Child Neurology Open
Volume 8: 1-8

DOI: 10.1177/2329048X211022196
journals.sagepub.com/home/cno
SAGE

Cognitive linguistic Treatment in Landau Kleffner Syndrome: Improvement in Daily Life Communication

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Abstract

We report a case study of cognitive linguistic treatment in a teenager with chronic severe Landau Kleffner Syndrome. The effect of speech and language therapy in LKS is rarely examined and our case is unique in that we use an effective approach in adult aphasia to treat language deficits in aphasia in LKS. The results show successful acquisition of a considerable amount of new words as well as improved communication in daily life. However, auditory verbal agnosia, the most prominent feature in LKS, persisted. Cognitive linguistic treatment seems a promising treatment to improve spoken language production in LKS, but more research is needed to optimize speech and language therapy of auditory verbal agnosia and auditory language comprehension in children with LKS

Keywords

Landau Kleffner syndrome, speech and language therapy

Received August 13, 2020. Received revised December 30, 2020. Accepted for publication April 28, 2021.

Introduction

Landau Kleffner Syndrome (LKS) was first reported in 1957 as "acquired aphasia with convulsive disorder".[1] LKS is an extremely rare form of acquired childhood aphasia (ACA). With the exception of one study, the literature does not provide information on the incidence or prevalence of LKS. Kaga and co-workers estimated the incidence at about 1 case in a million children in Japan.[2]

Typically, LKS manifests itself between the age of 3 and 7 years. Normally developing children show an acute or gradual breakdown of language, in association with epileptic symptoms. Not all children have clinically manifest seizures, but their EEG during sleep shows typical abnormalities with bilateral or unilateral spike waves originating in language-related areas. Epileptic symptoms may precede the aphasia, but the aphasia may also be the first symptom. The epilepsy is treated with anti-epileptic medication, with steroids or adrenocorticotrophic hormone (ACTH). This may lead to a rapid improvement of language functioning.[3]

The epileptic discharges are thought to disrupt the language system at different locations and at different stages of language development. In contrast to aphasias caused by structural brain damage, patterns of language breakdown and recovery after LKS are variable and difficult to predict. Some children show marked fluctuations in language functioning, with more than one phase of language breakdown, followed by a period of recovery.[4] In severe cases, the language disorder is permanent, with limited or no recovery.[5] However, complete recovery is also possible, sometimes after several years. In a long-term follow-up study of 29 patients, 8 patients recovered completely; the remaining patients had persisting language deficits of different degrees.[6] In a review paper, Baumer and colleagues

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noted that 45-70% of children with LKS suffer from permanent language deficits.[7]

In the vast but scattered literature, with more than 400 English case descriptions[2], the focus is on medical issues such as etiology, epileptic manifestations and medication.[8] In contrast, the issue of rehabilitation, and especially the contribution of Speech and Language Therapy (SLT) has received surprisingly little attention.[9] As a result, clinicians have very little or no evidence to base their treatment decisions upon, and it remains unclear whether or when language training is a realistic rehabilitation option.

The most prominent and often also the first sign of language breakdown in LKS is auditory verbal agnosia or word deafness, i.e. the inability to process auditory language input, in the context of normal hearing. Several studies have shown that this core deficit in LKS results from an auditory temporal processing impairment.[9–11] Children with LKS are unable to process rapidly changing frequencies in an auditory signal, resulting in the inability to identify strings of phonemes or words in speech. In adults, auditory verbal agnosia is a modality specific disorder that affects auditory processing of language, without affecting the production of spoken language, and without affecting written language. In children with LKS, the initial selective breakdown of auditory processing of language is followed by a further breakdown of language, affecting language production as well as language development. The spontaneous speech of children with LKS is characterized by short and syntactically simplified utterances, severe word finding difficulties, articulatory difficulties and severe phonological paraphasias, such as cluster reductions and omission of syllables.[4,12] Often, their speech is unintelligible. Some children do not speak at all for a period of time. For children who are still developing language, intact auditory processing is crucial. For instance, in the process of expanding vocabulary, a child acquires new words by hearing them being used in context. Auditory processing is also a basic component for phonological processing, and thus for reading and writing.

