The novel cognitive flexibility in aphasia therapy (CFAT): A combined treatment of aphasia and executive functions to improve communicative success

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Abstract

Purpose: Over and above language deficits, persons with aphasia can present with impairments in executive functions, including deficits in cognitive flexibility. Cognitive flexibility constitutes the ability to update behaviour quickly and flexibly in a changing environment. Its deficits can restrict communicative ability, e. g. the ability to change a topic. To date, these deficits have been neglected in aphasia therapy, even though their consideration regarding language treatment may be beneficial for the persons affected. The present study aimed to evaluate whether aphasia therapy including cognitive flexibility leads to more improvement than conventional aphasia therapy.

Method: A pilot group study with ten patients was conducted. The patients received both the novel Cognitive Flexibility in Aphasia Therapy (CFAT) and conventional aphasia therapy in a cross-over design. Each therapy method was delivered for 20 sessions within two weeks. An assessment battery was applied five times, including language skills, communicative ability and verbal/nonverbal cognitive flexibility.

Result: Patients profited from CFAT regarding language skills, communicative ability and verbal cognitive flexibility. Furthermore, compared to conventional therapy, CFAT was more effective for verbal cognitive flexibility.

Conclusion: This pilot study indicates that CFAT offers a novel opportunity to directly train cognitive flexibility in communicative settings and complements conventional therapy for optimal patient outcome.

Keywords

aphasia, communication, executive function, speech therapy, language

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Introduction

Communicative ability in aphasia According to the International Classification of Functioning, Disability and Health (ICF), a framework of the WHO (World Health Organization, 2001), the goal of rehabilitation should be to enhance participation in social life. Therefore, one of the main goals of aphasia therapy is to improve the patient's ability to take part in everyday communication (Helm-Estabrooks, 2002; King, Simmons-Mackie, & Beukelman, 2013;Worrall et al., 2011).

In order to reach this goal, conventional speechlanguage pathology usually focuses on improving linguistic skills (Helm-Estabrooks, 2002; King et al., 2013). However, there is increasing evidence that the ability to take part in conversations does not only depend on linguistic skills, but also on other cognitive abilities (Frankel, Penn, & Ormond-Brown, 2007; Fridriksson, Nettles, Davis, Morrow, & Montgomery, 2006), including working memory, inhibition or cognitive flexibility (Fridriksson et al., 2006). These functions are part of the umbrella term of "executive functions" and enable goal-directed planning, finding and maintaining problem-solving strategies and adapting these strategies quickly and flexibly to novel everyday situations (Helm-Estabrooks, 2002). Ramsberger (2005) emphasised that executive functions play an important role in the complex task of human communication. Some studies evaluated the status of executive functions in people with aphasia (PWA) and frequently found impaired inhibition, working memory or cognitive flexibility (Frankel et al., 2007; Fridriksson et al., 2006; Lee & Pyun, 2014), with significantly lower performance levels compared to healthy persons (Fonseca, Ferreira, & Martins, 2017; Purdy, 2002). The study by Fridriksson et al. (2006) complemented these findings by showing a high correlation between impaired executive functions and communicative ability. For a more intensive analysis on how deficits in executive functions can influence everyday communication in PWA, Frankel et al. (2007), Penn, Frankel, Watermeyer, and Russell (2010) and Beckley et al. (2013) conducted single case studies using a conversation analysis and standardised tests on executive functions. All of them revealed correspondences between impaired executive functions and communicative deficits: the PWA made digressions, showed poor performance in topic control and many perseverations (Frankel et al., 2007; Penn et al., 2010), revealed less repair skills (Penn et al., 2010) and did not spontaneously shift to alternative communication in case of word finding difficulties (Beckley et al., 2013). The origins of these difficulties cannot be attributed to language deficits only, but rather involve impaired cognitive flexibility, i.e. the inability to update behaviour quickly and flexibly to the changing environment (Diamond, 2013).

Cognitive flexibility in persons with aphasia

Cognitive flexibility is one of the most important executive functions in everyday communication (Beckley et al., 2013), including the conversation with family members. Since the course of conversations is not predictable, the communication partner must act and react in a highly flexible manner. To improve the communication between PWA and their family members, Beckley et al. (2013) developed a conversation training including self-reflecting methods to improve the flexible and spontaneous use of communication strategies of PWA, like writing or drawing. The single case study revealed that the participant reached a better acceptance of his strategy use after training, but he was not able to choose the strategies spontaneously and at his own initiative. The authors supposed that the reason for these remaining deficits was impaired cognitive flexibility, which was not trained explicitly in their study. In turn, this indicates that for improvements of cognitive flexibility, this function needs to be explicitly trained, as in the multimodal communication treatment (MCT) by Purdy and Wallace (2016). These authors tried to improve the flexible use of non-verbal strategies by practicing and alternating three response modalities (verbal, gesture and communication board). After MCT, some PWA showed increased effective use of non-verbal modalities, although others did not profit from this kind of treatment (Purdy & Wallace, 2016). Therefore, it is relevant to be able to determine which PWA profit from such cognitive-control based therapy. It is important to note that the use of non-verbal strategies is only one of the key characteristics of cognitive flexibility in communication. In everyday communication, other aspects of cognitive flexibility are also relevant, namely response to change of topic (Frankel et al., 2007) or response to misunderstandings by means of circumlocutions (Hernandez-Sacristan, Rosell-Clari, Serra-Alegre, & Quiles-Climent, 2012). The function of cognitive flexibility also plays a role in changing perspectives in the sense of the theory of mind. However, studies indicate that the theory of mind tends to be unaffected in PWA (Varley, Siegal, & Want, 2001; Varley & Siegal, 2000). Thus, the aim of the present study was to develop and to evaluate a novel aphasia therapy which integrates three key characteristics of cognitive flexibility: i) response to change of topic, ii) response to misunderstandings and iii) use of nonverbal communication. The novel therapy was designed to integrate realistic everyday communication to address real-life communication and to achieve better participation of PWA in everyday life. In order to compare the novel therapy to a conventional aphasia therapy, we performed a pilot group study with ten PWA in a cross-over design, and evaluated which PWA profited from the novel therapy, in order to be able to identify optimal candidates for this approach. In the counter-balanced design, PWA received the two therapy approaches in a block design, controlling for order of methods delivered. This pilot study intends to provide initial results on the effectiveness of our novel therapy, based on which a future Randomised Controlled Trial (RCT) could be conducted.

