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**Article:**

https://doi.org/10.1016/j.ijporl.2017.06.031

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Abstract

Objective: The aim was to explore PA skills German-speaking preschool children with cochlea implants (CIs) and how these skills may be related to their speech and language skills.

Methods: Three monolingual German-speaking pre-school children aged 5;04 – 6;01 with bilateral CIs were tested. Their cognitive, speech and language skills were assessed. Six subtests of a standardized PA test battery were administered (i.e. rhyme identification, rhyme production; phoneme identification- input and -output; phoneme blending-input and -output).

Results: All three children showed distinctive PA profiles. One boy, who had no spoken language deficits, struggled to complete the rhyme tasks but performed well on three phoneme tasks. However, he showed a discrepancy between expressive and receptive phoneme blending skills, scoring poorly on the expressive subtest. The second boy, who displayed grammar comprehension and expressive vocabulary difficulties, showed a mixed profile, with a below average performance on rhyme production. The girl who had significant speech and language deficits scored below average on all six PA subtests.

Conclusions: PA profiles in children with CI vary considerably and PA testing should include a range of different PA tasks. The assumed link between spoken language deficits and PA difficulties shown in children with normal hearing could be confirmed.

Keywords: children with CI, phonological awareness, German-speaking
BACKGROUND

There are a considerable number of people with sensorineural hearing loss. To date approximately 300,000 people world-wide have been provided with cochlear implants (CI) of which more than 25,000-30,000 people received CIs in Germany. Current numbers show that each year approximately 3,000 people in Germany undergo CI-surgery [1]. The German Federal Statistical Office reports that in total 3,707 people between birth and the age of 95 received a cochlear implant in 2015. 62 of them received a cochlear implant with several electrodes (e.g. double array), 238 with signal electrode, but not hearing preservative, 396 with a signal electrode hearing preservative and 9 received a cochlear implant without any specific information. In total, 705 of them were children with sensorineural hearing loss aged between birth and fifteen years [2]. Studies which examined the speech and language development of children with and without sensorineural hearing loss, report different developmental trajectories [3-7]. For example, there is empirical evidence that a considerable number of children with CI show vocabulary deficits [7]. Vocabulary growth is an important factor in the storage of word forms [8, 9]. The more words a child learns, the more detailed the stored information about the words need to be. For example, the words <house> and <mouse> can only be differentiated if the child can recognize their different onsets (i.e. /m/ and /h/).

Another skill which is often affected adversely is phonological awareness (PA). PA is the skill to manipulate the word form independently of its meaning [10]. It is a complex metalinguistic skill which can be assessed on different linguistic units, i.e. syllable, rhyme, and phoneme level. In addition, different levels of explicitness can be differentiated, i.e. identification, segmentation, blending, and manipulation of linguistic units. The developmental progression continues from syllable, to rhyme, and phoneme level and children are first able to identify sounds (e.g. “Fish, dog, foot – which words sound the same at the beginning?”), followed by segmentation and blending. The most difficult task is to manipulate the word form (e.g.
“What is the word you get when you say ‘cat’ without /k/”? PA, in particular phoneme awareness, is an important prerequisite for literacy acquisition and therefore a crucial skill to acquire during preschool and school age. Children with speech processing difficulties and expressive phonological impairments are at risk for PA deficits [11, 12]. There is a considerable number of children with CI who show speech perception and production difficulties and empirical studies found that children with CI show weaker PA skills than typically developing children [13-15]. However, large variability in test performance for PA and other language variables are evident as well. Factors such as age of implantation and parental/carer language support may have a considerable influence on children’s language performance [16-19]. Therefore, children with CI need close monitoring of their spoken language acquisition, including PA, to allow early identification of speech and language difficulties and, if needed, to be provided with specific speech and language therapy. Until recently most of the research has focused on English-speaking children. Little is known about PA skills in preschool CI-children speaking a language other than English and there is a lack of data on PA development in German-speaking preschool children with CI. Therefore, this pilot study aimed to assess PA performance in German-speaking preschool children with CI and to explore their individual PA profiles in relation to their language skills. Three preschool children aged 5;04 – 6;01 with profound hearing impairment and bilateral CIs were included. They had not entered formal education at the time of testing (note: children are on average around the age of six when they start formal school tuition in Germany).

