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RESEARCH ARTICLE



## Clinical trial of the D.E.L.P.H.I.N. speech treatment for children and adolescents who stutter

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### ABSTRACT

**Purpose:** The present trial investigated the D.E.L.P.H.I.N. (Deblockierungsimpuls, Entspannung, Logopädie, Phonetik, Hör-Wahrnehmungstraining, Intensität, nasaler Schwingungssakzent) speech treatment for children and adolescents who stutter, an approach using fluency shaping with an intensive time schedule and group sessions.

**Methods:** Participants were 42 male and 14 female persons who stuttered with a median age of 13.0 years. Participants and their parents completed the strength and difficulties questionnaire at the beginning of the therapy and 1 year later. Participants completed the Speech Questionnaire, an instrument to measure the impact of stuttering, twice before and twice after completing the therapy. The stuttering rate and mean length of the three longest stuttering events were studied four times in videos of interviews, reading texts and three telephone calls. Outcomes are reported for the 56 children and young adults who stuttered and who completed D.E.L.P.H.I.N. The main analysis was performed using linear mixed models.

**Results:** Quality of life as measured in the Strength and Difficulties Questionnaire (SDQ) – filled in by the participants/their parents – increased significantly from before the start of the therapy to 1 year after completion of the therapy. The subjective rating of the impact of stuttering also improved significantly. The stuttering rate and mean length of the longest stuttering event decreased significantly for all three modes from pre-treatment to 12 months post-treatment.

**Conclusions:** After D.E.L.P.H.I.N., a significant improvement of the relevant endpoints quality of life, impact of stuttering, and severity of stuttering is observed.

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Stuttering; children; adolescents; fluency disorder; therapy

## Introduction

Stuttering is a common speech disorder in all ages. A lifetime incidence of 5% is the most consistently reported statistic. Recent data suggest an even higher incidence, close to 10% [1]. The incidence of this disorder varies according to age group. Up to 90% of children who stutter will recover during childhood. During adulthood, <1% is affected [1]. Stuttering treatment is most successful during pre-school years. With increasing age, outcomes are less positive [2–8].

Treatment of children who stutter has changed during the last 30 years. In earlier years, a “hands-off” attitude was preferred. Now, there is a consensus that early intervention should be practised [9] although there is still a debate about the preferred approach. Multifactorial treatment strategies try to identify the stressors of the child and to modify the environmental stressors. Many pre-school children who stutter have been treated based on the Demands and Capacities Model [10]. This approach aims to decrease the demands set by the environment and the child him- or herself, and to increase the child’s capacities for speaking fluently. In contrast, the Lidcombe Program [11] uses operant conditioning techniques to teach parents to verbalise positive and negative responses to their child’s speech.

As stuttering has a widespread impact on the life of the affected, a treatment is mandatory. Stuttering children have more negative attitudes to communication than their peers and those attitudes to communication worsen progressively during the school years [12,13]. Adolescents who missed the window of opportunity for treatment during pre-school and early school age or underwent one or more therapies without sufficient effect experience or anticipate problems entering professional life or beginning university studies.

School children who stutter are often chronically bullied because of their stuttering. Blood and Blood found that 11- to 12-year-old children who stutter have a 63% risk of bullying compared to 22% for controls [14]. Another report of 28 stuttering children aged 7–15 years found that 59% of them reported being bullied and 38% reported bullying to have occurred on most days or every day [15]. Gladstone and his colleagues [16] found that bullying during the school years is strongly associated with subsequent anxiety emerging during early adolescence. Adolescents who stutter score more highly than adolescents who do not stutter on the measures of communication apprehension and view themselves as being less able communicators than their fluent peers [17]. Craig and Tran [18] concluded that

adolescents who stutter are more likely to become more shy, socially avoidant, and fearful of communication than their fluent peers, and to develop pervasive negative attitudes and beliefs about themselves as communicators. As persistent stuttering in adolescents and adults can have a serious mental and social impact [14,15], therapy should also be considered for older children and adolescents who still stutter if the patient is willing to participate. Treatment of adolescents who stutter is an under-researched area [3].

Earlier studies about adolescents reported improvements after therapy based on speech restructuring using either smooth speech [19] or prolonged speech [20]. Speech restructuring has also been shown to be effective in the frame of intensive treatment programs [21], or integrated with cognitive-behaviour therapy [22].

Speech therapy remains the treatment of choice, also for adults. Other treatment options, for example, pharmacological treatment, have not been shown to be effective in any well-designed study.

In addition, speech-restructuring treatments are generally not well accepted by adolescents and young adults. Clients are often inconsistent in applying new speech patterns, and thus decreasing the effectiveness of the treatment. Hearne et al. [20] conjectured that adolescents who stutter might prefer to stutter rather than to use a technique to control stuttering that involved decreasing rate and/or naturalness of speech. Adolescents may be reluctant to use an unnatural speech pattern with their peers or at school as this draws attention to the speaker, which in turn may increase social anxiety.

Twenty-three adolescents were treated with the Comprehensive Stuttering Programme, which included prolonged speech, cognitive and social training, self-generated home maintenance programmes, and refresher clinics. This led to a reduction in stuttering severity (69% maintained a satisfactory level of post-treatment fluency, an additional 7% a marginally satisfactory level) and 80% of the subjects rated their speech fluency as good or fair 12–24 months after treatment [21].

A question often discussed is whether the treatment of stuttering should be intensive or spaced over intervals. Intensive therapy is said to accelerate the acquisition of a new speech technique, but with a higher risk of relapse, whereas interval training may lead to slower acquisition, but also slower extinction of the new skills. James et al. [23] included 20 adult and adolescent stutterers (age, 19–50 years) and compared 16 2-h sessions of fluency training administered within 4 days to 2-h sessions twice per week for 8 weeks. Both treatment formats were equivalent on all measures.

Ryan and Ryan [24] conducted a treatment comparison study. Twelve children and adolescents (age, 7–17 years) were assigned to the “Gradual Increase in Length and Complexity of Utterance” programme, with nine of them completing the programme. Of these, six children achieved a near zero stuttering rate. Twelve other subjects aged 7–17 years were assigned to Delayed Auditory Feedback, with five of them completing the therapy. The authors

report that 11 subjects from both groups, who completed the Transfer and Maintenance Programs, reduced their stuttering from 7.9 stuttered words per minute to 0.8 stuttered words per minute at a 14-month follow-up.

Craig et al. [19] conducted a prospective study and compared four groups of 9- to 14-year-old children with stuttering. Twenty-seven of them received an intensive fluency-shaping technique using a variant of prolonged speech called smooth speech; 25 a home-based smooth speech, a less intensive fluency-shaping technique combined with child/parent interaction therapy; and 25 received electromyography feedback. The 20 children who were in the no-treatment control group received therapy after 3 months. All three experimental groups were significantly better than the control group and there was no significant difference between the experimental groups up to 1 year after the treatment. In the study conducted by Hancock et al. [25], 62 of the 77 subjects who had participated in the first study could be examined 2–6 years after treatment. The percentage of stuttered syllables (SS) scores was similar to those recorded 1 year post-treatment. Nobody’s treatment had superior treatment efficacy. The speech rate per minute was significantly higher than 1 year post-treatment, again with no differences between the treatment groups. Speech naturalness was the same for all treatment groups and over time in the view of the patients and the therapists; only the parents tended to rate the children’s speech as less natural during follow-up.

