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Master's Thesis

A Multi-Level Modeling Approach of Speech Perception after Cochlear Implantation.

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Title: A Multi-Level Modeling approach of speech perception after cochlear implantation.

Summary:

- Objective: To evaluate the long-term development of patients with a cochlear implant of the University Medical Centre in Utrecht; with regard to three potential predictive factors: severity of preoperative hearing loss, duration of preoperative deafness and cause of deafness (the 'bony disorders' meningitis and otosclerosis vs. all other causes of deafness).
- Study design: Retrospective longitudinal clinical study. Predictors of speech perception, after cochlear implantation surgery, included preoperative hearing loss, duration of deafness and effect of a bony disorder as cause of deafness (meningitis or otosclerosis); with use of Multi-Level Modeling analysis.
- Patients: 247 adult patients with a cochlear implant.
- Interventions: Unilateral multichannel cochlear implantation.
- Main outcome measures: Postoperative speech perception (CVC) scores.
- Results and conclusion: Perception of CVC words after cochlear implantation is significantly predicted by duration of deafness, preoperative hearing loss and presence or absence of a bony disorder as cause of deafness. There is no effect of interaction of these prediction variables, nor among themselves nor with the time predictors (=duration of implant use). The development of speech perception over time is best described by a linear and a negative quadratic growth model. As Multi-Level Modeling has been demonstrated in previous studies to be more powerful in hypothesis testing than other analysis tools, our unexpected result of cause of deafness being a significant predictor of speech perception might be due to the sensitivity of the Multi-Level Modeling method.

Introduction

Cochlear implantation (CI) has become a common intervention for patients with profound sensorineural hearing loss, giving them the ability to perceive sounds. Although postoperative open set speech perception is a common result for many CI-users, performance of speech perception varies widely between them. 'The causes of this enormous variation in outcomes are only partly understood at the present time.' (Peterson, Pisoni & Miyamoto, 2010, p237). Many efforts have been made to determine the sources of this variability, such as age at onset of deafness, age at implantation, type of implant and processor used, type and intensity of rehabilitation, etiology, residual hearing, previous hearing aid usage, duration of implant use, etcetera. Many predictors behaved inconsistently over investigations. Gantz, Woodworth, Knutson, Abbas & Tyler (1993) concluded that variability among CI users is not attributable to differences between different implant systems, but should mainly be attributed to the characteristics of patients. In the next part of this introduction results from earlier studies to three characteristics of patients, being possible predictors of success, will be summarized.

Duration of deafness is consistently identified in the literature as highly predictive of postoperative performance. The shorter the duration of deafness, the better the speech understanding with the CI. (Shea, Domico & Orchik, 1990; Blamey et al., 1992; Gantz et al., 1993; Battmer, Gupta, Allum-Mecklenburg & Lenarz, 1995; Blamey et al. ,1996; Albu & Babighian, 1997; Van Dijk et al., 1999; Friedland, Venick & Niparko, 2003; Blamey et al, 1996,; Rubinstein, Parkinson, Tyler & Gantz, 1999; Gomaa, Rubinstein, Lowder, Tyler & Gantz, 2003; Oh et al., 2003; Green et al., 2007; Klop et al., 2008). Each year of additional deafness results in a loss of 0.8% - 1% on postoperative word perception testing (Blamey et al., 1996; Rubinstein et al., 1999; Friedland et al., 2003; Green et al., 2007). Green et al. (2007) concluded that duration of deafness accounted for 9% of the variability.
Based on this literature, we expect to find duration of deafness as being a predictor of speech perception after CI.

Preoperative residual hearing ability in one or both ears has also been stated as a predictor for CI benefit in several investigations. In literature different measures of residual hearing were used. Gomaa et al. (2003), Summerfield & Marshall (1995), Rubinstein et al. (1999) and Adunka, Buss, Clark, Pillsbury & Buchman (2008) used speech perception scores as measure of the preoperative residual hearing to predict postoperative performance, whereas others used pure-tone hearing loss as measure of the residual hearing (Blamey et al., 1992; Gantz et al., 1993, Van Dijk et al., 1999; Francis, Yeagle, Bowditch & Niparko (2005). Others used both types of measures

(Neumann et al., 2008; Roditi, Poissant, Bero & Lee, 2009). The manner of measuring the residual hearing appeared in most studies not to influence the effect of the degree of hearing loss on postoperative performance. Most of these studies confirm that patients with some residual hearing appear to have better auditory scores. (Summerfield & Marshall, 1995; Rubinstein et al., 1999; Van Dijk et al., 1999; Gomaa et al., 2003; Francis et al., 2005; Neumann et al., 2008). Preoperative residual hearing seems to protect the spiral ganglion and/or or the central auditory pathways from degeneration (Gomaa et al., 2003).

