The treatment of apraxia of speech

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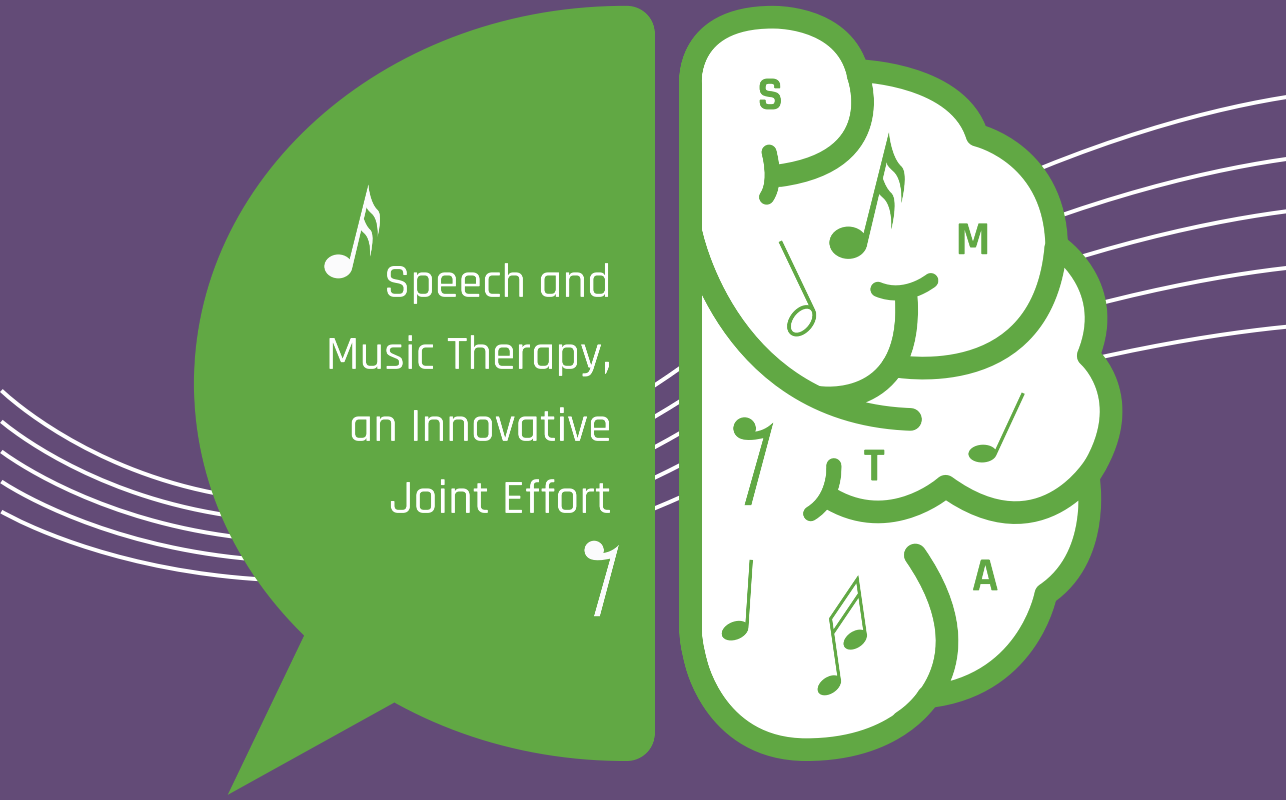
Chapter 7

Prognostic factors of recovery after SMTA treatment in non-fluent aphasia and Apraxia of Speech



7.1 | Language and speech recovery

Recent research of speech recovery after therapy using musical elements focusses on lyric type in relation to brain regions. Stahl, Henseler, Turner, Geyer, and Kotz (2013) propose a two-path model of speech recovery in which formulaic speech (i.e., common phrases, such as "How are you?") has been differentiated from propositional speech. Figure 7.1 represents Stahl et al.'s (2013) two-path model. In this model, cortico-stratial areas of the right hemisphere support features of singing (e.g., Özdemir, Norton, & Schlaug, 2006) and processing of formulaic speech production (e.g., Sidtis, Canterucci, & Katsnelson, 2009). Conversely, left perilesional regions are involved with propositional speech in standard speech therapy (e.g., Saur, Lange, Baumgaertner, Schraknepper, Willmes, Rijntjes, & Weiller 2006; Meinzer, Flaisch, Breitenstein, Wienbruch, Elbert, & Rockstroh, 2008). Therefore, Van Lancker-Sidtis (2004) suggests that formulaic and propositional speech may be lateralised differently in the brain. Stahl et al. (2013) studied lyric type in patients with non-fluent aphasia (i.e., Broca's aphasia and global aphasia). Propositional speech was used in standard therapy and formulaic speech was applied to singing and rhythmic therapy. The results showed that patients who received standard therapy improved their production of propositional speech, in contrast to patients who received singing and rhythmic therapy and improved in formulaic speech. Stahl et al. (2013) found that only patients who received standard therapy showed generalisation to the production of unknown phrases. Therefore, they concluded that their results were in line with the suggested neural pathways of propositional and formulaic speech (Van Lancker Sidtis, 2004) and proposed the two-path model of speech recovery.