Laiho and Klippi [26] studied 21 children/adolescents who stutter and 29 of their mothers/fathers in intensive therapy courses including stuttering modification and parental guidance. The percentage of syllables stuttered dropped in the case of two-thirds of the participants and most participants and parents reported that the positive changes in stuttering behaviour had continued up to 9 months after the therapy.

An adaption of the Camperdown Programme that had previously been used in adults was used in three adolescents aged 13, 14, and 16 years by Hearne et al. [20]. One of them responded well to the treatment, with the percentage of stuttered syllables falling to approximately 1% in the period up to 1 year after the treatment. The second adolescent, who withdrew from the treatment during maintenance, still had a marked reduction. The third demonstrated a worsening of the stuttering. The question of whether the Camperdown Programme can also be applied by webcam without clinic attendance was studied in three adolescents aged 13, 15, and 16 years [27]. The webcam service delivery was appealing to participants and their parents. With a mean of 11 clinician hours, two participants achieved large stuttering reductions that did not diminish in the year after the treatment; one participant failed to maintain treatment gains during the 12 months follow-up period.

In the phase II trial [28], 16 adolescents aged 12–17 years were included. For the group as a whole, stuttering was reduced in terms of frequency and severity, but it was only efficacious and efficient for half of the group.

Hewat et al. [29] developed another treatment programme for adolescents and adults who stutter. Based on the operant procedure of self-imposed time-out (SITO), the participants learned to stop speaking for a short period after each stutter. More than half of the participants reduced their stuttering by more than 50%. Of the 30 participants, four were adolescents (age, 12–17 years). One of these failed to respond to SITO; another was unable to learn SITO. The two other adolescents both reached a significant reduction of the stuttering rate.

Maintenance of treatment effects after a period of intensive training was studied by Cream et al. [30]. Eighty-nine participants aged between 12 and 74 years received five consecutive days of intensive group speech-restructuring treatment followed by a maintenance programme of seven weekly clinic visits. For each participant of the experimental group, an individual target video of 4 min 30 s was produced. The participants were instructed to watch the whole video each day for 4 weeks and to talk parallel without stuttering, but only half of the participants actually watched their video every day. As participants in the experimental group demonstrated no advantage in the percentage of SS, but did report a better self-rating of the stuttering and their quality of life, the authors concluded that additional video self-modeling could be a useful adjunct to standard speech-restructuring treatment.

Nejati et al. [31] studied whether children with developmental stuttering could benefit from attention training. Fifteen children in the case group were compared to a control group in a randomized design. Those from the case group showed not only a lower reaction time after the intervention but also a lower stuttering severity. The authors concluded that training of inhibitory control and better emotion regulation may contribute to this effect [31].

Fry and her group [32] investigated whether an intensive treatment programme for older teenagers who stutter is effective in reducing overt and covert aspects of stuttering. They included three males aged 17;7, 17;11, and 18;10 years. One participant completed all required recordings. They found a significant trend of reduced frequency of stuttering for all three participants during the intensive therapy phase. This trend continued throughout the consolidation phase and remained significant up to 9 months after the intensive therapy in the case of the first participant. Participants also reported increased self-efficacy about speaking and reduced overt and covert aspects of stuttering.

Early reviews about stuttering treatment were published by Yairi and Ambrose [33] and Bloodstein and Bernstein-Ratner [34]. Nye and Hahs-Vaughn [35] provided an analysis of the methodological quality of experimental and quasi-experimental group designed studies in stuttering treatment research. They complained of a lack of clarity and completeness of reporting issues affecting the replicability of the studies. Blomgren [36] gave a brief overview of stuttering and provided examples of stuttering therapies. Nye et al. [37] conducted a systematic review about behavioural stuttering interventions for children and adolescents and included nine studies representing 327 treated participants

across seven different intervention types. They found treatment effects but no difference between two treatments. Wallace et al. [38] reviewed the evidence of screening and treating children for speech and language. They found no evidence for an impact of screening on speech and language outcomes. With regard to stuttering they found only two trials that examined the Lidcombe Program of Early Stuttering Intervention with significant treatment effect [39,40]. A systematic review of the literature of effectiveness of non-pharmacological interventions for stuttering was published by Baxter et al. [41,42].

Information about the effect of stuttering treatment in Germany has been sparse up to now.

Baumeister et al. [43] studied the effect of a German stuttering therapy summer camp under practice conditions. Forty participants aged 9–19 years were taught a more open handling of their stuttering and received fluency shaping and stuttering modification therapy. Participants <12 years stayed in the camp for 2 weeks; older participants for 3 weeks. The intention of the camp was to lay a basis for future therapies, not to replace other modes of therapy. In terms of percentage of SS, speech rate and naturalness of speech, an improvement could be measured up to 2 months after the camp.

Metten et al. [44] investigated 19 children and adolescents aged 9–19 years who participated in an intensive stuttering treatment in a summer camp for 3 weeks (treatment acc. to Rosenberger). The severity of stuttering and the impact of stuttering on life quality showed improvements in the 10 months after the intensive therapy compared with the results before the treatment.

Lattemann et al. [11] studied the Lidcombe Programme in 46 German pre-school children who were randomly assigned to a waiting group or to the experimental group which received the Lidcombe Programme for 16 weeks. Compared to the waiting group, the treatment group showed a significantly higher decrease in SS in home-measurements and clinic-measurements.

The Kassel Stuttering Therapy (KST) is a computer-assisted German adaptation of fluency shaping. Out of 400 clients (age, 9–65 years) who completed the 2- to 3-week inpatient intensive treatment, long-term data after 1 year ( $n=238$ ), 2 years ( $n=69$ ), and 3 years ( $n=69$ ) were reported [45]. The changes of disfluency from before therapy to 1 year or more after therapy showed effect sizes of  $d>1.1$ . The results after 2 and 3 years were correlated to those after 1 year. Studying the effects of the KST in 32 children aged 9–13 years, the average reduction in dysfluencies in the children was similar to the effects in adults, but the spread of outcomes was wider. The parents rated the success lower than the children themselves [46].

The D.E.L.P.H.I.N. speech treatment for children and adolescents who stutter [47] now has a long tradition in Germany, but its efficacy and efficiency have not yet been assessed. In order to assess the development of the stuttering symptoms, the impact of stuttering on quality of life, but also the drop-out rate, we planned a prospective study (compare Table 1) using questionnaires, standardised videos

**Table 1.** Design of the study.

	6–2 Months before treatment T1	10–6 Weeks before treatment T2	First day of the intensive treatment T3	Last day of the intensive treatment T4	3–6 Months after treatment T5	11 Months after treatment T6	12 Months after treatment T7
SDQ-D			x				x
FzS	x		x	x			x
Text reading	x		x	x			x
Interview	x		x	x			x
Telephone call		x			x	x	
Speech naturalness	x	x	x	x	x	x	x

All measures were used repeatedly.

produced by the therapists and telephone calls made by a speech therapist of the clinic to the home of the patients before and after the treatment.