In contrast to the previous studies, Blamey et al. (1992), Gantz et al. (1993) and Adunka et al. (2008) did not find preoperative residual hearing as being a significant factor influencing CI performance. In Adunka's study, the fact that both groups of patients not only differed according to hearing loss but also appeared to differ according to duration of deafness, might have confounded the results. There might be a trade off between a negative influence of longer substantial hearing loss and a positive effect of better preoperative hearing.

We conclude that most studies report CI patients with some preoperative residual hearing to reach better postoperative perception scores. For that reason we also expect to find preoperative residual hearing as being a predictor for auditory success after CI.

Cause of deafness as predictive variable has been found to be inconsistent between investigations. Blamey et al. (1996) reported the relation between speech perception and the aetiology of deafness to be 'relatively weak'.

Meningitis and otosclerosis both can lead to deafness. Because both disorders can affect the state of the cochlea through bone growth, leading to cochlear bone anomalies, these disorders will be grouped in this study to a single term 'bony disorder'. In meningitis, bacterial labyrinthitis can cause labyrinthitis ossificans. The inflammation of the membranous labyrinth then progresses to fibrosis and ossification (Brodie, 2008). In otosclerosis, the normal bone of the otic and labyrinthine capsule of the inner ear is resorbed and, as the disease progresses, replaced by thick, irregular bone. Advanced otosclerosis is associated with cochlear ossification (Quaranta et al, 2005). This often leads to a progressive and eventually profound bilateral sensorineural or mixed hearing loss, tinnitus and episodes of sudden hearing loss (Arnold, Kau & Schwaiger, 1999). Also meningitis often results in uni- or bilateral sensorineural hearing loss, varying from mild hearing loss to total deafness.

We expect that the cochlear bone anomalies in both disorders have a similar influence on speech perception after CI. For that reason these disorders are combined into one single ‘bony disorder’ group. Combining the results of both groups will lead to a larger study population and will increase the power of this study. In this paper the results of bony disorders will be compared against all others possible causes of deafness.

In the literature, trends of poorer postoperative speech performance of patients with meningitis as cause of deafness are found. These trends however appeared not to be statistically significant (Dorman, Hannely, Dankowski, Smith & McCanless, 1989; Lehnhardt & Aschendorff, 1993, in Van Dijk et al, 1999; Waltzman, Niparko, Fisher & Cohen, 1995; Battmer et al., 1995). Blamey et al (1996) reported a group of patients with bacterial labyrinthitis to perform significantly worse than patients with other causes of deafness ($p = .004$). But because bacterial labyrinthitis does not always result in meningitis, these results cannot be considered as significant evidence of worse post meningitis speech performance, although it supports the trend. Eshraghi, Telischi, Hodges, Odabasi & Balkany (2004) found no significant differences in mean performance between the meningitis group and the control group. The post meningitis group, however, required progressively higher electrical stimulation levels and more complex programming modes over time as compared to the control group. These observations suggest a more complicated development in the post meningitis groups.
In the literature, trends of poorer postoperative speech performance of patients with otosclerosis as cause of deafness are also found. Also these trends appeared not to be statistically significant (Blamey et al., 1992; Quaranta et al., 2005; Sainz, Valdecasas, Garofano & Ballesteros, 2007; Sainz, Valdesacas & Ballasteros, 2009). Rotteveel et al. (2010) compared speech perception scores within CI patients with otosclerosis and observed wide variability in the results. The rate of progression of the otosclerosis did not influence speech perception.
So, trends of worse speech perception in CI patients with a bony disorder (meningitis or otosclerosis) as cause of deafness are found, but without significant evidence. For that reason we do not expect to find significant differences in speech perception of patients with and without a bony disorder as being their cause of deafness.

Based on the previous literature, we have formulated the following hypotheses:

1. Duration of deafness is a significant predictor of speech perception of adult CI patients: the shorter the period of deafness, the better the results.
2. Preoperative hearing loss is a significant predictor of speech perception of adult CI patients: the less severe the hearing loss before implantation, the better the results.
3. The distinction between a bony disorder and all other causes of deafness is no significant predictor of speech perception of adult CI patients.

