. PET measurements are generally limited with respect to spatial and temporal resolution and the invasiveness of the procedure, which requires injection of oxygen-15-labelled water into the bloodstream of the participant. However, anatomical and temporal specificity could not have been improved by using functional magnetic resonance imaging (fMRI), as the auditory implants are not MRI compatible. In addition, PET is a completely noiseless imaging modality, which is useful for both CI-participants and for the study of speech. Finally, because only the head of the participant is positioned in the tomograph, compared with the whole body imposition of fMRI, it is possible to communicate visually with the participant during tomography.

We measured raised or reduced cerebral activity as the change of the brain uptake of $H_2{}^{15}O$ oxygen-15- labeled water, which matches the distribution of cerebral blood flow (CBF), using an ECAT EXACT HR 47 Tomograph (Siemens/CTI). Emission scans were initiated at 60,000 true counts per second after repeated intravenous bolus injections of doses of tracer with an activity of 500 MBq (13.5 mCi), which equals a radiation dose of 0.465mSv. Activity decayed for 10 min before each new tomography session. The tomography took place in a darkened room with participants' eyes closed.

The babble and running speech conditions were duplicated, generating a total of four tomography sessions. The uptake lasted 90 seconds (single frame) at intervals of 10 min. Each frame registered 47 3.1 mm sections of the brain. After correction for scatter and measured attenuation, each PET frame was reconstructed with filtered backprojection and smoothed with a post-reconstruction 10 mm Gaussian filter resulting in a resolution of 11 mm full-width-at-half-maximum (FWHM).

Restrictions

Rules of regulation mean that participants who volunteer for scientific experiments may receive a total maximum radioactive radiation of 6 millisieverts (mSv) within one year. Here, the total radiation dose administered over the three times of scanning was approximately 5.58mSv. Due to these restrictions, no preoperative baseline scans could be acquired.

Image pre-processing

Participants' MR images were co-registered to an MR template averaged across 85 individual MR scans in Talairach space ((Talairach & Tournoux 1988), using a combination of linear and non-linear transformations (Grabner 2006). Each summed PET emission recording was linearly co-registered to the corresponding MR image using automated algorithms. In the three participants for whom no MR scan was available, the PET scans were directly registered to an MR atlas brain in Talairach space. To smooth the PET images for individual anatomical differences and variation in gyral anatomy, images were blurred with a Gaussian filter resulting in final 14 mm at full width half maximum (FWHM) isotropic resolution.

Data analysis
All images were processed using Statistical Parametric Mapping 8 (SPM8; Wellcome Neuroimaging Department, UK [http://www.fil.ion.ucl.ac.uk/spm]). Local maxima of activation clusters were identified using the Montreal Neurological Institute (MNI) coordinate system, and then cross-referenced with a standard anatomical brain coordinate atlas (Talairach & Tournoux 1988). Differences in global activity were controlled using proportional normalization (gray matter average per volume). Significance threshold for task main effects was set to $P < 0.05$, family wise error (FWE) corrected for multiple comparisons. We tested the effect of side of implant and type of implant in a separate pre-analysis. As we found neither main effects nor interactions with functional data involving these variables, we concluded that these factors had no significant effect on the results. They were thus not included in further analyses.

Three analyses were performed as described below:

Analysis 1:
The first analysis identified the main effects of time, speech/babble contrast, and history of hearing loss (POST HL vs. PRE HL), and possible interactions between these effects. This analysis was performed as a single SPM-matrix in a factorial 3-way design with time, contrast and group (post vs. pre) as factors. To define a region-of-interest (ROI) we created a mask based on the main effect of contrast.

Analysis 2:
The second analysis identified possible main effects of contrast, time, or interactions between these factors, in the inferior frontal gyrus (IFG), more specifically Broca's area (BA 44/45). This analysis was performed as two 2-way factorial analyses of the POST HL and the PRE HL subgroups separately. To define a region-of-interest, we created a mask based on bilateral inferior frontal gyri (Broca's region), including putative Brodmann regions 44, 45, and 47 using the WFU pick-atlas (Tzourio-Mazoyer et al. 2002).

A*nalysis 3:*
The third analysis identified the main effects of contrast and group (CI vs. NH) at the CI baseline and possible interactions between these effects. This analysis was performed as a single SPM matrix in a factorial 2-way design with condition and group as factors. To define a region-of-interest, we created a mask based on main effect of contrast.

Behavioral measures:
As part of the music training study, the CI subjects were tested with the Hagerman (HAG) speech perception test (Hagerman et al. 2001) at the same three milestones as selected for PET data acquisition (BL, MP, and EP). The NH group performed the test along with their PET scan session. The HAG data was analyzed and plotted with Sigmaplot for Windows 11.0 (Systat Software Inc).

Results

Analysis 1: In the first analysis, we found a main effect of contrast across subjects, regardless of subgroup, in bilateral superior temporal gyri. A t-test confirmed that the effect was driven by higher activity during running speech (Table 2; Figure 1). There was no significant main effect of time, nor any interaction between the effects. The ROI analysis revealed significant interaction between the effects of contrast and group in BA 21/22 in the left superior temporal gyrus (Table 2; Figure 2). A plot of contrast estimates showed a larger difference between running speech and babble in the postlingual than in the prelingual subgroup (Figure 2).

Table 2. Experimental main effects postlingual HL subgroup vs. prelingual HL subgroup

POST HL subgroup vs. PRE HL subgroup	**Coordinates**					
Analysis	***x***	***Y***	***z***	**Z score**	**Region**	**Brodmann area**
Main effect of contrast (RS vs. BA)	-58	-20	0	6.55	L STG	BA 21/22
	58	0	-6	5.67	R STG	BA 21/22
	64	-10	0	5.47	R STG	BA 21
Main effect of time					NS	
Interaction time x contrast					NS	
Main effect of group					NS	
Interaction contrast vs. group					NS	
Interaction time vs. group					NS	
ROI analyses	***x***	***y***	***z***	**Z score**	**Region**	**Brodmann area**
Main effect of group (ROI)					NS	
Main effect of time (ROI)					NS	
Interaction contrast x group (ROI)	-58	-26	0	4.09	L STG	BA 21/22
	-56	-16	-3	3.24	L MTG	BA 21

Table 2: Main effects and interactions found in POST HL subgroup vs. PRE HL subgroup. Top: main effects on whole brain. Bottom: main effects of ROI mask based on main effect of contrast. *L STG* left superior temporal gyrus, *R STG* Right superior temporal gyrus

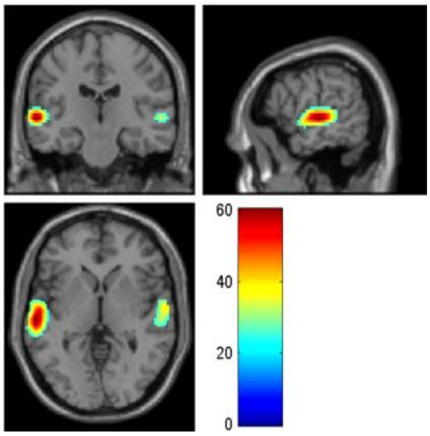

Figure 1: Activation map for main effect of contrast across subgroups in the whole brain analysis showing greater activity in superior temporal gyri (BA 21/22) during speech comprehension
(F (1,78) = 60.14).

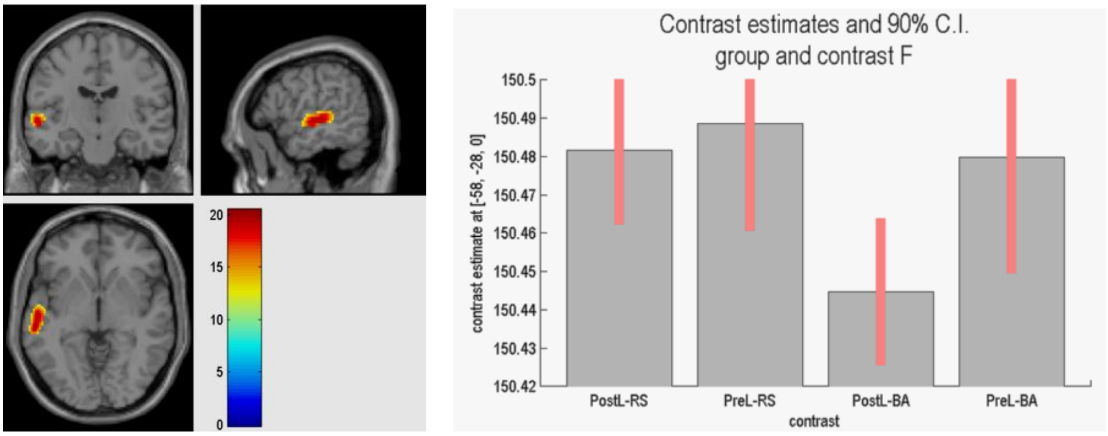

Figure 2: Left: Activation map for interaction between effect of contrast and effect of PRE/POST subgroup in the ROI analysis (L BA 21/22) showing greater activation during speech for the post-lingual group (F (1,78) = 20.42). Right: Bar plot showing contrast estimates of conditions in the two subgroups. PostL: post-lingual group; PreL: pre-lingual group; RS: running speech condition; BA: multitalker babble condition.

Analysis 2: In the second analysis, the bilateral IFG ROI analysis, we found a main effect of speech/babble contrast in the postlingual subgroup (BA 47). A t-test confirmed that the effect was driven by higher activity during running speech. We found a main effect of time in left IFG (Broca's area BA 45) at p = 0.006 (FWE corrected, Figure 3), with no significant interaction between contrast and time. The prelingual subgroup had no main effects in the bilateral IFG ROI analysis (Table 3).

