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Long-Term Recovery From Chronic Global Aphasia: A Case Report

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In 2000, a 57-year-old man had a cerebral hemorrhage that resulted in spastic hemiparesis and global aphasia with agraphia and alexia. He received continuous speech therapy, with no significant improvement in speech performance. Three years after the event, a directed resource-orientated and music-supported training (SIPARI[®]) was initiated that had been developed especially to support speech rehabilitation for chronic nonfluent aphasia patients. The effects of this treatment on language capabilities were tested by independent and experienced investigators with the Aachener Aphasie Test (standardized procedure). Tests were carried out 20 months after onset of treatment in 2004, and in two follow-up studies in 2005 and 2008. After the first treatment period of 20 months, the patient showed clinically significant improvements in

I n Germany, the demarcation line for the attribution of a chronic condition in aphasia patients in rehabilitation is 12 months after the event (e.g., stroke, brain tumor, head injury). While intensive logopedic treatment aims at an immediate improvement, chronic aphasias are estimated as relatively therapy resistant. The extent of any progress is expected to be limited and in most cases restricted to a few language modalities (Huber, Poeck, & Weniger, 2002; Weniger, 2003).

Although contrary reports were published early in the 20th century, and experienced clinicians always confirmed them (Franz, 1924; Grötzbach, spontaneous speech as well as in the token test (a measure to evaluate severity of aphasia), repetition, and naming. The first follow-up study in 2005 as well as the last assessment in 2008 revealed further clinically significant improvements in speech performance. At the time of this writing, the patient is able to talk in everyday situations and to participate verbally in social life. The fact that he regained confidence and enjoyment in using speech has improved his own quality of life and that of his family and friends considerably.

Keywords: speech rehabilitation; chronic global aphasia; long-term rehabilitation; directed music-supported training (SIPARI[®]); Aachener Aphasie Test; significant improvement

2004, 2005; Gutzmann, 1901; Holland & Beeson, 1995; Holland, Fromm, DeRuyter, & Stein, 1996; Leischner, 1998; Taylor Sarno, 1991), many of the patients and their families are still confronted with the discouraging assessment that no changes are to be expected once the chronic condition has set in. Recent research results do not appear to change this opinion, although they suggest that changes in the sense of the functional cortical reorganization of, for example, the motor system take place over many months after the event (Platz, 2005).

Several research findings validate that certain compensatory mechanisms are activated years after the event (Karbe et al., 1998; Naeser & Palumbo, 1998; Schlenck, 1990), for example, the recruitment of right-hemispheric regions for speech processing when the left-hemispheric centers are permanently impaired.

It is necessary to revise the traditional view that activation patterns of remaining speech areas in the

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left hemisphere suggest a fast recovery, whereas activation patterns in the right hemisphere predict insufficient compensation over long periods. Reorganization patterns seem to differ very much individually. Patients use remaining healthy areas of the left hemisphere, but they also use remaining speech areas of the right hemisphere, and this to a far greater extent than traditionally stated (Grande & Huber, 2005; Huber, 2002; Risse, Gates, & Fangman, 1997; Silvestrini, Troisi, Matteis, Cupini, & Caltagirone, 1995; Thompson, 2000).

A number of therapy forms have emerged over the years, developed specifically for seriously affected aphasia patients, for example, Visual Action Therapy (Helm-Estabrooks, Fitzpatrick, & Barresi, 1982), Melodic Intonation Therapy (MIT; Albert, Sparks, & Helm, 1973), or from the field of pragmatic approaches, Promoting Aphasic Communicative Effectiveness (Davis & Wilcox, 1985).

Examples from the German-speaking area are MODAK (Modalitätenaktivierung in der Aphasietherapie; Lutz, 1997) and constraint-induced aphasia therapy (Pulvermüller et al., 2001; Meinzer et al., 2007).

Therapy Concept

Apart from traditional interventions like logopedics, physiotherapy, or ergotherapy, there are almost no other outpatient therapies available for seriously affected chronic aphasia patients in Germany. Longstanding experience from musical work with aphasia patients in self-help groups, and the observation of improving speech performance, prompted us to develop a method and to assess the treatment effects.