Duration of implant use also seems to be a predictor of speech performance (Blamey et al., 1996; Kiefer, et al., 1996). The last hypothesis therefore is:

4. Duration of implant use is a significant predictor of speech perception of adult CI patients.

To our knowledge, in all previous retrospective longitudinal studies of predictors of speech perception in CI-users statistical analyses has been done by use of general linear models (GLM) analyses. However, there are reasons to doubt whether GLM is the most appropriate way to analyze data consisting of several measurements per person.
In a longitudinal study as described in this paper, in which every individual person has been tested at several postoperative times, the observations must be regarded as repeated measures. The scores of these repeated measures are not independent but correlated, because they are nested within individuals. The different CVC scores (repeated measured within-persons) can be seen as level 1. Due to individual differences in speech recognition, the different scores of person A may be more similar to each other than to the scores of person B. This between-person variation can be regarded as level 2 (higher-level).
Such a hierarchical data structure results in multiple random factors affecting the data. The present design has two random factors and three fixed factors. The Intra class Correlation Coefficient (ICC) can be used as a measure of dependency between scores. ICC is the ratio of the between-person variance to the total of variance and represents the proportion of the total variability in the outcome that is attributable to the between-persons situation (Field, 2009). A large ICC means that the within-subject variability is minimal and the between-subject variability is large. This implies independency of the scores and makes a conventional ANOVA inappropriate, because independency of the observations is an important assumption of uni- or multivariate ANOVA-analysis. (Field, 2009)

A repeated-measures analysis of variance (RM-ANOVA) is able to handle a hierarchical data structure, by separating the random variance into a between-subjects variance and a within-subjects variance (Quené & Van den Bergh, 2008).
However Multi Level Modelling (MLM), also known as the hierarchical linear model or variance component model (Quené & Van den Bergh, 2008; Buxton, 2008; Field, 2009; Garson, 2011), has several advantages over RM-ANOVA and proved to be superior over RM-ANOVA. (Quené & Van den Bergh, 2004, 2008; Baayen, Davidson & Bates, 2008; Gelman 2006). MLM is a general form of classical ANOVA and linear regression and can also handle a design consisting of both random factors and fixed factors, but it does not require sphericity, it is more powerful in finding effects and contrasts in data and it is capable of analyzing incomplete data (Quené & Van den Bergh, 2004). Sphericity means that all within-subject variations of scores are equal (Field 2009; Quené & Van den Bergh, 2004), resulting in homogeneity of regression slopes with differences of intercepts, for all residual scores have homogeneous variances. In practice many data sets lack sphericity, which can be tested by Mauchly's test (Field, 2009). The different individual scores result then in individual growth patterns which differ in intercepts and in slopes. MLM allows the intercept and slope to vary from person to person. So it accounts for the variation in the individual measurements (the within-subjects variation) and for the variation between subjects, resulting in a better fit than with a simple linear regression model. All multiple random effects can be considered simultaneously.

In this paper the long-term development of the UMC Utrecht CI patients is evaluated with use of the relatively new Multi-Level Modeling. The effect is examined of several potential predictive factors that have been found to play a role in other studies: cause of deafness (bony disorders: meningitis or otosclerose), hearing loss before implantation, and duration of deafness.

Materials and methods

Subjects

The CVC-participants phoneme scores were extracted from the cochlear implant database of the UMC Utrecht, consisting of data from all persons that applied for cochlear implantation. Excluded were all persons that had not been operated for different reasons, children (implanted before the age of 18;0 years), bilateral CI-users and persons that had been re-implanted for whatever reason. The remaining cohort of 401 persons appeared to be quite incomplete with regard to information about preoperative hearing loss, cause of deafness and/or duration of deafness. For that reason all cases with lacking information about these three variables were excluded from analysis, resulting in a cohort of 247 patients, of which n=111 (44.9%) was male and n=136 (55.1%) female. All persons had been implanted after the age of 18;0 years. The mean age at implantation was 51y6m (SD 14y1m, range 18y;6m-85y;3m). The patient characteristics are summarized in Table 1.

The duration of deafness has been defined as the period between the onset of deafness of the best ear and the CI-operation. The age at onset of deafness has been defined as the time of onset of sudden hearing loss, or in progressive cases, the time since failure to benefit from hearing aids, being determined from the patients' history and by reviewing audiograms. The mean age at onset of deafness was 38y10m (SD 20y;9m, range 0y;0m – 84y;4m). Of all 247 persons, n=18 (7.3%) had become deaf before the age of 2 years, being defined as 'pre-lingually' deaf, whereas n=229 (92.7%) had become deaf after the age of 2, being defined as 'post-lingually' deaf. When the arbitrary age criterion of pre-post lingually deafness was extended to the age of 4 years, n=26 (10.5%) of the persons were pre-lingually deaf and n=221 (89.5%) were post-lingually deaf.