Our main research questions were whether

1. general quality of life in the view of the participants and in the view of the parents changed from before treatment (T3) compared to 1 year after treatment (T7).
2. the impact of stuttering changed from before treatment (T3) compared to 1 year after treatment (T7).
3. stuttering rate changed from before treatment (T1 + T2) compared to 11–12 months after treatment (T6 + T7).
4. the mean length of the three longest stuttering events changed from before treatment (T1 + T2) compared to 11–12 months after treatment (T6 + T7).

As patients who do not adhere continuously to the therapy are often omitted from the statistics, we included all patients who agreed to take part in the therapy during 1 year. We got their names from the therapists and were able to follow nearly all of them during the whole year after the weeks of intensive therapy. As stuttering therapies should lead to a stable effect, we conducted the last examination 1 year after the intensive therapy.

## Materials

### D.E.L.P.H.I.N. speech treatment for children and adolescents who stutter

This speech treatment aims at older children and adolescents who stutter severely with a high psychological strain and a high motivation to work continuously on their speech. Additionally, a relative (often a parent) must be prepared to follow the therapy and the training at home. This speech treatment is rooted in an in-patient stuttering therapy for children with special needs as practised in a residential school in Hamburg, Germany. There the patients stayed for 1 year without their parents and pupils had to repeat the year at school. Schütz copied this kind of therapy in a comparable institution, the “Sprachheilzentrum Gieszen” and then modified it into an intensive therapy with integration of the parents.

The treatment consists of an initial assessment session, 24 days of in-patient intensive treatment in groups of 7–10 similarly-aged persons, and follow-up care about 6 and 12 months afterwards. In between, patients must carry out

daily homework and conference calls (via skype) with other participants. The D.E.L.P.H.I.N. method is named after the initials of its components: Deblocking impulse, relaxation (German: Entspannung), speech-language pathology (German; Logopädie), Phonetics, Hearing perception training, Intensity, and Nasal swinging accent [47].

The patient learns a completely new speech pattern in three main steps:

The first goal is the acquisition of costo-abdominal breathing, as many patients suffer from tension in the chest region. Then the patient is taught in one-on-one-sessions what Schütz calls the “deblocking impulse,” a kind of sigh, a letting go. The deblocking impulse is formed without pressure. It leads to a relaxation of the region around the larynx and impedes a cramping of the false and true vocal folds. This deblocking impulse should be used in every syllable with the main accent. This technique is practised throughout the whole therapy, during meals, talking to the parent or to other patients. It impedes stuttering and physical movements. Approximately, 5 days later, the second step follows. Now the patient learns what Schütz calls “nasal swinging accent”: a gentle voice onset with nasal character in words beginning with a vowel. The nasal character facilitates a soft voice onset. The patient combines the “deblocking impulse” and the “nasal swinging accent,” in the beginning exaggerating both. Using these techniques, patients realise that they can prevent and impede stuttering when they use this speech modification technique, but that it demands constant awareness and performing of the speech pattern. During this second step, patients are exposed to stressful situations such as shopping, telephone calls, lectures, reciting of poems, discussions, speaking in noise, questions to unknown persons, interviews, or ordering in a restaurant to practise the technique. During the third week, the length and magnitude of the “deblocking impulse” and of the “nasal swinging accent” are decreased, leading to a more and more natural speech. In addition to the one-on-one therapy and group sessions, patients also work in the speech laboratory. Here they wear headphones to dampen the ambient noise. Patients have texts with marks for the “deblocking impulse” and the “nasal swinging accent” and read these texts aloud using the speech technique. This is recorded and then patients listen to themselves to monitor their management of the technique. Additionally, videos of the training in different situations with high stress levels are analysed. Hearing perception training is used to get auditory feedback for correctness of

breathing, deblocking impulse, and timing of speech. During the intensive therapy, a patient attends 140 sessions (mostly group sessions). Additionally, every day patients participate in sessions of progressive muscle relaxation according to Jacobson. These instructions are given in the speech technique. Furthermore, the therapy includes European drum sessions to foster sense of community and to improve concentration and coordination skills. In contrast to African drum playing, where the right hand is mainly used, here the player alternates from one hand to the other after each beat; both halves of the brain are activated and are said to be better connected. During the drum sessions, the speech technique is used, and for some patients the movement of the hands facilitates the speech technique. The patients internalise the speech technique more rapidly. The patients take part in three to four sessions a week and continue playing at home.

Back at home, participants are tasked with performing daily exercises. Each patient receives an individual training programme. To guarantee consistent use of the D.E.L.P.H.I.N. speech modification technique at home, patients are furthermore required to be supervised by an adult. During intensive treatment, adults are taught how to monitor and correct the patient's speech in order to prepare them for their supervisory role at home. The patient's school or workplace is informed about the speech technique to help facilitate the transfer to daily life there. The patient gives a lecture on the first day after the intensive therapy. During the 2 years after the intensive therapy, the patient has maximally 2 weeks and four single days for the stabilisation phase, usually 50–80 logopedic sessions. The speech technique the patients use is the same for all age groups. The texts used are adapted to the specific age group. In the therapy of children, the work of the parents is more relevant as children can monitor themselves less reliably. In adolescents, psychosocial burden and anxiety are more important. For this reason, training of real situations is more often practised. To date, this intensive therapy has only been delivered by the developer and her daughter. A treatment protocol is not publicly available. The courses of instruction for other therapists lasting 1 year are planned for the future. So far, the developer and her daughter have also brought their therapy to South Africa, where treatment was conducted in English.

## Participants

Participants were 56 school-age children, adolescents, and mainly young adults aged 8–36 years (mean, 15.3; median, 13.0; SD, 6.4 years), most of them previously diagnosed as stuttering severely. Forty-two participants were males and 14 females. All 56 participants who attended the treatment between 1 January 2014 and 15 February 2015 gave their consent to take part in the study. Three participants stopped the treatment prematurely. Patients were conjoined in groups of similar age irrespective of their gender or the severity of the stuttering. Group 1/14, 2/14, 3/14, and 1/15

comprised 10 participants, group 4/15 7 participants, and group 5/15 9 participants.

Nearly, all participants had previously had undergone stuttering treatment (49 out of 51; for five participants no information about preceding therapy was available), 29 of whom had had more than one course of treatment. In most cases (20) therapy had been conducted once a week, in 17 cases twice weekly, in 11 cases three times weekly and in four cases more often than that. For 27 of the participants, a history of receptive and expressive language disorder was reported. Other comorbidities were attention-deficit hyperactivity (HA) disorder in three, perception disorders in four, learning disability in six, and motoric disorders in six participants. The study was approved by the Ethical Committee of the Federal Medical Association. All participants and all parents signed informed consent forms.

## Methods

A repeated-measures design was used. As summarized in Table 1, four outcome measures were used.