Table 3. Experimental main effects of IFG ROI analyses of POST HL subgroup and PRE HL subgroup

POST HL subgroup	**Coordinates**					
ROI analysis based on bilat. IFG	***X***	***y***	***z***	**Z score**	**Region**	**Brodmann area**
Main effect of contrast (RS vs. BAB)	-46	14	-6	4.91	L IFG	BA 47
	52	16	-6	3.85	R IFG	BA 47
	46	16	-9	3.74	R IFG	BA 47
Main effect of time	-42	20	9	4.29	L IFG	BA 45
Interaction contrast x time					NS	
PRE HL subgroup	***X***	***y***	***z***	**Z score**	**Region**	**Brodmann area**
Main effect of contrast (RS vs. BAB)					NS	
Main effect of time					NS	

Table 3: Main effects and interactions found in IFG ROI analyses of the POST HL subgroup and the PRE HL subgroup respectively.

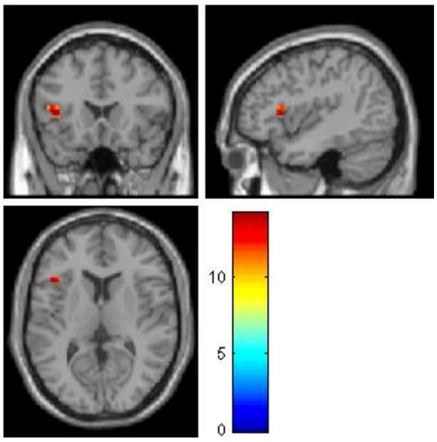

Figure 3: Activation map for main effect of time (Broca's area) in the separate analysis of the POST HL group, with ROI based on bilateral IFG (F (2,60) = 14.19).

Analysis 3: In the third analysis, we found a main effect of speech/babble contrast across the CI and NH groups bilaterally in superior temporal gyri, in the left middle temporal gyrus, and in the right inferior parietal lobule. T-tests showed that the superior temporal gyri bilaterally and the left middle temporal gyrus were more active during running speech, while the right inferior parietal lobule was more active during babble, possibly because of heightened attention (Johannsen et al. 1999). We found a main effect of CI vs. NH exclusively in the caudate nucleus. A t-test showed that this effect was due to higher activity of this area in the NH group than in the CI-group. No interaction was found between the effect of contrast and the effect of group in whole-brain analysis (Table 4).

The ROI analysis based on main effect of contrast yielded a main effect of CI vs. NH in secondary auditory cortex including Wernicke's area (BA 22) in the right superior temporal gyrus. A t-test showed that this effect was due to higher activity of this area in the NH group than in the CI-group. Furthermore, in the ROI analysis, we found an interaction between the effect of speech/babble contrast and the effect of group in the right inferior parietal lobule (Table 4).

Table 4. Experimental main effects Cochlear Implant group vs. NH group

Cochlear Implant group vs. NH group	**Coordinates**					
Analysis	***x***	***y***	***z***	**Z score**	**Region**	**Brodmann area**
Main effect of contrast (RS vs. BAB)	-58	-18	0	6.7	L STG	BA 22/21
	-54	4	-9	5.25	L MTG	BA 21/38
	62	-8	0	6.53	R STG	BA 21/22
	54	-44	36	4.64	R IPL	BA 40
Main effect of group	12	20	3	4.84	Caudate	
Interaction contrast x group					NS	
ROI analyses	***x***	***y***	***z***	**Z score**	**Region**	**Brodmann area**
Main effect of CI vs. NH (ROI)	58	-8	6	3.37	R STG	BA 22
Interaction contrast x group (ROI)	52	-46	39	3.45	R IPL	BA 40

Table 4: Main effects and interactions found in CI-group vs. NH group. Top: main effects on whole brain. Bottom: main effects of ROI mask based on main eff. of contrast. *R MTG* Right middle temporal gyrus, *R STG* Right superior temporal gyrus, *L MTG* left middle temporal gyrus, *L STG* left superior temporal gyrus. *R IPL* right inferior parietal lobule.

Behavioral measures: The mean Hagerman performance of the entire CI-group increased 31 percentage points from baseline to midpoint and again by 10.5 percentage points from midpoint to endpoint, for a total increase of 41.5 percentage points, although this average increase reflected major differences between the two subgroups. In the postlingual subgroup, the mean Hagerman performance increased by 37.6 percentage points from baseline to midpoint and again with 13.6 percentage points from midpoint to endpoint, for a total of 51.3 percentage points. In the prelingual group, the mean Hagerman performance increased by 12.8 percentage points from baseline to midpoint, and by 1.8 percentage points from midpoint to endpoint for a total of 14.5 percentage points (Figure 4).

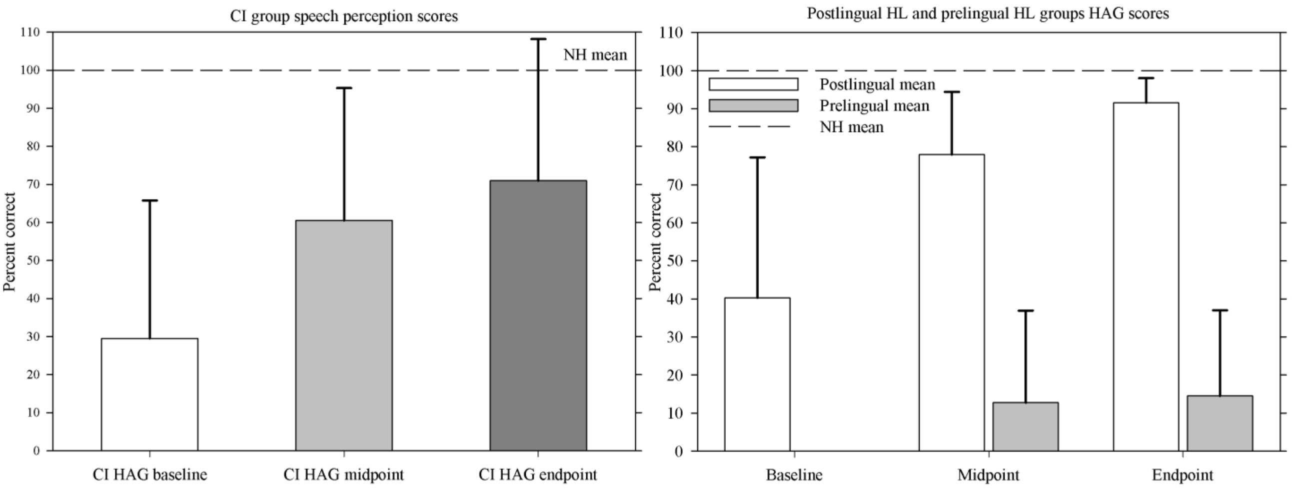

Figure 4: Left: Barplot showing mean percent correct Hagerman speech perception scores for the entire CI group at BL, MP, and EP. Right: Barplot showing mean percent correct Hagerman speech perception scores for the POST HL subgroup and the PRE HL subgroup respectively at BL, MP and EP. Dashed line shows NH mean. Error bars show standard deviation.

Discussion

Implant users with postlingual hearing loss versus implant users with prelingual hearing loss

Our investigation of brain activation following cochlear implantation revealed a difference in the way CI-recipients with postlingual HL and recipients with prelingual HL distinguish speech and babble. Due to a differential processing of the stimuli in the two subgroups, the CI-users with postlingual HL displayed a greater contrast between activation for speech and babble in BA 21 and 22 in the left superior temporal gyrus than the CI-users with a prelingual HL. Implant users with postlingual HL had greater activation during speech than during babble, and implant users with prelingual HL had comparable levels of activity during speech and babble. We speculate that the postlingually deaf listeners disengage attention when they are presented with the incomprehensible babble stimulus. This disengagement is then reflected in decreased temporal brain activity. In contrast, the prelingually deaf CI-listeners may be equally attentive to the two stimuli, regardless of their nature, as reflected in undifferentiated activity.

This difference between postlingual and prelingual HL is mirrored in the behavioral measures. The postlingual subgroup not only possessed a moderate level of speech perception at baseline (i.e., 1-2 days after switch-on of the implant), but also sustained remarkable gains in performance, the majority of which occurred in the first three month period. In contrast, the prelingual subgroup had no baseline speech perception and only modest progress during the study period. This finding is consistent with expectation and implies an association between behavioral performance and brain activity related to the history of hearing loss. In prelingual deafness, the neuronal connections of the auditory pathways may not be established in the appropriate time window of opportunity. The subsequent electric stimulation at some time in adulthood may produce some hearing sensation, but the discriminations of sounds and time intervals remain defective (Mortensen et al. 2005). Follow-up studies in the present population may provide interesting insight into the degree, to which speech perception progresses in the prelingual subgroup on the long term.

We found a main effect of time exclusively in Broca's area, and only in the postlingual subgroup. This is an indication that the changes in the process of adapting to the implant most profoundly are manifested in this specific area, which is associated with speech perception and production. Surprisingly, we found no interaction between the speech/babble contrast and time. This suggests that the area becomes increasingly activated, regardless of whether the stimulus makes semantic sense or not, or is active in the distinction between sense and nonsense.

In the entire CI-group, the bilateral middle and superior temporal gyri were significantly more active when participants listened to running speech than when they listened to multi-talker babble, across all points of measurement, including, more specifically, Brodmann areas 21 and 22. Thus, on average, the auditory brain regions in newly implanted CI-recipients clearly distinguished between speech-like noise and speech, confirming that both hemispheres are involved in the speech perception process, also during unilateral stimulation.

Cochlear implanted participants at baseline versus normally hearing participants

We found significant activation of bilateral superior temporal gyri and left middle temporal gyrus during speech, both in CI-participants at baseline and in NH individuals. Furthermore, right inferior parietal lobule was significantly more active during babble, and significantly more so in the CI-group than in the NH group. Analysis of the CI-group versus the NH group showed a significantly higher activation in the caudate nucleus in the NH group. Finally, ROI analysis showed that during speech, the NH group involved the right STG more than did the CI-group.