The mean duration of preoperative deafness was 13y2m (sd 14y9m, range 0y;1m-61y;2m). In Figure 1 duration of deafness is expressed in months.

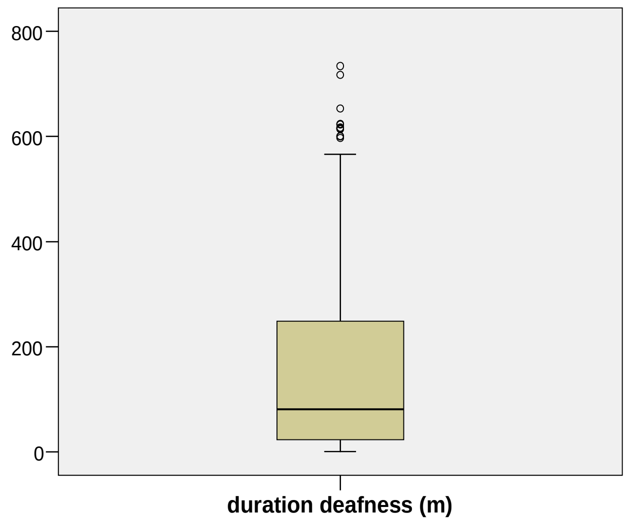

Fig 1. Duration of deafness (m)

To quantify ***preoperative hearing loss*** the mean unaided pure-tone average (PTA) threshold (dB HL) was used, averaging over 0.5, 1, 2 and 4 k Hz of both ears before operation, as it has been shown to be important to incorporate the hearing history from both ears (Roditi et al, 2009). Value 125 dB was used for 37 patients (=15.0%) who did not respond to sound at the upper limits of the audiometer.

In 11 cases a special audiometer was used, with an extended range, showing thresholds up to 140 dB HL. Because of the preference not to change the original data, the effect of these deviated hearing loss data on the average hearing loss was inspected, by comparing the average of the real data with 11 cases >125 dB and the average of the (temporarily) changed data with the maximum of 125 dB HL for all cases. The mean of the first group was 115.0 dB and the mean of the second group was 114.9 dB, which was not significantly different (*p= .773)*. The original data with the data of 11 cases with hearing loss > 125 dB were therefore used for this study. Of the 11 cases with hearing loss > 125 dB 3 cases (= 27.2%) had meningitis or otosclerosis as cause of deafness. This percentage of cases with a bony disorder is quite similar to the percentage of all bony disorders of the total group (= 22.7%).

The pure tone average (PTA) of the left ear was 115.1 dB (sd 11.0 dB; range 78-133 dB), of the right ear 115.0 dB (sd 11.1 dB; range 73-138), with a mean of both ears of 115.0 dB (sd 9.4 dB; range 75-136 dB).

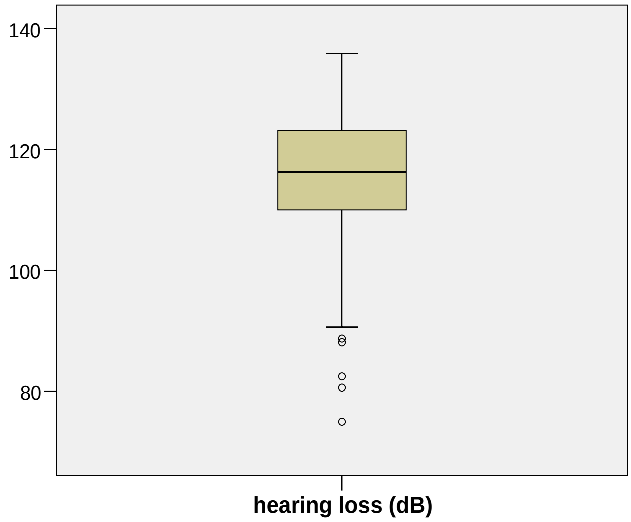

Fig. 2. Hearing loss (dB)

Of all 247 patients, 56 persons (22.7%) had a bony disorder as ***cause of deafness***. For 43 persons (17.4 % of all 247 patients) this was meningitis and for 13 persons (5.3% of all 247 patients) it was otosclerosis. For 191 persons (77.3%) there was a different cause of deafness or the cause was unknown.

Comparison of the mean durations of deafness of patients with a bony disorder and patients with another cause of deafness show that durations between these groups differ significantly (*p*= .004). The mean duration of deafness of persons with a bony disorder was 215.8 months (sd 219.5m) and for the other patients 139.4 months (sd 160.1m).

