Journal of Communication Disorders 89 (2021) 106073

Contents lists available at ScienceDirect

Journal of Communication Disorders

journal homepage: www.elsevier.com/locate/jcomdis

Speech restructuring group treatment for 6-to-9-year-old children who stutter: A therapeutic trial

Harald A. Euler[a,*], Anna Merkel[b], Katja Hente[b], Nicole Neef[c], Alexander Wolff von Gudenberg[b], Katrin Neumann[a]

[a] Department of Phoniatrics and Pediatric Audiology, University Hospital Münster, Westphalian Wilhelms University of Münster, Kardinal-von Galen-Ring 10, 48149, Münster, Germany
[b] Institute of the Kassel Stuttering Therapy, Feriendorfstr. 1, 34208, Bad Emstal, Germany
[c] Department of Diagnostic and Interventional Neuroradiology, Georg-August-University, Robert-Koch-Str. 40, 37075, Göttingen, Germany

ARTICLE INFO

Keywords:
Stuttering
Treatment
Effectiveness
Speech restructuring
Fluency shaping
Children

ABSTRACT

For children who stutter (CWS), there is good evidence of the benefits of treatment for pre-school age, but an evidence gap for elementary school age. Here we report on the effectiveness of a fluency shaping treatment for 6- to 9-year-old children. The main treatment component is the reinforcement of soft voice onsets. An intensive in-patient group treatment phase lasts 6 days, followed by a 6-month maintenance phase with 3 in-patient weekend group refresher courses. Child and a parent participate together in various treatment activities. In this controlled intervention study (waitlist control, intention-to-treat design) assessments were performed before treatment (T1), 4 weeks after the intensive phase (T2), at the end of the maintenance phase (T3), and 1 year later (T4). Participants were 119 children (108 boys, 11 girls, age 5.5-10.4 years). Control conditions included a subgroup with delayed treatment (N=25) as well as the assessment of complexity of utterances, inter-rater reliability, and speech naturalness. From before treatment to 1-year follow-up, percent stuttered syllables and OASES-S (Overall Assessment of the Speaker's Experience with Stuttering - School-age) scores decreased with large effect size. Speech naturalness improved during this period but did not reach the level of non-stuttering children. Complexity of utterances increased during the intensive phase, but only temporarily. Twenty children (16.8 %, including dropouts) showed no demonstrable treatment benefit. Fluency shaping treatment can be effectively applied to young school children. It is assumed that parental support, group therapy, intensive treatment, and regular exercises at home are essential.

1. Introduction

Evidence-based stuttering treatment methods show a mixed picture. There are efficacious treatment options for preschool children (e.g., Harris, Onslow, Packman, Harrison, & Menzies, 2002), and the same can be said for adolescent and adult persons who stutter (Neumann et al., 2016, 2017). But solid evidence-based treatment (EBT) options for school children are rare if current criteria for good

* Corresponding author.
E-mail addresses: euler@uni-kassel.de (H.A. Euler), anna.merkel@kasseler-stottertherapie.de (A. Merkel), katja.hente@kasseler-stottertherapie.de (K. Hente), nneef@gwdg.de, neef@cbs.mpg.de (N. Neef), awvgudenberg@kasseler-stottertherapie.de (A. Wolff von Gudenberg), Katrin.Neumann@uni-muenster.de (K. Neumann).

https://doi.org/10.1016/j.jcomdis.2020.106073
Received 28 March 2020; Received in revised form 4 October 2020; Accepted 5 December 2020
Available online 29 December 2020
0021-9924/

evidence (e.g., Oxford Centre for Evidence-Based Medicine [OCEBM], Levels of Evidence Working Group, 2011) are applied. Case studies or efficacy studies with one-digit sample sizes, for example, would not constitute good evidence, nor would studies without follow-up. In a systematic review, Nye et al. (2013) evaluated the evidence quality of existing treatment studies of children who stutter (CWS) using the checklist by Downs and Black (1998) of methodological quality and found nine studies with a quality rating of 14 points or higher, of 27 possible points (Craig et al., 1996; Franken, Boves, Peters, & Webster, 2005; Harris et al., 2002; Harrison, Onslow, & Menzies, 2004; Jones et al., 2008; Lattermann, Euler, & Neumann, 2008; Lewis, Packman, Onslow, Simpson, & Jones, 2008; Riley & Ingham, 2000; Ryan & Van Kirk Ryan, 1995). Of these nine studies with good evidence, seven focused on the treatment of CWS in preschool age. Two studies (Craig et al., 1996; Ryan & Van Kirk Ryan, 1995) had included some elementary school children in their sample of children and adolescents but did not provide separate results for these children.

The lack of solid treatment studies, particularly for school-aged children, has been noted before (Bloodstein & Bernstein Ratner, 2008) and has been explicitly criticized (Nippold & Packman, 2012; Nippold, 2011, 2012). The situation has improved only moderately since then if strict inclusion criteria are applied, namely two-digit sample sizes, follow-up data of at least three months, control of key variables, and reporting of effect sizes or data which allows their subsequent calculation. With these inclusion criteria, only few EBT options for school-age CWS are available (Andrews et al., 2012, 2016; Koushik, Shenker, & Onslow, 2009). Several other studies must be disregarded here because treatment results for young school children are not reported separately (Keilmann, Neumann, Zöller, & Freude, 2018; Metten, Zückner, & Rosenberger, 2007; Millard, Zebrowski, & Kelman, 2018; Senkal & Ciyiltepe, 2018) or because of other methodological deficits like missing follow-up data or insufficient sample sizes (Budd, Madison, Itzkowitz, George, & Price, 1986; Laiho & Klippi, 2007; Wolff von Gudenberg, Neumann, & Euler, 2006).

Treatment gains may be estimated either by efficacy studies, that is, treatment under well-controlled research conditions, or by effectiveness studies, that is, treatment under everyday conditions in the field (Cochrane, 1972). Efficacy studies determine whether a treatment *can* work, whereas effectiveness studies determine whether it *does* work (Haynes, 1999). Effectiveness studies are the rarer ones, but they are particularly valuable for determining the efficiency of health care services and for the assessment of practical value for the client, as they take into account the availability of treatment and its application in everyday practice.