The observation that during speech stimulation, the NH participants involved the caudate nucleus more than the CI-participants may be explained by a reduction of the effort needed by the NH participants to deal with the well-known task of receiving a message. The caudate nucleus is a part of the striatum, which subserves among other tasks the learning of slowly modulated skills or habits (Gabrieli 1998). To the normally hearing listener, the reception of auditory information is an every-day experience similar to following a known route, e.g. see Wallentin et al. (2006) for a similar argument. In contrast, to CI-listeners, auditory stimuli are non-habitual in the strongest sense of the words, thus relying on other sources of processing.

In contrast to Giraud et al. (2001a), we found no significant activation of the left insula. Furthermore, that study showed a left-lateralized activation of temporal and frontal regions in NH controls versus the significantly higher right-lateralized activation seen in NH individuals in the present study. However, direct comparison between the two studies is difficult, as several differences in study design and implant experience of the participants exist. The CI-participants' involvement of the right parietal lobule suggests that at this initial stage of the CI adaptation, CI-listeners, unlike NH individuals need to pay attention to the speech-like noise to determine its possible character. Mortensen et al. (2006) found increased activity in right cerebellar cortex when running speech was comprehended relative to babble, but only in CI-listeners with high speech comprehension. The authors speculated that this could be due to cognitive work of cerebellum subserving verbal working memory, or a contribution of the right cerebellar hemisphere to precise representation of temporal information for phonetic processing. However, this finding was not replicated in the present study, which may reflect differences in duration of implant use and a mixture of speech comprehension levels.

Cross-modal plasticity

Giraud and colleagues (2001c) consistently demonstrated activation of areas BA 17/18 in the visual cortex when CI-users responded to meaningful sounds. The authors argued that the process was associated with improvement of lip-reading proficiency. Such cross-modal interaction between vision and hearing was not replicated in the current study. Differences in the methodology used in the two studies may explain this discrepancy. The Giraud study involved repetition of words and syllables and naming of environmental sounds, contrasted with noise bursts, as opposed to the current study, which involved passive listening to a story contrasted with speech-like noise. Furthermore, sound was presented in free-field, whereas in the current study, the auditory stimuli bypassed the microphones of the speech processor and was fed directly to the auxiliary input. Finally, the strict conservative statistical methods used here preclude reporting of results that are not statistically significant when corrected for multiple comparisons.

Conclusion

The present PET study tested brain activation patterns in two groups of newly implanted adult CI-recipients, who listened to speech and non-speech stimuli. The groups were similar with respect to implant experience, but heterogeneous with respect to onset of hearing loss. Unlike CI-listeners with postlingual hearing loss, CI-listeners with prelingual hearing loss showed undifferentiated activation of left superior temporal gyrus during speech and speech-like stimuli. This difference was reflected in behavioral data. Furthermore, Broca’s area was activated as an effect of time, but only in CI-listeners with postlingual hearing loss. This confirms the key role of Broca’s area in restoration of speech perception, but only in individuals in whom Broca’s area has been active prior to the loss of hearing. The study clearly demonstrates that adaptation to the electrical stimulus of the cochlear implant is highly related to history of hearing loss. Patients whose hearing loss occurred after the acquisition of language involve brain areas associated with speech comprehension, which is not the case for patients whose hearing loss occurred before the acquisition of language.

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III

Musical Methods for Little Digital Ears -music training and testing in preschool children with cochlear implants

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ABSTRACT

Objective: A population of young children with cochlear implants has emerged because of neonatal hearing screening and early implantation. Supplementary methods of habilitation to expand auditory, social, and cultural competences of these children should be developed and evaluated. Music offers an enjoyable learning environment that may improve musical discrimination skills and generalize to the linguistic domain. In this study, we evaluated the effect of a music training program and the utility of new music tests directed toward early-implanted preschool cochlear implant users.

Methods: Twenty-one children with cochlear implants (36–72 months) were assigned to two groups: the music group, which received weekly music training for 3 months, and the control group, which received no formal music training. A group of normal hearing children provided music test reference data. Training included various music-making and listening activities. Music tests were developed to measure musical instrument identification, pitch change detection, and familiar melody identification. Furthermore, linguistic skills were assessed. Testing was performed immediately before and after the 3-month study period. Parental feedback was obtained regarding possible changes in the musical behavior of the music group children.

Results: The music group demonstrated higher average scores than the control group in all music tests at the end of the study period. Mean performance gains were larger for the music group, but the difference between groups was non-significant. On an average, the music group children performed pitch detection task similar to normal hearing peers, while the controls performed significantly poorer. Furthermore, the mean music group linguistic performance was slightly better than the mean control group performance. Parental feedback indicated that training had a stimulating impact on the everyday musical behavior of the music group children. Finally, musical performance strongly correlated to age at testing, but not to age at implantation.

Conclusion: Pediatric cochlear implant users benefit from musical training and enjoy participating in structured singing, dancing, and playing activities. The test results, participant responses, and parental feedback all indicate that the proposed training offers a stimulating environment and substantial listening practice. This training may support long-term musical, linguistic, psychosocial, and cultural development of these children.

Introduction

A cochlear implant (CI) is a device designed to re-establish hearing abilities in individuals who are severely deaf and can help the average adult CI user recognize about 90% of words in sentences in quiet listening conditions [1] and [2]. However, because of the implant's poor transmission of pitch information, music listening and enjoyment remains problematic [3] and [4]. Poor pitch perception affects music enjoyment and complicates speech perception, especially in noisy surroundings. This can cause difficulties when using pitch cues to separate a target stimulus from the surrounding noise [5]. Therefore, improved pitch perception may be beneficial for implant users not only for music enjoyment but also for other aspects of listening.

In the United States, as many as 12,000 newborns with hearing impairments are identified every year, with an estimated additional 4000–6000 infants acquiring late-onset hearing loss [6]. Approximately 40% of these children will be severely or profoundly deaf [7] with little or no benefit from a hearing aid (HA). Due to the introduction of newborn hearing screening programs (since 2004 in Denmark), identification of hearing loss has accelerated, dropping to 1–3 months of age, making cochlear implantation possible at around 12 months. The result is a new and growing population of prelingually deaf infants, to whom the prospects of developing spoken language, reading, and academic skills remain positive. In general, younger age at implantation has been associated with better language progress [8], [9] and [10]; however, the impact of early implantation on music perception and enjoyment remains unknown. Enhanced development in the central auditory system and greater cortical plasticity in early-implanted compared to late-implanted children may have favorable implications for music processing as well [11] and [12].

Studies involving adult CI users have shown positive behavioral results from music training. For example, Galvin et al. [3] demonstrated significant improvement in the ability of six CI users to identify melodic contours after a short period (~2 months) of daily computer-assisted exercises. The effect was most notable in contours with large pitch changes, but the benefit of the training interestingly generalized to the ability to identify a familiar melody (2007). In a similar study, postlingually deaf CI users had significantly improved recognition and appraisal of timbre of musical instruments [13].

For pediatric CI, studies of the effects of musical training have been sparse. Abdi et al. [14] studied CI users aged 3–12.5 years following a music training program that involved either simple perceptual tasks or learning to play a musical instrument. Brief reports of the musical development of children suggested that the training may have been beneficial, although no objective evaluation, such as music tests, was described. In a case/control study including 18 pediatric CI users, Yucel et al. [15] investigated the outcome of a longitudinal music training program based on a take-home electronic keyboard. According to parent reports, the music group (MG) children showed significant improvement in almost all areas of music perception. However, parents may lack objectivity and accuracy in assessment of the musical skills of their children, and this may have affected the results. Recently, Chen et al. [16] examined pitch perception in a group of children with CIs (5–14years), half of whom attended music classes. Despite existing confounds, the authors concluded that duration of music education positively correlated with the performance of pitch perception.

Numerous studies comparing musically trained and untrained children have shown that music lessons may benefit non-musical domains [17]. Music training, for instance, positively affects the child's linguistic abilities such as phonological processing, early reading, and sensitivity to speech prosody [18] and [19]. Furthermore, language development can be strongly facilitated by use of song, which indicates that children's songs and lullabies may support acquisition of linguistic prerequisites [20]. Because associations between music and language rely on shared resources such as melody (intonation), rhythm (timing), and dynamics (stress), music training may affect the linguistic development of pediatric CI users too. In addition, it has been demonstrated that auditory coaching is imperative for children with CIs to develop age-appropriate communication skills, and that educational environments that emphasize the development of speech, auditory, and spoken language skills are particularly beneficial [21], [22] and [23]. Since music offers a learning environment that is multisensory and enjoyable, it is plausible to believe that music can play a valuable supplementary role in auditory/oral habilitation programs.

In a sample of preschool children with early CIs, the present study aimed to evaluate the outcome of a music training program designed to focus on pitch-related skills and develop a music test battery that would properly measure the child's perception of pitch and timbre as well as be brief, comprehensible, and enjoyable. We hypothesized that intensified active exposure to music and live musical sounds would expand the ability of children to perform musical discrimination tasks, especially for pitch, melody, and timbre. Furthermore, we expected that the musical experience would be noticeable in the everyday life of the children, and would be expressed as increased musical interest and activity observed by their parents. Finally, we expected that the improved musical competences would generalize to the linguistic domain.

Materials and Methods

Ethical approval: The study was conducted in accordance with the Helsinki Declaration and approved by the Research Ethics Committee of the Midlands Province of Denmark in September 2008. Informed consent was obtained from the parents of all participants.