We hypothesised that (i) the novel therapy would improve performance of PWA for the following functions: cognitive flexibility measured by Regensburger Wortfluessigkeits-Test (RWT), German version ([Regensburg Test of Fluency], Aschenbrenner, Tucha, & Lange, 2000), Screening of Category Shift as well as Cognitive Flexibility in Aphasia Screening (CFA-Screening); language skills measured by Screening of Naming as well as Bielefelder Wortfindungs-screening (BIWOS), German version ([Bielefeld screening of word finding] Benassi, G€odde, & Richter, 2012); communicative abilities measured by the Szenario-Test, German version of the Dutch Scenario Test (Nobis-Bosch et al., in press; van der Meulen, van de Sandt-Koenderman, Duivenvoorden, & Ribbers, 2010; see Table I). Additionally, we hypothesised that (ii) the novel therapy would be more effective than conventional aphasia therapy, whereby we also expected that the order of therapy methods would not influence therapy outcome, and (iii) individual therapy outcome would relate to aspects of initial PWA performance as a predictive factor, so that eligibility criteria can be derived.

Overview of study assessments.

Test (German versions)	Function assessed	Timeline
Core formal language tests		
BIWOS[a]	Word retrieval	T1-T5
Szenario-Test[b]	Multimodal everyday communication	T1-T5
Peripheral formal language test		
AAT[c]	Severity and type of aphasia, distribution of speech and language deficits	T1 and T5
Token Test	Auditory comprehension	
Repetition	Repetition of spoken words and sentences	
Written Language	Reading and writing	
Naming	Naming performance	
Comprehension	Auditory and reading comprehension	
Core formal tests for cognitive flexibility		
WCST-64[d]	Cognitive flexibility, problem solving, concentration, planning, organisation, working memory and inhibition	T1-T5
Total Number Correct	Problem solving: correct application of the problem solving strategy	
Perseverative Errors	Tendency to persevere on old problem solving strategy	
Non-perseverative Errors	Probing new problem solving strategies, which still lead to incorrect responses	
Concept Level of Responses	Conceptual ability: insight into sorting principles	
Number of Categories completed	Overall success	
Trials to complete First Category	Conceptual ability for the first problem solving strategy	
Failure to Maintain Set	Maintenance of the correct problem solving strategy	
RWT[e]	Word fluency: Cognitive flexibility, word retrieval, problem solving	T1-T5
Semantic category shift	Shift between two semantic categories	
Word form shift	Shift between two initial letters	
Therapy screening		
Language screening		
Screening of Naming	Naming performance on high frequency nouns, low-frequency nouns, verbs	T2-T4
Screening of cognitive flexibility		
CFA-Screening[f]	Cognitive flexibility, everyday communication	T1-T5
Screening of Category Shift	Cognitive flexibility, word retrieval	T2-T4

Table 1: [a] Bielefeld screening of word finding (Benassi, G€odde, & Richter, 2012); [b] Szenario-Test (Nobis-Bosch et al., in press); [c] Aachen Aphasia Test (Huber, Poeck, Weniger, & Willmes, 1983; Huber, Poeck, & Willmes, 1984); [d] Wisconsin Card Sorting Test-64 (Kongs, Thompson, Iverson, & Heaton, 2000); [e] Regensburg Test of Fluency (Aschenbrenner, Tucha, & Lange, 2000); fCognitive Flexibility in Aphasia Screening. T: test time.

Method

Participants

Participants were considered for inclusion if they (i) were in the late post-acute or chronic phase of aphasia after stroke (6 months post-onset), (ii) had German as their mother tongue, and (iii) were less than 75 years of age. The latter was important to exclude cognitive decline and dementia in old age. Exclusion criteria were (i) a psychiatric disease, (ii) intensive aphasia therapy (>5 sessions per week) less than 6 months ago, (iii) significant difficulties in hearing or eyesight, and (iv) severe apraxia of speech or dysarthria, which were taken from speech therapy reports. The latter two were excluded since, otherwise, the accuracy and speed of articulation could be compromised. Both would hinder the accomplishment of the tasks in diagnosis and therapy, and prevent optimal analysis of responses and outcomes. Following pretesting, PWA were excluded if they revealed high or mastery performance (i) for naming as assessed by the subtest Naming of the Aachener Aphasie Test (AAT), German version, (percentile rank (PR) 79) ([Aachen Aphasia Test] Huber, Poeck, Weniger, & Willmes, 1983; Huber, Poeck, & Willmes, 1984); (ii) for cognitive flexibility as assessed by the Wisconsin Card-Sorting Test-64 (WCST-64) (PR >30) (Kongs, Thompson, Iverson, & Heaton, 2000) or an assessment of word fluency (RWT; PR >16) (Aschenbrenner et al., 2000); or if they revealed severe difficulties (iii) in speech comprehension (AAT subtest Comprehension; PR < 13) or (iv) in verbal learning (Verbaler Lerntest, VLT, German version [verbal learning test], PR <25) (Sturm & Willmes, 1999; see Table I for overview). An assessment of PWA's speech comprehension performance was important to ensure sufficient understanding of test and therapy instructions. The examination of verbal learning was relevant to exclude memory disorders for verbal stimuli, as these are related to difficulties in learning and long-term storage of new verbal information. It can be assumed that PWA with verbal learning difficulties cannot benefit from the novel therapy, but rather need specific therapy to improve verbal learning first.

After a preliminary pilot study with one person with aphasia to establish feasibility of the treatment protocol, ten PWA with left-hemisphere aphasia were recruited from local private practices and rehabilitation centres (see Supplementary Table SI for details). The ten PWA were selected from a larger pool of 33 potential participants (see Supplementary Figure S1 for details). Twenty-three PWA had to be excluded after pre-testing, and ten PWA were treated and completed the trial, so there were no drop-outs. All PWA provided informed written consent and the study was approved by the Ethics Committee at the Medical Faculty of the RWTH Aachen University (EK 248/14).