The mean hearing loss of these groups also differed significantly (*p*= .000). The mean hearing loss of persons with a bony disorder as cause was 119.6 dB (sd 6.9 dB) and for the other patients 113.7 dB (sd 9.6 dB).

Table 1: **Summary of the characteristics of the 247 patients**:

	N	**%**
Gender:		
Male	111	44.9 %
Female	136	55.1 %
Cause of deafness:		
Bony disorder	56	22.7%
Other cause	191	77.3%

	MEAN	**SD**	**RANGE**
Age @ onset of deafness	38y;10m	20y;9m	0;0m - 84y;4m
Age @ implantation	51y;6 m	14y;1m	18y;6m - 85y;3m
Duration of deafness	13y;2 m	14y;9m	0;1m - 61y;2m
Preoperative HL			
AS	115.1 dB	11.0 dB	78-133 dB
AD	115.0 dB	11.1 dB	73-138 dB
ADS	115.0 dB	9.4 dB	75-136 dB

Speech perception testing

The patients were evaluated postoperatively, at least once a year, with a set of speech tests. For this study the test scores of the NVA CVC test (Bosman & Smoorenburg, 1995) were extracted from the database and used as outcome measure. This test consists of lists of 11 monosyllable words, with a consonant-vowel-consonant structure (CVC). The speech material was presented in a free field condition at a level of 65 dB SPL, without lip-reading. The patient's score was the percentage of correctly repeated phonemes of two lists. The scores of six test moments were used in this study: 12, 24, 36, 48, 60 and 120 months after implantation. Data of the years in between were ignored, because of too many missing values.

In this study, when the term at or after *implantation* is used, at or after *switch on* of the cochlear implant is meant, which is generally 6 weeks after the operation.

Data analysis

Exploration of the data showed that CVC scores were not normally distributed. Furthermore, there were a lot of missing values in CVC scores and there was no sphericity (Mauchly's W = 0.146, p=0.000).

For a Multi-Level Model-analysis the R environment for statistical computing and graphics, version 2.12.0. was used (http://www.r-project.org). The linear mixed models were fit by maximum likelihood. The *p*-values (obtained by generating 25 000 simulations of the model) were computed with an extra function (pvals.fnc), which was applied to the fitted model using bootstrapping (MCMC) techniques. (Baayen et al, 2008, p397: 'see Andrieu, de Freitas, Doucet & Jordan, 2003, for a general introduction to MCMC'). A resulting *p*-value of less than 0.05 was considered significant (Baayen et al., 2008). The estimates were based on CVC scores interpolated to time point zero (= at implantation).

Results

The first multilevel linear regression model was constructed without any explanatory variables, resulting in a *random-effects-only* model (= 1st model). Intra-Class Correlation (ICC), used as measure of dependency between the CVC scores, equals the between-subjects variance component divided by the total variance (Garson, 2011). The ICC was 106.74 / (106.74+198.38) = 0.3498. So 35% of the total variance was associated with the CVC outcome variance between persons. This quite large ICC indicates that the CVC scores were indeed correlated within the different persons. Then, this model was extended with the explanatory variables and the effect of time, resulting in a *fixed-and-random-effects* model (= 2nd model).

Table 2, analogous to Janse (2009) and Quené & Van den Bergh (2004), shows the results of the MLM analysis and contains the estimated parameters for slope (= β-coefficients) for the fixed effects, the variances of the two random effects, and the evaluation characteristics of the 1st and the 2nd model.

Table 2:

Estimated parameters for slope for the fixed effects, variances of the two random effects, and evaluation characteristics (of the 1st and the 2nd model: (* = p<.05, ** = p< .01).

The first columns of the random effects represent the variance and the other columns of the random effects represent information about the standard deviation of the variance (mean sd, lower boundary and upper boundary of the 95% confidence interval), in both models.

