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RESEARCH ARTICLE

A combined therapy for limb apraxia and related anosognosia

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ABSTRACT

Left hemisphere stroke frequently leads to limb apraxia, a disorder that has been reported to impact independence in daily life and rehabilitation success. Nonetheless, there is a shortcoming in research and availability of applicable trainings. Further, to date, anosognosia for limb apraxia has largely been neglected. Therefore, we developed a *Naturalistic Action Therapy* that trains object selection and application with an errorless learning approach and which includes supported self-evaluation. The current study presents the results of two stroke patients participating in the training. The procedure entailed two baseline and one post-training sessions including standardized limb apraxia and anosognosia assessments as well as 18 naturalistic action tasks. The training consisted of 15 sessions during which 4–6 of the 18 naturalistic action tasks (e.g., pour water into a glass, make a phone call) were trained. Both patients showed improvement in trained and untrained tasks as well as in standardized apraxia and anosognosia assessments. Training effects appeared strongest for the trained items. The procedure is documented in detail and easy to administer and thus may have the potential to be applied by relatives. The results of this pilot-study are promising and suggest that the approach is suitable for further evaluation.

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Introduction

Limb apraxia is commonly defined as a motor-cognitive disorder leading to difficulties in imitating gestures, pantomiming tool-use and actually using real tools (Goldenberg, 2013). Despite its negative impact on independence in

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daily life activities (Goldenberg & Hagmann, 1998b; Goldenberg, Daumüller, & Hagmann, 2001; Unsal-Delialioglu, Kurt, Kaya, Culha, & Ozel, 2008) and rehabilitation success (Dovern, Fink, & Weiss, 2012; Hanna-Pladdy, Heilman, & Foundas, 2003; Wu, Burgard, & Radel, 2014), studies on the effectiveness of apraxia rehabilitation are scarce (Buxbaum et al., 2008; Buxbaum & Randerath, 2018; Cantagallo, Maini, & Rumiati, 2012; Dovern et al., 2012; van Heugten & Geusgens, 2017; Worthington, 2016). Further, to the best of our knowledge, there were only two new rehabilitation approaches published since the review by Buxbaum and colleagues in 2008 (see also Worthington, 2016). This apparent lack in novel training approaches may be explained by the persisting misbelief, that apraxia only occurs in lab settings but not in real life and further, that it recovers spontaneously (van Heugten & Geusgens, 2017). However, studies have shown that 88% of patients diagnosed with apraxia were still apraxic after 20 weeks of standard neurorehabilitation (Donkervoort, Dekker, & Deelman, 2006).

Findings like these demonstrate the necessity to generate training programmes for apraxia, and the need to build upon and advance existing approaches for rehabilitation. However, there exist only few rehabilitation studies that focus on limb apraxia affecting real tool-use. The most studied training approach was implemented by van Heugten and colleagues (Donkervoort, Dekker, Stehmann-Saris, & Deelman, 2001; Geusgens et al., 2006; Geusgens, van Heugten, Cooijmans, Jolles, & van den Heuvel, 2007; van Heugten et al., 1998; van Heugten, Dekker, Deelman, Stehmann-Saris, & Kinebanian, 2000). Their strategy training resulted in significant improvements of motor functions and apraxia in trained as well as in untrained ADL tasks. Effects of training were found directly after training and three months later (Geusgens et al., 2007; van Heugten et al., 1998). Goldenberg and colleagues also developed and evaluated different training approaches involving the application of tools (Goldenberg et al., 2001; Goldenberg & Hagmann, 1998a). In their direct training, ADL tasks were trained until they were solved without errors. This led to a significant improvement in the trained tasks but was only stable for 3 out of 6 patients after six months and did not generalize to untrained tasks (Goldenberg & Hagmann, 1998a). However, for all of those training interventions, Buxbaum et al. (2008) and Worthington (2016) claimed in their reviews, that apraxia was poorly diagnosed because the used assessments were not checked for reliability or validity. Further, Buxbaum et al. (2008) criticized in their review that the published training approaches frequently neglected the following points: (a) object recognition tasks were usually not performed, (b) changes in performance in standard apraxia tests were only seldomly evaluated, (c) the evaluation of maintenance and generalization of training effects were not standard practice, (d) the training was adapted to the individual's difficulties while omitting standardized procedures and (e) often the methods used in training were not sufficiently specified, making a replication of results impossible. Moreover, all of those trainings did not control for spontaneous recovery although the patients usually received training in the subacute phase, on average two months after stroke onset (e.g., van Heugten et al., 1998). Dovern et al. (2012) also criticized that only three randomized controlled trial studies were published between 1965 and 2011 (Donkervoort et al., 2001; Smania et al., 2006; Smania, Girardi, Domenicali, Lora, & Aglioti, 2000), whereby two of those focused on the training of gestures and not on real tool-use.