Participants: Twenty-one prelingually deaf preschool children (3–6 years) with CIs were assigned to two groups based on their availability to attend weekly music training sessions: MG (n = 10) and control group (CG, n = 11). The MG children (3 boys; 7 girls) were 45–74 months old (M = 61; SD = 11) at the start of the project and lived in the greater County of Aarhus, Denmark. Their average age at implantation was 35 (SD = 17) months. Four of the MG children had bilateral implants and three were bimodally aided with a HA in the non-implanted ear. The MG children attended weekly musical training sessions over the course of 3 months. The CG children (7 boys; 4 girls) were 33–79 months old (M = 58; SD = 13) at the start of the project and lived in Denmark. Their average age at implantation was 23 months (SD = 13). Seven of the CG children had bilateral implants and one was bimodally aided. The CG children were not exposed to formal musical training and parents were instructed to avoid changes in their musical environment till after the end of the 3-month study period (Table 1).

All children in the study followed an individual oral/aural habilitation program, which reflects local practice. All participants had hearing parents and exclusively communicated by auditory–oral means. Most of the children had used bilateral HAs prior to implantation. Four children, two in each group, had one or two parents without Danish as their primary language. All children were fitted with CIs having the most advanced processing strategy and had their implants and processors controlled on a regular basis. Two children originally recruited for the CG were disqualified after the initial test. One failed to complete the tests and the other possessed an uncharacteristically high level of musical performance. The two groups matched relatively well on musical background—a judgment based on questionnaire information about the passive and active musical habits of CI users and their families.

Normal hearing reference: To obtain a reference for the music test battery a group of age-matched normal-hearing (NH) children (3 boys; 7 girls) 42–74 months old (M = 63; SD = 7.3) were recruited. NH children attended the same preschool as four of the MG children, and they were considered by parents and caretakers to represent typical musical development and offered a fair comparison to the children with CI.

Table 1

Table 1. Background and clinical data for the subjects in the MG and the CG.

Group	Sex	Age at proj. start (months)	Number of implants	Side of implant	Hearing aid	Age at implant 1 (months)	Age at implant 2 (months)	Implant use (months)	Etiology of deafness	CI sound-processing strategy
1	M	74	2	L/R	-	19	39	55	[a]Cong. non spec.	ACE 1200 Hz
2	M	73	1	L	-	54	-	19	[b]Pen, [c]Mon	ACE 900 Hz
3	F	67	1	L	R	50	-	17	Pen, Mon, [d]AN	ACE 900 Hz
4	F	66	1	R	L	29	-	37	Cong. non spec	ACE 1200 Hz
5	F	66	1	L	N	54	-	12	[e]Gen. (conn 26)	ACE 900 Hz
6	F	67	1	R	L	53	-	46	Gen. (conn 26)	ACE 1800 Hz
7	F	57	1	L	R	38	-	19	[f]Her. non spec.	ACE 900 Hz
8	F	49	2	L/R	-	19	40	30	Pen, Mon	ACE 1200 Hz
9	M	47	2	L/R	-	18	37	29	[g]CMV	ACE 1800 Hz
10	F	45	2	L/R	-	14	14	32	Meningitis	ACE 900 Hz
Mean		**61** *(SD 11)*				**35** *(SD 17)*	**32** *(SD 13)*	**26** *(SD 13)*		
11	F	79	1	R	L	51	-	28	Her. non spec.	ACE 2400 Hz
12	M	78	2	L/R	-	17	41	61	Cong. non spec	ACE 900 Hz
13	M	67	2	L/R	-	34	47	33	Gen. (conn 26)	ACE 1200 Hz
14	M	65	1	R	-	8	-	57	Meningitis	ACE 900 Hz
15	F	57	2	L/R	-	20	31	38	Cong. non spec.	ACE 900 Hz
16	M	56	2	L/R	-	16	41	40	CMV	ACE 2400 Hz
17	M	52	1	R	-	35	-	17	AN	ACE 900 Hz
18	F	50	2	L/R	-	13	15	35	Her. non spec.	ACE 900 Hz
19	M	50	1	L	-	34	-	16	Meningitis	ACE 1200 Hz
20	F	53	2	L/R	-	15	34	38	Her. non spec.	ACE 2400 Hz
21	M	33	2	L/R	-	13	13	20	Cong. non spec.	ACE 900 Hz
Mean		**58** *(SD 13)*				**23***(SD 13)*	**32***(SD 13)*	**35***(SD 15)*		

Table 1: Etiology of deafness: [a]non specified congenital hearing loss, [b]Pendred's syndrome, [c]Mondini dysplasia, [d]auditory neuropathy, [e]genetic connexin 26, [f]non specified hereditary hearing loss, [g]cytomegalovirus. All children are users of the Nucleus implant (Cochlear ltd ®, Australia).

The music training program: The music program was scheduled for 3 months, and sessions took place weekly as 90-min modules in a shared preschool activity room. The room was acoustically dampened and well suited for musical activities. A teacher from the preschool, who knew several of the children, was involved in all sessions. On several occasions, parents or caretakers were also present. Two music teachers took turns in leading the musical activities according to a preceding agreement. One was a teacher of music at the Royal Academy of Music, Aarhus, Denmark and the other, a Master's student in music teaching finishing her education. All sessions were captured on digital video to document behavioral responses of the children to allow adjustment to the music program if necessary.

The training program included singing, playing (percussion), dancing, and listening activities. By establishing a familiar structure with recurrent activities in an appreciative and encouraging learning atmosphere, we sought to motivate the children to participate and complete the program. Our intention was to provide experiences with fundamental musical features such as high/low, up/down, weak/strong, start/end, pulse/tempo, and sound quality through music-making activities. To support these objectives the musical material had to meet the following criteria: (1) melodies should comprise between a fifth and an octave, and be short, logical, and preferably have a call/response or an echo character; (2) songs should not contain lengthy lyrics or odd grammar such as displaced verbs; (3) accompanying movements or gestures should be uncomplicated and easily imitable; and (4) recorded versions of songs should be available in indisputable quality, performed in vocal ranges suitable for preschoolers, preferably sung by female or child voices. On this basis, we selected 20 songs, which were compiled on a CD and distributed to the families along with a booklet containing musical scores and lyrics. Parents were instructed to play the CD at home at any convenient time, and if possible, sing the songs with their children focusing specifically on the lyrics and melodic contours.

New songs were taught stepwise, first articulating the words thoroughly, then with rhythm, and finally with melody. Phrases and stanzas were repeated at decreased tempo to strengthen the internalization and memory process, thereby taking advantage of the joy of recognition. As a complimentary method, we practiced imitation or call-response, a teaching tool involving short-term memory.

Music sessions followed a regular pattern:

1. Welcome song addressing all children by name
2. Singing two or three familiar songs (choice of the children)
3. "Choir" – vocal exercises; refreshing old songs; learning new songs
4. Singing game(s) – old and/or new games with dancing/movements
5. Intermission
6. Musical quiz games – "which instrument is playing behind the curtain?" (e.g., cowbell, triangle, wood block, cymbals, cajon, and rattle egg) or "is the melody rising, falling, or flat?" (short melodies played on guitar or xylophone)
7. Percussion ensemble – simple complementary rhythms played on conga drums, cowbell, wood block, cajon, and similar instruments with short distinct sounds.
8. "Goodbye song" – all children lying on the floor listening and relaxing.

Apparatus and stimuli: To assess development in the musical discrimination skills of the children, we created a battery consisting of three tests: (1) musical instrument identification (MII); (2) pitch change detection (PCD); and (3) familiar melody identification (FMI). The MII and PCD tests were presented in the computerized test environment MACarena (Waikong Lai, Zürich University Hospital). The stimuli played back at approximately 65 dB SPL through a small active loudspeaker (Fostex 6301B, Fostex Company, Japan) placed approximately 1 m in front of each child. FMI test was presented "live" to ensure direct communication with the child and motivation to complete the test. Testing took place in a quiet room in the children's preschool or, in rare cases, at their homes. Tests were performed before and after the musical intervention program, or, in the case of some control children, within an equal time span (3 months ± 1 week). The same examiner administered all musical tests.

MII: The MII test is a three-alternative forced-choice test featuring three different musical instruments: flute, piano, and double bass, the latter clearly distinguishable from the other two by playing one octave lower. For the musical material, we programmed a well-known Danish children's song "Mariehoenen Evigglad" (Ladybug Ever Happy fig. 1) in the MIDI-sequencer Cubase 4.1 (Steinberg Media Technologies GmbH, Hamburg, Germany). A track was made for each instrument and played back using the software sampler Halion 2.0 (Steinberg) with high quality instrument samplings. The three instrumental versions of the song were divided into four phrases (A, B, C, D, fig. 1.) and exported as 12 CD-quality sound files (44.1 kHz 16 Bit). We presented the melodic phrases in random order using MACarena. This way each instrument was presented four times in the test, playing different melody sections. The melody design was inspired by The Zurich Music Test Battery [24] and ensured that the instrument identification was not associated with a single melodic feature.

Figure 1

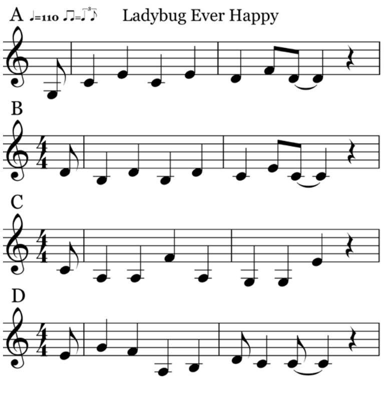



Fig. 1: Ladybug Ever Happy in musical notation. The melody comprises an octave from G3 (196 Hz) to G4 (392 Hz). Letters A–D categorize the four parts presented randomly in the test.