In adults with acquired language problems after brain injury, SLT is often based on cognitive linguistic language models, such as the one developed by Ellis & Young.[13] This approach starts with thorough diagnostics to determine which language components are impaired and to what extent. Based on these findings, disorder-oriented language treatment follows, targeting the impaired language components. In adult aphasia, this approach has proved to be highly effective.[14,15] With a few exceptions,[16–18] there is little attention for disorder-oriented auditory training in children with LKS and to our knowledge, a cognitive linguistic rationale in examining and treating language deficits has not been applied in LKS. In this case report, we focus on the benefits and limitations of cognitive linguistic language training in severe chronic LKS. We examine whether such a language therapy improves the affected linguistic components as well as daily life communication. The boy and his parents gave written informed consent to publish this case study.

Patient and Methods

Case Report

In this right-handed boy with normal early development, including language development, language started to break down at the age of 3;9 years, following the typical pattern seen in LKS. First, his comprehension of spoken language deteriorated, which was attributed to hearing loss. Audiological assessment at the age of 4;3 revealed normal hearing. At that time not only language comprehension was severely affected, but his language production had seriously deteriorated as well. At the age of 4;6, a 24 hr EEG registration plus video showed a well-developed background pattern with severe abnormalities (peak waves) left temporal, and less severe abnormalities more central and parietal. During sleep these sharp peaks became more frequent, but there was no complete pattern of continuous spike and wave during sleep (CSWS). The video registration showed no seizures. Based on the EEG pattern and the clinical picture of verbal agnosia and aphasia, he was diagnosed with LKS.

Medical treatment with Prednisolone (1mg/kg bodyweight, 6 weeks) had no effect on language functioning, also not after increasing the dose. Treatment with antiepileptic drugs (AED) and an experimental treatment with intravenous immune globulines also remained without effect. Over the years, he never had any seizures. At the age of 5;11, a MEG was performed to localize the focus of epileptiform activity before the subpial transection. The results revealed many interictal spikes, considerably more left than right, with foci in the superior temporal gyrus, near the supramarginal and angular gyrus. At the age of 7, subpial transection was performed. The EEG abnormalities disappeared. A follow up EEG 1,5 year after the subpial transection, as well as all other follow up EEGs were normal. However, there was no effect on language functioning. From then on, no AED or other medications were prescribed.

Because of his severe limitations in auditory comprehension, he was judged to function as a deaf child, in spite of his normal hearing. From the age of 5 till the age of 16, he received formal education at schools for children with hearing loss and disorders of language development. At the age of 13, he produced short, agrammatic utterances with occasionally recognizable words or automatic sentences. He was very communicative, and used "Total Communication" including spoken language supported by signed language, natural gestures, fingerspelling, and pointing. However, although he, his parents and siblings all learned Dutch supported sign language, he never accepted being treated as a deaf child and tried to communicate as much as possible without gestures. There were no behavioral problems and his non-verbal IQ (SON-R 6-40)[19] was 118.

At the age of 16, he was referred to the expertise team for Acquired Childhood Aphasia at Rijndam rehabilitation, Rotterdam, for diagnosis and treatment advice. Both he and his parents felt that he might be able to further improve his communication with extra training. After years without improvement, his mother had observed some language

Table 1. Test Scores at Intake.

Test	Score/max score
ASRS Spontaneous Speech Interview	2/5
Boston Naming Test-NL	
Spoken naming	8/60
Written naming	8/60
Auditory discrimination words	
PALPA 2	53/72
Word Comprehension	
PALPA 45, spoken word comprehension	31/40
PALPA 46, written word comprehension	30/40
Token Test	10/36

improvement over the last months, and unlike earlier in his life, he showed greater ability to learn new words. His language deficits severely affected communication in daily life and as such participation in society. He had a very limited social life and his dream was to become an assistant cook after finishing school. He currently held a part-time job as a dishwasher in a restaurant; working in the kitchen would require him to learn words frequently used in a restaurant kitchen. Further, his colleagues would have to adapt their communication with him (e.g. writing ingredients down or pointing to pictures).