Overall study design

The pilot group study was performed in a cross-over design with baseline, so that each person with aphasia underwent both Cognitive Flexibility in Aphasia Therapy (CFAT) and Everyday Language Therapy (ELT). PWA of group 1 received CFAT first, whereas PWA of group 2 received ELT first. The two methods were highly parallelised, and their order was counterbalanced across participants, with stratified randomisation of assignment to either group 1 or group 2 as good as possible by initial performance in AAT subtests

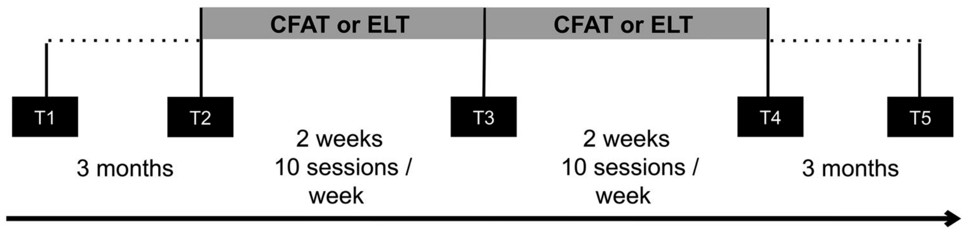

Figure 1: Overall study design. T: test time; CFAT: Cognitive Flexibility in Aphasia Therapy; ELT: Everyday Language Therapy.

Naming and Comprehension and WCST-64 perseverative errors.

Therapy sessions were delivered at high frequency, i.e. two to three sessions per working day and method over two weeks (see Supplementary Table S II for details according to TIDieR). As depicted in Figure 1, baseline performance was assessed twice (T1, T2) with a time interval of three months (baseline effects); assessments were repeated after the first (T3) and second therapy phase (T4) to assess specific treatment effects (pre vs. post CFATand ELT) and overall treatment effects (T2 vs. T4), as well as in a follow-up three months later (T5) to examine follow-up effects (T4 vs. T5).

Language testing

In order to allow for time-effective assessment of language performance, we performed the core formal language test battery at all test times (T1-T5), but a peripheral formal language test only at first pre-test (T1) and follow-up (T5) (see Table I). Moreover, a therapy screening of naming was delivered before (T2), intermediate (T3) and directly after therapy (T4).

The core formal test battery consisted of (i) the BIWOS (Benassi et al., 2012) to evaluate word retrieval and (ii) the Szenario-Test (Nobis-Bosch et al., in press; van der Meulen et al., 2010) to evaluate multimodal everyday communication. The peripheral formal language test entailed (iii) the AAT (Huber et al., 1983; Huber et al., 1984) to determine severity and type of aphasia including the distribution of speech and language deficits. The AAT consists of six aspects of spontaneous speech ratings (communicative behaviour, articulation and prosody, automated language, semantic structure, phonological structure, and syntactic structure) as well as the five subtests Token Test, Repetition, Written Language, Naming and Comprehension.

In order to consider individually relevant stimuli for training and assessment, 14 everyday topics were chosen for each person with aphasia; thus, the PWA were asked about their interests, hobbies and important themes of daily living at time point T1. PWA were supported in determining suitable topics as appropriate. Thirty possible relevant topics were chosen in advance by the authors from the entire set of the Activity Card Sort test (Baum & Edwards, 2001) and each topic was illustrated by a symbol card. If required, these symbol cards were presented one after the other to the PWA with the question whether the topic was relevant for them or not. Four of those topics were part of the CFA-screening at T1 (shopping, watching television, cooking a soup and washing clothes) (see Spitzer, Binkofski, Willmes, & Bruehl, 2019). The 14 most relevant topics for each person with aphasia were chosen and used for the therapy screenings at T2 and later time points (Screening of Naming; Screening of Category Shift; Cognitive Flexibility in Aphasia Screening). Moreover, five of these topics were used as training material for each therapy phase (see therapy regimen) (Supplementary Figure S2 for details).

For T2 to T4, two trained and two untrained topics out of the 14 topics were chosen for the Screening of Naming, with ten words of each topic selected according to their frequency (for each topic four high-frequency nouns, three low-frequency nouns, three verbs). For each word, a suitable picture was chosen. To make sure each picture fitted to the word, they were consecutively presented to three healthy persons for overt confrontation naming beforehand. If one of three persons did not correctly produce the target word, the picture was replaced by a more appropriate one.

Testing the functioning of cognitive flexibility

To examine cognitive flexibility, the core formal tests WCST-64 (Kongs et al., 2000) and the RWT (Aschenbrenner et al., 2000) were used at all test times (T1–T5). The RWT determines verbal performance in cognitive flexibility by counting the number of words the PWA can deliver from two alternating semantic categories (subtest semantic category shift) or with two alternating initial letters (subtest word form shift) (Aschenbrenner et al., 2000). The test-retest reliability of the four subtests ranges from rtt¼0.72 to 0.89. In contrast, the WCST-64 (Kongs et al., 2000) evaluates the nonverbal performance in cognitive flexibility, which minimises the influence of language deficits (average *generalisability coefficient* 0.74). The patient is to attribute 64 cards to four desk cards, while the sorting principles must be inferred by the patient assessed. The patient only receives feedback on whether an assignment was correct or incorrect. After ten consecutive correct attributions, the sorting principle changes without any further information. For this task the patient must develop an appropriate problem-solving strategy and must modify it flexibly when the stimulus conditions change (Kongs et al., 2000), for which different functions are relevant like sustained attention, working memory, inhibition and cognitive flexibility (Fridriksson et al., 2006). The latter is especially assessed by scoring perseverative errors (Greve, Bianchini, Hartley, & Adams, 1999), which mean that the patient persists in responding to a sorting principle that is not correct.