A recent controlled study illustrated how the use of musical components as a directed intervention adapted to the specific needs of aphasia patients is beneficial (Jungblut, 2005; Jungblut & Aldridge, 2004). This resource-orientated and trainingcentered combination treatment is based on parallel processing strategies between music and language, which suggests the use of music specifically in aphasia therapy. As this article aims at reporting research results, it should be mentioned that details on the neurophysiological and neuropsychological background and the method have been described elsewhere (Jungblut, 2002, 2005; Jungblut & Aldridge, 2004; Jungblut, Gerhard, & Aldridge, 2006). Based on the well-known phenomenon that even seriously affected aphasia patients are often able to sing the words of known songs—although they are unable or almost unable to speak (Keith & Aronson, 1975; Morgan & Tillduckdharry, 1982; Ustvedt, 1937; Yamadori, Osumi, Masuhara, & Okubo, 1977)—this rhythmic-melodic training addresses residual melodic abilities.

Parallels are recognizable with MIT, which also uses musical elements in the treatment of aphasia (Albert et al., 1973; Sparks & Deck, 1994; Sparks, Helm, & Albert, 1974; Sparks & Holland, 1976). However, MIT is neither applied as group therapy nor suitable for global aphasia patients (Benson, Dobkin, & Gonzales, 1994; Naeser & Palumbo, 1995; Sparks & Deck, 1994). The SIPARI[®] method has been successfully applied in a group context with patients suffering from Broca's aphasia and global aphasia (Jungblut, 2002; Jungblut & Aldridge, 2004).

SIPARI[®]

The directed music-supported training approach (SIPARI[®]) used in this study comprises the components of Singing, Intonation, Prosody, breathing (German: Atmung), Rhythm and Improvisation as essential elements. Similar to MIT, the starting point represents the activation of remaining righthemisphere speech abilities in aphasia patients. Mental preparation, termed inner singing (Perry, 1994, and Perry, Zatorre, & Evans, 1995, cited in Marin & Perry, 1999, p. 688), is an important training element and expands the concept of melody. It also comprises breathing exercises that encourage elementary vital processes as the basis of any vocal utterance and prepares for phonation. In addition, the treatment is extended by rhythmic exercises (instrumental and/or vocal) to support the phonological and segmental capabilities of the left hemisphere. Improvisations enable the patient to practice communication on a nonverbal level. In the group context, this therapy component allows the stimulation of spontaneous verbal interaction and the training of perceptive and expressive skills that form the basis of every communication.

Our treatment objectives are to improve linguistic, motor, and cognitive functions and thereby support speech-motor processes and also those speech-systematic processes that encourage planning



Figure 1. Cranial computer tomography (from 2000) showing the extension of the hemorrhage.

and sequencing performance. Single therapy sessions allow us to focus on the specific problems of the patient in each case. Group therapy comprises practicing all components of the SIPARI[®] method in a proportionally balanced manner.¹

Case Description

In February 2000, at the age of 57, Mr. Benz (pseudonym) suffered from an extensive cerebral hemorrhage in the area of the left thalamus reaching up to the radiate crown (being about 3 centimeters in diameter), massive bleeding into the plexus choroidei, and rupture of the second, third, and fourth ventricle. The clinical symptoms were spastic hemiparesis on the right and severe global aphasia with agraphia and alexia (see Figure 1).

Mr. Benz is married and has two grown children. Up to the present date, he lives in his own house with his wife. Before his stroke, he was a lawyer in a leading position, open-minded, highly communicative, and well educated. The skilled use of language constituted a considerable part of his quality of life because he was also active as a lecturer and teacher. Mr. Benz is right-handed. Neuropsychological findings showed considerable cognitive deficits, specifically in short-term memory. His attention and concentration were also assessed as severely impaired. Psychological findings showed loss of temporal and spatial orientation, reduced drive, and much restrained affectivity. Formal thought processes were slowed down in general.

In the initial stage, Mr. Benz slumped apathetically in his wheelchair. Questions had to be asked repeatedly before he noticed them. Reactions concerning facial play, gesture, or vocal utterances were absent or presented with a delayed response of approximately several seconds up to a minute.

After several rehabilitative interventions and continuous speech therapy (45 minutes twice per week over a period of 2 years), experienced speech therapists diagnosed, in a final report after 6 weeks of day hospitalization in a neurological center in 2002, global aphasia as mainly unchanged in this patient two years after the event. Only auditory comprehension was assessed as *temporarily noticeably* improved.

SIPARI[®] Intervention

We started the SIPARI[®] training in March 2003. The described therapy process was a research project. Mr. Benz produced almost no spontaneous speech in the beginning of the therapy. Apart from single-word productions, which were uttered essentially on demand and with significant impairments in initiating them, he produced mainly stereotyped answers. Mr. Benz had no musical background.