Language assessment showed very limited spontaneous speech production. He produced short and agrammatic utterances that were often incomprehensible and unintelligible, due to distorted articulation. Further, he produced many phonological paraphasias. He also used some complete "ready-made" utterances, e.g. "what is it called", "do you know this", which he spoke fluently and well-intoned. Often, he appeared unable to understand the speech-language therapist.

Formal language testing revealed severe disturbances of auditory processing as well as auditory comprehension (Table 1, auditory discrimination words, word comprehension and Token Test[20]). This is in line with his verbal auditory agnosia. His vocabulary was extremely limited and at the level of children aged 6;6-6;11 years (measured with the Peabody Picture Vocabulary Test (PPVT).[21] He scored slightly higher for written words (score PPVT average age level for 7;0-7;11 years). With this pattern, there was some evidence that his vocabulary for written words was somewhat larger than for spoken words, but still considerably smaller than that of children of his own age.

His aphasia was also manifest in severe word finding difficulties and word production difficulties (Table 1, Boston Naming Test (BNT).[22] We did not test his use of sign language, but his parents reported that his use of gestures was similar to his spoken language: he also had a small vocabulary and very limited use of grammatical structures in sign language.

Treatment

Our cognitive linguistic treatment was aimed at improving daily life communication in areas important to this boy, such as work. To this end, 4 subgoals were defined:

1. Enlarging vocabulary: we selected words that were functionally relevant in his daily life, such as names of food and kitchen utensils. For each word, its meaning (semantics) as well as its form (phonology) were trained. Semantic judgment tasks and word webs were used to process semantic aspects. The word's phonological form was trained by giving attention to stress pattern, number of syllables and phoneme order. In all these exercises, both the picture and the written word of the target item were shown.
2. Facilitating oral production of personally relevant words and utterances: we used melody, stress and rhythm in the context of naming tasks (words) and in role play situations (utterances). These exercises were based on Melodic Intonation Therapy (MIT)[23], an effective language production treatment in adult aphasia.[24] Similar to MIT, the boy and the SLT together repeatedly said words or utterances, exaggerating the stress and intonation pattern. We also followed the MIT steps: from joined repetition to spontaneous production of the target item. However, we did not use 2 of the key elements of MIT: singing and indicating the rhythm with a motor movement (e.g. tapping the hand). The boy was unable to sing and he did not want to use hand movements while speaking. Therefore, the SLT presented the targets melodically intoned and indicated the stress pattern with a rhythmical hand movement, but the boy was not asked to produce melody or hand tapping.
3. Improving auditory processing: we used discrimination tasks comprising of the newly learned words. These words were presented in the context of a story. For instance: the therapist read a story with the phonologically similar words "sauce" and "salt". The boy was given a picture of "sauce" and a picture of "salt" and each time he heard one of these words during the story, he had to point to the corresponding picture.
4. Facilitating sentence production: we trained several simple, functionally relevant sentence templates. Thus, we taught him to start sentences with "can I . . . ." or "I would like . . . ". Further, we trained several function words. For instance, he did not know the meaning of the words 'with' and 'without', so we trained these in role playing situations (e.g. asking for coffee with milk).

All 4 treatment components were systematically addressed in an intensive experimental treatment block of 3 weeks of face to face cognitive linguistic therapy: 5 days a week, 2 hours per day. After 3 months without SLT, this treatment was repeated in a second treatment block of 9 weeks (2 days a week, 2 hours per day).

Outcome measures and follow-up

We evaluated the treatment effects of our cognitive linguistic treatment at 3 levels: (I) improvement of trained items, (II) generalization to untrained items and (III) generalization to

Table 2. Outcome Measures Per Treatment Goal

Test	Therapy goals
Naming task: trained items	• Vocabulary • Word articulation
Boston Naming Task	• Vocabulary: generalization to untrained items • Word articulation: generalization to untrained items
Auditory discrimination sentences	• Auditory analysis
PALPA 45	• Auditory word comprehension
Spontaneous speech	• Communicative functioning (all therapy goals)
Scenario Test	• Communicative functioning (all therapy goals)

daily life communication. Table 2 lists the standardized aphasia tests we selected as outcome measures for each treatment goal. Since there is no standardized aphasia test to examine auditory processing of sentences (treatment goal 2), we developed an ad-hoc test.