To our knowledge, there are two studies that provide some indirect evidence of the effectiveness of stuttering treatment in children of school age. Kalinowski, Saltuklaroglu, Dayalu, and Guntupalli (2005) asked speech and language therapists who worked in schools about the evaluation of their effectiveness in treating CWS with respect to complete recovery. Despite a median time of three years spent on a caseload, the median reported recovery rate was no higher than the one to be expected by the chance of unassisted ("natural") recovery alone. However, the authors had not asked for an improvement in fluency or other progress, which might be a more realistic treatment goal for school children than a complete and undoubted recovery.

Euler, Lange, Schroeder, and Neumann (2014) asked 88 adult PWS about their retrospective effectiveness evaluation of each of the 231 treatments they had received during their lifetime. Of the most popular treatments in Germany, two approaches performed well: fluency shaping and stuttering modification. The other three approaches were evaluated as having been of little effect: hypnosis, breathing regulation, and "unspecified logopedic treatment". In Germany, CWS are rarely treated at school, but are usually sent to a speech-language pathologist ("logopedist") in private practice, a treatment whose costs are regularly covered by health insurances. In most cases, this outpatient treatment is an individual rather than a group treatment and is extensive (usually one session per week) rather than intensive. This "unspecified" logopedic treatment is the one that school children receive most often of all treatment options available in Germany. The more effective treatments, stuttering modification and fluency shaping, are mainly used in adolescent and adult PWS.

The aim of this paper is to report the effects of a group treatment with a speech restructuring method in 6- to 9-year-old German CWS. The International Classification of Functioning, Disability and Health (ICF) model of the World Health Organization (2001) provides an ideal clinical framework for the assessment and treatment of stuttering which does justice not only to its manifold negative effects on speech, but also on social communication and the working and living world of those affected (Yaruss, 2007). Because fluency, rhythm, and speed of speech are related to body function and structure which belong to the essential constituents of the model, improving speech fluency is recognized by societies such as the American Speech-Language-Hearing Association and guidelines on speech fluency disorders such as the German guidelines (Neumann et al., 2016, 2017) as one of the primary goals of the treatment for stuttering This goal is in accordance with Nippold (2012), has strongly recommended that improving speech fluency should be the main goal of treatment for school children as well. Such an improvement should not be bought at the expense of a reduction in speech naturalness but should be accompanied by a reduction of psycho-emotional stress and an improvement in quality of life.

Therefore, the two primary outcome parameters of the current study are defined by objectively assessed stuttering frequencies and by scores on the OASES-S questionnaire for school children (Yaruss & Quesal, 2006; Yaruss, Coleman, & Quesal, 2010, 2014). The OASES quantifies the subjectively perceived impact of stuttering in everyday life and on life quality. The speech restructuring treatment employed here is a child-tailored adaptation of the Kassel Stuttering Therapy (Euler & Wolff von Gudenberg, 2000; Euler, Wolff von Gudenberg, Jung, & Neumann, 2009), with parents as co-therapists.

In stuttering research, a random allocation of children to a wait-control condition that lasts as long as the treatment is ethically unacceptable. A comparison of two randomly assigned treatments could be an alternative in efficacy research, as illustrated in the study by de Sonneville-Koedoot, Stolk, Rietveld, and Franken (2015), but such a design would be difficult to install in effectiveness research, and it would leave open the question about the treatment effect as compared to no treatment and would thus not control for spontaneous recovery. For these reasons, a delayed treatment control (see Method, Section 2.6.) was chosen here. Various other control measures are desirable, common, and employed here, namely (1) a check of the inter-rater reliability of the stuttering counts, (2) assessments of speech naturalness, and (3) an outcome assessment after the last therapeutic intervention, preferably one year later and in an out of clinic setting.

In this study, an additional control check was used which, to the authors' knowledge, has not yet been reported, namely the maintenance of speech complexity during a treatment that restructures the child's way of speaking. The length and the grammatical complexity of utterances are associated with stuttering (Logan & Conture, 1995; Melnick & Conture, 2000; Yaruss, 1999), and possible treatment-induced reductions in speech complexity may circumvent speech disfluencies in CWS (Riley & Ingham, 2000; Ryan & Van Kirk Ryan, 1995). It is therefore necessary to ensure that a reduction in stuttering frequencies is not achieved or facilitated by reducing the linguistic complexity of the child's speech, but that the speech complexity is maintained during and after the speech restructuring treatment.

2. Method

2.1. Participants

All children who were registered for the FranKa treatment between March 2010 and March 2013 were included in the study. At the beginning of treatment, the 119 children (108 boys, 11 girls) were between 5.5 and 10.4 years old (mean for boys 7.7 years, for girls 8.3 years). To be eligible for treatment, the parent had to sign a treatment agreement that said that repeated assessments would be performed for clinical and research purposes, including unannounced telephone calls, and that the anonymized patient data would be stored and processed for treatment evaluation. The parents thus provided full written consent.

The study had been approved by the Ethical Board of the Hesse State Medical Association, Frankfurt/Main, Germany, registration number FF 89/2017.

2.2. Inclusion/exclusion criteria

The treatment was tailored for children aged six to nine years. Minor deviations from this age window were accepted if a child younger than six years was considered cognitively and socially able to participate, and if a child older than nine years was considered not yet able to participate in the treatment for the next age group of 9 to 12-year-olds (not reported here), a treatment that takes place without parental company.

The only inclusion criterion, apart from age, was the willingness of parents and children to adhere to the treatment format (e.g. in-patient, group treatment, exercise at home after the intensive phase). This willingness was implied by the registration for treatment. Kind and severity of the stuttering symptoms were not exclusion criteria.

2.3. Treatment

The treatment used in this study is a child-tailored adaptation of the Kassel Stuttering Therapy. The adult version, described in Euler et al. (2009), is a modification of the Precision Fluency Shaping Program (Webster, 1980). The main modification is the use of a biofeedback software (*flunatic*) during the individual practice session to train soft voice onsets and continued phonation. The child version of the treatment is named FranKa, derived from the cities of Frankfurt and Kassel where the treatment was developed. The biofeedback program in the children's version is *flunatic! junior*, which trains soft syllable onsets at the beginning of an utterance.