Some findings showed that PWA might have difficulties with categorisation per se. Since categorisation is an essential requirement for solving the WCST-64 (Purdy, 2002), the PWA performed a categorisation task beforehand, in order to control for categorisation difficulties. Based on Purdy (2002), the PWA had to classify nine pictures according to the categories (i) vehicles, (ii) animals, and (iii) furniture. All of the PWA completed this task successfully.

Cognitive flexibility for therapy materials was screened at T2, T3 and T4 by the Screening of Category Shift. The PWA were asked to alternately produce 20 words from two trained topics and 20 words from two untrained topics. For each word, the PWA had 10s time. To avoid bias, the four topics of the Screening of Category Shift were different from the topics of the Screening of Naming (see Supplementary Figure S2 for details).

Testing cognitive flexibility in communication

In order to evaluate cognitive flexibility in communication, it was necessary to create a special screening – the Cognitive Flexibility in Aphasia Screening (CFA-Screening) – which was performed at all test times (T1–T5). Our previous diagnostic study found good inter-rater reliability, specificity and sensitivity for the CFA-Screening and showed that it is well able to determine impaired cognitive flexibility in communicative situations (Spitzer et al., 2019). The CFA-Screening assesses the three key characteristics of cognitive flexibility in communication, which are the *a) response to change of topic, b) response to misunderstandings and c) use of nonverbal communication*. The communicative setting in which the Speech-Language Pathologist (SLP) provoked flexible responses at T1 comprised four pre-defined topics; at T2 and each later test time, two trained and two untrained everyday topics were individually selected from the pool of 14 relevant topics. Each of the provocations was followed by a conversation. Responses were triggered by a series of eight photos per topic depicting the everyday situation (Supplementary Figure S3 for details). During each conversation, each of the three key characteristics was provoked five times by the SLP, namely by a) posing an unexpected question so that the PWA must change to another topic, b) reproducing a purposely incorrect utterance from the PWA, so that the PWA must re-express their utterance in other words, and c) asking to present the message in a nonverbal way. To guarantee a standardised process, the provocations in each topic were determined in advance and noted on protocols by the SLP during the conversation.

After each provocation, the SLP scored the corresponding PWA performance in cognitive flexibility dichotomously. Different objective criteria derived from the literature were determined and compiled into a scoring system (Spitzer et al., 2019): (a) *Response to change of topic* was taken to be accomplished if the PWA showed an adequate answer to the unexpected question within two seconds without any word-finding difficulties or perseverations. When the PWA corrected the SLP within two seconds, an adequate response to (b) *misunderstandings* was assumed. The pilot study revealed that patients tended to repeat the misunderstood word or just answered "no". Thus, in these cases the person with aphasia was asked to explain the word in a different way, and the response was successful if the explanation was correct or an adequate synonym was provided within two seconds. c) *Use of nonverbal communication* was taken to be successful if the PWA switched to gestures or used pencil and paper within two seconds. It was not essential how appropriate and specific the gestures or drawings were, the PWA only needed to clearly have initiated nonverbal means of communication. The rating system was used by the SLP online during communication, and the screening was video-recorded to check the ratings offline later.

Therapy regimen

Each therapy day of CFATand ELT consisted of two steps, impairment-based exercises (step I) and communicative exercises (step II), both of which were based on two of the individual everyday topics. Therefore, from the 14 individual everyday topics chosen at T1, five were randomly assigned to CFAT and five to ELT, and for each therapy day two of them were randomly selected in varying combination (see Supplementary Figure S4 for details). The material of CFAT and ELT was the same: for step I, Aphasia and executive functions 5 materials from the trained topics of the Screening of Naming were used (see above). For step II, materials from the trained topics of the CFA-Screening were used. Impairment-based exercises in each type of therapy (CFAT and ELT) focussed on word finding or verbal descriptions in response to target pictures. The communicative exercises added conversational interactions about target pictures, resembling real-life conversations. The type of exercises for both steps differed between the therapy regimen: while ELT constituted common aphasia therapy techniques of word finding difficulties, CFAT explicitly targeted cognitive flexibility as well. All other methodological aspects, including number of exercises and structure of the communicative exercises (initiation and course of the conversation), were the same in CFAT and ELT.

Procedure for CFAT

Step (I) Impairment-based exercises

In three impairment-based exercises, each key characteristic of cognitive flexibility was addressed: (a) *change of topic* constituted a verbal fluency task in which the person with aphasia was to produce any word within each of the two alternating individual everyday topics; (b) *response to misunderstandings* not only alternated topics, but also alternated the tasks of overt naming and circumlocution in response to a picture; (c) *use of nonverbal communication* alternated confrontation naming with nonverbal expression of the depicted meaning, over and above alternating topics. To support the PWA in performing the task, a predefined hierarchy of assistance was delivered by the SLPs fostering reasoning about and self-contained use of appropriate strategies (Hinckley, Carr, & Patterson, 2001). The increasing hierarchy consisted of: (i) repeating and elaborating the task instruction; (ii) indicating that a change of task/topic had occurred; (iii) jointly considering possible adequate responses and strategies, which activated the acquisition and the use of problem-solving strategies (see Supplementary Table S III for detailed description and examples for both therapy methods).

Step (II) communicative exercises

For communicative exercises, the SLP provoked all three characteristics of cognitive flexibility in line with the CFA-Screening: (a) *change of topic* was provoked by the SLP by asking an unexpected question; for (b) *response to misunderstandings* the SLP reproduced an utterance from the PWA incorrectly, and for (c) *use of nonverbal communication* the SLP asked the PWA to explain something in a nonverbal way. To ensure standardisation and fidelity of treatment procedures, the provocations in each topic and their order were determined in advance, and they were constructed to have a similar level of difficulty. Therefore, for (a) an unexpected open question has always been formulated; for (b) and (c) the target word has always been a concrete noun or a high-frequent main verb. Their use and according PWA responses were noted on protocols during the conversation by the SLP. As in the CFA-Screening, the responses were additionally triggered by a series of eight photos per topic (Supplementary Figure S3 for details), with a similar arrangement within each photo series, also to ensure a standardised process. For example, the first photo of the series often showed the materials involved in the topic or the ingredients. Contrary to the CFAScreening, the PWA received increasing assistance as in step I.

