

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

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## ABSTRACT

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

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

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

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

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**TRIAL REGISTRATION:** not relevant.

The implementation of universal neonatal hearing screening (UNHS) and the increased use of bilateral cochlear implants (CI) have provided more and more children with profound hearing loss access to bilateral auditory signals during their first year of life [1, 2]. Recent reports indicate that unilateral cochlear implantation in prelingually deaf children within the first year of life may result in speech and language skills comparable to those of children with normal hearing [3-6]. In Denmark, UNHS was implemented in January 2005, and children have been offered bilateral implantation either simultaneously or sequentially as from September 2006. It has

previously been reported that outcomes of CI were associated with the Danish Region from which the children originated [7], and it is therefore of great interest to study whether such regional differences still exist after the introduction of UNHS and bilateral implantation. This new group of children with CI has now reached an age where testing of outcomes of audition, speech and language is possible. The aim of this study was to investigate whether regional differences in CI outcome still exist between East and West Denmark.

## METHODS AND MATERIAL

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

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TABLE 1

Characteristics of all 94 cochlear implant recipients

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

TABLE 2

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

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

CAP = capacity of auditory performance

CI = cochlear implant

PPVT-4 = Peabody Picture Vocabulary Test-4

SIR = speech intelligibility rating

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

The participating families came to one of the two paediatric CI centres for testing accompanied by one or both parents. Four speech- and language pathologists, two from each centre, carried out the tests and the parental interviews. All testers used spoken language and

all test results were scored according to standards from normally hearing children. **Table 2** summarizes all tests and assessments used, the age criteria for each test, number of responses and standardised test categories for scoring.

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

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

### Data analysis

The data distribution for all tests and the regional differences are presented. For comparison of categorized outcome between East and West, the  $\chi^2$  test or Fisher's exact test (when n > 5 in any category) was used. A stat-

istical significance level of 5% was chosen. Potential rater variability within either of the centres was assessed using logistic regression models.

*Trial registration:* not relevant.

## RESULTS

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

## DISCUSSION

Children with CI in Denmark can hear and discriminate just as well as children reported in international studies

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

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



**TABLE 3**

Distribution of test results in relation to geographical region.

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



**TABLE 4**

Regional differences of rehabilitation of the 83 tested recipients.

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



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

in East) may partly explain their poorer outcome [13, 14]. The documented differences in parental communication mode, parental participation, educational placement, number of rehabilitation and learning support assistant hours, however, suggest that the most likely explanation for the significant differences in CI outcome in Denmark lies in the different organisations of the Danish paediatric CI population. In East Denmark, the primary guiding comes from the CI centre in close collaboration with the local speech and language therapist. In West Denmark, the former schools for the deaf play an important role in the guiding of the parents. In the West, children are referred to the paediatric advisory services at the former schools for the deaf once the diagnosis of hearing impairment has been established at the audiological hospital clinic. These advisory services at the former schools of the deaf with a core tradition of teaching sign language to deaf children are thus the parents' first-hand rehabilitation contact; their contact is not with the team at the CI centre, which focuses on and stresses the evidence-based importance of the use of spoken language and parental involvement in the rehabilitation of the children with CI. This fact probably explains why 24% of the parents in West Denmark as opposed to 7% in East still were using a combination of spoken language and signs for their child, even though the detrimental impact of this on the outcomes of speech/language and social well-being is well documented [7, 15]. Some may argue that West children do not get enough speech and language therapy after implantation, and that they do not have enough learning support assistance. This study does, however, question the value of the contents and the absolute amount of provided hours of learning support assistance, as well as speech and language therapy provided by advisory services at the former schools for the deaf. The West population received markedly more hours of rehabilitation and support teaching, even though they do not have

more frequent or more severe additional handicaps and although they hear and discriminate as well as their East counterparts. However, as presently indicated, this has no positive effect on their outcome. This questions the contents, quality and socio-economic value of these efforts.

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

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