**METHOD**

**Participants**

All three children were monolingual German-speaking pre-school children, two children are male, one is female (referred to in the following as M1, M2, and F1 respectively). M1 was
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6;01 at the time of testing, M2 5;04, and F1 6;0. All children suffer from sensorineural hearing loss but had no additional physical or neurological disabilities. All children were fitted with bimodal hearing aids and later with bilateral cochlear implants (MED-EL SONATATI100, speech processor OPUS 2). Detailed characteristics of the children, including age of implantation and their audiological data, can be found in Tables 1 and 2 respectively.

Table 1

Table 2

Material

Cognitive, speech, and language skills

The following assessments were administered to test the children’s cognitive, speech and language skills:

1. **SON-R 2½-7 [20]**: This standardized [21], nonverbal assessment for children aged 2;6 – 7;11 was used to assess cognitive skills. It comprises six subtests, each with 15 items, assessing spatial visualization (subtests: Mosaics, Patterns), concrete reasoning (subtests: Puzzles, Situations) and abstract reasoning (subtests: Categories, Analogies). Participants receive feedback after each item, with the examiner providing support if a task is not solved correctly. The test takes between 50 and 60 minutes.

2. **TROG-D [22]**: The German version of the well-stablished TROG (Test for Reception of Grammar, Bishop, 2003) was chosen to assess receptive grammar skills. The test is standardized for children aged 3;0 – 10;11 and comprises 21 grammatical constructs, each of which is tested by a four picture identification paradigm. Participants choose the picture which matches the examiner’s statement. In addition to the target picture, there are also grammatical and lexical distracters for each item.

3. **PLAKSS [23]**: This speech assessment tool provides an overview of a child’s phonetic and phonemic speech inventory, including phonological processes. Normative data are available for children aged 1;6 – 6;0. There are two subtests: a picture naming task and
a 25-word inconsistency test. The former tests 99 items, which cover all sounds of the
German phoneme inventory in all positions (word-initial, -medial, and -final), the
latter comprises 31 picture maps, which participants are asked to describe.

4. **WWT** [24]: The test assesses semantic and lexical skills via two components: WWT-
expressive, a picture naming test, and WWT-receptive, a follow-up task in which
participants’ receptive vocabulary is tested based on the items that could not be named
in the first task. Normative data for children aged 5;6 and 10;11 is available and its
internal reliability varies between $\alpha = .90$ and $\alpha = .92$ depending on age group. WWT-
expressive was used as an expressive vocabulary measure for M1 and F1. Since M2
was younger and age norms for his age were not available, an alternative standardized
assessment for expressive vocabulary was administered (i.e. AWST-R, [25]).

**Phonological awareness skills**

Six out of eleven subtests from a standardized PA test battery for German-speaking preschool
children were administered [26]. Input and output tasks were included to allow comparisons
between receptive and expressive PA skills. For each subtest three practice and twelve test
items were presented:

1. **Rhyme-identification-input** (RhymeIDin): A stimulus word is depicted at the top of the
page and three choice pictures underneath it. Children point to the picture of the word
that rhymes with the stimulus word. Apart from the correct answer, a phonological
distracter and a semantic distracter are depicted.

2. **Rhyme-production-output** (RhymeProdout): Children are asked to produce as many
words as possible that rhyme with the stimulus word. There is a time limit of fifteen
seconds for each item. The children are instructed to produce real rhyme words or
rhyming pseudowords.

3. **Sound-identification-input** (SoundIDin): The stimulus word is depicted at the top of
the page and the three possible choices underneath it. Children point to the picture of
the word that shares the initial sound(s) with the stimulus word. Apart from the correct answer, a phonological distracter and a semantic distracter are depicted.

4. **Sound-identification-output** (SoundIDout): Children are presented with pictures of pairs of words. Both words in each pair share either a single consonant onset (C), a two consonant onset cluster (CC), or the first consonant of the consonant onset cluster (CC). Children pronounce the shared sound(s).