Testing procedure: As an introduction, each child was presented with a poster showing illustrations of the three instruments (fig. 2). The examiner highlighted each instrument while the corresponding sound of the melody's first phrase (A) was played back. This procedure was repeated twice for each instrument. When performing the test, the child was informed that the computer would now choose instruments itself, and it was his job to point out which one was playing. Responses were registered with a mouse click by the examiner. In general, the children participated with enthusiasm, as they would in a computer game. The test was easily understandable for all children, and no previous knowledge about musical instruments was required.

Figure 2

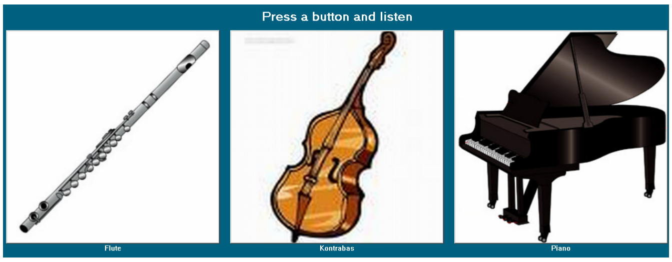


Fig. 2: Poster illustrating the three choices for the MII test. The poster is identical to the picture on the computer screen.

PCD: The PCD test is a two-alternative forced-choice test designed to measure the distance threshold at which children are able to detect a change in pitch. The test is based on a design adapted from Vongpaisal et al. and requires the children to tell whether one note in a sequence of five is different [25]. With Cubase 4.1, we programmed eight different five-note tunes. One of these (the standard) consisted of five repeating notes (C4 = 261.6 Hz), while the rest (deviants) were altered upwards in increasingly smaller intervals of 7, 6, 5, 4, 3, 2 or 1 semitones instead of the fourth note (fig. 3). All tones had an equal duration of 250 ms and an intertone pause of 200 ms. For the timbre, we used a modified digital sampling of a clarinet played through the Halion software sampler, similar to the method used in the MII test. As opposed to a piano tone, the clarinet sample has a long sustain, and thereby possible ambiguous pitch impressions as an effect of tone offset are avoided [24]. The eight sequences were exported as wave files for presentation in MACarena.

Testing procedure: The biggest challenge in administering this test was to make sure that the younger children (including those in the NH group) would comprehend their task. To accomplish this we introduced them to the standard melody by playing it three times, telling them to remember it well and to say "Yes" whenever they heard it (i.e., the "Yes melody"). If, on the other hand, they heard a *different* melody they were told to say "No" (i.e., a "No melody"). Example of a "No melody" was also given twice. The paradigm was adapted from "Audie" created by Edwin E. Gordon [26]. Having tested that the child fully understood this concept, we moved on to the actual test. As in the MII test, the examiner would register the responses by clicking the corresponding buttons on the computer screen. For further clarification, children were shown a small poster illustrating the two choices. In the case of a shy or quiet child, he or she could just point out the answer (fig. 4).

Figure 3

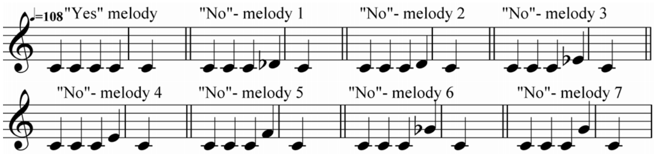



Fig. 3: Eight sequences of the PCD test in standard music notation.

Figure 4

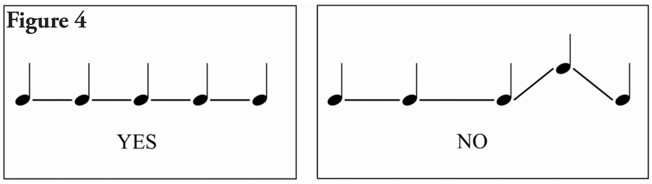


Fig. 4: Poster showing the two choices for the PCD test. The illustration is identical to the picture on the computer screen.

FMI: We designed the FMI test to examine whether the children were able to recognize well-known songs without accompanying lyrics. Four songs considered to be well-known by most Danish children were chosen for the test: (A) Mariehoenen Evigglad (Ladybug Ever Happy), (B) Lille Peter Edderkop (Itsy Bitsy Spider), (C) Se den lille kattekilling (See the Little Kitten); and (D) Hjulene på bussen (The Wheels on the Bus) (fig. 5). A multiple choice test in three parts presenting A/B, A/ B/ C, and A/B/C/D was designed as described below.

Testing procedure: The test was performed in three parts. First, the child was presented with a small poster showing pictorial representations of melodies A and B, and asked to identify the two songs by keyboard plus vocal presentations with lyrics. The procedure was repeated to assure the accuracy of recognition. Subsequently, three trials were given in which the examiner played the instrumental versions of the two songs in random order. The child identified the songs by pointing, and answers were registered on a score sheet. Second, melody C was introduced in the same manner as in part one, and the child was presented with a three-alternative poster. The test procedure was similar to part 1. Finally, melody D was introduced in the same manner as in parts one and two, and the child was presented with a four-alternative poster (fig. 6). The test procedure was similar to parts one and two. Note that melody A is in "swing" style and clearly distinguishable from melodies B, C, and D, which all feature similar simple straight rhythm. Due to time constraints, the FMI test was performed only at the end of the 3-month period. Therefore, we were not able to measure performance gains but rather the level of FMI performance after training/no training.

Figure 5

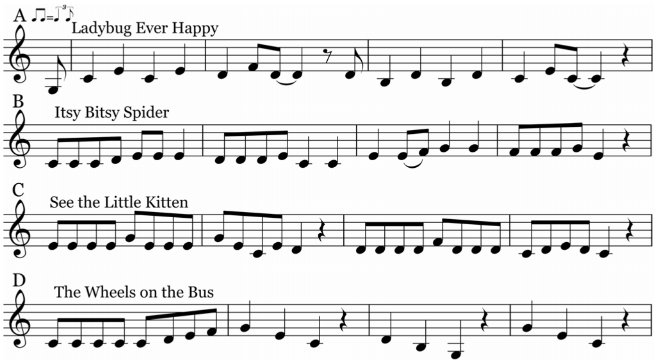



Fig. 5: Four melodies of the FMI test in standard music notation. All melodies are in C major within the range from G3 (196 Hz) to G4 (392 Hz).

Figure 6

Fig. 6: The small poster presented to the child at step three with pictorial representations of melodies A, B, C, and D.

Linguistic tests: Four existing tests were chosen for the language perception measurements. The Linguistic test 1[27] and Galker test [28] for children aged 3–4 years and the Viborg material [29] and Test for the Reception of Grammar (TROG) [30] for children aged 5–6 years. Considering the age variation, no single test could properly encompass the entire group. Tests were chosen for brevity, validity, and logistical ease.

Linguistic test 1 is an expressive vocabulary test that requires children to name a series of pictures presented to them in a booklet. Scores were differential, depending on the specificity of the children's answers: 1 = below normal (infantile words like choo-choo), 2 = normal (train), 3 = above normal (locomotive, freight train). A standard score table based on random sampling is available (http://www.spf-nyheder.dk/download/sproglig_test_1.pdf). The test duration was approximately 5 min and the maximum score was 63 points.

The Galker test is an audiovisual, computerized, word discrimination test in background noise and comprising 35 word pairs. Words are presented by a female speaker, and the task is to determine which of the two pictures fits the spoken word. The test is standardized for ages 3–5 based on results from a sample of 388 NH children. The signal/noise ratio was 0 dB. The test duration was 5 min and the maximum score was 35 points. The Viborg material is an expressive vocabulary test similar to Linguistic test 1 and adjusted for children aged 4–7 years. The test was standardized with 660 Danish children. The test duration was 7–10 min and the maximum score was 51 points.

TROG is a multiple-choice test designed to assess understanding of grammatical contrasts. The test consists of 80 four-choice items, from which each child is required to select the picture that corresponds to a sentence spoken by the examiner. Items are divided into blocks of four, with each block testing a respondent's understanding of a specific type of contrast. A block is completed when all four items in the block are correctly answered. The contrasts are arranged in order of increasing difficulty, thereby making it appropriate for an age range of 4–12 years. The Danish version was recently standardized utilizing a revised version (TROG-2). The test duration was 10–20 min and the maximum score was 20 points.

Parental feedback: We created a questionnaire to obtain parental feedback concerning possible changes in the children's musical behavior (Questionnaire 1). Questionnaire 1 was distributed to the families in MG at the end of the program. It had a response rate of 100%. The questions were divided into four categories: (1) general musical interest (response/attention to music), (2) interest in the actual program (talk about/refer to events/songs), (3) singing and dancing activities, and (4) linguistic development associated with musical exposure. In all categories, the parents stated the extent to which they agreed that they had observed changes in their children following the musical program by rating a series of statements [(1) Strongly disagree, (2) Disagree, (3) Neither agree nor disagree, (4) Agree, (5) Strongly agree].

Statistical methods: For each test, the percentage of correctly answered items was used for analyses. Data were analyzed and plotted with Sigmaplot for Windows 11.0 (Systat Software Inc). Paired t-tests were performed to compare within-group results, and t-tests were performed to compare results across groups. Variables with non-normal distribution were compared using the Wilcoxon/Mann–Whitney U-test. Mean, standard deviation, median, and range values were given as descriptive statistics. Significance threshold was set at 0.05.

Results

MII: Figure 7 shows MII scores for the two groups before and after the 3-month study period. The mean MG performance significantly improved from 60.7% to 93.3% correct (paired t-test: $p < 0.001$). Furthermore, the mean CG performance significantly improved from 68% to 85.5% correct (paired t-test: $p = 0.016$). While mean performance gains at the end of the study period were larger for MG than those for CG, significant difference in performance gains between the two groups (t-test: $p = 0.097$) was absent. In addition, significant difference between 3-month MG and NH peer performance levels (Mann–Whitney $p = 0.607$) or between CG and NH peer performance levels (Mann–Whitney: $p = 0.108$) was absent. Ceiling performance (100% correct) was observed in 4 of the 10 MG children and 3 of the 11 CG children at the end of the study period, and in 7 of the 10 NH children.