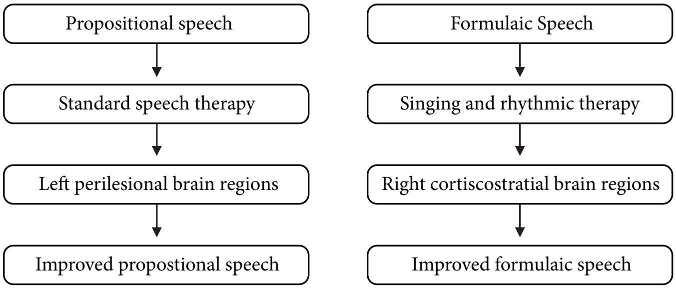


Figure 7.1 The two-path model of speech recovery (Stahl et al., 2013)

SMTA uses formulaic speech (such as, "Good morning") as well as propositional speech (such as, names of relatives). Furthermore, in Stahl et al.'s (2013) model, singing and rhythmic therapy are related to formulaic speech. SMTA also uses the musical parameters melody and rhythm. However, these musical parameters are used both in formulaic speech and in propositional speech. Therefore, it is unknown how various aspects of SMTA fit in Stahl et al.'s (2013) model.

Apart from lyric type and brain regions, little is known about prognostic factors influencing speech recovery after therapy in which musical elements are used. Therefore, the current study focuses on prognostic factors after SMTA treatment and speech-language therapy (SLT). First, general therapy related factors from motor learning related to AoS treatment and central issues in aphasia treatment are discussed in the following paragraph.

7.2 | Prognostic factors of speech recovery

Principles of motor learning give shape to various aspects of AoS treatment, as discussed in Chapter 3. Maas et al. (2008) reviewed the literature in which principles of motor learning were extended to the treatment of speech motor control. They found no evidence for practice amount (i.e., number of sessions) or practice distribution (i.e., treatment duration). However, limited evidence was found for practice variability (i.e., constant and variable practice), practice schedule (i.e., random and blocked practice) and target complexity (i.e., simple versus complex target items).

Central issues in efficacy studies of aphasia treatment are intensity and time post-onset at the start of therapy. In their review of treatment intensity, Bhogal, Teasell and Speechley (2003) showed that with intense therapy, (i.e., 8.8 hours per week across 11.2 weeks) treatment effects were significant, while no positive effects were found in more protracted interventions (i.e., averaging two hours/week across 22.9 weeks). Also, Sage, Snell and Lambon Ralph (2011) compared the outcome of intensive (i.e., every day for two weeks) and non-intensive naming therapy (i.e., twice a week for five weeks). Naming improved in both types of therapy; however, one month after therapy, accuracy was better for the items learned non-intensively than for those learned intensively. Regarding time post-onset, Robey (1998) found that treatment, started within three months of symptom onset, was more effective than no treatment. Finally, many clinicians assume that patients make less progress in therapy after the first year, but Moss and Nicholas (2006) demonstrated in their review of single-subject studies that even patients receiving treatment many years post onset responded positively. The authors found no correlation between time of post onset and magnitude of change with treatment in chronic patients (i.e., less than twelve months post onset).

Apart from intensity and the time post-onset of therapy, severity of the aphasia must be considered in the recovery process as well, because of its direct impact on outcome (Cherney & Robey, 2008). There is general agreement that the better the initial performance on aphasia tests is, the better the recovery will be (Basso, 1992). Still, De Riesthal and Wertz (2004) reported that the relationship between initial severity and total change on their measures was uncertain. Pederson, Jorgensen, Nakayama, Raaschou, and Olsen (1995) found a longer recovery process in patients with severe aphasia than in patients with a moderate or mild aphasia.

It can be concluded that the information about prognostic factors until now is inconclusive. Furthermore, it remains unknown whether various prognostic factors as discussed above, are applicable to treatments using musical elements in non-fluent aphasia and AoS. Therefore, a retrospective study on patients treated with SMTA in parallel with regular speech language-therapy (SLT) was performed.