Also, none of the trainings included supported self-evaluation as an approach to address a lack of insight into the impairment (anosognosia) of solving naturalistic actions, despite the fact that anosognosia has been reported to potentially co-occur with limb apraxia (Buchmann, Jung, Liepert, & Randerath, 2018; Kusch et al., 2018) and that the performance of apraxic patients could benefit when monitoring their errors (Morady & Humphreys, 2009). Anosognosia should be taken into account for therapy approaches. A lack of insight into the impairment leads to reduced rehabilitation motivation (Buxbaum et al., 2008; Fleming, Strong, & Ashton, 1998; Peskine & Azouvi, 2007) and less independence in activities of daily life after rehabilitation discharge (Pedersen, Jorgensen, Nakayama, Raaschou, & Olsen, 1996).

In light of the presented limitations of apraxia trainings so far, we aimed to develop and present an apraxia rehabilitation method which considers all of the above-described demands on efficient training interventions mentioned by Buxbaum et al. (2008). The *Naturalistic Action Therapy* approach was designed to provide a practicable training that enables the evaluation of training efficacy on a. limb apraxia impairments and b. anosognosia of limb apraxia. Here we present an early phase case study conducted in a neurorehabilitation clinic. The *Naturalistic Action Therapy* approach was tested in two subacute stroke patients showing impaired use of familiar tools and diminished insight into their impairment. In addition, five patients participated in a control group without training.

Materials and methods

Participants

Both, therapy patients ($N = 2$) and control patients ($N = 5$), were recruited from the neurorehabilitation centre "Kliniken Schmieder" in Allensbach, Germany. The patients did not require intensive care and were able to actively participate for one hour in therapy sessions. Patients were initially right handed but could only use their left hand due to stroke-induced hemiplegia. For specific demographic and descriptive data, see Table 1.

The study design was approved by the ethical committee of the University of Konstanz. All patients participated voluntarily. Informed consent was obtained from patients and their authorized relatives and privacy rights were observed. The study was conducted in accordance with the Declaration of Helsinki.

Table 1. Demographic and global assessment data.

		Case 1	Case 2	Control group *N* = 5
Gender		Male	Female	4 male, 1 female
Age		80	56	*M* = 67, range: 61–76
Weeks since lesion onset at t1		14	15	*M* = 14, range: 8–19
Barthel index		55	50	*M* = 75, range: 45–100
Aphasia (AAT)		Severe to massive	Severe to massive	Severe to massive
Hemiplegia (WMFT)		Severe in right face, arm and leg	Severe in right arm, slight in right leg	Slight to severe in right arm
Impaired visuo-spatial perception / Neglect (BIT)		Severe to the right side	No	No
Apraxia				
FTT	t1	15 (severe)	13 (severe)[a]	*M* = 15, range: 10–19
	t2	19 (mild)	17 (severe)	*M* = 17.6, range: 15–20
NAT–BT	t1	0 (severe)	1 (severe)[a]	*M* = 1.8, range: 0–6
	t2	3 (mild)	2 (moderate)	*M* = 2.2, range: 0–5
Anosognosia				
VATA-NAT	t1	25 (present)	Cannot be interpreted	*M* = 16.8, range: 9.5–32
	t2	15 (present)	Cannot be interpreted	*M* = 6.8, range: 0–16

Notes: AAT: Aachener Aphasia Test (Huber, Poeck, Weniger, & Willmes, 1983); BIT: Behavioral Inattention Test: line bisection, star cancellation (Plummer, Morris, & Dunai, 2003); FTT: Familiar Tools Test; NAT: Naturalistic Action Test – Breakfast Task; t1 = assessment of limb apraxia and related anosognosia directly before training (cases 1 & 2) / before normal rehabilitation setting (control group); t2 = assessment of limb apraxia and related anosognosia after training (cases 1 & 2) / after normal rehabilitation (control group); VATA-NAT: Visual-Analogue Test assessing Anosognosia for Naturalistic Action Tasks; WMFT: Wolf Motor Function Test.

[a] Assessed during screening session directly before training.

Case 1

Case 1 was an 80-year old man with an acute left hemisphere haemorrhagic stroke and right-sided haematoma caused by falling due to the incident. The CT scan showed the presence of an embolic stroke with a haemorrhagic transformation in the left territory of the middle cerebral artery mainly including angular and supramarginal gyrus. It additionally revealed a prior right infratentorial ischaemia in the pons. The patient's brain scan is depicted in Figure 1.

Case 1 was assessed for limb apraxia and related anosognosia 14 weeks after stroke onset and then received the *Naturalistic Action Therapy* (see Figure 2). Due

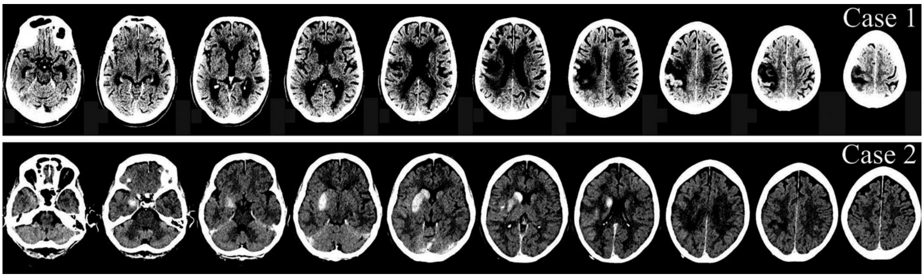

Figure 1. Multislice head CT images of Case 1 and 2. The images show axial slices of the patients' brains. Per individual, slices were selected to enhance visibility of the lesions.