Results of group analyses.

p Values/Cohen's d	Core formal language tests		Language screening	Core formal tests of cognitive flexibility			Screenings of cognitive flexibility	
	BIWOS[a]	Szenario -Test[b]	Screening of Naming	WCST-64 Pers.[c]	RWT[d]/ sem	RWT[d]/ form	CFA-Screening[e]	Screening of category shift
Baseline differences (T1 vs. T2)	0.100^{f}	0.098^{g}	—	0.105^{g}	0.296^{f}	0.447^{f}	0.301^{f}	—
Overall treatment effects (T2 vs. T4)	$0.002\uparrow^{**f}$	0.148^{g}	$0.001\uparrow^{**g}$	0.075^{g}	$0.042\uparrow^{*f}$	0.126^{g}	$<0.001\uparrow^{***f}$	$<0.001\uparrow^{***f}$
Follow-up effect (T4 vs. T5)	0.075^{f}	0.430^{g}	—	0.348^{g}	0.178^{f}	0.078^{g}	$0.016\downarrow^{*f}$	—
Specific treatment effect CFAT (pre vs. post CFAT)	$0.010\uparrow^{*f}/0.222$	$0.031\uparrow^{*g}/ 0.464$	$0.035\uparrow^{*f}/0.465$	$0.414^{f}/0.045$	$0.049\uparrow^{*f}/0.427$	$0.242^{g}/0.321$	$<0.001\uparrow^{***f}/0.907$	$<0.001\uparrow^{***f}/0.968$
Specific treatment effect ELT (pre vs. post ELT)	$0.111^{f}/0.117$	$0.500^{g}/-0.278$	$0.030\uparrow^{g}/0.519$	$0.035\uparrow^{*g}/-0.256$	$0.389^{f}/0.160$	$0.438^{g}/-0.067$	$0.136^{f}/0.184$	$0.113^{f}/0.230$

Table 2: *p < 0.05, **p ≤ 0.01, *** p ≤ 0.001; ↑ : improvement; ↓: decrease; T: test time; CFAT: Cognitive Flexibility in Aphasia Therapy; ELT: Everyday Language Therapy. aBielefeld screening of word finding (Benassi et al., 2012); bSzenario-Test (Nobis-Bosch et al., in press); cWisconsin Card Sorting Test-64, perseverative errors (Kongs et al., 2000); dRegensburg Test of Fluency (Aschenbrenner et al., 2000); sem: semantic category shift; form: word form shift; eCognitive Flexibility in Aphasia Screening; fpaired t-test, one-tailed; gexact version of Wilcoxon signed ranks test, one-tailed.

Procedure for ELT

Step (I) impairment-based exercises

Impairment-based exercises consisted of (a) a confrontation naming task, (b) naming from verbal description by the SLP, and (c) nonverbal expression of the spoken word delivered by the SLP. An increasing hierarchy of assistance which is well-known in aphasia therapy (Abel, Willmes, & Huber, 2007; Nickels, 2002) was used, which consisted of (i) a semantic cue, (ii) a phonological cue and (iii) a semantic closure sentence.

Step (II) communicative exercises

In the communicative exercises, the PWA and the SLP talked about the two individual everyday topics of the day based on a series of eight photos, like in the CFA-Screening. For each series, the person with aphasia was first to express the most important content of the photos. Consecutively, the SLP asked free and open questions on the current topic and gave assistance as presented for the impairment-based exercises (see Supplementary Table S III for details).

Data analysis of CFAT and ELT

All statistical analyses were calculated with IBM SPSS Statistics (version 21) on the group level. At first, baseline-effects (T1 vs. T2), overall treatment effects (T2 vs. T4) and follow-up effects (T4 vs. T5) were analysed with the paired t-tests for normally distributed data and the exact version of the Wilcoxon signed ranks test (one-tailed) and involved all outcome-measures of CFATund ELT (see below).

For the evaluation of specific treatment effects of CFAT (pre vs. post CFAT), the following outcome measures were analysed: for language skills and communicative ability the Screening of Naming, BIWOS and Szenario-Test; for cognitive flexibility the CFAScreening, Screening of Category Shift, RWTand the number of perseverative errors of WCST-64 (paired t-tests for normally distributed data and exact version of Wilcoxon signed ranks test, one-tailed). For all therapy screenings (Screening of Naming, Screening of Category Shift and CFA-Screening) we compared trained versus untrained topics. For specific treatment effects of ELT, the same assessments were evaluated, but improvements were only expected for Screening of Naming, BIWOS and Szenario-Test (paired t-tests for normally distributed data and exact version of the Wilcoxon signed ranks test, one- tailed).

Interaction effects and comparison of CFAT vs. ELT

Additionally, it was relevant to analyse possible interaction effects between the assignment to the orderrelated patient group and the improvements for both therapy methods (CFAT and ELT). Therefore, after testing the formal requirements for a mixed analysis of variance (ANOVA), we conducted an ANOVA with repeated measures for each outcome measure. In the design, the assignment to the group getting one specific sequence of treatment methods was the between subjects factor (GROUP), whereas the method (CFATor ELT) was the within-subjects factor (METHOD). In case of no interaction effects, i.e. no differential carry-over effects of the first on the second treatment method applied, we could utilise the result of the test for the main effect METHOD, to decide whether CFAT was more effective than ELT.

Analysis of PWAs' profile regarding therapy outcome

To analyse whether the degree of patient improvement for CFATor ELT was associated with the level of impairments at T1, we conducted a correlation analysis. Thereby, test scores of AAT subtests Naming and Comprehension, BIWOS, Szenario-Test as well as both subscales of RWT at T1 were correlated with the specific treatment effects of CFATand ELT in the following outcome measures: Screening of Naming, Screening of Category Shift and CFAScreening.

Main and interaction effects of mixed ANOVAs.