To examine whether he was able to learn new words (therapy goal 1), we developed a picture naming task consisting of 57 pictures he was unable to name before treatment. We used the Boston Naming Task (BNT)[22] to investigate generalization to untrained items. This test consists of 60 black-and white line drawings of objects. Naming difficulty increases gradually over the 60 items, with high frequency target words in the first items to very low-frequency target words in the last items. Because the test was extremely difficult for him, we administered only the first 40 items (words of high and middle frequency). Performance on these two picture naming tasks was also used to evaluate improvements in the articulation of words (therapy goal 2), both trained (picture naming task trained items) and untrained words (BNT). Crucially, the linguistic process of word *retrieval* differs from word *production*. Therefore, an incorrect or incomplete realization of the phonological form of a word (e.g. 'pineap juice' instead of pineapple juice) was counted as correct word retrieval (therapy goal 1), but as an incorrect articulation of the word form (therapy goal 2).

To examine the effect of this cognitive linguistic treatment on auditory processing (therapy goal 3) we developed a sentence discrimination task. The tester read aloud 2 utterances which were either identical or minimally different (e.g. 'The plates are hot' and 'The plates are not hot'). The boy's task was to indicate whether the 2 sentences were identical or not. The tests consisted of 22 phrases, of which 11 contained trained words or utterances and 11 were untrained. However, since the exercises for auditory processing were extremely difficult during treatment, we did not succeed in training all of the 11 sentences selected beforehand. As a result, the sentence discrimination task consisted mainly of untrained items. We used subtest 45 of the Dutch Psycholinguistic Assessments of Linguistic Processing in Aphasia (PALPA)[25] to examine generalization to the comprehension of spoken words.

Sentence production (therapy goal 4) was evaluated in the context of generalization to communicative functioning in daily life, measured by his spontaneous speech and the Scenario Test.[26] These two tasks were also used to measure generalization to communication of the other therapy goals. Spontaneous speech is a measure for verbal communication. We recorded a conversation between the boy and the tester on topics such as school, hobbies and work. We selected a sample of 3 minutes of the boy's spontaneous speech (excluding tester's speaking time) and counted the number of interpretable words as well as the mean length of utterance of the 5 longest utterances (ML5LU). Length of utterance refers to the number of words of an utterance. The ML5LU variable is often used in children with speech and language disorders.[27]

The Scenario Test measures verbal and nonverbal communication. The test is used in adult aphasia and contains 18 items presenting a daily life communicative situation. The patient is asked to react as if he were in that situation. He is free to use all possible communication channels (speaking, writing, gesturing, pointing, drawing). We selected 13 items representing potential communicative situations in this boy's life. Three of these items were communicative situations in a restaurant (eg. asking for the bill) and as such required the use of words and phrases trained during SLT.

Tests were administered before and after the two treatment blocks of cognitive linguistic treatment, with a follow-up at 18 and 30 months after the start of treatment (see Figure 1).

The boy received no SLT between the testing moments posttreatment and the first follow up session (3 months after the end of treatment). He followed a second intensive block (9 weeks, 4 hours a week) of cognitive linguistic treatment. Treatment was similar as described above, except that a new set of 67 words was trained. The first set of trained words was not addressed in the second block. After this second block, all tests were repeated, including the performance on the words trained in the first block (T4, 7 months). SLT was continued, but less frequent and no longer focusing on cognitive linguistic processes, but on the use of communication strategies and a communication device. We repeated all tests 1,5 years and 2,5 years after the first treatment block. The boy received no SLT in the year between these 5th and 6th testing sessions.