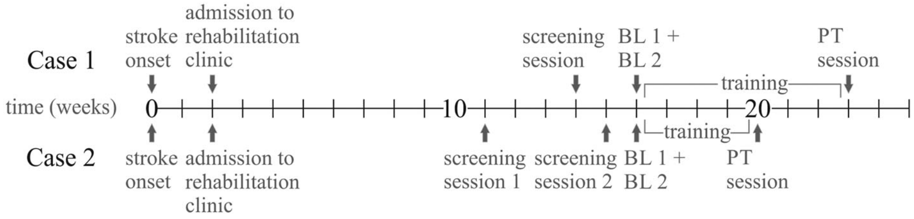

Figure 2. Timeline for Case 1 and 2 displaying the study design. Small vertical lines indicate one-week post-stroke onset. Baseline sessions are indicated as "BL 1" and "BL 2". Post-training session is indicated as "PT session".

to the limited length of hospital stay, a second screening session in order to assess the effect of spontaneous recovery was not feasible.

Case 2

Case 2 was a 56-year old woman with subacute left hemisphere stroke. The CT scan showed the presence of a left middle cerebral artery syndrome including basal ganglia (with putamen, pallidum, caudate nucleus, omitting the insula) and semioval centre. The scan additionally revealed lesions in the anterior horn of the lateral ventricle caused by a prior right embolic infarction. The brain scan is depicted in Figure 1.

Case 2 was assessed for limb apraxia and related anosognosia 11 and 15 weeks after stroke onset. The doubling assessment was used to measure the effect of spontaneous recovery in this patient before training her with the *Naturalistic Action Therapy* (see Figure 2). In the gap between week 11 (first assessment) and week 15 (second assessment), she received the normal rehabilitation programme (physical, occupational, speech therapy, neuropsychology) without any training specified for limb apraxia.

Control group

In order to evaluate spontaneous remission in apraxic deficits, 5 patients (sex: 4 male, 1 female; age: $M = 67.0$, range: 61–76 years) diagnosed with limb apraxia affecting real tool-use and related anosognosia in a first session 8–19 weeks after stroke onset ($M = 14$ weeks), performed the same tasks 4 weeks later again. In the 4-week gap they received the normal rehabilitation programme without any training specified for limb apraxia.

Assessment of limb apraxia and related anosognosia

Patients were selected based upon their performance in the *Diagnostic Instrument for Limb Apraxia – Short Version* (Buchmann & Randerath, 2017; Buchmann et al., 2019; Randerath, Buchmann, Liepert, & Büsching, 2017) and in the *Visual-Analogue Test assessing Anosognosia for Naturalistic Action Tasks* (VATA-NAT; Buchmann et al., 2018). To be included in this study, patients had to show difficulties in the application of familiar tools and objects (Familiar Tools Test and Naturalistic Action Test – Breakfast Task) and reduced insight into their tool-use difficulties.

The Familiar Tools Test (FTT) required the patient to select the correct tool out of three and show its application with the presented object (e.g., "scoop soup from pot to bowl"). Up to three practice items were used to secure task understanding. The evaluated test consisted of 5 actions.

The Naturalistic Action Test – Breakfast Task (NAT–BT) required the patient to prepare a breakfast consisting of a toasted slice of bread with butter and jam and a cup of tea with sugar. To facilitate task comprehension, photos of the final products were shown. To evaluate the performance, a combined score based on accomplished steps and error frequency was calculated.

To assess anosognosia for common tool-use we applied the VATA-NAT. In the VATA-NAT patients were asked to provide a self-evaluation upon their performance in a set of naturalistic actions. The questionnaire was developed to accommodate patients with aphasic deficits. Each task was described verbally and presented on a picture. The patients estimated their performance on a 4-point-Likert visual-analogue scale (0 = no problem, 1 = few difficulties, 2 = serious difficulties, 3 = impossible to plan and execute the shown task) with two smileys indicating the respective ends of the scale: smiling (no problem) versus with a straight mouth (impossible). Self-evaluation scores were captured once before and once after the patients were being tested with the FTT and NAT–BT and once after training. The experimenter evaluated the patients after their performance in FTT and NAT–BT. Further, in order to ensure task comprehension, the questionnaire includes a control question asked each time the questionnaire was applied ("Do you have difficulties to jump over a lorry?" taken from Della Sala et al., 2009). It is recommended that one should refrain from interpreting data of patients who failed to answer this control question at least at one timepoint. For analyzing the status of anosognosia, the evaluations of patients after being tested with FTT and NAT–BT were considered.

Training procedure

In the pre-pilot stage, the training procedure and evaluation methods were pretested with four patients suffering from neurological disorders and resulting apraxic symptoms in FTT and NAT–BT. The improved training procedure and evaluation method were applied here.