General quality of life in the view of the participants and in the view of the parents was assessed before treatment (T3) and 1 year after treatment (T7) using the Strength and Difficulty Questionnaire (SDQ) [48–50]. The Fragebogen zum Sprechen (FzS) – Speech Questionnaire [51] was completed twice (T1, T3) before treatment (the diagnosis schedule and the first day of treatment) and twice after treatment (T4, T7), once on the last day of intensive treatment and once 1 year later.

The number of syllables stuttered and the mean length of the three longest stuttering events were assessed during interviews (T1, T3, T4, T7), text reading (T1, T3, T4, T7), and telephone calls (T2, T5, T6). The interval between T2 (telephone call after agreeing to participate but before the beginning of the intensive treatment) and the beginning of the treatment was 35.0 days (mean and median, SD 17.6), that between the end of the intensive therapy (T4) and the first telephone call after this therapy was  $120.4 \pm 21.3$  days (mean  $\pm$  SD; median: 117), and that between the end of the intensive therapy (T4) and the last telephone call after this therapy was  $336.5 \pm 9.7$  days (mean  $\pm$  SD; median: 335). For the calculations of the percentage of syllables stuttered and the mean length of the longest stuttering events, both assessments before the treatment (T1 for interview, text reading, and T2 for telephone call) and both assessments in the long term (T6 for telephone call and T7 for interview and reading) were summarised.

## Strength and Difficulties Questionnaire

The SDQ is a short questionnaire that addresses positive and negative behavioural attributes of children or adolescents [48–50]. Its good psychometric properties have been confirmed by many studies worldwide. The 25-item rating scale addresses emotional symptoms (ES), HA, inattention-conduct problems (CP), peer problems (PP), and pro-social behaviour, (social strengths, e.g. altruism). During

development, special attention was given to a well-balanced inclusion of both positive and negative behavioural aspects, and hence the raw scores of several items need to be inverted before they are summed up to scale values. The scores of ES, HA, CP, and PP are summarised to the “Total Difficulty Score”. Strength and Difficulties Questionnaire versions are available for parents, teachers, and as self-ratings for children aged 7 years and older. There are three response categories: 0 = not true, 1 = somewhat true, and 2 = certainly true. As demonstrated by Becker and co-workers [50], the presence of any clinical diagnosis of psychopathology is mirrored by significantly elevated scores in all SDQ scales. Cut-off values (based on normative SDQ scoring bands) allow discrimination between clinical cases (high difficulties, distinct mental health disorders), non-cases (low difficulties, normal), and borderline cases (medium difficulties, between normal and clinical). About 10% of the participants in the norm sample were classified as clinical cases, 10% as borderline cases, and 80% as normal. The SDQ has been translated and validated for the German language [50] with German norms for the parent ratings [48,49]. For the scoring, actual norms taken from the SDQ homepage were used. We used the total score as described in the manual, summarising the four subscales “emotional,” “conduct,” “HA,” and “peer,” but not “prosocial.” We used only those questionnaires in which in each subscale at least 60% of the items were marked. For our further calculations, we did not use the classification as normal, borderline, or clinical case but instead the raw values.

### ***Fragebogen zum sprechen – speech questionnaire***

The Speech Questionnaire was developed and validated by Cook [51] to measure the impact of stuttering on the lives of children and adolescents aged 8–17 years. Cook aimed to develop an assessment based on the framework of the International Classification of Functioning (ICF). The ICF describes all health-related experiences in terms of structure and function of the body and the activities a person might engage in during participation in daily life.

Yaruss and Quesal [52] adapted the ICF framework to the study of stuttering. Following these authors, stuttering must be looked at in terms of several interacting components, not only the impairment in body function indicated by the observable characteristics of stuttering but also:

- the speaker’s affective, behavioural, and cognitive reactions to stuttering;
- the effects of the environment on stuttering, indicated by difficulty in different speaking situations and reactions of others;
- the overall impact of stuttering on the speaker’s life, indicated by limitations in communication activities and restrictions in participation in daily life.

Taking these components into account, Cook developed a short instrument of 27 items with a 6-point Likert scale [51]. The questionnaire covers attitudes towards life as a

person who stutters (five items), emotions when stuttering (seven items), different speaking situations (eight items), and the impact of stuttering on school performance, friendship, and the ability to achieve one’s own goals (seven items). The direction of the answers was changed – in some the highest agreement corresponded to a major handicap; in other items to the absence of a handicap. The absolute total score can vary between 27 and 162, lower values corresponding to a low psychosocial load of the stuttering and higher values as an expression of a high burden. In a study with 147 children and adolescents, Cook found that the FzS is a reliable and valid instrument for measurement of health-related quality of life. Comparing the results of the questionnaire to those of the Stuttering Severity Instrument-3, Cook proposed a classification of total scores into nine degrees of severity.

For the English language, the Assessment of the Child’s Experience with Stuttering (ACES) was developed by Yaruss and Quesal [52–54]. This questionnaire comprises 98 questions and collects information about general perspectives on stuttering, affective, behavioural, and cognitive reactions to stuttering, functional communication difficulties, and impact of stuttering on the speaker’s quality of life. This questionnaire was translated into German by Metten et al. [44], but was judged to be too long for our purposes. The German version of OASES-T for teenagers was not available at the start of the study. For these reasons, the Fragebogen zum Sprechen (FzS) was used for this study. Participants completed the FzS at baseline, on the first and last day of the intensive treatment and 1 year after completing the therapy. For our further calculations, we used the raw values.

### ***Percentage of SS and mean length of the three longest stuttering events***

As part of the clinical routine, standardised situations are video documented about 6–2 months before treatment (when the participants present themselves at the therapy centre to decide whether an intensive treatment is indicated; T1), on the first day of the intensive treatment (T3), on the last day of the intensive treatment (T4) and 1 year after the intensive treatment (T7). In this study, the videos of interviews with semi-standardised questions and text reading were used for further analysis.

Additionally, beyond-clinic audio recordings were made of the participant’s speech during telephone conversations with a caller unknown to the participant or parent (T2, T5, T6). All telephone calls were made to a home landline or to a mobile phone. The caller was a research assistant at the university clinic who recorded the conversation over the phone.

For each mode of speech, the percentage of stuttered syllables (% SS) was calculated for 250 syllables, but not the very first ones to allow accommodation to the situation. To determine the mean length of the longest stuttering event, the file was displayed on a computer with the programme “Computerized Speech Lab.” The longest stuttering events

were marked and measured, and then the three longest stuttering events were used to calculate the mean.

### **Speech naturalness**

Speech naturalness was judged by the speech therapist who performed the acoustic analysis. As proposed in the SSI-3 [55], we judged distracting sounds, facial grimaces and head movements in the videos of the interviews on a scale from 0 to 5 (0 = none; 1 = not noticeable unless looking for it; 2 = barely noticeable to casual observer; 3 = distracting; 4 = very distracting; 5 = severe and painful-looking).

In all samples, we also assessed the following three acoustic parameters on a scale from 0 = normal to 2 = markedly deviant:

1. prosody, which was classified as normal – minimally deviant – markedly deviant (e.g. monotone or exaggerated),
2. breathing, which was classified as normal – minimally deviant – markedly deviant (e.g. inspiratory phonation, speaking on residual air)
3. tonus during phonation, which was classified as normal – minimally deviant – markedly deviant (hypertonus, hypotonus).