Because group therapy would have been too demanding due to Mr.Benz's neuropsychological deficits, described earlier, therapy was started as single therapy. Diagnosis of the remaining musical abilities revealed that the patient had retained the ability to sing, but automated recall of known texts was not always possible and was clearly impaired by problems with phonological processing. In analogy to speech, phonemic paraphrases were produced in independent singing. He did well when called upon to repeat songs phrase by phrase. However, his attention span was short and was restricted to a maximum of four syllables at the start of the SIPARI[®] therapy.

Processing of melody components (e.g., intonation) appeared unimpaired, whereas processing of temporal components (e.g., duration) appeared to be difficult at times; for example, repeating tasks revealed that duration of sounds was often perceived incorrectly. Breathing and phonation were inconspicuous, and articulation appeared dysarthric. However, in this area the patient showed considerable progress in the case of exaggerated prosodic speech (a focus on pitch, or fundamental frequency, in contrast to duration). In the course of the therapy, Mr. Benz used fewer phonemic paraphrases as soon as stressed speech was achievable. In terms of the rhythmic aspects of speech, processing of even metric courses seemed rather unimpaired, whereas distinct sequencing problems occurred with rhythmic group-forming tasks. Longer sequencing (more than four beats) revealed deficits.

Musical communication in joint improvisation was characterized by much enjoyment and creative power. His creative resources were obvious, and the "former Mr. Benz" reappeared. His musical play was metrically even, and sometimes it appeared as if he was fixed on one theme. At the level of speech, this behavior corresponded with a clear trend to perseveration. Initially, attention and concentration were much reduced and unstable, and affectivity was impaired, so that only 20 minutes of purposeful intervention were possible in some of the first sessions.

After about 30 therapy sessions, joint improvisations revealed distinct changes that correlated with improvements of cognitive function. The initial metrically even play that had a tendency to perseveration began to show more rhythmic variations so that musical pieces emerged (e.g., in the form A-B-A). The musical context also changed significantly. Having been mainly a soloist who avoided contact, Mr. Benz gradually paid attention to another person. He started to arrange musical alternations of dialogical character that finally ended in a joint "conclusive discussion." After 8 months of single sessions, the first clear improvements occurred in his linguistic abilities; concentration and attention improved, and he produced longer items vocally and instrumentally and sequenced them more adequately on the levels of vocal sound and rhythmtempo. He recognized errors and increasingly tried to correct them. He needed fewer and fewer cues (initial sound) in doing so. The growing ability to "loosen up" helped him to perseverate less. He took far more time to prepare utterances internally. He made more frequent spontaneous utterances that were positively noticed by his family. Improved attention, concentration, and sequencing performance (i.e., from sound, word, and up to syntax level) as well as regained self-confidence even in his verbal capabilities enabled him to increasingly produce complex utterances.

After 45 single therapy sessions conducted over a period of a year, Mr. Benz also participated in group therapy sessions. His progress made participation in a group setting useful in order to encourage Mr. Benz to apply his regained capabilities with other equally impaired patients. Working together in the group influenced above all his drive and motivation considerably but also activated cognitive skills and social competencies.

Between 2005 and 2008, therapy was continued, and up to now Mr. Benz has taken part in 230 single sessions (45 minutes, once per week) and 130 group sessions (45 minutes, once per week). Therapy was conducted by the first author. Logopedic interventions went on continuously.

Assessment

The instrument used for assessment of the efficiency of the treatment was the Aachener Aphasie Test (AAT) 20 months after the onset of treatment (2004) and again in 2005 and 2008. The starting point was the AAT taken in November 2000 (9 months post onset).

The AAT is a standardized procedure for evaluating the severity of aphasia, developed and validated in the German language (Huber, Poeck, Weniger, & Willmes, 1983), subsequently translated into several European languages, and also validated and standardized in Dutch and Italian. The AAT can be applied repeatedly to assess the efficiency of speech therapy interventions. The presence and type of aphasia was established using the ALLOC classification procedure, a nonparametric discriminantanalysis computer program (Huber et al., 1983) using the normative data of the AAT. The AAT consists of six rating scales for spontaneous speech (communicative verbal behavior, articulation and prosody, automatized language, semantic structure, phonemic structure, syntactic structure) and five subtests (token test, repetition, written language, naming, comprehension) for the assessment of specific language impairments. Pre- and posttests were performed by experienced logopedists and clinical linguists of the Aphasia Center North Rhine Westphalia (Aphasiker-Zentrum NRW e.V.).