The intensive, in-patient phase of the FranKa treatment lasted six days, followed by a 6-month maintenance phase in which three weekend in-patient refresher courses were scheduled. During the intensive treatment, the child patients and their parent spent about eight hours daily in various treatment activities. In addition to specific speech trainings and exercises at the computer, speech games, activity games (preferably those which require verbal interactions), painting, short lectures on stuttering and speech physiology, the program includes exchanges of experience and leisure activities. Most of these activities take place in groups of varying size and composition, ranging from six to eight children per group, with three therapists and perhaps one or two interns.

During the intensive treatment week, child and parent together learn the technique of soft syllable onset at the beginning of a phrase (utterance). What constitutes a correct soft onset is explained in detail, also with the help of pictures, like a snail for slow, a chewing gum for drawn out, a hill for gentle incline. The parent is expected to use soft syllable onset all or most of the time, the child only in separate and explicitly designated situations, like soft-speaking rounds or in training sessions at the laptop. Audiosequences of speech, including the child's own speech, are used for illustrative feedback.

The daily schedule contains an individual session with child and parent at the laptop with the training software *flunatic! junior, which* displays images of objects or situations that prompt utterances of varying complexity. When the icon of an image is clicked, the corresponding sound is modeled with a soft voice onset. When the sound is repeated by the child (or parent), a voice curve indicates whether the sound onset was correct. If the voice onset was smooth enough, a collectable smiley drops down. At the end of the daily therapy work, the smileys can be exchanged for various trinkets or treats. These token swaps quickly become ritualistic and well received events.

The aim of the *flunatic! junior* exercise units is to enable the child to experience speech control, that is, to experience in a stress-free game setting that fluent speech can be produced actively and in a controlled manner. At the end of the week, most children easily access the new speech pattern with the soft phrase onset in various speaking situations such as playing, telephoning, or in structured situations such as the beginning of a meal.

The accompanying parent is instructed about the importance of regular practice (speech training at the laptop, doing speaking games, transferring fluency gains into various speaking situations), and about the importance of the parental role model and of social reinforcement.

In contrast to the Lidcombe treatment (Onslow, Packman, & Harrison, 2003), in which the child is socially reinforced for fluent speech, in the FranKa treatment the child is not systematically reinforced for fluent utterances, but instead for soft voice onsets. A shaping regime is employed in which the child is initially reinforced verbally (during work with *flunatic! junior* also visually) for approximations to an optimal soft onset. The reinforcement is gradually and intuitively changed from a continuous to a variable ratio schedule. The parent may be reinforced verbally for providing adequate feedback to the child. Many German parents feel uncomfortable praising the child for a behavior that does not seem particularly praiseworthy to them (Lattermann et al., 2008). Care is taken to use age- and context-specific praise, and often to praise only occasionally to criticize. For example, a younger child may be happy about being praised frequently, whereas an older child my feel annoyed. As with the Lidcombe treatment, gentle corrective feedback is occasionally used, for example, when an onset was repeatedly too sharp, but the parents are instructed to use positive feedback much more often than negative feedback. It goes without saying that these descriptions are formal ones, and informal social feedbacks (e. g. facial expressions, changes in prosody), both for onsets and for fluency of utterances, are inevitable and even welcome for a natural discourse.

After the week of intensive training, when child and parent are home again, the child is not required to use the soft voice onset all the time. Instead, special speaking situations are arranged, like speaking rituals, speaking games, sending voice messages, "soft speaking rounds", and similar situations which may generate in the child a joy of speaking. Outside these situations, the child is not required to monitor his or her speech and is not admonished to use the soft onsets. However, the parent may give corrective feedback or model the correct utterance. In the course of the therapy year, the encouragement to use the new speech technique is gradually expanded from structured speech situations to freer speech situations. In what way and how quickly the special speaking situations are extended to other everyday speech situations is largely left to the parents, with advice and support from the treatment team. The aim is to avoid that the soft speaking technique becomes a chore for the child.