### **Statistics**

We conducted all analyses in R V. 3.3.1 (R Foundation for Statistical Computing, Vienna, Austria, 2016) using the packages lme4 (V. 1.1-15) and lmerTest (V. 2.0-36). The visualisations were obtained using the package ggplot2 (V. 2.2.1). For descriptive analyses, mean values, SDs, and median and quartiles were calculated for the four primary endpoints at baseline (pre-treatment) and about 1 year after treatment. For the secondary endpoints regarding speech naturalness, absolute and relative frequencies were calculated at all time points. For confirmatory analyses, the four outcome variables SDQ, FzS, stuttering rate, and mean length of stuttering were tested for a difference at different points in time (pre-treatment vs. 1 year after treatment). The analysis was specified before closing the data base.

For the evaluation of statistically significant differences between the two points in time, a generalisation of ANOVA with repeated measurements was used. Specifically, separate linear mixed models for the four endpoints were fitted using the point in time as a fixed effect. The variables gender, age group (<17 vs.  $\geq 17$ ) and survey method (depending on endpoint: self-reported questionnaire/parent-reported questionnaire for SDQ, personal interview/reading text/telephone interview for stuttering rate, and mean length of stuttering) were used in the model as adjusting variables. The assumption that the effect of the treatment is the same in each age group and gender group was checked beforehand visually. Additionally, we included the information at all other points in time, but allowed for a non-linear effect of time. To model different temporal progressions between survey methods, we included an interaction between the last point in

time and the survey method. We expected the values of one individual and members of the same treatment group to be dependent. To account for these dependencies, we included random effects for each individual and each treatment group, allowing for different intercepts. The model assumptions (i.e. normality of residuals) were checked visually. Marginal  $p$  values over the different types of surveys per endpoint were obtained using a likelihood ratio test comparing models with and without considering the effect of the last time point. The estimated effects for the difference between pre- and 12 months post-treatment are presented together with standard errors and  $p$  values. The assumption that the treatment effect is the same in both genders and age groups was checked in a sensitivity analysis (results not shown).

We used the bootstrap method with 1000 bootstrap samples to estimate the estimated marginal mean values for a new individual based on the obtained models. The global level of significance  $\alpha$  was set to 0.05. Using the Bonferroni method to adjust for multiple comparisons, the four hypotheses were tested on a local level of significance of 0.0125 using confirmatory methods.

### **Ethical approval**

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. The study was approved by the Ethical Committee of the Federal Medical Association [Ethikkommission der Landesärztekammer Rheinland-Pfalz, Bearbeitungsnummer 837.366.13 (9051-F)]. Informed consent was obtained from all individual participants and all parents included in the study.

## **Results**

### **Results at baseline for the questionnaires**

The values for the SDQ and the FzS were judged visually as normally distributed.

### **Subjective rating of quality of life as given in the SDQ**

At baseline, participants reached a total score in the SDQ of  $11.7 \pm 5.4$  (median 11.0, range, 2–26); parents a mean  $10.7 \pm 7.4$  (median 9.5, range 0–36, 10 missing values).

In their representative study, Woerner et al. [48] determined cut-off scores and used these to assign each raw score to one of the three categories (normal  $\leq 15$ , borderline 16–19, abnormal  $\geq 20$  for self-assessment and normal  $\leq 13$ , borderline 14–16, abnormal 17–40 for parents). At baseline, 43 participants reached a normal total score in the SDQ, 8 a borderline one, and 5 an abnormal one. The total scores of the parents were normal for 29 participants, borderline in 11 cases, and abnormal in 6 cases (10 missing values).

**Speech questionnaire (FzS).** The FzS was used to assess self-reported speech pathology and influence of stuttering on different life situations. The mean value for the whole group was 106.08 ( $\pm 24.20$ , 20 missing values); the median 107. Five participants had a value of max. 80 points, corresponding to a maximally moderate stuttering (percentile rank, 1–60), four participants reached 81–91 points (moderate to severe, PR 61–77), six were between 92 and 98 (severe, PR 87–88), three between 99 and 106 (severe to very severe, PR 89–95), and 18 had 107 or more points (very severe, PR  $\geq 96$ ).

For both the SDQ and the FzS, the values for males were lower on average than for females (SDQ: -1.31, SE 1.64,  $p$  value .43, FzS: -10.07, SE 3.92,  $p$  value .013) and the values for younger participants were lower on average than for older ones (SDQ: -1.39, SE 1.20,  $p$  value <0.001, FzS: -18.68, SE 4.72,  $p$  value <0.001).

### Variation in time for the questionnaires

General quality of life was measured using the SDQ, which was completed by participants and their parents. The results for children and adolescents (SDQ self) and their parents (SDQ parents) on the first day of the intensive treatment and 12 months after treatment are summarized in Table 2. These are raw values and should be interpreted with care, as the correlation between values due to the fact that the same individuals were observed is not accounted for and the values are not adjusted for group effects.

We observed on average higher values for older participants, corresponding to a more reduced quality of life, than those for younger participants (+1.39, SE 1.86,  $p$  value .46), and higher values on average for females compared to males

(+1.32, SE 1.64,  $p$  value .43). For the calculations, an adjustment for age, gender, and method of gathering (self-reported or parent-reported) was applied. We found a significant difference ( $p < .001$ ) between the values assessed on the first day of the treatment and 12 months after the treatment, both for the results of the participants (-2.93, SE 0.80) and for those of the parents (-1.66, SE 0.87) adjusted for age and gender.

The Fragebogen zum Sprechen, the speech questionnaire (FzS, [51]) was completed by each participant twice before the therapy (T1, T3), after the intensive therapy (T4), and 1 year later (T7). The values for the whole group are listed in Table 2. These are raw values, the correlation between values due to fact that the same individuals were observed is not accounted for and the values are not adjusted for group effects. During the study, it turned out that after the initial treatment some of the test persons did not answer all questions but noted instead on the questionnaire that they no longer stuttered and thus found these questions inappropriate for them now. For this reason, the main analysis was conducted by setting these answers to the best possible answers meaning that we assumed the participant felt no impact by his speech in that situation. In a separate sensitivity analysis, the corresponding answers were set to "missing," but the results of this analysis (data not shown) did not differ from those of the main analysis.

On average, the answers of the older participants showed more severe effects of stuttering than those of younger ones (+18.68, SE 4.72,  $p$  value <.001); those of females had higher values on average than those of males (+10.07, SE 3.92,  $p$  value .0130). Between the diagnostic stage before therapy and 1 year after the therapy adjusted for gender and age, a significant reduction of problems with stuttering was found (-36.04, SE 3.80,  $p < .001$ ) although there was some

**Table 2.** Results in the Strength and Difficulty Questionnaire for children and adolescents (SDQ self) and their parents (SDQ parents) on the first day of the intensive treatment and 12 months after treatment and in the Speech Questionnaire (FzS) 2–6 months before treatment, on the first and last day of the intensive treatment and 12 months after treatment.