The current study focused on prognostic factors influencing recovery after therapy for patients with non-fluent aphasia and AoS. First, it was assessed whether patients improved after therapy on language tests. The Dutch Aachen Aphasia Test (AAT; Graetz, De Bleser, & Willmes, 1992) was used as the general aphasia measure. The Amsterdam-Nijmegen Everyday Language Test (ANELT; Blomert, Koster, & Kean, 1995) was used to measure verbal communication. Next, associations were studied between the AAT and ANELT scores and selected prognostic factors of recovery.

7.3 | Methods

7.3.1 | Subjects

The datasets of 78 patients treated with SMTA were available for this study. The data of 37 patients were excluded because there were no post-treatment scores, leaving 41 datasets for analysis. Apart from the data pre- and post-treatment on AAT, 10 of these 41 patients had also completed the ANELT before and after therapy. Characteristics of the 41 included patients are shown in Table 7.1.

Table 7.1 Patient characteristics (n = 41)

Predictors	Sub-category	n (%)	Predictors	mean (SD)
Gender	Male	24 (59)	Age (years)	56.32 (12.02)
	Female	17 (41)		
			MPO	2.33 (1.47)
Handedness	Right	35 (85)	Duration (weeks)	30.56 (16.91)
	Left	6 (15)		
			# sessions SMTA	35.95 (20.19)
Education	High	9 (30)	# sessions SLT	111.83 (52)
	Intermediate	9 (30)		
	Low	12 (40)		
Aetiology	ICVA left	36 (88)		
	Other	5 (12)		
Lesion location	MCA	33 (87)		
	Other	5 (13)		
Diagnosis	Aphasia	7 (17)		
	Aphasia + AoS	34 (83)		
Syndrome	Global	16 (39)		
	Broca	19 (46)		
	Other	6 (15)		
Type	Nonspeaking	16 (39)		
	Nonfluent speaking	25 (61)		
Severity of aphasia	Severe	24 (59)		
	Moderate/mild	17 (41)		
Cognitive functioning	Impaired	26 (63)		
	Normal	15 (37)		

ICVA = ischaemic cerebrovascular accident, MCA = medial cerebral artery, MPO = months post onset, AoS = apraxia of speech, n= number, SD = standard deviation, SMTA = speech-music therapy for aphasia, SLT = speech language therapy, # = number

Patients were suffering from non-fluent aphasia (39% global aphasia and 46% Broca's aphasia, as indicated by the AAT) and in 83% AoS accompanied the aphasia. The speech disorder was in 88% of the subjects caused by a left ischaemic CVA. The most common lesion location was the middle cerebral artery region (i.e., 87%). Apart from the speech disorder, 63% had impaired cognitive functioning. Almost 60% of the participants were male, 85% were right-handed and education-level was equally distributed over the subjects across high, intermediate and low education. All participants received SMTA and SLT.

All participants had been treated at the rehabilitation centre "Revalidatie Friesland" with a rehabilitation programme of on average six hours per week, consisting of SMTA, individual SLT and group treatment aimed at improving their writing performance and general communication skills.

7.3.2 | Primary outcome measures

For this study, the scores on the Dutch version of the Aachen Aphasia Test (AAT; Graetz et al., 1992) and the Amsterdam Nijmegen Everyday Language Test (ANELT; Blomert, et al., 1995) were selected as outcome measures.

The *AAT* is a comprehensive, psychometrically valid and reliable language test-battery. It provides an evaluation of spontaneous speech, naming, repetition, comprehension, reading and writing. For the purpose of this study, only subtests that required speech production were included, since SMTA is targeted at speech production. Comprehension, reading and writing were thus excluded from the analysis. The AAT allows a psychometric single-case analysis of each subtest for each patient (Code, Torney, Gildea-Howardine, & Willmes, 2010). The overall severity of the language impairment is tested with the AAT Token Test.

The *ANELT* assesses functional verbal communication. Verbal responses to 10 everyday scenarios are scored on two 5-point scales for *comprehensibility* and *intelligibility*. An example of a scenario is: “Your neighbour’s dog barks all day long. You are really fed up with it and want to discuss this situation with your neighbour. What do you say to him?” The ANELT has adequate ecological and construct validity, adequate inter-rater reliability and test-retest reliability (Blomert et al., 1995).

The AAT and ANELT were administered by the patient’s speech therapist before and after treatment (no longer than 4 weeks post treatment).

7.3.3 | Prognostic factors of speech recovery

In order to identify variables that were most likely to explain recovery from aphasia and AoS, eleven potential prognostic factors were selected:

(1) *aetiology* (left ischemic CVA and other; e.g., traumatic brain injury), (2) *lesion location* (Middle Cerebral Artery and other; e.g., posterior artery), (3) *diagnosis* (aphasia and aphasia + AoS), *syndrome* (global, Broca and other), (4) *type* (non-speaking and non-fluently speaking), (6) *severity* of aphasia (severe and moderate/mild as defined by the AAT Token Test scores), (7) *cognitive functioning* (impaired or normal as defined by psychological report), (8) *months post onset* (MPO), (9) *duration* (length of treatment in weeks), (10) *number of sessions SMTA*, and (11) *number of sessions SLT* (individual and group sessions; communication and writing).