Results

Significant clinical improvements were ascertained in spontaneous speech, in the token test, and in repetition and naming. For an assessment of the degree of speech impairment related to the entire group of aphasia patients, the AAT assesses percentile scores from the score values of the subsections. The percentile score found for one test value



scores (see Figure 2). The improvement exceeds the critical difference a factor of 1.9. Comparison of Test 3 and Test 2 revealed an improvement of 4 percentile scores; a further improvement could be assessed with 2 percentile scores in comparison of Test 4 and Test 3.

measure to evaluate the severity of the aphasic disor-

der. Comparison of Test 2 and Test 1 revealed a clinically significant improvement of 32 percentile

Repetition

Comparison of Test 2 and Test 1 revealed a clinically significant improvement of 57 percentile scores (see Figure 2). The improvement exceeds the critical difference a factor of 3.6. Comparison of Test 3 and Test 2 revealed an improvement of 3 percentile scores; a further improvement could be assessed with 4 percentile scores in comparing Test 4 and Test 3.

Naming

Comparison of Test 2 and Test 1 revealed a clinically significant improvement of 49 percentile scores (see Figure 2). The improvement exceeds the critical difference a factor of 4.9. Comparison of Test 3 and Test 2 revealed an improvement of 23 percentile scores; this result remained unchanged in a comparison of Test 4 and Test 3.

The written language and comprehension subtests had to be ended prematurely due to the patient's agraphia and alexia.

Profile

In the AAT, the reliability-weighted sum of the five subtest scores is used to determine the profile level, an overall and clinically relevant measure of the severity of aphasia. Differences in profile are then seen as overcoincidental if the absolute difference of the two profiles exceeds 1.41 (Huber et al., 1983). The improvements mentioned above lead to a clinically significant improvement in profile level. The difference between profiles in comparison of Test 2 and Test 1 is 12.25. The third test compared to the second again shows a clinically significant difference in profile levels of 2.96. Comparison of Test 4 to Test 3 revealed again a clinically significant increase in profile, with a difference of 1.73 (see Figure 3).

Figure 2. Evaluation of test results in percentile scores.

indicates the percentage of patients of the exercise sample (n = 376) who have achieved the same or a lower score (Huber et al., 1983). To simplify the description, the results are presented in percentile scores (see Figure 2).

Spontaneous Speech

Compared to the first test, Mr. Benz improved significantly in the second test, on 6 out of 6 levels of spontaneous speech. According to Huber et al. (1983), a substantial improvement in spontaneous speech requires at least 2 points out of a scale of 5 points per description level. A comparison of Test 3 and Test 2 shows almost no changes in the assessment of spontaneous speech, whereas the comparison of Test 4 and Test 3 revealed an improvement of 1 point on each description level (Table 1).

Token Test

This subtest of the AAT distinguishes aphasic disorder from nonaphasic disorders and represents a

	pre.	2000- post.	2004 diff	[sig.		pre.	2004- post.	2005 diff]sig.		pre.	2005- post.[2008 diff	sig.
	point		points					points						
spontaneous speech									-					
communicative behavior	1	3	2	S		3	3	0	1		3	4	1	1
articulation and prosody	1	3	2	S		3	3	0	1		3	4	1	1
automated speech	1	3	2	S		3	4	1	1		4	5	1	1
semantic structure	1	3	2	S		3	3	0	1		3	4	1	1
phonematic structure	1	4	3	S		4	3	-1	1		3	4	1	1
syntactic structure	0	2	2	S		2	3	1	1		3	4	1	1
pre. pretest post. posttest diff change pretest - posttest S significant change according to AAT														

Table 1. Comparison Profiles of Spontaneous Speech



Figure 3. Changes in profile level in percentile scores.

The most important result of this study can be summarized as follows: Mr. Benz has clearly regained joy in verbal communication and participates increasingly in verbal social contact; despite his remaining impairments, he rises to speak in the company of several persons, he starts talking with somebody he meets, or he makes phone calls. Transfer into everyday communication has been successful in his case; his quality of life has considerably improved in regard to self-confidence and with social contacts. With respect to expressive language, he replies to questions precisely, with minimally reduced speech rate. His syntax is in most cases complex; agrammatical symptoms occur rarely. Remaining problems are word finding and sometimes lack of motivation and persistent severe problems in reading and writing.