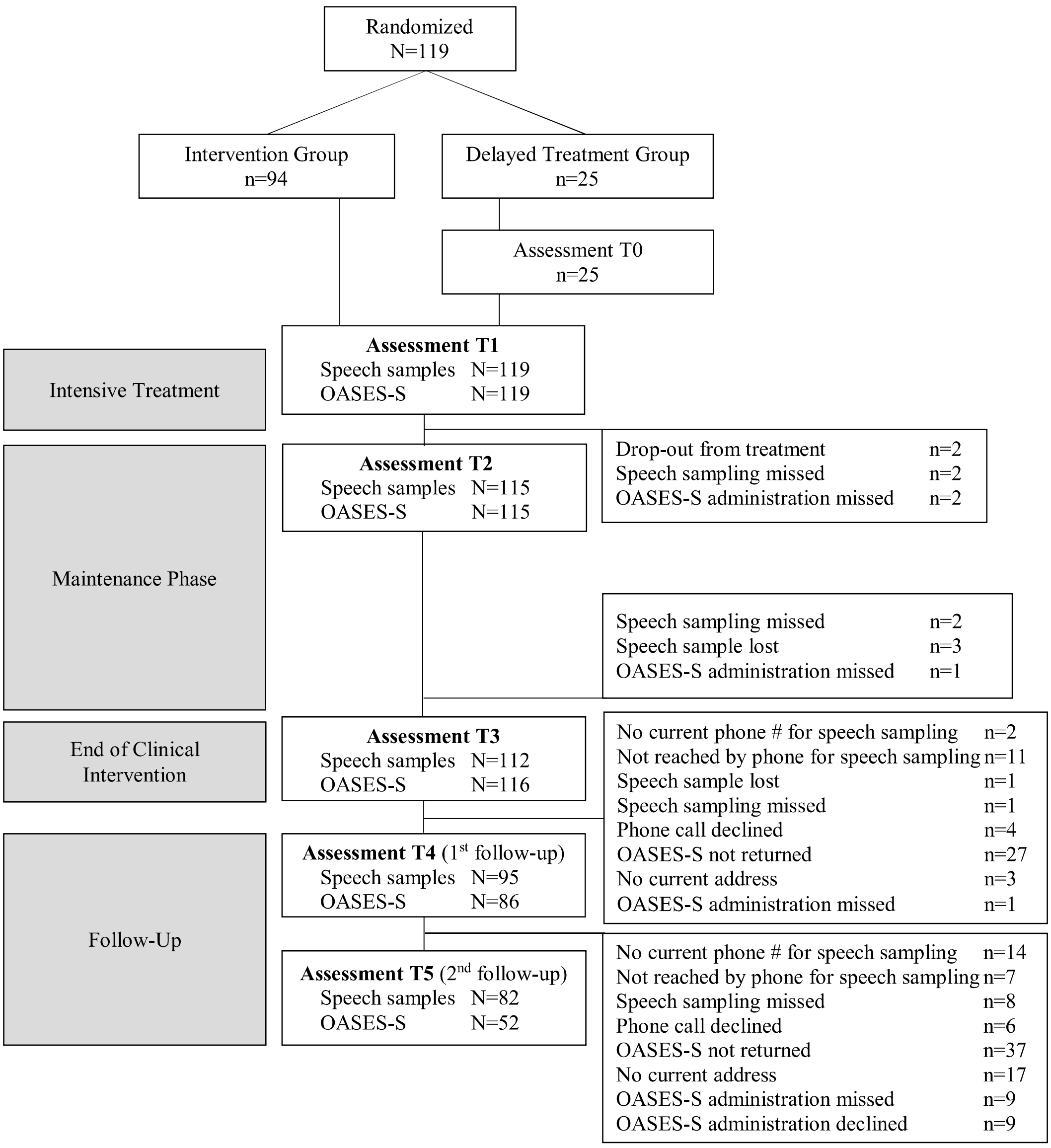

Fig. 1. Participant flow through treatment phases.

Parents and children are instructed to complete at least one ten-minute daily training session with the speaking software in the first 6 months. These practice sessions on the computer are automatically recorded and sent to the institute by e-mail once a month. Most people in Germany are covered by statutory health insurance (others have compulsory private insurance), and the cost of the stuttering treatment, other than travel, board and lodging, is covered by statutory health insurance. A compliance contract with the various insurance companies stipulates that the cost for the *flunatic! junior* software will only be covered if the training documents are sent in regularly and completely. The time spent in practice without the computer is not recorded, but participants are encouraged to practice daily.

During the maintenance phase, the treatment groups meet again for a weekend. The participants report and discuss their progress and problems, especially with the transfer into new speaking situations. Care is taken to sensitize the parents to the individual and situation-specific speech demands, including those set by the parents themselves. Social and speech anxieties of the children are a main topic in the group.

2.4. Primary outcomes

Primary outcome measures were the objective severity of stuttering in a conversation with the therapist and the subjective impact of stuttering, defined as the mean score on the OASES-S questionnaire (*Overall Assessment of the Speaker's Experience with Stuttering*; Yaruss & Quesal, 2006, 2014). The severity of stuttering was quantified by the three scores of the *Stuttering Severity Instrument* (SSI-4, Riley, 2009), that is, the percentage of stuttered syllables (%SS), the mean duration of the three longest stuttering events, and the ratings of physical concomitants. These three scores were analyzed separately and were not combined into the SSI severity score because the latter is only ordinal and therefore not best suited for data analysis.

The outcome measures were taken on five occasions (see Fig. 1): T1, pre-treatment; T2, four weeks after the intensive treatment; T3, six months later at the end of the maintenance phase; T4, another 12 months later, that is, 18 months after the intensive treatment; and T5, an additional two years later, that is, three years after the intensive treatment. For the delayed treatment control group (see below), %SS was reported at an additional earlier time point, that is, directly at or shortly after registration (T0). The ratings of physical concomitants were only collected during times T1 to T3, when child and parent were in the clinic. At later time points the speech samples were collected by telephone because at these times the child and parents were at home.

During the first three assessment points (T1, T2, T3), speech samples of a conversation with the therapist were video recorded. One year and three years after the maintenance phase (T4, T5), the speech samples were taken in a passive telephone call outside the clinic. Such a call saves travelling to the institute, and earlier studies (Block, Onslow, Packman, Gray, & Dacakis, 2005; Carey et al., 2010; Karimi et al., 2013) have shown that telephone calls provide valid data. To get a family member to answer the call, at least three calls were attempted, usually more and up to 10 calls. If still no contact could be established, the data was listed as missing at that time.

The %SS was tallied with the CSSS-2 app (Bakker & Riley, 2009) by independent observers who were not involved in the treatment. Speech samples of at least 500 syllables (occasionally less, especially before treatment) were sent to trained raters and scored according to the SSI-4 manual.

The OASES questionnaire assesses the self-reported impact of stuttering in different life situations with a 5-point rating and is considered to be the most comprehensive and useful tool of its kind. It has the added advantage that it has been translated into several languages including German, thus enabling intercultural comparisons.

The version for school children contains 60 items, arguably quite long for elementary school children. For the assessments T1, T2, and T3, the questionnaire was completed with the help of a therapist. At T4 and T5, a parent assisted the child. The OASES-S is designed for children between 7 and 12 years. In this study, almost a third of children were younger than 7 years. In view of parental assistance, it was decided to also administer the OASES-S to these younger children instead of using a different questionnaire, in order to ensure comparability of data.

The administration of the OASES-S after the intensive treatment was delayed by four weeks, comparable to the equivalent post-treatment assessment of stuttering frequencies, to allow the reduction of disfluency symptoms to start showing up in daily life.

2.5. Control for complexity of utterances

Twenty children were randomly selected and their speech samples from three assessment times (before and immediately after intensive treatment, four weeks after intensive treatment) were examined for two criteria: at least 500 recorded syllables, and the use of a soft syllable onset (treatment objective met) in at least one of the two assessments after treatment. Sixteen children met both inclusion criteria.

Complexity of utterances was assessed by mean length of utterance (MLU) and by clausal density. An outpatient speech-language pathologist transcribed orthographically the 48 speech samples and calculated MLU and clausal density for each sample. Only words, but no morphemes, entered the MLU calculation. Imitations and repetitions as well as aborted, incomprehensible, ambivalent, and stereotypical expressions were not counted as utterances. Compound words and proper names were counted as a single word, auxiliary words and verb prefixes were counted separately, and ellipses were also counted. Filler words and interjections were not counted.