		1ST MODEL					2ND MODEL				
			MCMC mean	*p* MCMC	CI 95% LB	CI 95% UB		MCMC mean	*p* MCMC	CI 95% LB	CI 95% UB
Fixed effects	Intercept (CVC-score time point 0:										
	1st Model: all patients	53.67	55.09	< 0.001**	53.42	56.69					
	2nd Model: bony disorder-group						42.50	42.04	0.000**	36.06	47.83
	Cause (= Group 'other cause')						10.47	9.88	0.004**	3.25	16.56
	Duration of deafness						-0.03	-0.03	0.000**	-0.04	-0.02
	Hearing loss						-0.48	-0.44	0.006**	-0.75	-0.13
	Time						0.23	0.37	0.002**	0.15	0.59
	Time²						-0.001	-0.002	0.007**	-0.00	-0.00
	Time * cause						-0.11	-0.12	0.362 ns	-0.37	0.13
	Time² * cause						0.00	0.00	0.312 ns	-0.00	0.00
	Time * hearing loss						-0.01	-0.01	0.211 ns	-0.02	0.01
	Time² * hearing loss						0.00	0.00	0.100 ns	0.00	0.00
Random	Patients variance (= between patients)	106.74	10.33		9.48	11.19	89.92	9.48		8.71	10.25
	CVC-scores variance (= within pat.)	198.38	14.09		13.23	14.96	146.37	12.88		12.10	13.69
Evaluation	Log likelihood	-3481					-3387				
	Chi-square (compared to 1st model)						188				
	p=deviance (compared to 1st model)						< 0.01				

The results of the upper part of Table 2 (fixed effects) show that perception of CVC words after CI is significantly predicted by cause of deafness, by duration of deafness and by preoperative hearing loss. Scores are expressed in percentage of correctly repeated phonemes. With the 2nd model, the estimates of the fixed effects, interpolated to time zero (= at implantation), result in the following predictor model:

$$_{pred}CVC\% = 42.50 + 10.47*OC - 0.03*Dur - 0.48*HL + 0.23*T - 0.001*T^2$$

Explanation: *OC= other cause of deafness than bony disorder*
Dur= duration of deafness, in months
HL= degree of preoperative hearing loss, in dB
T = time (= duration of implant use, by linear growth model)
T^2= time ^{2} (= duration of implant use, by quadratic growth model)

The intercept value in the 1st model represents the estimated average CVC score of all patients at time zero, whereas the intercept value in the 2nd model represents the estimated average CVC score of the bony disorder-group at time zero. The other cause group, consisting of persons with a different cause of deafness than meningitis or otosclerosis, has an estimated average CVC score of 42.50 + 10.47 = 52.97 at time zero.
There is a negative relationship between the duration of deafness and the CVC scores and between the preoperative hearing loss and the CVC scores. The longer a person has been deaf before implantation, the lower the average speech performance after implantation. The more profound the hearing loss before implantation, the lower the average speech performance after implantation.
The development of speech perception over time is best described by a linear and a negative quadratic growth model.
There is no effect of interaction of the explanatory variables, nor among themselves nor with the time predictors.

The second part of Table 2 (random effects) shows that the total residual variance decreased with adding the fixed effects to the model by 68.83 units of variance. The between-subjects variance was reduced when the explanatory variables were added, by 16.82 units of variance. The within-subjects variances were reduced when the effect of time was added, by 52.01 units of variance. The effects of time described only part of the variance between CVC scores. There still remains a large amount of unexplained variance (146.37 units).

The evaluation part of Table 2 shows that the 2nd model has a much better fit than the 1st model. As already mentioned, the linear mixed models were fit by maximum likelihood. The goodness of fit was evaluated by the likelihood ratio test, for this only covers the variance explained by the fixed effects instead of the R^2 that should also cover the random effects (Janse, 2009). 'The difference between the two log likelihoods, multiplied by 2, follows a chi-square distribution with the difference in number of parameter as number of degrees of freedom' (Janse, 2009, p 2369). The results of this calculation show that the 2nd model has a much better fit than the first model ($p < 0.01$).

The results of Table 2 are visualized in Figure 3, at percentiles 25, 50 and 75, being for hearing loss respectively 110, 116 and 123 dB, and for duration of deafness being respectively 23, 81 and 254 months.