Figure 7

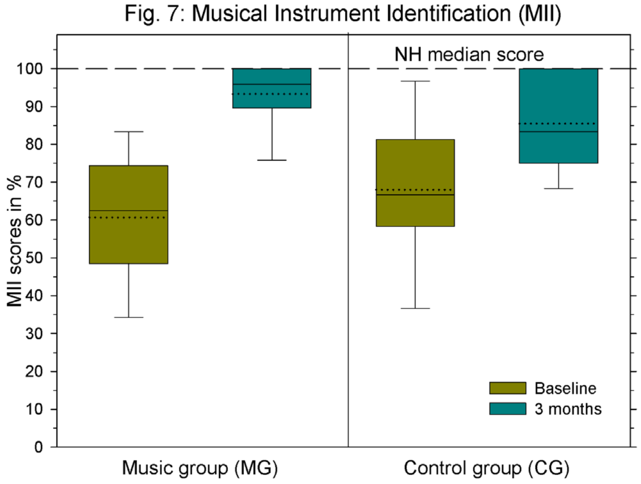


Fig. 7: Box plot of MII scores before and after the 3-month study period for MG and CG, respectively. Error bars show 10th/90th percentile. Solid box lines: median. Dotted box lines: mean. Horizontal dashed line: NH median score.

PCD: Figure 8 shows PCD scores for the two groups before and after the 3-month study period. The mean MG performance significantly improved from 65.1% to 82% (paired t-test: $p = 0.007$). Furthermore, the mean CG performance significantly improved from 60.7% to 71.6% (paired t-test: $p = 0.016$). While mean performance gains at the end of the study period were larger for MG than those for CG, significant difference in performance gains between the two groups was absent. The difference between 3-month MG and NH peer performance levels was not significant (Mann–Whitney: $p = 0.277$), while a significant difference was observed between 3-month CG and NH peer performance levels (Mann–Whitney: $p = 0.004$). Ceiling performance (100% correct) was observed in 4 of the 10 MG children and 5 of the 10 NH children.

Figure 8

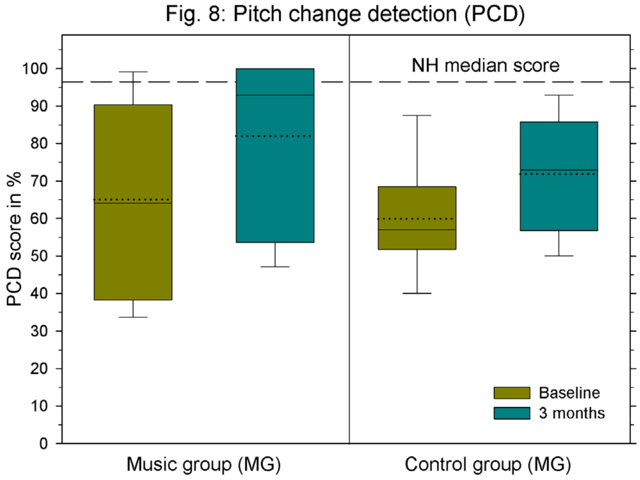


Fig. 8: Box plot of PCD scores at baseline and after 3 months for MG and CG. Error bars show 10th/90th percentile. Solid horizontal box lines: median. Dotted horizontal box lines: mean. Horizontal dashed line: NH median score.

FMI: Figure 9 shows MII scores for the two groups after the 3-month study period. The mean MG performance was slightly higher (M = 69.6; SD = 16.68) than that of CG (M = 63; SD = 17.22). The difference between the two groups was not significant. A significant difference between MG and NH peer

Figure 9

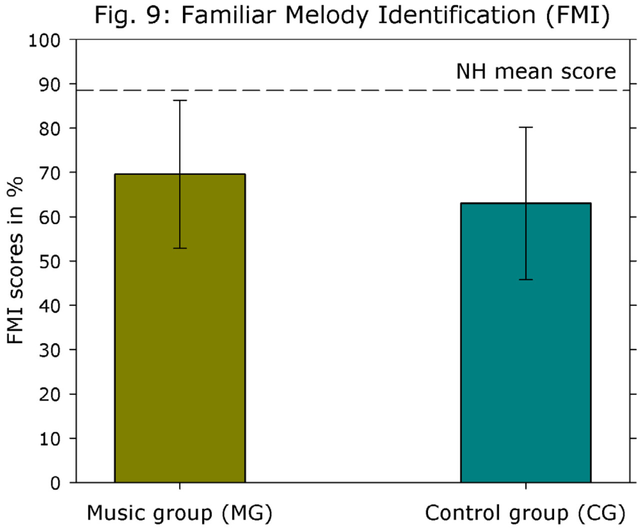


Fig. 9: Bar chart showing FMI scores of MG, CG, and NH groups. Error bars show standard deviation. Horizontal dashed line: NH mean score.

performance levels (t-test: $p = 0.010$) as well as between CG and NH peer performance levels (t-test: $p = 0.002$) was observed. Ceiling performance (100% correct) was observed in 1 of the 10 MG children at the end of the study period and in 4 of the 10 NH children. Two children in CG were unable to complete the test (N = 9).

Linguistic test results: Table 2 shows NH reference and the individual and mean scores of the younger children in the two linguistic tests before and after the 3-month study period. In Linguistic test 1, the mean MG performance improved from 68% to 86% correct (paired t-test: $p = 0.089$). Furthermore, the mean CG performance improved from 67.7% to 74.7% correct (paired t-test: $p = 0.168$). While mean performance gains were larger for MG than those for CG, significant difference in performance gains between the two groups (t-test: $p = 0.181$) was absent. At the end of the study period, the MG children scored significantly higher than the NH reference (t-test: $p < 0.001$), which was not the case for the CG (t-test: $p = 0.327$). In the Galker test, the mean MG performance improved from 70% to 75% correct (paired t-test: $p = 0.069$). Furthermore, the mean CG performance improved from 67.7% to 74.7% correct (Wilcoxon: $p = 0.938$). While mean performance gains were larger for MG than those for CG, significant difference in performance gains between the two groups (t-test: $p > 0.05$) was absent. A significant positive difference between 3-month MG and NH peer performance levels (t-test: $p = 0.03$) as well as those between CG and NH peers (Mann–Whitney: $p = 0.026$) was observed.

Table 2

Table 2. Linguistic test results for younger children (aged 3-4 years)

Linguistic test 1							**Galker test**				
Group	Age	NH ref.	Base-line	3 mths.	Dif.	P (t-test)	NH ref.	Base-line.	3 mths.	Dif.	P (t-test)
MG							**MG**				
A7	57						62.9	85.7	91.4	5.7	
A8	49	69	66	84	18		57.1	62.9	71.4	8.6	
A9	47	69	78	86	8		57.1	60.0	65.7	5.7	
A10	45	69	60	88	28		57.1	71.4	71.4	0.0	
Mean	**55**	**69.0**	**68.0**	**86.0**	**18.0**	**N.S.**	**58.6**	**70.0**	**75.0**	**5.0**	**N.S.**
MG vs. NH ref.					**17.0**	**<0.001**				**16.4**	**0.030**
CG							**CG**				
B5	57						62.9	45.7	74.3	28.6	
B6	56	71	66	62	-4		60.0	77.1	74.3	-2.9	
B7	52	71	56	60	4		60.0	42.9	37.1	-5.7	
B8	50	71	84	78	-6		60.0	80.0	74.3	-5.7	
B9	50	71	78	90	12		60.0	60.0	80.0	20.0	
B10	53	69	72	90	18		57.1	88.6	77.1	11.4	
B11	33	59	50	68	18		37.1	65.7	62.9	-2.9	
Mean	**50**	**68.7**	**67.7**	**74.7**	**7.0**	**N.S.**	**56.7**	**65.7**	**68.6**	**2.9**	**N.S.**
CG vs. NH ref.					**6.0**	**N.S.**				**11.4**	**0.026**
MG gain vs. CG gain					**11.0**	**N.S.**				**2.1**	**N.S.**

Table 2: NH, individual and mean scores of the younger children for Linguistic test 1 and Galker test. NH ref. = NH reference scores derived from standard tables. N.S. = not significant.

Table 3 shows the individual and mean scores of the two linguistic tests for the older children before and after the 3-month study period. In the Viborg material test, the mean MG performance significantly improved from 60.2% to 70.6% correct (paired t-test: $p = 0.007$) and the mean CG performance improved from 63.7% to 69.6% correct (paired t-test: $p = 0.154$). While mean performance gains at the end of the study period were larger for MG than those for CG, significant difference in performance gains between the two groups (t-test: $p = 0.306$) was absent. Both groups scored very close to the NH reference. The distribution of increased score across children was stable, which reflects reduced test uncertainty with this older age group.

TROG registers completed four-item blocks. Only three CG children had sufficient stamina to complete the test. In TROG, the mean MG performance improved from 49.2% to 55% correct (Wilcoxon: $p = 0.125$), and the mean CG performance improved from 43.3% to 53.3% correct (paired t-test: $p=0.074$). While mean performance gains at the end of the study period were larger for CG than those for MG, significant difference in performance gains between the two groups (Mann–Whitney: $p = 0.381$) was absent. Note that the CG children scored at a lower baseline level than the MG children, and the final MG scores were slightly higher than those of CG. A significant negative difference between the NH reference and both final MG TROG performance (t-test: $p = 0.026$) and CG TROG performance (t-test: $p = 0.008$) was observed.