The study design of baseline assessment, training procedure and post-training assessment for both patients is shown in Figure 2.

Materials, such as a brief training manual (Randerath, Buchmann, & Löser, 2019) and evaluation sheets are made available on https://www.moco.uni-konstanz.de/publikationen/assessments/ or https://kops.uni-konstanz.de/.

Baseline assessment

Patients were presented with 18 naturalistic action tasks, which were neither part of the FTT, the NAT–BT nor the VATA-NAT. Tasks were presented by giving a verbal instruction (e.g., “Please sharpen a pencil.”) and by showing a picture of the final action product simultaneously (e.g., a sharpened pencil and the rest of its shavings). The photos were created using the materials which were presented during the training. The experimenter asked the patients to perform each action by themselves without support. All materials were in one cupboard with drawers, each containing the sorted materials for one task. The patients were requested to select the correct materials for the task and then demonstrate the appropriate tool and object use. The tasks included different number of steps, in order to vary the demands on the correct selection of objects and tools and the sequencing of the single intermediate steps which has been suggested for example by Bienkiewicz, Brandi, Goldenberg, Hughes, and Hermsdörfer (2014), Harrington and Haaland (1992) or Weiss, Rahbari, Hesse, and Fink (2008).

Points were given for the correct selection of materials, for every correct intermediate step towards the final product and for tidying up the material. The patients got no help in accomplishing the steps. To be able to compare the tasks, percentage scores were calculated. To ensure that executing the tasks once does not improve the patients’ performance, baseline assessment was assessed twice on two consecutive days in reversed order of tasks.

For both baseline assessments interrater data were obtained. The agreement was very high (Case 1: baseline 1: $\alpha = .979$, baseline 2: $\alpha = .977$; Case 2: baseline 1: $\alpha = .999$, baseline 2: $\alpha = .990$).

Training

Out of the 18 baseline assessment tasks, training tasks were selected. All tasks, on which the patient scored higher than 90% on average, were excluded from the list of possible training tasks as wells as from the list of untrained tasks evaluated for later comparison. To rank the remaining tasks according to the individual’s performance, average scores of each task across both baseline measures were calculated. Based on the ranking and on daily relevance, every second (Case 2) or third (Case 1) task was trained and the other half/ two-thirds of tasks were not trained. The number of tasks selected for training was based on the speed and capacity of the respective patient. Each training session took 60 min. Since Case 1 was showing a reduced attention and workload capacity as well as avolition (based on observational therapist notes), he was only able to train four tasks, whereas Case 2 was able to be trained with six tasks within the predetermined time of one hour.

To circumvent training order effects, the task which was trained last in the previous session was the first one to be trained in the next session. The training sessions started one day after baseline assessments. Patients were trained in 15 training sessions within 4–7 consecutive weeks. Frequency of training sessions was based on their medical status and available time for training within the schedule of the normal rehabilitation programmme.

The *Naturalistic Action Therapy* is based on an errorless learning principle. Similar to the baseline assessments, patients were asked to perform a naturalistic action including tool selection and application. In contrast to the diagnostic baseline assessment, patients received support when necessary. Using a shaping principle, help was offered in five stages:

1. Specification of action outcome: If the patient was not able to start an action by him-/ herself, the examiner first showed the photo of the final product.
2. Verbal instruction of next step: If the cue with the final product did not help the patient, the examiner explained verbally what to do next.
3. Specification of interim outcomes: If verbal instruction didn't suffice, a photo of the actual interim goal was shown to the patient.
4. Correct movement demonstrated by experimenter: If visual and verbal cues did not succeed, then the participant was asked to imitate the correct movement shown by the experimenter.
5. Movement guided by experimenter: If all these cues did not help the patient in performing the task, the examiner guided the movement of the patients' arm and hand.

If necessary, these supportive measures were provided for each step in the same order including selecting and tidying up the materials of the requested action. For each step per task, five credits could be achieved. For every supportive intervention, the patient lost one credit point. Total performance was again transformed in percentage scores to be able to compare tasks with a differing number of steps.

To train self-evaluation, the patient was asked directly after completion of a task to rate the amount of difficulty he or she had with each intermediate step, as well as with the entire task. Afterwards, the experimenter estimated the patients' performance and gave feedback. Similar to the VATA-NAT, the evaluation was indicated on a 4-point-Likert visual-analogue scale (see above).

Post-training assessment

Within one-week post training, diagnostics were repeated. All 18 naturalistic action tasks were assessed within one session. The apraxia assessments FTT and NAT–BT were also repeated. Furthermore, the VATA-NAT was assessed to evaluate training effects on anosognosia. The evaluation procedure was the same as in screening sessions.

Interrater data were also obtained for post-training assessment and revealed high agreement as well (Case 1: $\alpha = .927$; Case 2: $\alpha = .989$).

Results

Case 1

Baseline assessment

In the screening session the performance of Case 1 was interpreted to be severely apraxic (see also Table 1 and Figure 3). In the VATA-NAT questionnaire, the patient judged his ability to solve such actions to be fine (see also Table 1).