The clausal density was quantified here according to the method proposed by Nippold, Mansfield, Billow, and Tomblin (2008). These authors define clausal density as the number of subordinate clauses in relation to all clauses and and calculate it by dividing the number of all clauses (main clauses plus subordinate clauses) by the number ofT-units, which equals the number of main clauses. A T-unit ("minimal terminable unit") is a sentence that can stand alone and consists of one main clause and any subordinate clause or nonclausal structure that is attached to or embedded in it (Hunt, 1970). If there are no subordinate clauses in a speech, the clausal density is 1.00. If each sentence of a speech contains both a main and a subordinate clause, the clausal density is 2.00.

2.6. Delayed-treatment control

A no-treatment control condition is not ethically justifiable in treatment research of stuttering in childhood. However, a certain waiting period is often necessary anyway, especially when participants are assembled for a group treatment which only starts on a few yearly dates.

A delayed-treatment control not only provides information about the extent of spontaneous recovery during the waiting period, but controls for a factor that is often ignored in stuttering treatment research. If the amount of stuttering is assessed once before treatment and again after treatment, the child is familiar with the assessment situation in the second assessment. Possible fluency gains from increased familiarity might mask genuine treatment gains, which would constitute a bias in estimating the treatment effect.

Speech samples were taken from all children at the time of registration. However, these samples were intended for clinical use only and their %SS are not reported here, with the following exception. A subsample of 25 children was quasi-randomly block selected to serve as a delayed-treatment control group. Of these children, %SS was additionally recorded for analysis from the first assessment at registration (T0) in order to allow comparison of stuttering frequencies at T0 and T1. The mean waiting time of these 25 children from registration (T0) to the start of intensive treatment (T1) was 90 days, with a range of 16–247 days.

2.7. Inter-rater reliability

Pre-treatment (T1) speech samples from 24 children were randomly selected. The %SS of these samples were counted by a second and independent raters and compared with those of the routinely collected samples. The scores from both raters correlated with a Spearman rho of .84. The mean %SS were very similar for both observers (6.3 vs 6.1), and the mean difference between the two was 0.21 %SS. The occurrence of two outliers, that is, two of the 24 cases with large differences between both observers, accounted for the mediocre inter-rater agreement.

2.8. Speech naturalness

The audio speech samples from assessments at T1, T2, and T4 were compiled from 20 randomly selected children. In addition to these 60 samples, audio speech recordings were taken from 20 5- to 10-year-old normal speaking, non-stuttering children. These 80 samples were randomly mixed and presented to four independent observers who were not professionals in speech-language matters and who rated the speech naturalness according to the 9-point scale of Martin, Haroldson, and Triden (1984, 1 = highly natural, 9 = highly unnatural).

2.9. Drop-outs

Drop-outs as well as other missing data are shown in Fig. 1, which also sketches the study design.

2.10. Data analysis

The changes between selected assessment points were examined with the *t*-test for matched pairs for the stuttering frequencies and the OASES-S scores. For this purpose, the positively skewed distributions of the %SS had to be normalized to allow for parametric testing, which could be achieved by a log10-transformation, with the exception of the distribution of %SS at T5. The OASES-S scores were normally distributed, as shown by the Kolmogorov-Smirnov test. The scores for the mean duration of the longest three blocks and for the physical concomitants were heavily skewed and were left untransformed.

Change scores were tested inferentially only for theoretically meaningful and clinically relevant paired comparisons. The most important comparison is the one between the assessment immediately before treatment (T1) and at the end of the maintenance phase (T3) as well as 18 months after treatment (T4). The comparison between T1 and 42 months after treatment (T5) was considered less

Table 1
Mean and standard deviation (SD) for percentages of syllables stuttered (%SS, not transformed) and total OASES-S scores over assessment points; second lines: N, 95 %-confidence interval.

	Assessment Point				
	Pre-treatment (T1)	Post-treatment (T2)	6 months, end of maintenance (T3)	18 months post-treatment (T4)	42 months post-treatment (T5)
Mean % SS (SD)	9.4 (7.1)	5.6 (5.5)	5.6 (5.5)	4.7 (4.4)	3.9 (5.0)
N, 95 %-CI	119, 8.1–10.7	115, 4.6–6.7	112, 3.8–5.5	95, 3.5–5.3	82, 2.8–5.0
Mean OASES-S (SD)	2.42 (0.50)	2.20 (0.51)	2.09 (0.50)	1.96 (0.53)	1.96 (0.62)
N, 95 %-CI	119, 2.33–2.51	115, 2.10–2.29	116, 2.00–2.18	86, 1.85–2.08	52, 1.79–2.14

informative because of the high follow-up loss (see Fig. 1), particularly for the OASES-S scores. These data are reported here (Table 1), but not interpreted further.

In addition, it is of interest to know which changes from one assessment point to the next were significant. Such comparisons involve several double dippings (circular analysis) that require a correction for multiple comparisons. Therefore, after a Bonferroni correction the significance level for paired comparisons was set to $p = .0125$ to reach the conventional level of significance.

The effect sizes for treatment changes were quantified with Cohen's d for paired comparisons (Dunlap, Cortina, Vaslow, & Burke, 1996) in the case of parametric scales. For nonparametric scales the standardized response means (SRMs) were calculated as effect size indicators. Samples of $N > 25$ were tested for normality of distribution with the Kolmogorov-Smirnov test, samples of $N \leq 25$ with the Shapiro-Wilk test.

3. Results

Table 1 shows mean pooled %SS and OASES-S total scores at each assessment.

3.1. *Percentage of syllables stuttered*

The drop of %SS (log10-transformed scores) from T1 to T2 was significant ($p < .001$), with an effect size of $d = 0.70$. The change from T2 to T3 was not significant. The change from T3 to T4 was significant at $p < .001$ ($d = 0.44$). The change from T4 to T5 was tested non-parametrically with the Wilcoxon-test on the non-transformed scores, because the distribution at T5 could not be normalized easily. With $p = .045$, this change missed the set significance level of $p = .0125$.