Table 3

Table 3. Linguistic test results for older children (aged 5-6 years)

Viborg material							**TROG**				
Group	Age	NH ref.	Base-line	3 mths	Dif.	P	NH ref.	Base-line	3 mths.	Dif.	P
MG											
A1	74	72.7	74.5	78.4	3.9		75.0	50.0	60.0	10.0	
A2	73	72.7	56.9	58.8	1.9		75.0	55.0	55.0	0.0	
A3	67	70.8	68.6	88.2	19.6		65.0	55.0	65.0	10.0	
A4	66	70.8	49.0	64.7	15.7		65.0	35.0	35.0	0.0	
A5	66	70.8	62.7	68.6	5.9		65.0	45.0	50.0	5.0	
A6	67	70.8	70.6	80.4	9.8		65.0	55.0	65.0	10.0	
A7	57	65.3	39.2	54.9	15.7						
Mean	**67**	**70.6**	**60.2**	**70.6**	**10.4**	**0.007**	**68.3**	**49.2**	**55.0**	**5.8**	**N.S.**
MG vs. NH ref.					**0.0**	**N.S.**				**-13.3**	**0.026**
CG											
B1	79	75.1	70.6	80.4	9.8		75.0	50.0	55.0	5.0	
B2	78	75.1	62.7	66.7	3.9		75.0	35.0	50.0	15.0	
B4	67	70.8	72.5	70.6	-1.9		65.0	45.0	55.0	10.0	
B5	65	65.3	49.0	60.8	11.8						
Mean	**70**	**71.6**	**63.7**	**69.6**	**5.9**	**N.S.**	**71.7**	**43.3**	**53.3**	**10.0**	**N.S.**
CG vs. NH ref.					**-2.0**	**N.S.**				**-18.4**	**0.008**
MG gain vs. CG gain					**4.5**	**N.S.**				**4.1**	**N.S.**

Table 3: NH, individual and mean scores of the older children for the Viborg material and the TROG. NH ref. = NH reference scores derived from standard tables. N.S. = not significant.

Parental feedback: Table 4 lists the combined mean Likert scale scores of the parents' responses in the four different categories. On an average, parents reported that their child expressed a general increased interest in music during the program and

Table 4

Table 4. Quest. 1 results - observed benefit from music program

Question category	1. Increased general musical interest	2. Great interest in the actual program	3. Increased singing and dancing activity	4. Improved linguistic skills
Mean	4.23	4.28	4.53	4.08

Table 4: Mean Likert scale scores for observed benefit from the music program derived from responses to Questionnaire 1 by parents in the MG. Maximum score=5.

Table 5

Table 5. Quest. 2 results: Parents' expectations to their child’s outcome from various musical activities

Question category	1. General benefit from basic music activities	2. Boost in social competences from basic music activities	3. Boost in linguistic competences from basic music activities	4. Benefit from singing lessons	5. Benefit from learning a musical instrument	6. Benefit from singing in a choir
Mean	4.9	4.5	4.5	4.5	4.4	3.9

Table 5: Mean Likert scale scores for the expected outcome from various musical activities derived from responses to Questionnaire 2 from the parents to all participants. Maximum score=5.

frequently referred to the music program, listened to the CD, and spontaneously sung songs from the program. They observed increased and improved singing and dancing activities. Furthermore, the parents found that their children in general had improved sound discrimination, developed more prosody, and become more attentive to language nuances and rhymes.

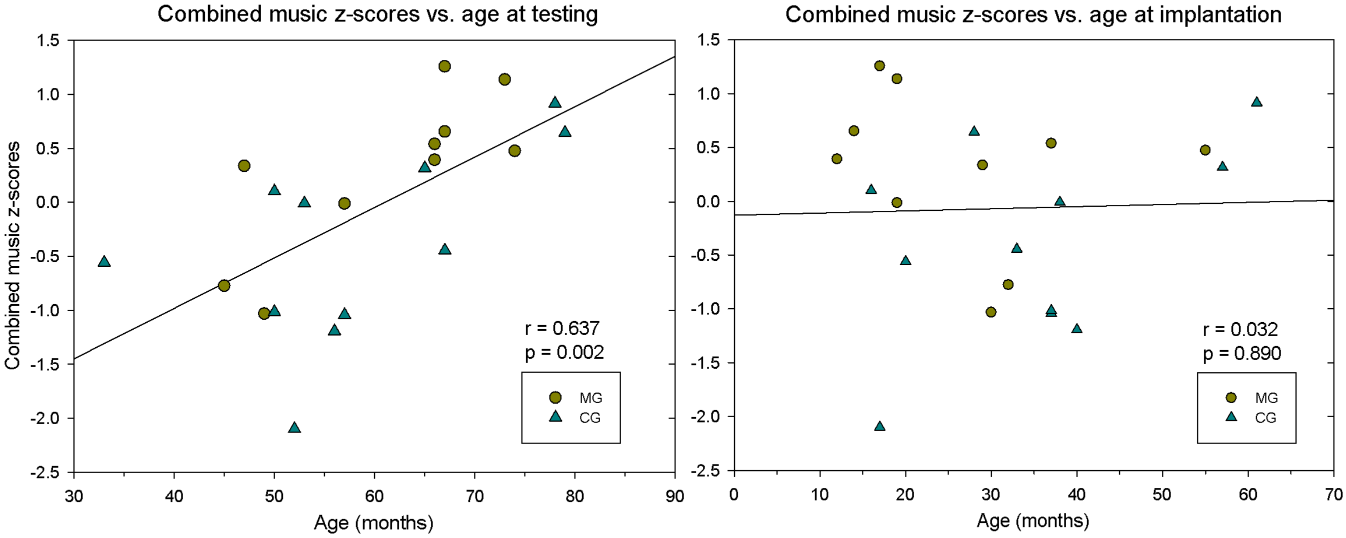
In a follow-up to the initial parent questionnaire (Questionnaire 2), we additionally asked parents about their possible plans for their child’s participation in music programs, and to what extent they expected that their child would benefit from various forms of musical activity. Questionnaire 2 was provided to all 21 families and had a response rate of 100%. Thirty-eight percent of the families had plans, 10% had no plans, and 52% considered letting their child participate in a public musical teaching program. Ninety percent stated that they would be more inclined to do so if they knew that the program was especially arranged for children with CIs. As shown in Table 5, parents on an average expected that their child would benefit to a large extent from musical activities in general and even from learning to sing and play an instrument.

Video observation: Our comprehensive video material provided supplementary documentation about the responses of the MG children to the musical training. Observations showed that the children, after some initial reluctance, tended to joyfully engage themselves in the different musical activities. Furthermore, we found that their participation intensified as their familiarity with persons and repertoire grew stronger. We did not observe any child showing signs of unpleasantness associated with the sound level resulting from the musical activities.

Correlation analyses: To investigate whether age at testing or age at implantation associated with individual test results, we performed linear regression analyses. We transformed the music scores from the concluding tests to normalized Z scores and calculated a single combined music score for each child. As shown in the linear regression plot (fig 10), we found a strong correlation between combined music scores and age at testing ($r^2 = 0.406$, $p = 0.002$), while there was no correlation between combined music scores and age at implantation (fig. 10) ($r^2 = 0.001$, $p = 0.890$). To examine the relationship between the individual scores within the three musical domains, we performed a Spearman rank order correlation analysis. The analysis showed a strong correlation between all music test scores: MII vs. PCD ($r = 0.64$, $p = 0.004$), PCD vs. FMI ($r = 0.48$, $p = 0.049$), and MII vs. FMI ($r = 0.64$, $p = 0.002$).

Similar linear regression analyses of the linguistic test results showed no correlations between individual combined linguistic scores and age in any of the groups. A correlation analysis of the relationship between music and linguistic performance showed

Figure 10


Combined music z-scores vs. age at testing

Combined music z-scores vs. age at implantation

Fig. 10: Linear regression plot of individual music scores vs. age at testing and individual music scores vs. age at implantation.

only weak correlation between combined music and combined linguistic scores (Spearman rank order: r = 0.331, $p = 0.151$).

Uni- or bilateral implants: Regression analyses showed a weak negative correlation between combined music scores and number of implants (r2 = 0.120, $p = 0.125$), indicating that children with one implant on an average scored slightly higher than those with two. Analyses of linguistic test scores and number of implants showed a weak positive correlation among the younger children ($r^2 = 0.135$, $p = 0.267$), but not among the older ones ($r^2 = 0.0000385$, $p = 0.987$).

Discussion

The purpose of the present study was to develop a music training program aimed at early-implanted pediatric CI users and evaluate its effect on musical and linguistic skills and everyday music behavior. We hypothesized that the musically exposed children would improve their musical discrimination skills and that this would possibly generalize to their linguistic skills as well. Furthermore, we expected that the MG children would willingly and joyfully participate in the music-making activities and possibly display increased musical interest and activity when at home.

Main findings: The mean musical performance of MG was significantly better after training, and their mean performance gains were larger than CG, although not significantly. Furthermore, the CG children also increased their musical discrimination skills significantly, but in general they scored lower than the MG children and significantly below the NH level in the final PCD test. In contrast, the MG children almost equaled the NH level in MII and PCD at the end of the training period. In addition, parental feedback revealed that the music training had a strong positive impact on the children's musical interest and musical activity. Finally, the overall mean linguistic performance of MG was slightly better than the mean CG performance at the end of the study period. Although the study represents many challenges and some limitations associated with size and heterogeneity of the sample and shortcomings of tests, the results are very encouraging. This is emphasized by the fact that the approach of the training program was to teach and make music in the real world environment rather than within pure laboratory tasks.

The remarkable progress in the ability of the MG children to identify a musical instrument compared to the CG children is consistent with Gfeller et al. [13], who demonstrated significant improvement in identification of musical instruments in a group of adult CI users who followed a computerized training program. Though the MG children may have had a small advantage in understanding the test, it is important to note that the test stimuli were exclusively presented during testing and that training did not directly aim to improve this skill. This indicates that the difference between groups reflects a generally increased attention toward musical sounds in the MG children as an effect of the training.