Results

Figure 2 presents the results on trained items. Before treatment, the boy was unable to name any of the 57 pictures. After treatment, there was a marked improvement in *naming* trained items, with good maintenance over 3 months. This indicates that he has learned new words (treatment goal 1) and was able to retain these newly acquired words during several months without SLT. After the second block of cognitive linguistic treatment, he successfully learned new words again: before treatment he was able to name 57.4% of a new set of 67 words. After treatment, he correctly named 88.1% of these items. In figure 2, we merged the results on the 2 sets of trained items. After the second treatment block, he correctly named >90% of

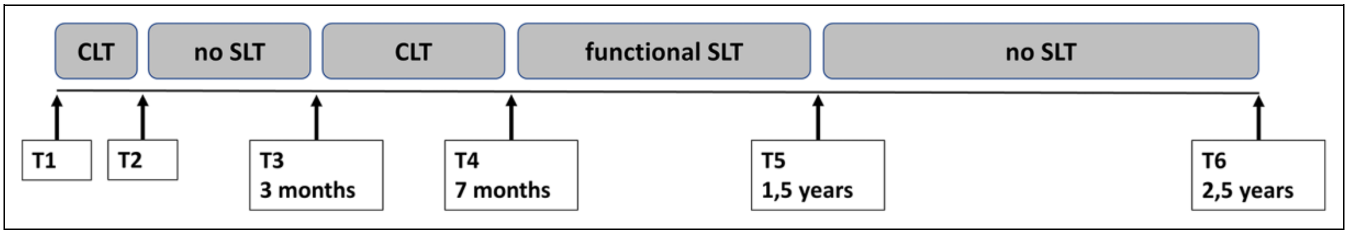

Figure 1. Testing moments.

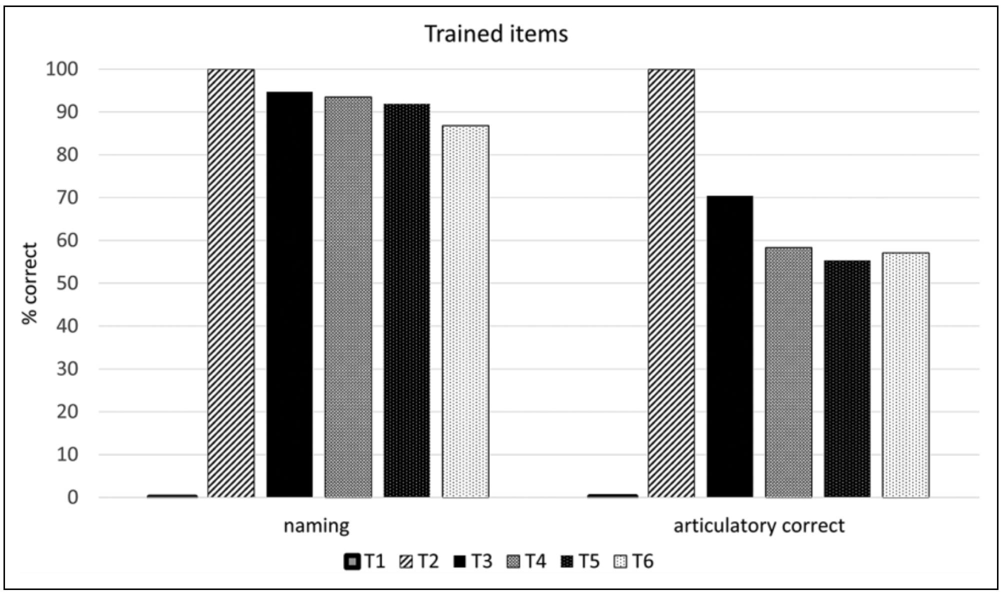

Figure 2. Results for trained material: naming task.

all trained items, i.e. the items trained during the first treatment block *and* those trained during the second block (T4, Figure 2). As is clearly visible, he retained more than 80% of all treated words even after years without cognitive linguistic treatment (T5, T6 Figure 2).

For the *articulation* of words (treatment goal 2), we also found a treatment effect: after treatment, the boy correctly pronounced all of the 57 trained items (Figure 2). However, at follow-up, his performance slightly dropped and his pre-treatment struggles producing unstressed syllables and clusters were again evident. These struggles remained during long-term follow up (Figure 2).

Figure 3 illustrates the performance on tests used to examine generalization of treatment effects. Due to time limitations at the post therapy test session, the BNT and PALPA 45 were only assessed at follow up and not immediately after therapy. As is visible in Figure 3, the considerable improvement on naming trained items did not generalize to untrained material: there is no improvement on the Boston Naming Test. In contrast to the absence of an effect on the articulation of trained items, the articulation on the retrieved items of the Boston Naming Test does improve. However, the items he is able to name on the BNT are articulatory much less complex than the items in the two set of trained words.