Of the overall changes, that from T1 to T4 seems to be a more appropriate indicator for the longer-term treatment effect than the change from T1 to T5 due to the high loss to follow-up at T5. The change from T1 to T4 was significant ($p < .001$), with an effect size $d = 0.86$. By convention, an effect size of 0.80 or larger is considered large (Sawilowsky, 2009).

The changes in frequencies of stuttering over the assessment points are shown in Fig. 2A.

3.2. *OASES*

In Table 1, the changes in OASES-S total scores over the assessment points look small. However, the OASES only provides item scores between 1 and 5. Thus, small absolute changes in the overall mean scores can be clinically meaningful, which is reflected in the relatively small standard deviations associated of these scores.

The decrease of OASES-S total scores from T1 to T2 was significant ($p < .001$), with an effect size of $d = 0.46$. The change from T2 to T3 was significant ($p = .005$, uncorrected) with an effect size $d = 0.24$. The change from T3 to T4 was not significant, nor was the change from T4 to T5. The change from T1 to T4, as the most appropriate indicator of the effectiveness of therapy, was significant ($p < .001$, uncorrected), with a $d = 0.86$.

The changes in mean OASES-S scores over the assessment points are shown in Fig. 2B. The OASES-S is divided into four subtests which showed the following changes from T1 to T4: General information about one's own speech and about stuttering $d = 1.33$; One's own negative reactions to stuttering $d = 0.61$; Communication in everyday situations $d = 0.58$; Quality of life $d = 0.30$.

The correlations between the overall OASES-S scores and the %SS was not significant before or during treatment and maintenance phase (T1, T2, T3), but were significant at T4 ($r = .37$) and T5 ($r = .30$).

3.3. *Speech naturalness*

For the six possible dyads of the four speech naturalness judges, the ratings correlated between $r = .59$ and $r = .85$. As this was

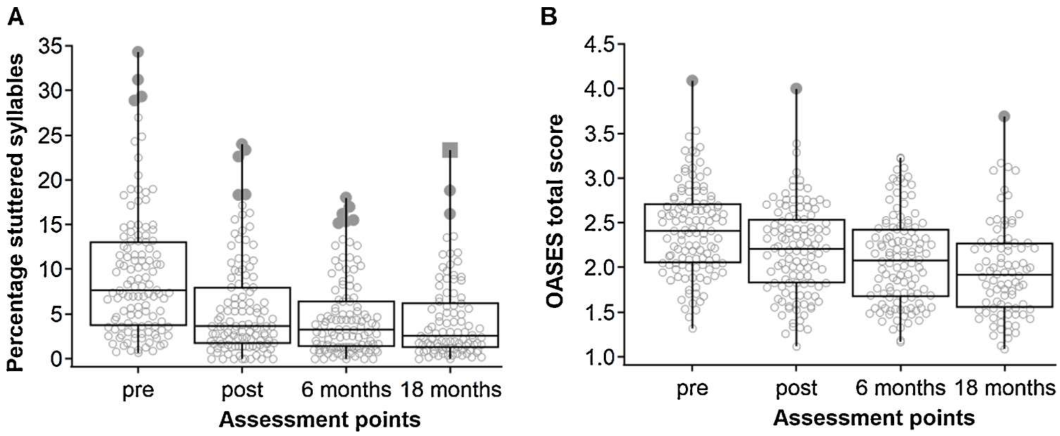

Fig. 2. **A** Percentage of stuttered syllables (%SS) and **B** mean OASES-S scores over assessment points. Gray data points: outliers; square gray data point: outlier >2 SD.

regarded as sufficient agreement between the judges, the naturalness ratings for each sample were averaged over the four judges.

The 20 fluent speaking children received a mean rating of 2.03. The 20 pre-treatment samples (T1) had a mean rating of 4.43, the post-treatment samples (T2) 3.93, and one year after the last therapeutic intervention (T4) 3.31. The change in speech naturalness over these three assessment points yielded a significant linear effect as tested with a univariate analysis of variance for repeated measurements ($F_{1,19} = 7.518$, $p = .013$). The change from T1 to T2 was not significant as tested with a *t*-test for paired comparisons, but the change from T2 to T3 was significant. The change from T1 to T4 was significant ($p = .013$), with an effect size of $d = 0.85$. The children's speech thus was judged by listeners to become more natural during treatment and follow-up. However, the speech of the treated children did not become as natural as the speech of non-stuttering children, as the difference between the samples from non-stuttering children and the samples from one year after the last therapeutic intervention was significant and very large ($t = 4.05$, $p < .001$, $d = 1.28$).

One of the OASES-S items asked about the subjectively perceived speech naturalness. The means for this item on the three assessment points T1, T2, and T4 were 2.70, 2.40, and 2.35. The change from T1 to T4 amounted to an effect size of $d = 0.72$. The rank correlations between the OASES-S scores for this item and the average ratings of the external judges were $rho = -.06$, .26, and .37. None of these correlation coefficients reached statistical significance.

3.4. Delayed-treatment control

The 25 children in the delayed treatment control group reduced their stuttering from 10.6 %SS at T0 to 9.1 %SS at T1. This reduction was not significant ($p = .297$) as tested with the *t*-test for paired comparisons on the log-transformed distributions.

3.5. Control for complexity of utterances

The 16 children in the subsample for the control of complexity of utterances had the following MLUs: 6.19 before treatment, 6.92 immediately after intensive treatment, and 6.61 one month after intensive treatment. The respective values for clausal density were 1.23, 1.33, and 1.20. The increase from before to immediately after intensive treatment was significant for MLU ($t = 2.47$, $p = .026$, $d = 0.50$) but missed significance for clausal density ($t = 2.13$, $p = .051$, $d = 0.63$). The decrease from immediately after to one month after intensive treatment was not significant for MLU and was very significant for clausal density ($t = 3.06$, $p = .008$, $d = 0.69$). Thus, the complexity of utterances did not diminish during intensive treatment, but increased temporarily.