	Core formal tests		Language screening	Formal tests of cognitive flexibility			Screenings of cognitive flexibility	
	BIWOS[a]	Szenario-Test[b]	Screening of Naming	WCST-64 Pers[c]	RWT/ sem[d]	RWT/ form[d]	CFA-Screening[e]	Screening of Category Shift
Main effect METHOD	$F(1,8) = 0.934$; $p = 0.362$	$F(1,8)=2.681$; $p=0.140$	$F(1,8)=0.006$; $p=0.941$	$F(1,8)=0.012$; $p=0.915$	$F(1,8)=0.093$; p=0.768	$F(1,8)=0.409$; $p=0.541$	$F(1,8)=10.436$; $p=0.012$	$F(1,8)=18.458$; $p=0.003$
Main effect GROUP	$F(1,8)=1.005$; $p=0.345$	$F(1,8)=1.231$; $p=0.299$	$F(1,8)=3.606$; p=0.094	$F(1,8)=1.729$; p=.225	$F(1,8)=0.401$; $p=0.544$	$F(1,8)=0.000$; $p=1.00$	$F(1,8)=0.037$; $p=0.851$	$F(1,8)=0.042$; p=.854
Interaction effect GROUP*METHOD	$F(1,8)=5.292$; $p=0.050$	$F(1,8)=0.262$; $p=0.623$	$F(1,8)=2.296$; $p=0.168$	$F(1,8) =0.993$; $p=0.348$	$F(1,8)=0.165$; $p=0.695$	$F(1,8)=0.102$; $p=0.757$	$F(1,8)=3.678$; $p=0.091$	$F(1,8)=0.831$; $p=0.389$

Table 3: *p < 0.05, **p ≤ 0.01, *** p ≤ 0.001; aBielefeld screening of word finding (Benassi et al., 2012); bSzenario-Test (Nobis-Bosch et al., in press); cWisconsin Card Sorting Test-64, perseverative errors (Kongs et al., 2000); dRegensburg Test of Fluency (Aschenbrenner et al., 2000); sem: semantic category shift; form: word form shift; eCognitive Flexibility in Aphasia Screening.

Result

Evaluation of CFAT and ELT

Baseline effect, overall treatment effect and follow-up effect

A summary of performance is presented in Table II (see Supplementary Table S IV and Table S V for all raw scores, means and standard deviations). Baseline performance (T1 vs. T2) was stable. The overall treatment effects (T2 – T4) were (highly) significant for BIWOS, Screening of Naming, subtest semantic category shift from the RWT, CFA-Screening and Screening of Category Shift. The analysis of followup effects (T4 vs. T5) showed that therapy effects were maintained over a period of three months for all tests, except for the CFA- Screening which showed a decrease. Specific treatment effects of CFAT The specific treatment effects showed that PWA performed significantly better after compared to before CFAT in all outcome measures, except WCST-64 (Kongs et al., 2000) and subtest word form shift of RWT (see Table II). A detailed analysis of the Szenario-Test showed that three PWA improved numerically (PWA 5, 8, 10); the other PWA showed no or only slight numerical improvements (PWA 1, 2, 3, 4, 6, 7, 9) (see Table S I). The analysis of trained versus untrained topics of the Screening of Category Shift and CFA-Screening revealed significant improvements in trained (p<0.001 and p¼0.001) as well as untrained (p¼0.004 and p¼0.041) topics, respectively (paired t-test, one-tailed).

Specific treatment effects of ELT

To the contrary, significant treatment effects of ELT were present for WCST-64 and Screening of Naming (see Table II). For the latter, PWA showed improved performance for both trained (p¼0.008) and untrained topics (p¼0.040; exact version of Wilcoxon signed ranks test, one-tailed).

Interaction effects and comparison of CFAT vs. ELT

The formal requirements for the ANOVA were met. The ANOVA analyses revealed no interaction effects GROUP x METHOD (see Table III). Therefore, we analysed the main effects for METHOD, which showed that PWA profited significantly more from CFAT compared to ELTon both of our novel screenings of verbal cognitive flexibility (CFA-Screening: F(1,8)¼10.436; p¼0.012; Screening of Category Shift: F(1,8)¼18.458; p¼0.003).

Analysis of PWAs' profile regarding therapy outcome

The correlation analysis showed that the degree of improvement of PWA for CFAT was associated with the level of impairments at T1: there was a significant negative correlation between performance in AAT subtest Naming (Huber et al., 1983; Huber et al., 1984) at T1 and improvements in the Screening of Naming, and between performance in RWTsemantic category shift at T1 and improvements in the Screening of Category Shift. Significant positive correlations were found for performance in AAT subtest Comprehension at T1 with improvements in the CFA-Screening for CFAT, and for performance in AAT subtest Naming at T1 with improvements in the Screening of Naming for ELT (see Table IV).

Discussion

The present pilot study provides first evidence that the novel CFAT can improve verbal language skills, communicative ability and verbal cognitive flexibility of PWA and indicates that this linguistic-executive approach is especially effective for those PWA with impaired expressive but rather preserved receptive capabilities. Moreover, all PWA completed the trial (no drop-out), confirming feasibility of the study. These results can be substantiated by future studies.

As expected, PWA after CFAT showed significantly improved performance in all of our language outcome measures as well as in cognitive flexibility as measured by our screenings and the semantic word fluency subtest. The improvements were not restricted to the therapy settings or materials as assessed by the screenings,

Spearman rank (upper triangular) and Pearson correlation (lower triangular) between specific treatment effects (CFAT, ELT) and PWAs' initial profile.