Further, the auditory analysis of sentences (treatment goal 2) did not improve after treatment (Figure 3). The boy scored slightly higher on this test at follow up (Figure 3) and maintained this higher score on long-term follow up sessions. Qualitative analysis of his performance on the auditory sentence discrimination task at the 4 follow up testing sessions suggests that he became slightly better at detecting differences in syntactic structure (e.g. a statement versus a question), not completely focusing on the content words any more. However, since this test is not a standardized test, there are no norm data and it is impossible to determine whether this change in score reveals an actual improvement or is normal variation in score. His comprehension of untrained words (treatment goal 2) did not improve (Figure 3, PALPA 45).

Regarding the ultimate treatment goal, improvement of daily life communication, the results show a considerable improvement on the Scenario Test immediately after treatment, 3 months later as well as at long-term follow up (Figure 3). This improvement is above the cut-off of a clinically relevant improvement, as indicated in the test manual.[26] A qualitative analysis of his performance on the Scenario Test showed that before as well as after treatment, he completely relied on speaking in communicating, not using any strategy to support his spoken output. However, the quality of his spoken production improved: before

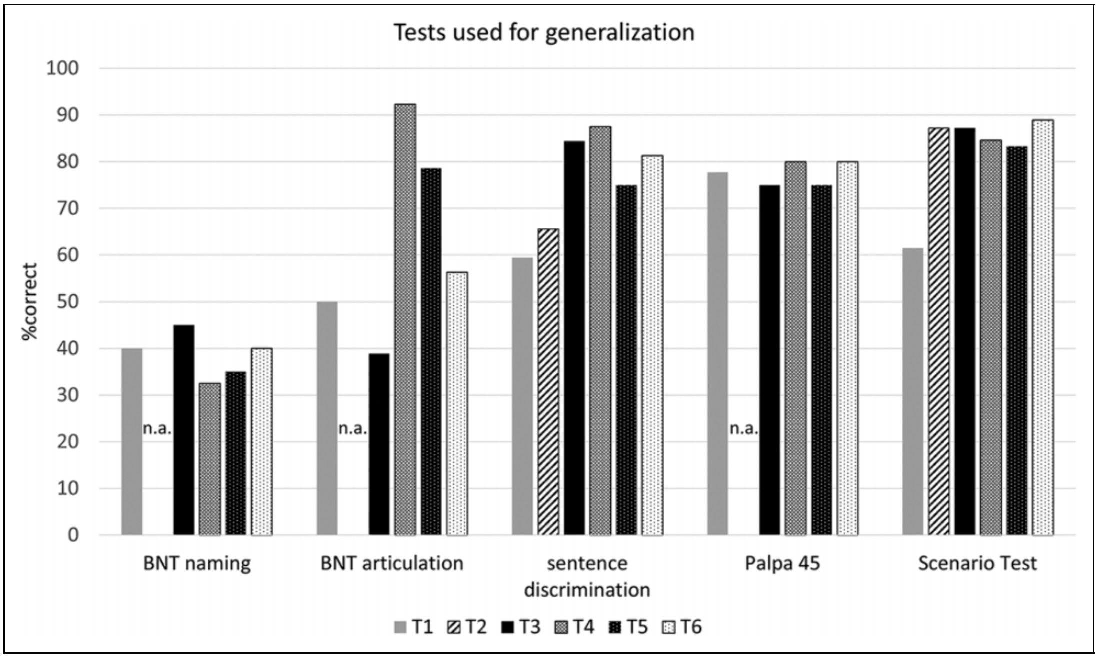

Figure 3. Results for generalization to untrained material.

treatment, he mainly used short, agrammatic utterances, which were sometimes difficult to understand due to poor articulation. After treatment, he produced slightly longer sentences, with more grammatical structure. He applied the grammatical structures and sentence templates trained during therapy to build new sentences, suggesting that he has not simply learned new utterances, but has acquired the ability to use the grammatical tools in relevant communicative situations. The results of the spontaneous speech measures point in the same direction: with the exception of the follow-up session at 7 months when he was extremely tired, he produced more interpretable words (range 205-263) and longer utterances (ML5LU range: 7.2-7.4) at all follow up sessions than he did before therapy (140 words; ML5LU 6.8).