Of the 14 tasks that revealed difficulties during the baseline assessment, four were selected for training: *stick a photo into an album, fill a pill organizing box with pills, prepare a letter to send and make a phone call*. All other tasks were not trained but re-evaluated in the post-training session. Please note, average baseline scores of untrained tasks (without tasks with at least 90% accuracy: $M = 51.35\%$) were higher than of trained tasks ($M = 29.88\%$; see Figure 6).

Performance in tasks selected for training

The performance scores of the patient in the baseline, training and post-training sessions for the trained tasks are shown in Figure 3. Only for two tasks, the patient demonstrated improved behaviour in baseline 2 (*sticking a photo into a. album* and *filling a pill organizing box with pills)*.

During training sessions, the patient was able to further improve in all four tasks. Across training sessions, the patient profited most from verbal, photographical and imitation cues. Compared to baseline sessions his performance improved by 13–23 percentage points post training. However, he remained unable to independently solve the intermediate steps of any task.

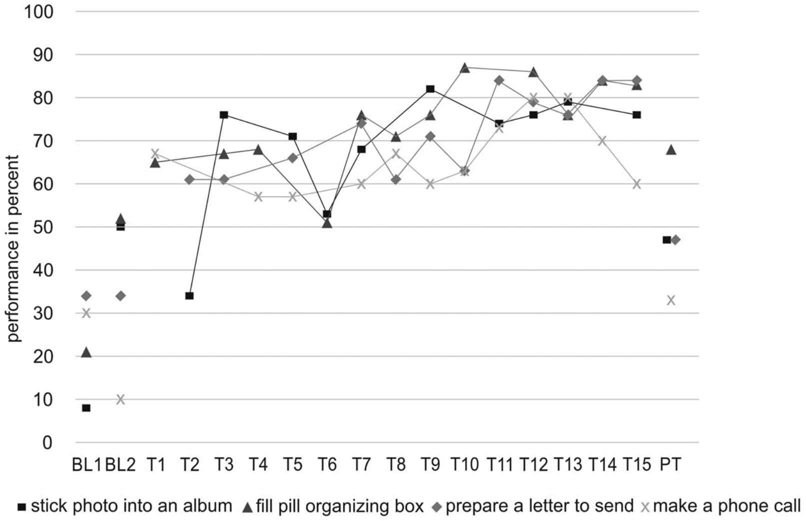

Figure 3. Performance of Case 1 in the trained tasks for baseline sessions (BL1 and BL2), training sessions (T1–T15) and post-training measure (PT). Since the patient was too slow, he was not able to execute all training tasks in all training sessions.

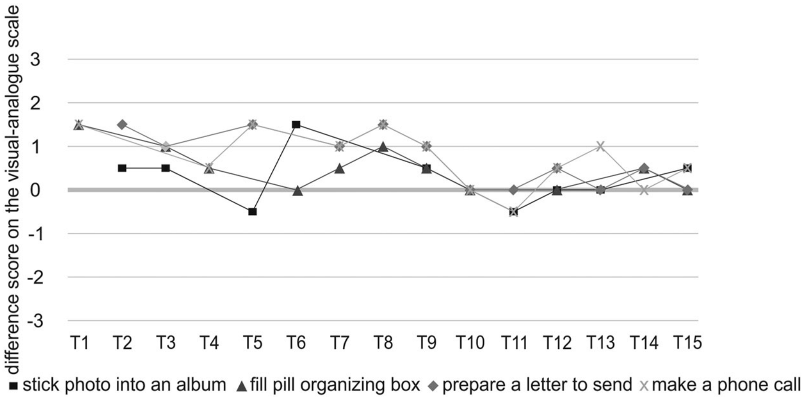

Figure 4. Difference scores (experimenter score – patient score) of Case 1 on the visual-analogue scale for rating his actual performance in the trained tasks.

From the beginning, the patient's self-evaluation was correct in one task (*sticking a photo into an album*), but he needed ten training sessions to adequately evaluate himself in the other tasks (*fill a pill organizing box with pills, prepare a letter to send and make a phone call*). The difference scores of his self-evaluation and the experimenters' rating of performance in the trained tasks are shown in Figure 4.

Post-training assessment

On average the patient improved his performance by 18.88 percentage points for the trained tasks, though even in the post-training assessment the patient failed to execute the trained tasks correctly. The patient also demonstrated improved behaviour in untrained tasks (without tasks with at least 90% accuracy) which on average increased by 11.65 percentage points in the post-training session (see Figure 6). Thus overall, five out of ten untrained tasks in which Case 1 scored less than 90% in the baseline sessions improved after 4 weeks, while performance in the other five untrained tasks stagnated at the same level or even worsened. For the tasks that were performed better than 90% in the baseline assessments, the patient achieved 100% in the post-training session.

In the post-training diagnostics, the patient also improved in standard diagnostic assessments of limb apraxia and related anosognosia. His performance was interpreted as being mildly apraxic. Before training the patient was severely apraxic. Further, his anosognosia improved compared to pre-training (see Table 1 and Figure 6).