3.6. Responders

An alternative way to indicate treatment effectiveness is to count the number of "responders" within a sample. Responders, also called "maintainers", are treated participants who meet certain criteria for treatment success. The criteria vary from study to study (for example, Langevin et al., 2006; Lewis et al., 2008). Here, responders are determined by three criteria: (1) The reduction from T1 to T4 in %SS of more than one half compared to pre-treatment %SS; (2) less than 3 %SS at endpoint T4; (3) reduction of the overall OASES-S score at T4 by more than one-half of the pre-treatment standard deviation, that is here by 0.25, compared with the pre-treatment (T1) score. A participant is called a responder if he or she meets at least one of the three criteria. By complement, a non-responder is a child who has not reached any of these three criteria. The endpoint is defined here as the mean of the values T3 and T4. This means that if a case has no entry at both evaluation points T3 and T4, the case is counted as non-compliance with the criterion. If a case has a miss at one of the assessment points, the entry at the other assessment point is counted. By this procedure, and in accordance with an intention-to-treat analysis, drop-outs are counted as failures.

The first criterion (reduction in %SS by more than half) was reached by 66 of the 119 children (55.5 %). The second criterion (< 3% SS at endpoint) was reached by 58 children (48.7 %), and the third criterion (reduction of OASES-S) by 68 children (57.1 %). A total of 99 children (83.2 %) were responders (at least one criterion reached); the remaining 20 children (16.8 %) were thus non-responders.

4. Discussion

The treatment of school children who stutter presented here is a speech-restructuring approach that reinforces a soft syllable onset and that is applied as a group intensive treatment. At the end of a year after the end of the maintenance phase, the children had reduced their stuttering frequencies with a large effect size. In addition, the subjectively evaluated negative effects of stuttering in daily life, which were assessed with the OASES questionnaire for school children, decreased with a large effect size. Speech naturalness increased with a large effect size but did not reach the level of non-stuttering children. The complexity of utterances was not affected by the treatment. Of the 119 children registered for treatment, 83.2 % had at least some benefit.

The treatment-induced reductions from before therapy to one year after the end of the maintenance phase were the same for the stuttering frequencies and the OASES-S score with a *d* of 0.86 for both measures. The treatment benefit was similar to that in adolescents and adults, which in earlier studies of the Kassel Stuttering Therapy ranged between $d = 0.75$ and $d = 1.13$ (Euler, Anders, Merkel, & Wolff von Gudenberg, 2016; Euler & Wolff von Gudenberg, 2000; Euler et al., 2009). Langevin et al. (2006) and Langevin, Kully, Teshima, Hagler, and Narasimha Prasad (2010) report on effect sizes of similar magnitude for a comparable speech restructuring treatment. The OASES reductions, however, were reported in Euler et al. (2016) for adolescents ($d = 1.36$) and adults ($d = 1.63$) to be larger than the reductions in stuttering frequencies. A similar difference between objective and subjective treatment measures was reported by Langevin et al. (2006).

The downward curves for the stuttering frequencies and for the OASES-S scores were shaped differently. As expected, the stuttering frequencies decreased strongly during the intensive phase and then only moderately, while the OASES-S score decreased steadily until follow-up. The immediate improvement of the speech fluency needs a certain amount of time to express itself in everyday life situations. The steady decline of OASES-S is clinically promising. At the same time, however, self-ratings such as the OASES must be interpreted with caution, as such ratings about treatment gains are subject to two biases, the self-justification effect and the response shift bias.

The self-justification effect is derived from the theory of cognitive dissonance (Festinger, 1961) and refers here to the following bias. If a treatment was costly in terms of time, effort, or money, a lack of treatment benefit would be cognitively dissonant for the treated person ("I was stupid enough to do that"). The incompatibility ("dissonance") between costs and benefits can be reduced by changing one or both components. Because the costs cannot be retrieved, the benefits are subjectively raised by sugarcoating the effect. This self-justification becomes greater the higher the costs are. An intensive stuttering treatment that lasts for months and requires frequent self-monitoring is indeed costly. The self-justification thus leads to an overestimation of the treatment effect. This could be the reason why for adolescents and adults the gains in OASES scores are more favorable than the gains in stuttering frequencies. For adolescents and adults, the comprehensive treatment costs might be more salient than for children (Euler et al., 2016).

The response shift bias (Sprangers & Schwartz, 1999) refers to the change in the patient's evaluating standards during treatment. Before treatment, a PWS may be satisfied if only few stuttering events occur. After treatment, the same person expects to be stutter-free and is now dissatisfied with the same frequency of stuttering. For instance, one item in the OASES-S questionnaire says, "I wish nobody would find out that I stutter". Before treatment, this desire may be unrealistic, leading to a response that this desire rarely or never occurs, but after treatment, understandably, the same desire may occur more frequently. The response shift bias leads to an underestimation of the treatment effect and requires recalibration.

The children in the delayed treatment control group reduced their stuttering frequencies only slightly and not significantly during the waiting period of an average of three months. Such a control group is usually installed to control for unassisted (spontaneous) recovery. However, there is another aspect worth controlling: the reduced symptom severity due to increased familiarity with the testing procedure. Stuttering is known to be associated with and influenced by social anxiety and anticipated communication demands (Bloodstein & Bernstein Ratner, 2008). In the study presented here, all children were tested twice before starting intensive treatment. At the second testing, shortly before the start of treatment, they were already familiar with the procedure. The delayed treatment group was established only to obtain an estimate of the magnitude of this familiarity effect. A possible spontaneous recovery assumedly did not take place during the waiting period because there was no correlation between the length of the waiting period and the reduction of %SS during the waiting period (rho = -.016).

Counterintuitively, OASES-S scores and stuttering frequencies showed no correlation before treatment ($r = .03$). After the intensive phase, however, the correlation increased to moderate values over the following assessment points, namely $r = .12$ (n.s.) at T2, $r = .13$ (n.s.) at T3, $r = .37$ ($p = .001$) at T4, and $r = .30$ ($p = .054$) at T5. The same effect of the absence of a correlation between OASES scores and stuttering strength before treatment and the later occurrence of such a correlation has been reported earlier in adolescents and adults (Cook, 2013; Euler et al., 2016)

Euler et al. (2014) reported that in Germany the most frequent stuttering therapy, especially for children of school age, is an unspecified and extensive individual treatment. "Unspecified" means that the essential elements of the treatment or the name of the treatment are not made clear to the child or the parents. "Extensive" means that the treatment frequency is usually one session per week. "Individual" means that in one session an individual child is treated by a speech-language pathologist. This most common treatment is not very effective despite its relatively long duration. The treatment approach presented here clearly appears to be the better alternative.