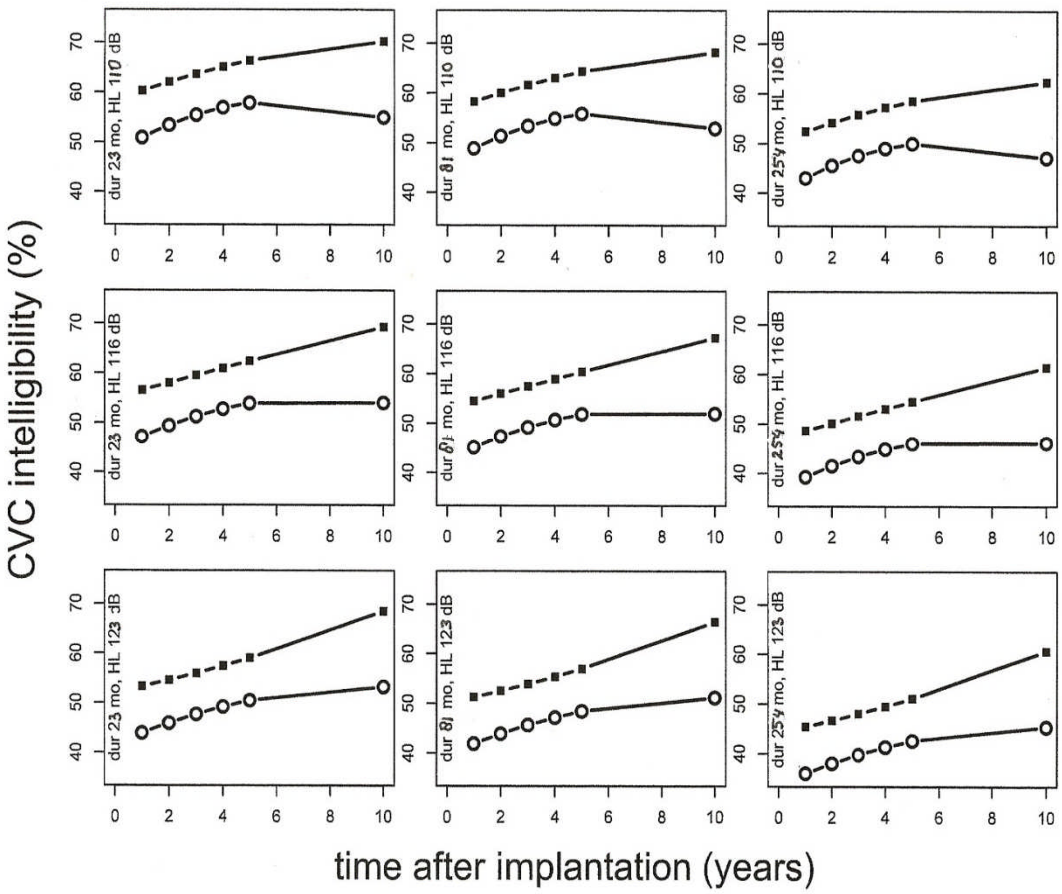

Fig. 3: Visualization of the model: predicted CVC scores against time after implantation, showing the effects of the factor preoperative hearing loss, being constant in rows and increasing in columns, and of the factor duration of deafness being constant in columns and

increasing in rows. The factor yes/no bony anomalies of the cochlea is symbolized by yes=circles and no=solid squares.

Figure 3 clearly shows the significant difference between the average CVC scores of both cause groups of about 10%, in accordance with the estimate of the intercept. The slight decrease of the CVC scores of the bony disorder group after 5 years of CI-use (in the upper three graphs) however, appeared not to be significant.

Discussion

The main predictors, viz. time, cause of deafness, duration of deafness and degree of hearing loss all yielded significant main effects on speech perception after cochlear implantation. None of the 2-way interactions between these variables were significant.

The 1st, 2nd and 4th hypothesis, as formulated in the Introduction, were therefore confirmed. The finding that time (Blamey et al., 1996; Kiefer et al., 1996), duration of deafness (Shea et al., 1990; Blamey et al., 1992; Gantz et al., 1993; Battmer et al., 1995; Blamey et al., 1996; Albu et al., 1997; Van Dijk et al., 1999; Friedland et al., 2003; Blamey et al., 1996; Rubinstein et al., 1999; Gomaa et al., 2003; Oh et al., 2003; Green et al., 2007; Klop et al., 2008) and preoperative hearing loss (Summerfield et al., 1995; Rubinstein et al., 1999; Van Dijk et al., 1999; Gomaa et al., 2003; Francis et al., 2005; Neumann et al., 2008) are significant predictors of postoperative speech perception after cochlear implantation, was in accordance with most studies in the literature.

The 3rd hypothesis, on the contrary, which was formulated as ‘The distinction between a bony disorder and all other causes of deafness is no significant predictor of speech perception of adult CI patients’, was rejected. The cause of deafness appeared to be a significant predictor of the success of speech perception after CI. This result was unexpected and it was not in accordance with most of other studies. Dorman (1989), Lehnhardt & Aschendorff (1993, in Van Dijk et al.,1999), Waltzman et al. (1995) and Battmer et al. (1995) found only trends of poorer postoperative speech performance of patients with meningitis, as did Blamey et al. (1992), Quaranta et al. (2005), Sainz et al. (2007 & 2009) for patients with otosclerosis. In all these studies, these trends were not statistically significant.