Case 2

Baseline assessment

The patient was selected for training because she showed severe apraxic behaviour in both the FTT and NAT–BT in her first as well as the repeated screening session after four weeks (see Table 1). It was not possible to interpret her VATA-NAT baseline scores since she answered wrongly at all three timepoints to the control question of the questionnaire.

In both baseline assessments, she was able to perform five of the 18 naturalistic action tasks without any difficulties. Of the remaining 13 tasks, six were selected for training: *set the table, fill a pill organizing box with pills, prepare a letter to send, make a phone call, take a photo* and *sharpen a pencil*. The baseline average scores of untrained (without tasks with at least 90% accuracy: *M* = 39.71%) and trained tasks (*M* = 37.50%) did not differ (see Figure 6).

Performance in tasks selected for training

Performance in baseline, training and post-training sessions for all trained tasks is shown in Figure 5. The patient showed a stable performance across both baselines in four out of the six trained tasks (*set the table, fill a pill organizing box with pills, make a phone call* and *take a photo)*. The other two tasks (*sharpen a pencil and prepare a letter to send*) were solved better in the second baseline but still were not solved correctly. While we did include the procedure of supported self-evaluation during training, we refrain from reporting and interpreting the anosognosia scores because task comprehension could not be secured (see above).

During training sessions, the patient improved in all six training tasks while reaching the 100%-level in two tasks in the last training session (*sharpen a pencil and fill a pill organizing box with pills*). Further, in one task she learned to interact with the respective tools but without taking social standards into account (*set the table*). Her main problem was to memorize the correct positions

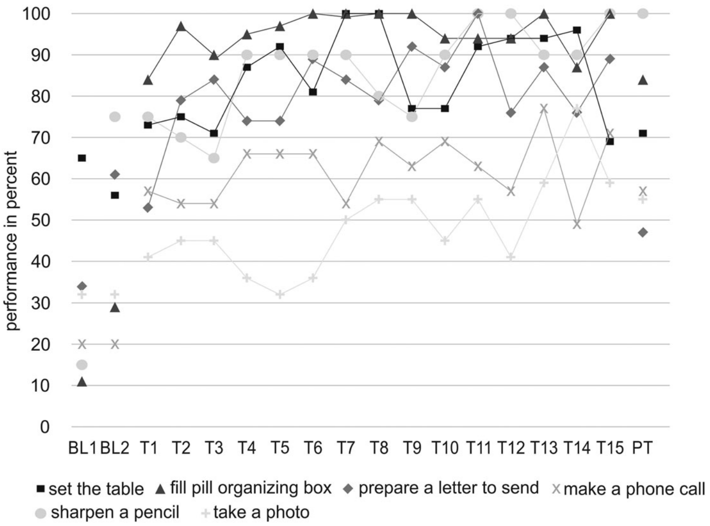

Figure 5. Performance of Case 2 in the trained tasks for baseline sessions (BL1 and BL2), training sessions (T1–T15) and post-training measure (PT).

of tools or objects. This may play an important role when filling a pill box but not when putting knife, spoon and table on the desk. Performance depended strongly on the requested steps. In some steps, the patient profited from the experimenters' help very quickly by correctly using the tool or object after the experimenter had shown its adequate use. For other steps, she was not able to copy the movement even after repeatedly showing her the adequate use. Most difficulties were shown in technical tasks (*make a phone call* and *take a photo*). Here, the patient only achieved a few small steps but was not able to perform functional relevant steps (e.g., dialing the number and look through the camera while taking a photo). Nonetheless, performance improved by 10.5–64 percentage points in trained tasks.

Post-training assessment

On average, the patient improved by 31.50 percentage points in trained tasks in the post-training measurement compared to the baseline sessions. For untrained tasks (without tasks with at least 90% accuracy), she was able to improve her performance by 14.57 percentage points (see Figure 6). Yet, *sharpening a pencil* was the only trained task, she could perform perfectly after training. The five tasks which were already performed on the 100%-level in both baseline sessions remained on this level in post-training measurement.

The data in Table 1 demonstrates that the patient was indeed able to improve her performance in the FTT and NAT–BT in total points in comparison to the