The gender ratio in the current sample, with only 9 % females, is unusual. All children treated in the specified time period were included into the study, irrespective of gender, and the Kassel Stuttering Therapy does not apply any gender-based criteria for acceptance for treatment. The more than typically lopsided gender ratio reported here is rather assumed to be due to a gender bias in the referral by parents of CWS. The reasons for this bias are unknown, but the bias is correlated with age. A comparison of gender ratios for the various age groups treated at the Kassel Institute reveals a trend. Among 9-to 11-year-olds, which are treated without an accompanying parent, 16 % of clients are females. Among teenagers the female percentage is 17 % and among adults it is 23 %.

Many theorists and practitioners criticize fluency reduction as a treatment goal as too narrow and prefer a comprehensive treatment approach instead (e.g. Yaruss, Coleman, & Quesal, 2012). In our opinion, such a preference is indeed desirable as long as the primary problem, stuttering, is not neglected. The German guidelines prescribe fluency gain as an indispensable treatment goal, as do the vast majority of parents who bring their child to stuttering therapy. The treatment approach of the Kassel Stuttering Therapy is not limited to mere fluency reduction, but naturally integrates many components that go beyond fluency training, like anxiety reduction, increasing self-confidence, and helpful attitude changes. The effect of these components is reflected in the improvement of the OASES scores, which is of the same order of magnitude as the improvement of speech fluency. For adolescents and adults, Euler et al. (2016) showed that the greatest treatment gains of the Kassel Stuttering Therapy could be observed in the reductions of speech-related anxieties and speech avoidance.

5. Limitations

The study presented here has several limitations. The stuttering frequency was assessed in only one speaking situation and only once at each assessment point. At the Institute of the Kassel Stuttering Therapy, the stuttering frequency in adolescents and adults is usually measured in four different speaking situations (talking to a therapist, reading a standard text orally, telephoning an unknown person, and interviewing passers-by on the street). Young schoolchildren are not yet able to read fluently, and they cannot be asked to call an unknown person or interview unknown people on the street. This unavoidable limitation to one speaking situation reduces reliability and increases the error variance in the outcome data.

A further limitation is the fact that the speech samples were recorded either in the clinic or with a passive telephone call by a therapist from the treatment team. This procedure is likely to produce a bias in favor of the treatment effects, showing to some extent temporary, situation-specific and attention-demanding speech *performance* instead of enduring, generalized and automatized speech *competence*. In a previous study (Euler et al., 2009) we were able to estimate the magnitude of this bias in a sample of 9- to 13-year-old CWS. A subsample of 20 children received a telephone call 6 months after the intensive treatment phase from an unfamiliar member of the treatment team, pretending to make a survey about children's TV preferences (covert call). She asked 12 questions which elicited longer answers. Within a few days, another unfamiliar team member called, identifying herself as an employee of the treatment institute and explained that the purpose of the call was to assess the speaking ability with the familiar procedure (overt call). The covert calls had a mean of 3.0 %SS, the overt calls 1.6 %SS ($p < .05$, $d = 0.15$). There is thus a bias favoring overt measurements, but the bias is relatively small compared to the overall treatment effect.

Another limitation is the lack of a no-treatment control, which is desirable in evidence-based treatment research. In the behavioral treatment of stuttering, however, such a control is ethically unacceptable. The recourse to alternative treatment as a control, such as de Sonneville-Koedoot et al. (2015), is unsatisfactory in this respect, as it only shows comparative but not absolute treatment effects. We tried to compensate for this limitation by applying several internal controls, including a novel one in stuttering treatment research, namely the complexity of utterances.

A further limitation is the application of the OASES-S questionnaire to young school children. This questionnaire is designed for 7- to 12-year-old children, but with its 60 items it is rather long for young school children and can only be completed with help of an adult. Furthermore, almost 31 % of the participating children (37 out of 119) were under 7 years old at the time of registration. These circumstances may have affected the validity of the questionnaire. The differences in OASES scores between younger (under 7 years) and older participants (7 years or older) were t-tested, and at no assessment point were the differences significant or near significant ($p = .10$). Furthermore, the small absolute mean differences over the assessment points varied in direction. Thus, there seems to be no apparent bias in the OASES scores due to age just below the limit prescribed by the test.

On the other hand, the OASES, which is also available in forms for adolescents and adults and has been translated into many different languages, enables a comparability across age groups and cultures that is not achieved by any other alternative subjective measure of the impact of stuttering in daily life. In addition, it provides information about the impact of stuttering in different life situations, such as family, school, or peer groups. For these reasons we decided to use the OASES test despite its limitations.

A final limitation is the small sample size of only 16 children for the control of speech complexity which prevented a robust detailed analysis of the relations between disfluency variables and variables of speech complexity.

6. Conclusion

This study fills a gap in contemporary research on the treatment of stuttering by demonstrating, on the basis of a relative large sample of children, that an intensive fluency shaping approach ("global" speech restructuring) using computer feedback and a subsequent maintenance phase is effective in the long term for most of the children treated, in terms of speech fluency, speech naturalness, and the reduction of negative emotional and social consequences of stuttering in school-age children. In this age group the reinforcement of only soft voice onsets is sufficient to restructure the speech. The intensive character of the treatment in the initial phase, the group character of the therapy, and the involvement of the parents as co-therapists seem to be important components, and this treatment seems to be generally superior to an extensive individual treatment.

CRediT authorship contribution statement

Harald A. Euler: Conceptualization, Methodology, Formal analysis, Writing - original draft, Writing - review & editing. **Anna Merkel:** Conceptualization, Methodology, Data curation, Visualization. **Katja Hente:** Investigation. **Nicole Neef:** Visualization. **Alexander Wolff von Gudenberg:** Funding, Resources, Project administration, Software, Supervision. **Katrin Neumann:** Investigation, Writing - review & editing.

Acknowledgement

We thank Patricia Purat for her help with the analysis of speech complexity.

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