One possible explanation for this unexpected difference with the literature is the use of Multi-Level Modeling in this study. As mentioned earlier, MLM has higher power in finding effects and contrasts in data as compared to other analysis tools (Quené & Van den Bergh 2004, 2008; Baayen et al, 2008; Gelman, 2006). Because MLM accounts for error non-dependency, it is possible to make more accurate inferences about the fixed effects (Albright & Marinova, 2010). As MLM is recommended in the literature as a method to increase the power in hypothesis testing (Quené & Van den Bergh, 2004), it is possible that the difference between the insignificant effects in previous studies and the significant effect in this study results from the higher sensitivity of the MLM method used in this study. Maybe the insignificant trends of poorer speech performance of patients with meningitis or otosclerosis as cause of deafness found in previous studies, would become significant effects if the MLM method for data analysis had been used.

A second possible explanation for this favorable difference with the literature is the fact that both cause groups in our study not only differed with regard to the cause of deafness, but also differed with regard to the preoperative hearing loss and duration of deafness. These two latter predictors both appeared to be related to speech perception performance after CI. This confounding fact might have affected the results. The significant difference between the CVC-results of both cause-groups might (partly) be due to their difference in hearing loss and duration, and not completely to the different causes (the size of this effect can be estimated with the model, which asks for another analysis).

A third possible explanation for this unexpected difference is the fact that the bony disorder group possibly was less homogenous than thought at first. Meningitis and otosclerosis were in this study combined to a single cause group, because we expect that the cochlear bone anomalies in both disorders have a similar influence on speech perception after CI. However, the effect of meningitis and otosclerosis on auditory performance after CI may also differ structurally. In contrast to otosclerosis, meningitis also seems to reduce the number of ganglion cells substantially (Nadol, Young & Glynn, 1989). Although the role of ganglion cell survival is up for debate and not found to be the critical factor in success of speech perception in CI as thought before (Blamey et al, 1996), Blamey et al concluded that ganglion cell survival ‘may play a role in determining auditory performance’ (1996, p 304). So, although there are valid reasons to combine meningitis and otosclerosis to a single cause group, there are also reasons to question this approach.

The development of speech perception over time was best described by a growth model with linear and a negative quadratic components. After a period of growth of speech perception, the results flatten out and decrease. Decrease of quality of the cochlear implant is no presumable explanation for this decrease of speech perception. Fittings of the speech processor take place yearly and the processor is being replaced every 5 year. A more presumable explanation might be the age of the patients. With a mean age at implantation of 51 years, the mean age at 5-10 years after CI use is 56-61 years. Age-related problems like general cognitive decline and decrease of information processing speed might play a role in speech perception decline. This is in accordance with Blamey et al. (1996), who declared the observed drop in performance of the CI patients as ‘a reflection of reduced central processing capabilities in patients after the age of 60 years’ (1996, p 304). This central processing component in speech processing of elderly persons is also found by Jerger, Chmiel, Allen & Wilson (1994), Van Rooij & Plomp (1990) and Janse (2009).

After the MLM-analysis, there still remains a large amount of unexplained variance (146.37 units). For that reason the outcome prediction of this study has to be applied with considerable care for individual cases. As already mentioned in the Introduction, the causes of the wide variation in outcomes between individual CI-users are only partly understood at the present time (Peterson, et al., 2010). Many already known and still unknown factors play a role in the complex process of speech perception of CI-patients.
As prelingually deafened adult implant recipients tend to achieve lower maximum performance of speech perception than postlingually deafened adults (Teoh, Pisoni & Miyamoto, 2004), an explanation for the large amount of unexplained variance in this study might be the fact that in this study no distinction was made between pre- and postlingually deafness. However, as only about 10% of the persons in this study can be defined as ‘pre-lingually’ deaf, the effect of this factor on the results is expected to be limited.

Conclusion

The main predictors, viz. cause of deafness, duration of deafness, preoperative hearing loss and time all yield significant main effects on speech perception after cochlear implantation. The 2-way interactions between these variables however are not significant, nor among themselves nor with the time predictors. The development of speech perception over time is best described by a linear and a negative quadratic growth model. The resulting predictor model is: $_{pred}CVC\% = 42.50 + 10.47*OC - 0.03*Dur - 0.48*HL + 0.23*T - 0.001*T^2$. (for explanation: see *Results*). We add that a large amount of the outcome variability was not explained by our model. So, the outcome prediction above has to be applied only with considerable care for individual cases.

As Multi-Level Modeling has been demonstrated in previous studies to be more powerful in hypothesis testing than other analysis tools, our unexpected result of cause of deafness being a significant predictor of speech perception might be due to the higher sensitivity of the Multi-Level Modeling method.

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