	CFAT[a]			ELT[b]			Performance at T1					
	Naming	Category Shift	CFA[c]	Naming	Category Shift	CFA[c]	AAT[d] Naming	AAT[d] Comp	BIWOS[e]	WCST- 64[f]	RWT sem[g]	RWT form[g]
CFAT[a]												
Naming	–	0.45	0.19	**−0.65***	−0.12	0.13	**−0.69***	−0.17	−0.30	0.09	−0.21	0.08
Category Shift	**0.64***	–	−0.30	−0.57	0.23	−0.21	−0.42	−0.32	−0.48	0.21	**−0.67***	−0.01
CFA[c]	0.23	−0.13	–	−0.26	0.42	−0.08	−0.30	**0.71***	−0.06	−0.03	0.33	−0.38
ELT[b]												
Naming	**−0.82****	**−0.67***	−0.21	–	−0.38	−0.01	**0.84****	0.02	0.51	0.13	0.32	0.01
Category Shift	−0.03	0.21	0.58	−0.18	–	0.01	−0.29	0.36	−0.19	0.04	0.17	−0.11
CFA[c]	−0.03	−0.16	−0.14	0.19	0.02	–	0.20	0.08	0.47	0.35	0.24	−0.12
Performance at T1												
AAT[d] Naming	**−0.71***	−0.63	−0.22	**0.85****	−0.25	0.25	–	0.28	**0.75***	0.09	0.28	−0.04
AAT[d] Comp	−0.18	−0.23	0.44	0.09	0.46	0.20	0.34	–	0.41	−0.24	0.40	−0.26
BIWOS[e]	−0.30	−0.38	−0.09	0.41	0.11	**0.65***	**0.67***	0.46	–	0.03	**0.71***	0.30
WCST- 64[f]	0.02	0.16	0.16	0.20	0.03	0.27	0.15	−0.26	0.11	–	−0.17	−0.49
RWT sem[g]	−0.26	−0.62	0.45	0.40	0.37	0.43	0.47	0.51	**0.76***	0.05	–	0.48
RWT form[g]	−0.06	−0.14	−0.09	0.10	0.18	−0.23	0.07	−0.10	0.24	−0.47	0.35	–

Table 4: Significant correlations given in bold.

*$p < 0.05$, **$p \leq 0.01$, *** $p \leq 0.001$; T: test time; CFAT: Cognitive Flexibility in Aphasia Therapy; ELT: Everyday; Language Therapy; PWA: person with aphasia.

aSpecific treatment effects, pre vs. post CFAT, as measured by the three screenings; bSpecific treatment effects, pre vs. post ELT, as measured by the three screenings; cCognitive Flexibility in Aphasia Screening; dAachen Aphasia Test, subtests Naming and Comp¼Comprehension (Huber et al., 1983; Huber et al., 1984); eBielefeld screening of word finding (Benassi et al., 2012); fWisconsin Card Sorting Test- 64 (Kongs et al., 2000); gRegensburg Test of Fluency (Aschenbrenner et al., 2000); sem: semantic category shift; form: word form shift.

but also generalised to other lexical items (BIWOS) and transferred to communicative settings (Szenario-Test). Thus, we assume that the CFAT may be one possibility to enhance the patient's ability to take part in conversation, which is one of the main goals of aphasia therapy (King et al., 2013; Worrall et al., 2011). A detailed analysis of the Szenario-Test showed that the improvements in the test after CFAT were mainly due to three PWA. We suppose that the other PWA also improved their communicative ability but these improvements could not be measured due to ceiling effects in the Szenario-Test.

Contrary to our expectation, however, nonverbal cognitive flexibility as measured by the WCST-64 did not improve. One reason could be that the WCST-64 does not measure cognitive flexibility only, but also other cognitive functions like working memory and problem solving (Kongs et al., 2000), which were not explicitly trained by CFAT. Furthermore, Fridriksson et al. (2006) discuss that the WCST-64 is too complex for PWA. Another reason could be that the linguistic-executive treatment exerted by CFAT targets verbal cognitive flexibility, but does not generalise to nonverbal cognitive flexibility. Moreover, while semantic fluency improves, formal word fluency does not, indicating that CFAT targets at both linguisticsemantic and verbal executive functions (see Bruehl and Willmes (in preparation), for a similar result for linguistic-executive treatment via interfered-naming therapy for aphasia) (see also below).

As expected and in line with previous studies (Brady, Kelly, Godwin, Enderby, & Campbell, 2016), the results indicate that the ELT leads to significant improvements in language skills. Contrary to our expectation, there were significant improvements for WCST-64 after ELT. However, an in-depth case analysis post-hoc revealed that only one person with aphasia improved substantially in this way; the reason is unknown. When this person was considered to be an outlier and excluded from this analysis, the effect did not reach significance (pre vs. post ELT; exact version of Wilcoxon signed ranks test, onetailed p¼.221).

The mixed ANOVAs indicate that the CFAT may be more effective than the ELT regarding verbal cognitive flexibility. This result is in line with our expectations and with the assumption of Beckley et al. (2013), who assume that verbal cognitive flexibility requires specific training to ameliorate. Furthermore, this result corroborates findings of Simic, Rochon, Greco, and Martino (2019) who reported that PWA with impaired executive functions profited less from conventional therapy. Thus, if PWA present with impaired verbal cognitive flexibility, our data suggest that CFAT might be the preferred choice over conventional therapy.

As expected, the ANOVA analyses showed no significant interaction effects between GROUP and METHOD, indicating that the order of therapy methods might not influence therapy outcomes differentially. However, applying a more lenient threshold revealed a marginally significant interaction effect ($0.05 < p < 0.10$) indicating some differential effect of order of therapy delivery for BIWOS and CFA-Screening. These differential order effects need to be investigated in statistically more powerful studies utilising larger samples.

The analyses of trained vs. untrained topics revealed generalisation effects for untrained topics regardless of therapy method and order. Further investigations are required to replicate and further illuminate generalisation in linguistic-executive treatments.

We hypothesised that individual therapy outcome is related to aspects of initial PWA performance as a predictive factor. The correlational findings indicate that PWA with lower performance in naming and category shift but better performance in comprehension may benefit from the CFAT, whereas PWA with good performance in naming may profit more from the ELT. Based on these results, we assume that CFAT allowed for improvements in various language and cognitive domains, both due to the fact that linguistic and executive processing are directly trained and that amelioration of the central executive control mechanism positively impacts the variety of control-dependent domains. These findings indicate for which PWA such linguistic-executive therapy is most appropriate (eligibility criteria). Especially PWA with extensive deficits may profit from our novel approach, whereas for PWA with milder deficits conventional aphasia therapy is sufficient. However, comprehension appears to be an exception here: the correlational findings indicate that CFAT is more successful for PWA with rather preserved auditory language comprehension. The most convincing explanation would be that CFAT requires metacognition, and Mayer, Mitchinson, and Murray (2017) argued that for such approaches sufficient comprehension is necessary.