5. **Sound-blending-input** (SoundBlendin): The target word is presented by the tester, segment by segment. The pause between each sound is one second. Three pictures representing the target word and two distracters are then presented to the children, who are asked to point to the right picture. One distracter matches the onset of the target word, the other its coda. They share the same number of syllables and, if possible, the same number of sounds as the target.

6. **Sound-blending-output** (SoundBlendout): Children are asked to produce a word by blending the sounds spoken by the tester. The pause between each sound is one second. The words used range from 2-5 segments in length. No pictures are presented.

For a detailed description of the design of the subtests see Schaefer et al. or Fricke & Schäfer [26, 27].

**Procedure**

The data for this pilot study were collected as part of a bachelor thesis of one of the authors and the project was reviewed by an internal ethics committee (Catholic University of Applied Sciences). The participants were recruited from the Department for ENT and Communication Disorders, Mainz, Germany. Information leaflets were sent to the parents/care givers, who were then contacted via telephone a few days later. All parents were happy for their child to participate and returned the signed consent form to the research team. The assessments were carried out in a quiet room at the children’s home. Parents or another care giver were allowed
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to attend the test session. Each child was seen for one session to test their phonological awareness skills, lasting 45-60 minutes. All sessions were scored online and audio recorded for later checking. Test scores for their cognitive, speech, and language skills were taken from their patient history with the permission from their parents/care givers and the clinic (Department for ENT and Communication Disorders, Mainz Germany).

RESULTS

Table 3 summarizes the results of the cognitive, speech and language assessments. M1 and M2 showed age appropriate cognitive skills. F1 could not attend the SON-R testing. However, her case history did not reveal any issues regarding her cognitive skills.

Table 3

M1 scored within normal range on both language measurements and showed age appropriate speech skills. M2 displayed language difficulties, scoring low on the comprehension test (TROG-D) and in the expressive vocabulary assessment (WWT). In addition to a language deficit F1 showed a speech impairment. She showed systemic speech errors, including fronting of velar plosives (/k, ɡ, ʪ / → [t, d, n]), fronting the postalveolar fricatives /ʃ/ to [s, z], backing the consonant cluster /tr, dr/ to [kʁ, ɡʁ] (e.g. /trepa/ <stairs> to [kʁɛpa]), and backing of the alveolar plosives /t, d/ to [k, ɡ]. Structural phonological processes were also observable, in particular assimilations and reductions of single consonants and consonant clusters in syllable onset or coda position. In summary, F1 showed a severe speech impairment, including delayed and deviant phonological processes.

Table 4

Table 4 summarizes the PA scores for all three children, including their percentile rank range. M1 struggles with both rhyme tasks. Scores for the Sound-identification subtests are age-appropriate. The Sound-blending tasks show a discrepancy between input and output. M1 scores highly on the input task but performs below average on the equivalent output task. M2
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shows a discrepancy between the input and output task on the rhyme tests. While his performance on the Rhyme-identification-input task is age-appropriate, he struggles to complete the Rhyme-production-output task. No problems are observable in any of the Sound-identification subtests. Some additional differences between input and output performance can be seen in the Phoneme-blending tasks. M2 scores higher and within age norms on the output task. F1 shows consistently poor PA skills across all six subtests. Her percentile scores are all significantly below average. Tables 5 – 7 provide an overview of the individual phonological awareness profiles.

DISCUSSION

The aim of this pilot study was to explore children’s individual PA profiles in relation to their speech and language skills. Three very different profiles could be observed. These differences cannot be explained by differences in IQ, since all children showed age-appropriate cognitive skills.