Discussion

This single case study describes the long-term recovery of a 57-year-old patient suffering from chronic global aphasia. Logopedic interventions were provided continuously from 2000 to 2008 with two therapy sessions per week. In 2002, experienced speech therapists diagnosed global aphasia as mainly unchanged in this patient two years after the event; only auditory comprehension was assessed as temporarily noticeably improved.

In 2003, 3 years after his stroke, a directed music-supported training (SIPARI[®]) was initiated. The efficiency of this treatment was repeatedly assessed with a standardized German language test (AAT) in 2004 and in two follow-up studies in 2005 and 2008. During this period, the patient received 230 single sessions and 130 group sessions of the SIPARI[®] training. Distinct improvements in linguistic performance could be assessed 20 months after the SIPARI[®] treatment had been started. The most outstanding improvements were achieved in expressive speech performance.

Spontaneous speech showed clinically significant improvements on all six descriptive levels in the second test. According to Huber et al. (2002), significant improvements in spontaneous speech are assumed to indicate successful transfer to everyday communication. The final profile of spontaneous speech (see Table 1), especially the development concerning the descriptive levels of communicative behavior, semantic structure, and syntax structure with a 4-point score on each level (out of 1 to 5 points) is an exceptional result.

Clinically significant improvements also appeared in the token test, in repetition, and in naming. These distinct improvements occurred in the chronic stage. Follow-up examinations confirmed that, even 8 years after onset, continual improvement can be achieved by a specific intervention. This single case study mainly confirms research results of a previous study on patients with chronic aphasia of the Broca and global type with a mean duration of aphasia of 11.5 years (Jungblut, 2005; Jungblut & Aldridge, 2004). However, the extent of such improvements is far larger in the case of Mr. Benz.

These results challenge studies from speech therapy that assess the chances for improvement in chronic global aphasia patients as limited (Huber et al., 2002; Poeck, Huber, & Willmes, 1989; Weniger, 2003).

Singing as an initial intervention with mostly right-hemispheric participation (Perry et al., 1999; Wildgruber, Ackermann, Klose, Kardatzki, & Grodd, 1996) is certainly a very effective therapy, particularly for disorders of articulatory-motor processes (Keith & Aronson, 1975). Yet recent studies have been unable to confirm an advantage of singing over speaking in brain-damaged patients suffering from speech disorders. Compared with speaking, singing did not improve word production (Hébert, Racette, Gagnon, & Peretz, 2003; Peretz, Gagnon, Hébert, & Macoir, 2004).

A deliberate use of the voice and its possibilities to systematically change the balance of acoustic elements, however, supports a process that uses the melodic speech elements as a starting point to stimulate phonological and segmental capabilities of the left hemisphere step-by-step through focusing on temporal-rhythmic components. Other studies also confirm that an accentuation of temporal-rhythmic components is effective in therapy (Belin et al., 1996; Boucher, Garcia, Fleurant, & Paradis, 2001).

An interesting point for future research is the question of whether a transferred focus to the auditory channel, with a deliberate accent on first melodic and, later, temporal components of linguistictonal memories, may regulate cognitive activities. Such an approach might help to store verbal material better and also support organization and planning of expressive speech. This hypothesis would reflect current research in the field of speech therapy (Baumgärtner, 2008).

Work with rhythmic structures is an essential factor in the improvement of cognitive performance as the basis of the significant improvements in the token test, in repetition, and particularly, in naming. Metrically even and rhythmically grouped patterns were developed on instruments, vocally, or in a combination of both in order to stimulate phonological and segmental abilities and specifically to improve impaired sequencing functions.

Rhythmic structures contain many anticipatory patterns that facilitate the timing of movement coordination and render movements more fluid and even (Aldridge, 1999; Altenmüller, Wiesendanger, & Kesselring, 2006; Thaut, 2005). Grouping exercises carried out vocally as well as on drums or in combination represent an excellent training for memory, planning, and synchronizing.

Further studies will have to show whether the success of this specific music-supported intervention may be explained not only by positive reactions to a treatment that involves the principles of motor learning but also by an influence of speech systematic or cognitive skills in the sense of an activation and reorganization of speech processes. Functional imagery might help to explain brain activity and specify potential new links created through directed music-supported interventions. Our actual fMRI study will hopefully serve to clarify some of these fundamental questions.

Declaration of Conflicting Interests

The authors have declared that there are no conflicts of interests in the authorship and publication of this contribution.

Note

 Methodological details and a collection of exercises of SIPARI[®] will be published in an upcoming issue of this journal.

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