	Before treatment T1/T2	First day of the intensive treatment T3	Last day of the intensive treatment T4	12 Months after treatment T7
SDQ self				
Mean		11.70		8.47
SD		5.43		5.24
Median		11.00		8.00
Q1		7.00		4.75
Q3		15.00		11.00
<i>n</i> (% out of <i>N</i> = 56)		56 (100%)		46 (82.14%)
SDQ parents				
Mean		10.72		8.86
SD		7.42		7.70
Median		9.50		6.50
Q1		5.00		3.00
Q3		14.75		14.25
<i>n</i> (% out of <i>N</i> = 56)		52 (92.29%)		46 (82.14%)
FzS				
Mean	106.08	101.50	63.38	66.28
SD	24.20	20.90	17.99	23.88
Median	107.50	98.00	60.00	62.00
Q1	91.50	87.00	50.50	48.50
Q3	122.25	116.50	76.50	82.25
<i>n</i> (% out of <i>N</i> = 56)	36 (64.29%)	55 (98.21%)	55 (98.21%)	51 (91.07%)

High values stand for high burden.

**Table 3:** Results (mean, standard deviation, median, first and third quartile, number of completed questionnaires) of the analysis of the frequency of SS and the mean length of longest event in the interview, the text reading, and the unannounced telephone call for the whole group and all assessments.

	Before treatment T1/T2	First day of the intensive treatment T3	Last day of the intensive treatment T4	3–6 Months after treatment T5	11–12 Months after treatment T6/T7
<b>Percent SS</b>					
Interview					
Mean	12.34%	12.50%	2.32%		4.20%
SD	11.00%	11.17%	4.98%		6.51%
Median	9.62%	10.20%	0.80%		1.60%
Q1	4.70%	5.00%	0.40%		0.40%
Q3	14.40%	16.30%	2.04%		4.40%
n (% out of N = 56)	56 (100%)	56 (100%)	55 (98.21%)		53 (94.64%)
Text reading					
Mean	15.54%	13.50%	1.18%		2.25%
SD	13.70%	11.98%	1.86%		5.61%
Median	11.60%	10.00%	0.40%		0.40%
Q1	4.40%	3.10%	0.00%		0.00%
Q3	22.00%	19.80%	1.60%		1.60%
n (% out of N = 56)	55 (98.21%)	56 (100%)	55 (98.21%)		52 (92.29%)
Telephone call					
Mean	12.66%			3.94%	4.32%
SD	8.95%			5.27%	5.17%
Median	10.80%			2.00%	2.00%
Q1	5.60%			0.80%	1.00%
Q3	17.20%			4.40%	6.00%
n (% out of N = 56)	53 (94.64%)			55 (98.21%)	55 (98.21%)
<b>Mean length of longest event</b>					
Interview					
Mean	2.92	3.21	1.25		2.05
SD	2.63	2.41	1.99		5.88
Median	2.05	2.65	0.53		0.60
Q1	1.23	1.69	0.10		0.10
Q3	3.53	3.90	1.22		1.53
n (% out of N = 56)	56 (100%)	56 (100%)	55 (98.21%)		53 (94.64%)
Text reading					
Mean	3.51	3.55	0.91		1.26
SD	3.93	3.16	1.66		4.15
Median	2.33	2.75	0.20		0.10
Q1	1.33	1.34	0.00		0.00
Q3	4.15	4.49	1.13		0.42
n (% out of N = 56)	55 (98.21%)	56 (100%)	55 (98.21%)		52 (92.29%)
Telephone call					
Mean	2.03			0.70	0.76
SD	1.40			0.74	0.93
Median	1.67			0.43	0.43
Q1	0.93			0.30	0.30
Q3	2.73			0.90	0.80
n (% out of N = 56)	53 (94.64%)			55 (98.21%)	55 (98.21%)

heterogeneity, indicating that some participants had experienced a worsening and others a further amelioration since the end of the intensive treatment T4.

#### Variation in time for the stuttering rate and mean length of the three longest stuttering events

The stuttering rate and mean length of the three longest stuttering events were determined via an interview and text reading twice before therapy, at the end of the intensive therapy and 1 year after the therapy. Additionally, the participants were called on the telephone before the start of the therapy, and 3–6 months and 11 months after the intensive therapy. Raw results are presented as the medians, quartiles, means, and standard deviations in Table 3, but again these values do not account for correlation and group effects and must be interpreted with due care. Although the values for

stuttering frequency and the mean of the longest stuttering events were not normally distributed, we have calculated means and standard deviations in addition to median and quartiles to allow for comparisons as this has also been done in most previous publications.

For the stuttering rate, older participants were estimated to present slightly more severe problems than younger ones (+0.0034, SE 0.0191, *p* value, .86) and females slightly less severe problems than males (-0.0024, SE 0.0195, *p* value .90). With adjustment for age and gender, the amelioration between the diagnostic stage 2–6 months before therapy and 12 months after the intensive therapy proved to be statistically significant (Interview: -0.08, SE 0.01, Reading: -0.12, SE 0.01, Telephone: -0.08, SE 0.01, *p* < .001).

The difference in the mean length of the three longest stuttering events, adjusted for age and gender, was statistically significant (Interview: -1.12, SE 0.46, Reading: -1.95, SE

**Table 4.** Primary results of treatment efficacy after 1 year for SDQ, FzS, stutter rate, and mean length of stuttering.

	Type	Estimate	Std. Error	Marginal p value*
SDQ	Self-survey	-2.927	0.800	<.001
	Parent-survey	-1.656	0.866	
FzS Stutter rate	Questionnaire	-36.036	3.791	<.001
	Interview	-0.084	0.011	<.001
Mean length of stuttering	Reading	-0.122	0.011	
	Telephone	-0.084	0.012	
	Interview	-1.117	0.458	<.001
	Reading	-1.947	0.462	
	Telephone	-1.311	0.505	

\*The effect is measured as the mean difference between 12-month post-treatment and prior treatment (value before treatment minus value 12-month post-treatment) separately for the types of measurement per endpoint and adjusted for age group and gender. This is listed under "Estimate." The corresponding standard error of the estimate is given under "Std. Error." The marginal *p* value is obtained using a likelihood-ratio test comparing a model with consideration of the effect of the last time point versus a model without consideration of the effect of the last time point and is calculated marginal over the types of measurement.

0.47, Telephone: -1.31, SE 0.51, *p* < .001). We observed on average longer stutter events for females (+0.688, SE 0.492, *p* value .168) and shorter stutter events for older people (-0.271, SE 0.484, *p* value .58).

Effect estimates for the difference 12 months post-treatment to prior treatment, adjusted for age group and gender, using the information of all time points with a mixed model are listed in Table 4. Here, the estimated mean difference is evident. For the FzS, the raw value is 36 points lower. For the stuttering rate, the effect was 0.084 for the interview and the telephone call and 0.122 for text reading.