Identification of musical instruments playing at the same pitch and loudness relies on perception of timbre or sound quality, which is largely related to differences in the harmonic spectrum. Our results show that though these frequencies are not naturally represented, CI delivers stimuli that are precise enough to permit recognition of timbre and that this ability may be affected by training. Exactly which strategy the pediatric CI users use in this task remains unclear and other acoustic cues such as tone on- and offsets or temporal envelope may be of assistance. Apart from an expected benefit for music appraisal, improved timbral discrimination may also have a positive impact on non-musical tasks such as voice gender recognition and perception of speech in noise involving competing talkers. Future studies on timbre discrimination should look further into this potential association.

With regard to PCD, the MG children showed an average progress that was eight percentage points higher than the CG children and a final score that significantly approached the NH level. This finding shows that majority of the MG children became able to identify changes as small as a semitone, the smallest interval in Western music. Such improvement in pitch detection as an effect of exposure to active music-making could potentially transfer to the general perception of melody and melodic contour. This is supported by observations from Galvin et al. [3] who found that an improved perception of melodic contour generalized to melody recognition. However, in line with the findings of Vongpaisal et al. [25], we found that the enhanced ability to detect small pitch changes seemingly did not facilitate recognition of melodies. One reason for this discrepancy may be that other cues than actual pitch changes assist the child in this detection task.

The MG children identified familiar melodies slightly more accurately than the CG children did after training, though the test stimuli were not included in the training program. The difference was non-significant, but suggests that the increased focus on melodic contour through guided singing practice and glissando exercises may have helped improve musical prerequisites for music identification. Nevertheless, despite the fact that all CI users easily recognized the songs when lyrics were presented, their average performance was significantly below the NH level in the instrumental versions, even with available rhythm cues. Identifying songs relies on detection of small pitch changes in the context of variable tones, and our results match previous research on song recognition in child implant users [31], [32] and [33], and demonstrates that current implant technology induces a pitch processing deficit very similar to the phenomenon of amusia in NH individuals as suggested by Nakata et al [31].

One single case of extraordinary familiar song identification may contrast this general view: a congenitally and profoundly deaf girl, who had been bilaterally implanted at the age of 11 months made perfect and immediate recognition of all songs on the first occasion of testing, for which reason she was excluded as a control child. Her exceptional abilities were accompanied by examples of spontaneous solo singing with precise melodic and lyrical renditions of children's songs. The girl, who was 34 months old at the time of testing, had received intense family-based and professional auditory training and participated in musical activities on a regular basis. The case suggests that musical stimulation in large doses and from a very early stage may help child implant users to outperform the technological limitations of the implant.

Linguistic test results: We saw significant progress in the expressive vocabulary development of older MG children compared to older CG children. In addition, we saw a trend toward a larger and more consistent progress in the expressive vocabulary development and phonetic discrimination skills of younger MG children compared to younger CG children. Although differences between the groups indicate some benefit from the musical program, significant differences were absent in the linguistic performance gains between the groups. The observed age-appropriate average levels in three of the four linguistic tests are consistent with Hayes et al., who found indications that children implanted before 2 years of age can achieve receptive vocabulary skills within the average range for hearing children [34].

In TROG, the CG children showed a larger gain than the MG children, although from a lower baseline level, resulting in a final score just below that of the MG children. Since TROG tests grammatical contrasts that are typically characterized by prosodic differences in the melodic and temporal speech pattern, we had expected to observe a larger increase in children who had been exposed to the music training. In turn, we saw that the children in both groups scored significantly below the level of hearing age peers. This shows that pediatric CI users, despite early diagnosis and implantation as well as vocabulary development and phonetic awareness at or above the NH level, have difficulties in perceiving complex linguistic information when presented in a context.

This in/out-of-context problem may be associated with the inability of CI users to use their PCD skills when required in the context of recognizing a familiar melody. Similar discrepancies between age-appropriate vocabulary development and below-average complex language scores have been documented by Geers et al. [35], implying that assessment of pediatric CI users' linguistic progress should include comprehensive and thorough tests like TROG. Furthermore, auditory training that emphasizes listening and speaking should be maintained throughout childhood to help these children keep pace with their hearing peers. Interestingly, we observed various genuine local dialects in our sample of children from all over the country. This shows that while child implant users may face trouble in perceiving the smaller interval changes of music, they appear able to perceive and produce the greater range of pitch changes in speech. Future research should examine this phenomenon in detail to see if child implant users acquire age-appropriate detection of prosodic speech and vocal emotion, and whether this is associated with music perception.

Our correlation analyses suggest that with respect to pitch-related musical performance, age at testing is a stronger precondition for implant outcome than age at implantation. This is in line with Sharma et al. [36] who found that children with a short period of auditory deprivation (<3.5 years) show age-appropriate P1 latency responses within 6 months after the onset of electrical stimulation. It is also partly consistent with Vongpaisal et al. [33], who demonstrated that age and working memory were significantly associated with song recognition performance in 8–18-year-old CI recipients. Another explanation of the association between age and test performance may be that tests, which require the child to hold stimuli in memory, may favor older children who have developed their memory and attention as part of maturation. The unexpected significant progress in musical performance of the CG children may also partly be explained by this correlation. Time and daily use bring great changes in perceptual and cognitive capabilities in pediatric CI users, even within domains that are not explicitly trained.

If age at implantation is not decisive for the musical performance in early implanted children, then why did we see no or only weak associations between age and linguistic scores? One reason could be the division of children into two groups with a small age span and small samples. Furthermore, it may reflect inter-child variability associated with differences in background variables like age at onset of deafness, pre-implant hearing levels, and parental support. Finally, different cognitive processes may be required for some aspects of music listening than for speech perception and production. In our study, we found internal consistency between performance on the different musical tests, but no correlation between music and language tests. This is in agreement with Singh et al. [37] who found no significant correlation between melody recognition and phoneme recognition in a study with adult CI-users. Similarly Gfeller et al. [38] found only weak correlations between speech perception measures and general attitude and involvement in music in a survey on children and adolescents with CIs. Music ratings, however, significantly correlated with composite communication scores, suggesting that older children, who successfully communicate via listening and speaking, use these skills to understand music.

Bilateral/unilateral implant: Correlation analyses of musical performance and number of implants showed that children with one implant scored slightly higher than those with two. This finding may be due to some residual hearing on the non-implanted ear in the unilaterally implanted children. Combined acoustic and electric hearing significantly improves melody recognition [39] because aided residual hearing provides fine-structure cues in low-frequency ranges (<1000 Hz), crucial for pitch perception. In support of this, we can report a case of one child, who scored 100% correct on all final music tests. The girl had some residual hearing and was bimodally aided as well as musically inclined. Interestingly, we found no correlation between linguistic performance and number of implants, though a similar benefit from residual hearing has been documented for speech perception as well [39].

The limitations of this study were absence of random assignment to groups and small sample size. However, since the MG children necessarily had to be available for training (i.e., live in the proximity of the training location), neither random assignment nor a larger training group was an option. Furthermore, it should be noted that the CG children were not a group *per se*, but rather individuals living across the country, who met the criteria for inclusion and whose parents responded to the recruitment letter. Thus, equivalent enrichment activities (like theater or painting) for the CG children, similar to those in studies on transfer effects, were not possible. Although the groups were relatively well matched on variables such as age at testing, age at implantation, communication mode, implant type and settings, and musical background, differences in etiology, hearing history, number of implants, and parental support make the results less robust than desired. However, perfectly matched groups are not likely to be found in CI research in general and child implant users in particular. While educational background may affect the degree to which parents can assist the auditory development of their children, economic status is hardly predictive for the included children's habilitation opportunities, since in Denmark as in many European countries, auditory training is mandatory and publicly paid for (as is the implant and the operation).

Another limitation of this study was parental feedback. In assessing their child's musical skills parents are not necessarily objective or accurate, and the lack of supplementary feedback from parents of the CG children for comparison weakens the impact of these measures. The MII and PCD tests showed unexpected ceiling effects, which may have precluded better results for some children. This suggests a need for tests that not only have a narrow age target but also consider the speed at which skills develop in this age group. Future revisions of the test battery should take this into account by differentiating it agewise and adding subtests. If implemented as a part of the general monitoring routines, such objective measurements of music discrimination skills could be a valuable supplementary tool in the assessment of CI outcome and tuning adjustments in preschool children.

Conclusion

In summary, our study shows that prelingually deaf pediatric CI users benefit from musical training and enjoy participating in structured singing, dancing, and playing activities. The video documentation, participant responses, test results, and parental feedback all indicate that the proposed music program offers a stimulating environment and substantial listening practice, which may support the musical, linguistic, psychosocial, and cultural development of these children. Furthermore, we found improved music perception skills and a tendency towards better linguistic skills after training compared to those in controls. However, future studies should examine these important aspects more profoundly with an extended training period and more solid musical tests. In the sample of early-implanted CI users studied here, we observed significantly poorer song recognition skills compared to NH peers, even after training. This indicates that in tasks relying on pitch processing in a context, early implantation is not advantageous in overcoming implant limitations. It also confirms that despite large pitch processing difficulties, children with CIs can derive considerable pleasure from music, primarily from features that are effectively transmitted such as rhythm and lyrics. Based on our findings, we conclude that music training with carefully selected material and well-considered methods is relevant to pediatric CI users and may provide a valuable supplement to other auditory habilitation initiatives. The commitment of parents and remarkable confidence in the potential benefits of music for their children indicates that such a post-operative habilitation effort could be relatively easy to implement.

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Conflict of interest statement

The authors declare no conflicts of interest.

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