M1 struggled considerably with the Rhyme-identification-input and Rhyme-production-output tasks and scored well below average. It seems that he has not acquired the principle of the onset-rhyme level yet. He is neither able to identify the rhyme words within a choice of three possible answers, nor does he manage to produce rhyme words independently. Since his vocabulary and speech skills are age appropriate and he is able to complete PA tasks on the phoneme level, it is assumed that the problem is not caused by speech or language deficits, but that it may be a problem related to the acquisition of the metalinguistic knowledge of the onset-rhyme level. In contrast, he scored highly on the Phoneme-identification-input/-output and Phoneme-blending-input task. This shows that he could perform equally well as his age-equivalent normal-hearing peers on those subtests and that children with CI do not necessarily
struggle to acquire PA skills. This supports earlier studies which found no differences in children with CI and normal-hearing children [28]. Three aspects might have contributed positively to that result. Firstly, his CIs were fitted during his first year of life, i.e. at a young age. Previous research has demonstrated that early implantation impacts positively on children’s speech and language development [29]. Secondly, M1 showed age appropriate speech and language skills, providing a good basis for the acquisition of metalinguistic skills. Thirdly, he had received extensive intervention from an early age, focusing on speech perception and production. Many speech perception and discrimination tasks would require the identification of onsets/sounds of different words. Hence, his good scores might be a result of intensive speech sound training during speech and language therapy. Despite his good results on three of the four phoneme tasks, he significantly struggled on the Phoneme-blending-output task. In this subtest no pictures are provided and therefore the auditory stimulus must be kept in short term memory in order to successfully complete the task (e.g. the child hears the stimulus word /z-a-l-a:-t/ and has to blend the single sounds to the word /zala:t/, <Salat>, i.e. <lettuce>). One explanation for M1’s poor performance might be short-term memory difficulties. However, since no short-term memory task was included in the test battery, this assumption could not be confirmed. The discrepancy between input and output shows that test design can considerably impact on PA performance. Moreover, it is important to account for memory load and to differentiate between subtests that require a verbal versus nonverbal response.

M2 showed a more mixed profile than M1. He scored within normal range on the Rhyme-identification-input task but struggled with the Rhyme-production-output task. This task requires children to produce rhyme words matching the provided stimulus word. As pictures are presented, this reduces the working memory load and potential working memory problems could not be assumed for M2. In addition, he performed well on the Phoneme-blending-output task (the task without pictures), assuming that holding a word in his short-term memory to
complete a task was not a problem for him. However, the rhyme output task not only tests rhyme production skills but also includes lexical retrieval. Although children are allowed to produce both words and pseudowords (e.g. “what rhymes with fish?” – correct answers may be: “gish, wish, lish”), children with a large, well-organized lexicon might have an advantage in comparison to children with vocabulary deficits. M2 showed impaired expressive vocabulary skills which might have contributed to the poor results on the expressive rhyme task. This assumption is in line with research showing that vocabulary skills are closely linked to PA performance [32-34].

F1’s PA performance was consistently below average on all six PA subtests. Similar to M2, her language deficits might have adversely influenced her outcomes. Moreover, it is assumed that her distinct and persistent speech impairment has significantly impacted on her PA performance. This is in line with research focusing on children without hearing impairment but with speech difficulties, which showed that children who displayed delayed or impaired phonological processes are likely to show PA deficits [12, 35]. F1’s results also corroborate the assumption that a combination of speech and language difficulties adversely affects PA skills [36, 37].

In sum, the presented case examples reflect the variability in PA performance that children with CI may display, even in a small population sample. It also corroborates the view that comprehensive PA profiles (i.e. a range of PA subtests) are needed to identify strengths and weaknesses within this complex metalinguistic skill. Task demands must be considered and both receptive and expressive PA subtests must be differentiated to establish a comprehensive profile of PA skills. Results from M2 and F1 have supported earlier research which suggests that children with additional speech and language deficits are likely to show PA deficits and, consequently, are at higher risk for literacy difficulties. However, group studies with substantially more participants are needed to further investigate PA skills in children with CI to confirm that co-morbid speech and language deficits impact similarly on PA as has been
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shown in cohorts of children without hearing impairment.

Acknowledgements

The authors gratefully acknowledge Prof. Dr. med. Annerose Keilmann and Dr. Karl Lippert for their assistance during the recruitment on behalf of the Department for ENT and Communication Disorders, Mainz, Germany. In addition, they would like to thank the parents, carers, and the children who took part in the project for their support.

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