### Speech naturalness

The results for distracting sounds, facial grimaces, and head movements in the videos the interviews were judged on a scale from 0 (normal) to 5 (markedly deviant). Additionally, data on three perceptive parameters (prosody, breathing, and tonus) are reported here (classified 0–2). For these data, no statistical calculations were done and the results are presented only descriptively. The values need to be interpreted with due care as the intra-patient correlations and group effects were ignored.

Figure 1 shows the development of distracting sounds, facial grimaces, and head movements. Distracting sounds apparently improved from the beginning of the therapy to the end of the therapy and had even improved over the period of 1 year after the therapy. Here the number of patients who were classified as normal (0) or not noticeable unless looking for it (1) increased.

Figure 2 shows the classification for prosody, breathing, and tonus for all assessment points. Prosody seemed to worsen from the beginning of the therapy to the end of the therapy; 1 year later it resembled the status before therapy. Tone and breathing seemed to worsen from the beginning of the therapy to the end of the therapy; after 1 year both parameters were judged as improved compared to before the therapy.

### Discussion

Our data present evidence for the efficacy of the investigated therapy in that we found significant improvement of the symptoms, evident in reduction of the stuttering rate, and an amelioration of general quality of life (SDQ) and psychosocial self-esteem of a person who stutters (FzS).

The patients accepted at the treatment centre were older children, adolescents, and young adults with severe stuttering, in other words those with a persisting stuttering. Nearly all of them had undergone stuttering treatment before without long-lasting effect. Patients and their relatives therefore suffered from a severe communication disorder and were willing to invest time and energy in the therapy. Several earlier studies have shown that an intensive treatment of stuttering is more effective compared to a therapy with low intensity [3].

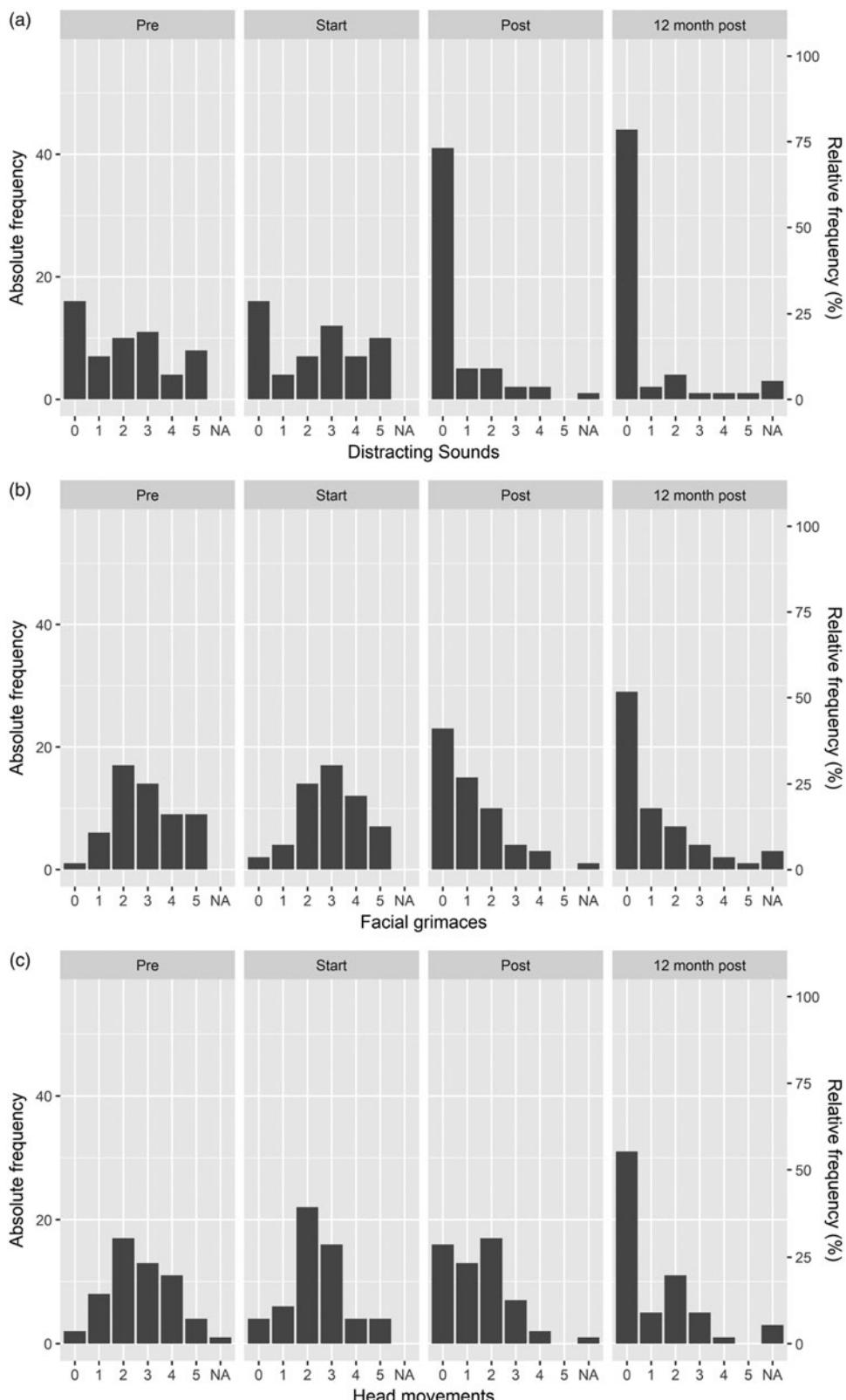
The present trial investigated the fluency shaping stuttering therapy as developed by Schütz [47]. Stuttering rate was shown to be reduced significantly for the comparison between before and 1 year after the intensive treatment. Although the effect was seen to be stronger at the end of treatment compared to 12 months after treatment, the effect was still significant after 12 months and the decrease of the effect is moderate. The same was the case for the three most extended stuttering events.

In addition, we also looked at the quality of life as measured with the SDQ completed by participants and their parents and the subjective assessment of stuttering by the participants themselves in the speech questionnaire, both of which demonstrated significant improvement.

We examined all patients who were willing to participate in the therapy. None of the patients who were treated in the centre between 1 January 2014 and 28 February 2015 declined to participate in the study. For this reason, we had a broad range of age. The FzS and the SDQ are questionnaires developed for subjects under 18 and 17 years, respectively [48,49,51]. For this reason, we conducted the calculations adjusted for subjects <17 years and above and for males and females. Older subjects and both genders reacted to the therapy in a comparable way (sensitivity analysis, results not shown), but we observed that older subjects complained on average of a more severe handicap and the SDQ demonstrated more severe stuttering symptoms. However, we observed a relatively high *p* value and thus it cannot be ruled out that the effect was observed by chance.

As in most other studies about stuttering, we included substantially more male participants. Interestingly, male participants <17 years reported on average less subjective impact of the stuttering but they demonstrated more severe stuttering symptoms compared to female participants. Here, the observed *p* values indicate a statistically notable effect. As this analysis was solely performed exploratively, this needs to be validated in a separate study.