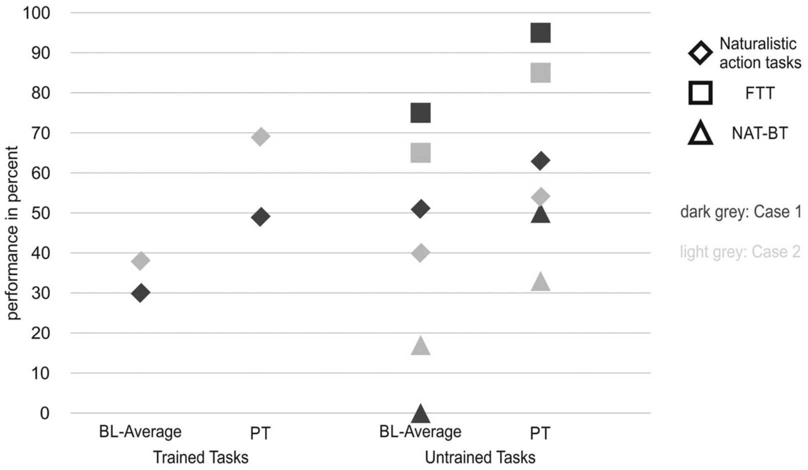

Figure 6. Performance of patients Case 1 (dark grey) and Case 2 (light grey) in percentage scores in trained (left) and untrained (right) tasks. For the naturalistic action tasks the untrained tasks only represent tasks, on which the patients scored lower than 90% across both baselines (BL-Average). These tasks are also displayed for post-training (PT) measure. The Familiar Tools Test (FTT) and Naturalistic Action Test – Breakfast Task (NAT–BT) were performed in apraxia screening session (here displayed in BL-Average) before and post-training. For Case 2, FTT and NAT–BT performance scores of the second screening session are displayed.

screening session that was conducted directly before the training interval. However, despite her improvements, she remained classified as being severely apraxic in the FTT and moderately apraxic in the NAT–BT. As mentioned earlier, results of the VATA-NAT questionnaire could not be interpreted due to the patient's repeatedly inadequate answer to the check question.

Control group

The control group was assessed with the FTT, NAT–BT and VATA-NAT twice with a four-week gap. The four-week gap included standard rehabilitation treatment only. Within-subject comparisons of initial and post-assessment scores revealed that short term spontaneous improvements for these patients could not be significantly established ($Z \leq -0.365$, $p \geq .250$).

Discussion

To the best of our knowledge, the here introduced *Naturalistic Action Therapy* is the first training intervention for apraxic patients that was designed to combine therapy for apraxic and anosognosic deficits. Based on the recommendations of Buxbaum et al. (2008) the training consisted of a variety of ADL tasks with the option to apply shaping to every single task by making the selection of tools more difficult and introducing different stages of supportive cues. By use of the support options for each task, the therapy approach implemented the principle of errorless learning. The training included explicit descriptions of defined tasks with distinct intermediate goals. Emphasis was placed on the detailed description of methods as well as on the standardization of procedure and evaluation in order to facilitate replication and further evaluation of the training. Furthermore, standard diagnostics were used by applying the FTT, NAT–BT and VATA-NAT in order to allow for a reliable and valid assessment of the generalization of training effects.

Although, of course, the presented pilot-data of only two single cases needs to be interpreted with caution, the results give rise to optimism that the training intervention may be a suitable approach for rehabilitation of limb apraxia affecting real tool-use and related anosognosia.

Both studied cases represented typical patients in a subacute post-stroke phase in a german rehabilitation clinic. In only 15 training sessions both cases were able to increase their performance from baseline sessions to post-training sessions in all tasks. While trained tasks seemed to profit the most from training, untrained tasks were solved better as well in post-training session. Furthermore, improvements were also shown in standardized limb apraxia assessments, which may also suggest generalization of training effects. Moreover, also the anosognosia training showed some effects when Case 1 needed to reevaluate his performances in other ADL tasks after training. He improved in recognizing the errors he made in the trained tasks and in part he was able to recognize difficulties in unrelated tasks.

This is in line with other general or holistic training approaches showing some effects on untrained tasks (Geusgens et al., 2007; van Heugten et al., 1998). Clear knowledge of the underlying mechanisms of functional recovery in cognitive neurorehabilitation is still being unravelled (Berlucchi, 2011; Cicerone et al., 2005; De Luca, Calabrò, & Bramanti, 2018; Harvey, 2009). There may be several explanations for why rather holistic training approaches combining high frequent practice, shaping and errorless learning principles with feedback strategies to improve awareness may be effective and even contribute to generalized improvements.

The holistic approach may offer several sources for a careful stimulation of specific functions (e.g., retrieval of object knowledge, visuo-spatial interaction), but also of more general mechanisms (e.g., attentional and working memory processes). Congruent with the cognitive praxis model described by Buxbaum and Randerath (2018) the proposed action working memory system, for example, may be stimulated by the repeated use of it in training via different routes needed for interacting with objects. The shaping strategy and its several support measures may reduce frustration and a capacity overload may be avoided.

In addition, spontaneous recovery may serve as an explanation for the positive effects. Although such an explanation cannot be excluded and may at least hold in parts, we doubt that the improvements can be entirely interpreted in the light of spontaneous remission. The strongest effects of spontaneous recovery are most likely to occur in earlier stages of the disease (Kollen, Van de Port, Lindemann, Twisk, & Kwakkel, 2005). Both patients started the *Naturalistic Action Therapy* greater than three months post-stroke onset (14–15 weeks after stroke onset). Further, the five tested control patients did not show significant improvements in FTT and NAT–BT after a 4 week waiting period. These time intervals were planned in accordance with the predicted minimum length of stay (4–6 weeks). Thus, patients were pre-tested directly after entering the rehabilitation centre (or after being transferred to the respective rehabilitation ward). Comparisons of screening measures before and after training were locked to the training start (screening 2) and end (post-training assessment). It needs to be critically noted that the training period of Case 1 was extended to 7 weeks due to the patient's low capacities and high rehabilitation needs. Last, before participating in the training Case 2 was pre-tested two times with a four-week interval between the diagnostic measures and did not show significant change between the two pretests.