In terms of the treatment of executive functions of PWA, Mayer et al. (2017) differentiate between process-based intervention and metacognitive strategies. The former means that underlying cognitive processes are directly activated and trained; the latter means the acquisition and the use of metacognitive strategies. The CFAT includes both: whereas the exercises in Step I and II are a more process-based intervention, we guided the PWA through the hierarchy of assistance to find and apply problem-solving and metacognitive strategies. Nonetheless, in the future the CFAT could be extended by introducing a period of reflection, in which the PWA and the SLP reflect about the use and acquisition of strategies. Such reflection can stimulate metacognitive and selfregulating behaviour and can improve self-monitoring and motivation of the PWA, like Lee and Sohlberg (2013) showed in their study. Therefore, such reflection could support the transfer of metacognitive strategies in daily live even more, and would be a worthwhile supplement for the CFAT.

There are six methodological aspects of the study which require further consideration. First, since our aim was to develop a novel linguistic-executive therapy, which integrates realistic everyday communication to address real-life communication, the topics for the dialogues of CFAT were selected according to their relevance in PWA's everyday life, and for each topic individualised material was prepared. Thus, the therapy tasks of CFATare relevant and interesting for the PWA, as generally postulated by King et al. (2013) for aphasia therapy. However, it is important to note that the individual creation of photo series for each PWA is not time-efficient for clinical practise. To make the individualised approach more feasible, we recommend the use of existing materials, like private photos of the PWA, and materials from journals, books or the internet as appropriate.

Second, it is important to note that the CFAT is limited to cognitive flexibility. However, other executive functions, like working memory, are also relevant for everyday communication (Fridriksson et al., 2006), and deficits of these functions should also be integrated into the therapy of PWA. To accomplish this, the CFAT procedure could be modified and extended to cover training of other executive functions, adapting the provocations and questions by the SLP in the guided everyday communication as featured by CFAT. Thus, given the theoretical and clinical need to understand and ameliorate executive impairments in PWA, the innovative linguistic-executive approach developed could be a starting point for further therapies which target language and cognitive functions in combination to improve everyday communication.

A third aspect refers to the need for suitable assessments to be able to actually measure therapy outcomes and to plan therapy (Helm-Estabrooks, 2002). Therefore, it is relevant to develop and evaluate valid and standardised assessments. Analyses indicate that the CFA-Screening is a means of assessing cognitive flexibility in realistic everyday communication in a feasible, standardised and time-efficient way (Spitzer et al., 2019). The CFA-Screening therefore complements non-linguistic assessments for PWA developed by Helm-Estabrooks (2002) and Kalbe, Reinhold, Brand, Markowitsch, and Kessler (2005). Further standardised assessments to assess the whole range of executive functions in PWA are necessary to be able to detect changes in PWA performance.

A fourth aspect is the development and maintenance of therapy effects. After a stable baseline, there were significant treatment effects due to the training as presented above. Performance after therapy remained stable in all tests but the CFA-Screening, which revealed decreased performance at follow-up. Thus, to reach more robust performance, an extension of therapy duration over and above the two weeks per method may be beneficial, or to integrate family members in CFAT, like in the communication partner training (Beckley et al., 2013; Simmons-Mackie, Raymer, Armstrong, Holland, & Cherney, 2010). In such a training, the communication partner could learn to support the PWA by offering the hierarchy of assistance, fostering reasoning about and self-contained use of appropriate strategies. Simmons-Mackie et al. (2010) showed that such partner trainings can improve communication activities of persons with chronic aphasia. Along the same lines, family members could be made aware of the PWA's cognitive flexibility disorder, and be informed about what reduces cognitive demands, e.g. by preventing rapid changes of topics. Manders, Mari€en, and Janssen (2011) showed that most family members value to be informed about optimal ways of communication. However, further studies are required to evaluate the positive effect of such information on everyday communication with PWA.

Another aspect concerns a possible confound: the treatment session and the assessments were conducted by the same SLP, whereby the SLP knew which treatment the PWA had received; thus, the investigator was not blind to the methods applied.

Finally, the small sample size of ten PWA constitutes a limitation of our study and therefore the results are still preliminary. However, results show a clear impact of CFATon language skills, communicative ability and verbal cognitive flexibility of PWA. Another limitation is the possibility of carry-over effects from the previous therapy phase to the following one in cross-over designs (Hills & Armitage, 2004). To avoid carry-over effects, Hills and Armitage (2004) recommend a wash-out phase after the first therapy phase so that the therapy effects can wear off. Nevertheless, even though we did not use a wash-out phase, we did not find significant differences in sequence effects, indicating that there were no differential carry-over effects.

Conclusion

In the present pilot study, we evaluated a novel aphasia therapy which targets both cognitive flexibility and language processing in realistic everyday communication. The results indicate that this linguistic-executive approach might improve language skills, verbal cognitive flexibility and communicative ability of PWA, and thus has high potential to enhance PWA's social participation. Furthermore, the results indicate that the novel therapy is especially effective for patients with more severe deficits in expressive language, but sufficient receptive abilities to understand instructions and process the metacognitive demands of the approach. These findings should be substantiated by future work.

Acknowledgement
The authors thank all cooperating private practices and rehabilitation centres for their support in patient recruitment and all persons who participated in our study. Furthermore, we thank the Neuroscience and Aphasia Research Unit (NARU) group at the University of Manchester for their feedback during a short visit of the first author in Manchester. We also thank the reviewer and the editor for their very helpful comments.

Declaration of interest
The authors report no declarations of interest.

Supplementary material
Supplemental data for this article can be accessed at https://doi.org/10.1080/17549507.2020.1757152

Funding
This work was supported by the German Academic Scholarship Foundation.

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