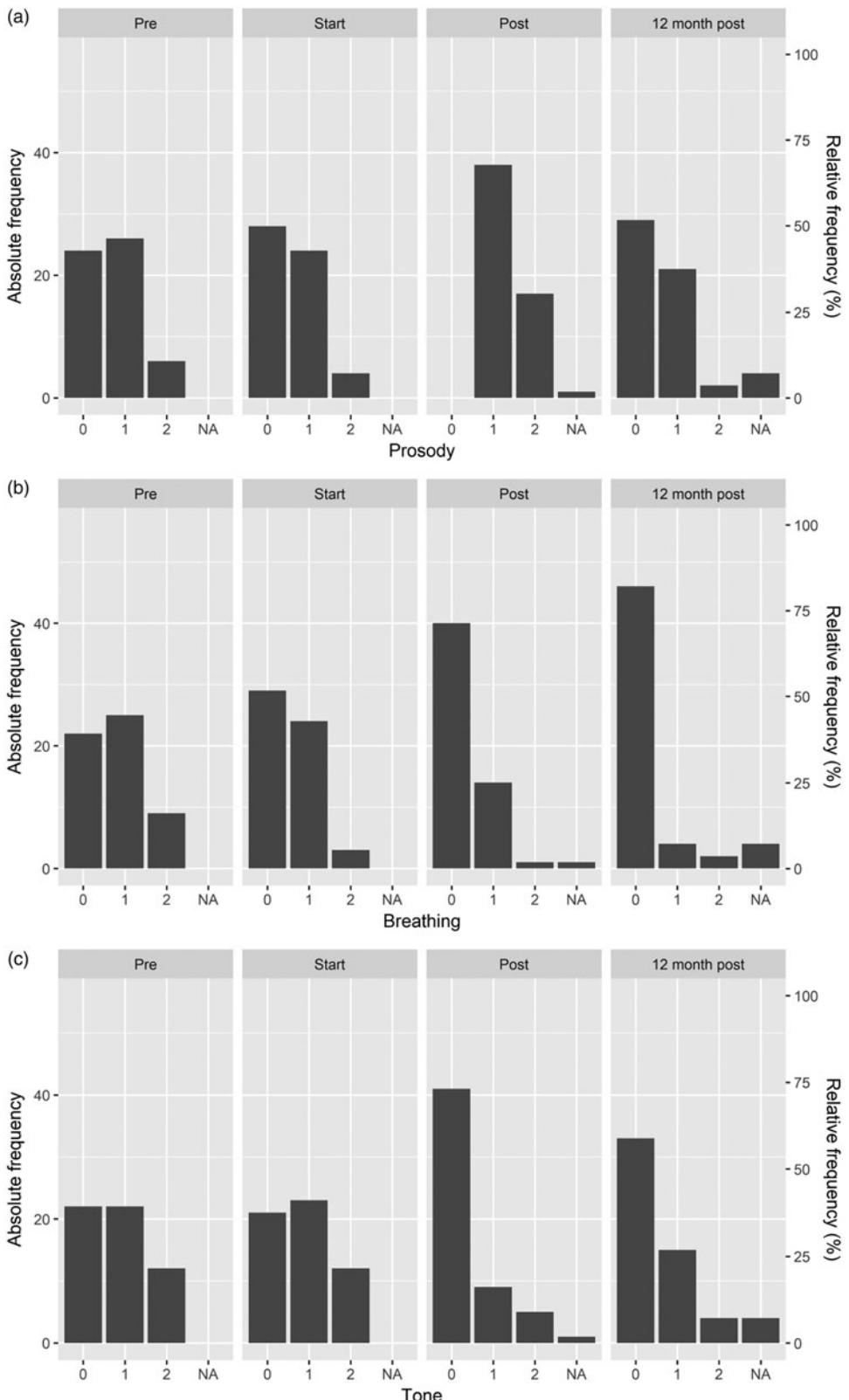
Euler and his group [56] conducted a retrospective evaluation of stuttering treatment by the clients themselves. Clients judged two methods (stuttering modification and fluency shaping) as effective and three methods (breathing treatment, hypnosis, and unspecific logopedic treatment) as



**Figure 1.** Development of the assessment of distracting sounds (a), facial grimaces (b), and head movements (c) for the interview, classified as 0 = normal to 5 = severe and painful-looking, and NA for missing value. The results presented as absolute (left, y-axis) and relative (right, y-axis) frequencies.

ineffective. Stuttering modification and fluency shaping were perceived as equally effective. Further analysis of the data showed that group treatment was generally superior to individual treatment [56]. The authors also stressed the intensity

of treatment and advocated at least periods of intensive treatment, something which contrasts strongly with common treatment approaches in Germany. Most patients nominally receive a single session per week, in reality less due to



**Figure 2.** Development of the classification for prosody (a), breathing (b), and tonus (c) for all assessment time points separately for the interview, classified from 0 = normal to 2 = markedly deviant and NA for missing value. The results presented as absolute (left, y-axis) and relative (right, y-axis) frequencies.

holidays and illness on the part of either patient or therapist. Relevant characteristics of treatment such as the use of fluency shaping techniques, the intensive character of therapy over weeks, and the interval character are realised both in the Kasseler Stutter Therapy [45] and in the therapy

studied here [47]. Euler et al. [45] reported on about 400 clients and followed 69 of them for more than 1 year. Disfluency rates collected in four different speech situations were reduced from 12.6% SS before therapy to 1.6% after therapy and remained in the following 3 years between 3.2

and 3.8% SS in those who could be examined. Subjective stuttering data (self-rating of stuttering severity and of avoidance of speech or speech situations) reflected the same trends, but to a lesser extent.

In contrast to other long-term outcome studies, we were able to collect almost complete data. The original sample consisted of 56 participants who could afford the costs of the treatment, either by themselves or with the support of their health insurance company, and who were assessed at the treatment centre as suffering from psychological strain due to severe stuttering. In total, 53 of these participants followed the complete therapy programme and we were able to collect all data including examination 1 year after the completion of the intensive therapy. We collected 601 out of a possible 616 samples; 2.4% were missing. We had no missing data for the SDQ, all patients and all relatives answered both times, nor for the FzS, as all participants gave back two questionnaires before the therapy and two after the therapy.

Bloodstein and Bernstein-Ratner [34] proposed 12 criteria to judge stuttering treatment. With the present investigation, most of these criteria are fulfilled. We present prospective data including objective measurements and data on self-evaluation of quality of life in a repeated-measures design through to 1 year after treatment. We did not fulfil the sixth criterion because of the lack of a control group, but the repeated measurements before the start of the intensive treatment demonstrate a relatively constant symptomatology. Another limitation (criterion 11 in Bloodstein and Bernstein-Ratner) is the fact that the described therapy has so far only been practised by the developer and her daughter. They plan to teach her method to other speech-language pathologists in which case future studies will be able to evaluate the efficiency of this method when applied by other therapists.

## Conclusions

In our study, we found that the therapy studied was associated with an improvement of the relevant endpoints quality of life, impact of stuttering, and severity of stuttering. We found the most relevant effect sizes for impact of stuttering on life quality, but also for the stuttering rate and mean length of the three longest stuttering events in the interview.

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## Disclosure statement

No potential conflict of interest was reported by the authors.

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