Interestingly, results demonstrated that tasks involving technological devices like the phone or camera were the most difficult to handle for both patients and revealed the lowest training effects. One explanation may be the reduced affording character of these devices compared to classic physical tools, which means that perceiving the object's properties does only provide a low level of prompting for a certain action. This idea is supported by the observation that other tasks, which appear rather low-afforded (e.g., *packing a bag* or *sticking a photo into an album*) were generally performed poorly by both patients compared to tasks that appear rather high-afforded (e.g., *sharpening a pencil* or *pour a cup of water*). This is in line with previous results of Randerath, Goldenberg, Spijkers, Li, and Hermsdörfer (2011) or Barde, Buxbaum, and Moll (2007) who showed that higher affordances facilitate actions and recognition. Another explanation for the differences in performance between tasks could be the frequency of occurrence in daily life and the complexity of tasks (Bienkiewicz et al., 2014; Harrington & Haaland, 1992; Weiss et al., 2008). Interestingly, Case 2 was not able to dial a number with the lab phone but was observed to frequently use her own phone. When asking her about this discrepancy, she claimed that she mostly received calls and therefore just needed to press the green button to pick up.

Object recognition was supported by showing the final-product photos with exactly the same material that the patient was supposed to solve the tasks with. Either patient never had difficulties with selecting the correct material.

Regarding anosognosia therapy, Case 1 showed a satisfactory effect after training. While the patient evaluated his performance quite adequately within the training sessions, in the post-training assessment using the VATA-NAT only little improvement was seen. He correctly evaluated his performance from the 10th training session on in the trained tasks and also showed a better insight into his difficulties with other activities of daily life. But his answers on the VATA-NAT questionnaire were still classified as anosognosic. This is in line with other cognitive behavioural trainings for anosognosia of hemiplegia that were able to improve the patients' insight into the specific activity performed at this moment but not when at rest (Ownsworth, Fleming, Desbois, Strong, & Kuipers, 2006).

One explanation for why Case 2 was able to understand task instructions for practical tasks but not for the VATA-NAT questionnaire is that she may not have understood the concept of scales and thus was unable to answer on the visual-analogue scale. This argument is strenghtened by the observation, that she was not able to recognize numbers when attempting to make a phone call.

Although the results of the present study are promising, the interpretability is limited as there were only two patients tested. Future studies need to apply controlled designs with larger samples. For example, the 18 tasks should be diagnosed in a non-training control group of apraxic patients. Further, patients in a chronic stage outside a regular rehabilitation setting should be considered as a target group. Also, larger group studies will be able to elaborate on more specific aspects such as disentangling the effects on object selection and tool-use production abilities. Additionally, follow-up measures should be implemented in order to check for variability as well as longterm effects, e.g., a second post-training session to measure performance of the 18 tasks should be implemented because the patients' performance may vary from day to day. Future studies should consider evaluating the efficacy of the training in the patients' private environment, and when conducted by relatives or caregivers. Further, potentially increased effects due to a prolongation or intensification of the training may have to be evaluated.

Unfortunately, the anosognosia assessment could only be interpreted for the patient who understood the instructions. Despite explaining the task in each training it remains possible that some aphasic patients (like Case 2) will have difficulties comprehending the instructions for self-evaluation by use of the visual-analogue scale – or may indeed believe they can jump over a lorry (although this then may rather be a problem rooted in delusions of grandeur).

Another approach to enhance the awareness for own deficits would be to show the patient immediately after training the video of their performance and to stop the video after each interim goal and ask the patients to rate their difficulties. Like Besharati, Kopelman, Avesani, Moro, and Fotopoulou (2015) have shown in their study about anosognosia of hemiplegia, such video analyses helped to improve the patients' awareness for their deficits in motor functions. However, the clear drawback is that this procedure is time-consuming and may require specific data protection measures and therefore may be too elaborate to be implemented into the clinical setting.

Last, for future therapy studies, modern technologies like transcranial direct-current stimulation (tDCS) could also be a useful addition that may enhance training effects. For example, Bolognini et al. (2015) showed that tDCS stimulation helped patients in planning imitation movements. Further, new cognitive assistive technologies like COACH (Mihailidis, Boger, Craig, & Hoey, 2008) or CogWatch (Hermsdörfer et al., 2013) have been proven effective for memory aid. Combined approaches may increase the effects of ADL trainings.

However, with the here introduced *Naturalistic Action Therapy*, an important step is done towards testing a method that facilitates conducting a standardized training of daily life activities as well as related anosognosia. A tremendous advantage of this training procedure is that it is easy to administer. It may even be suitable for being applied by healthy relatives of the patients in their home setting and the procedure can be flexibly adapted to other multistep activities of daily life, which may be of particular interest for the individual patient. This option would address previous suggestions, that apraxia therapy should be adaptable to the individual patient's abilities and needs in daily life (Goldenberg et al., 2001).

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