Discussion

In this case study, we examined the effects of cognitive linguistic therapy in a teenager with chronic Landau Kleffner Syndrome. In adult aphasia after stroke, there is ample evidence sustaining the effectiveness of this type of speech and language therapy.[14,15] However, to our knowledge, this is the first study investigating the effect of cognitive linguistic therapy in LKS.

The results suggest that our approach was successful for some, but not all impaired linguistic processes. Our treatment yielded acquisition of words and standardized phrases: in only a couple of weeks this teenager with LKS was able to acquire *and* retain more than 100 new words. He also learned several grammatical constructions and typical ways to start a sentence. Moreover, he was able to use these words and utterances correctly in daily life communication.

The lack of generalization to untrained words stresses the importance of training functionally relevant words. Speech and language therapists working with children with LKS need to carefully select the items to train and make sure that their client needs these words in his or her daily life communication. Training functionally relevant words and utterances helped our patient in working in a restaurant kitchen, although he and his colleagues still encountered many communicative obstacles. Obviously, participating in society as independently as possible is important for all children with LKS. They might therefore need lifelong SLT, not weekly year after year, but rather a regular intensive block of SLT training words and utterances that are relevant for their present situation in life.

One of the limitations of our study is that we had to design an ad-hoc test for examining the treatment effects on auditory analysis of sentences. As a consequence, it is impossible to know whether the observed improvement on this test reveals an actual improvement of linguistic functioning or is simply normal variation in score.

A further limitation is that we only examined his auditory processing at *sentence* level and not at the level of *words.* Hence, we are unable to draw firm conclusions on the effect of our treatment on auditory analysis. Anecdotal reports from his parents as well as observations during follow up suggest that he still suffered from severe auditory analysis deficits: if someone asked him to pass the salt, he would frequently give the sauce. We also observed no improvement on auditory comprehension of words. We therefore assume that his verbal auditory agnosia persisted and continued to hamper his communication.

In treating verbal auditory agnosia, we developed exercises known to be effective in adults with auditory analysis deficits after stroke. It is possible that this approach failed to work here because of different etiology and types of brain damage in stroke versus LKS. To our knowledge, the effect of such auditory processing training has rarely been examined before.

Language therapy in LKS typically focusses on the use of other linguistic modalities (e.g. reading) or types of communication (e.g. sign language) to circumvent the impaired auditory processing.[17,28] Probably, these children do not receive auditory comprehension treatment in the early stages of LKS, because they are educated at schools for children with hearing disorders where the focus is on using total communication, rather than improving auditory comprehension. We found one case study of verbal auditory training similar to ours in a 6-year old boy, 2;6 years post onset: 2 years of this training yielded only small improvement of auditory language comprehension. The boy remained unable to understand spoken utterances longer than 3 content words and his level of auditory comprehension was far below his chronological age level.[17] Moreover, a later study revealed that this same child, now 14 years old, suffered from severe auditory processing impairments.[18] This suggests that SLT does not improve the auditory verbal agnosia. This is in line with other studies showing that the core deficit, the auditory processing deficit, remains in children with LKS refractory to medical therapy, even in the context of improved auditory language comprehension.[9]

Conclusion

The cognitive linguistic approach is a promising approach to language treatment in LKS. We developed a treatment program targeting specific linguistic components and as such enabling us to disentangle the effects of our treatment: in a teenager with severe chronic LKS, for whom many believed that he had reached his plateau, spoken language production and communicative functioning improved after this type of SLT. Short blocks of cognitive linguistic treatment with functionally relevant words and phrases resulted in long term improved communication in daily life.

Although case studies provide limited evidence, we believe that cases like these are important in gaining insight in language rehabilitation in rare conditions such as LKS and as such in optimizing treatment.

Declaration of conflicting interests

The authors declared no potential conflict of interests with respect to the research, authorship, and/or publication of this article.

Funding

The author(s) received no financial support for the research, authorship, and/or publication of this article.

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