Each factor needed to be adequately distributed in order to be included in the analyses. Therefore, a cut-off criterion was established and the level was set at 25% (i.e., 10 patients). This means that each subcategory of a factor needed to be reflected in at least 10 patients. For example, *gender* was adequately distributed; 24 males and 17 females. However, *handedness* was not adequately distributed; 35 right-handed and 6 left-handed. If this condition was not met, the factor was not included in the analyses.

7.3.4 | Procedure

Inclusion criteria were: participation in at least five SMTA sessions and administration of the pre- and post-treatment assessments (AAT and ANTAT). The medical files of 78 patients having received SMTA between 2001 and 2010 were reviewed to see whether they met the inclusion criteria. The data of eligible patients (i.e., with AAT and ANELT scores before and after therapy) were assessed using the factors described above.

7.3.5 | Statistical analysis

Paired t-tests were used to compare the AAT and ANELT pre- and post-treatment scores. The dependent outcome variables were the pre-to-post differences in the scales' subtest scores. Apart from the paired t-tests, clinically relevant changes were analysed using the norms of the test manuals (Graetz et al., 1992; Blomert et al., 1995).

To analyse associations between the outcome variables and the various prognostic factors, Spearman or Pearson correlation coefficients were used depending on the type of data. A multiple backward stepwise regression analysis was used for those outcome variables that were influenced significantly by more than one independent variable. The level of statistical significance was set at p <0.05.

7.4 | Results

Table 7.2 provides the means and standard deviations of the overall scores on the different AAT and ANELT subtests. The results revealed significant changes on all AAT measures and the comprehensibility measure of the ANELT. No significant change was found on the intelligibility measure of the ANELT.

Table 7.2 Scores on the AAT and ANELT for the patients included in the study

Subtest (maximum score)	Pre-treatment mean score (SD)	Post-treatment mean score (SD)	t	*p*
AAT (n = 41)				
SP COM (5)	1.29 (1.12)	2.05 (1.34)	-5.83	0.001
SP ART (5)	2.02 (1.63)	3.02 (1.69)	-4.13	0.001
SP AUT (5)	1.95 (1.80)	3.02 (1.73)	-4.46	0.001
SP SEM (5)	1.80 (1.50)	2.56 (1.76)	-4.36	0.001
SP PHON (5)	1.54 (1.38)	2.54 (1.64)	-4.58	0.001
SP SYN (5)	1.12 (1.33)	1.93 (1.52)	-3.99	0.01
Token Test (50) *	39.83 (11.67)	29.85 (15.62)	6.54	0.001
Repetition (150)	59.05 (42.66)	90.68 (37.58)	-6.58	0.001
Naming (120)	28.78 (34.06)	59.73 (40.68)	-7.29	0.001
ANELT (n = 10)				
Comprehensibility (50)	24.50 (8.30)	33.60 (11.01)	-3.48	0.007
Intelligibility (50)	36.80 (9.87)	36.90 (10.51)	-0.03	0.97

SP = spontaneous speech, COM = communication, ART = articulation and prosody, AUT = automatic speech, SEM = semantic structure, PHON = phonological structure, SYN = syntactic structure, *= number of *in*correct responses, t=t-value of the t-test, *p*=level of statistical significance

Table 7.3 provides an overview of the proportion of patients with clinically relevant changes on the AAT and ANELT scores according to manual instructions (Graetz et al., 1992; Blomert et al., 1994). Significant positive change on the AAT spontaneous speech measures ranged between 19% and 34%. All other subtests of the AAT (i.e., Token Test, repetition and naming) revealed significant positive changes ranging between 51% and 73%. For the ANELT, 50% of the patients significantly improved on the comprehensibility measure and 33% on the intelligibility measure.

Table 7.3 **Clinically relevant pre-to-post changes (in percentages) on the primary outcome measures**

Outcome measures (critical significance in raw scores)	No change	Negative change	Non-significant positive change	significant positive change
AAT (n=41)				
SP COM (2)	46.3	0	34.1	19.5
SP ART (2)	41.5	7.3	22.0	29.3
SP AUT (2)	34.1	7.3	24.4	34.1
SP SEM (2)	48.8	4.9	22.0	24.4
SP PHON (2)	39.0	4.9	26.8	29.3
SP SYN (2)	48.8	2.4	24.4	24.4
Token Test (8)	12.2	2.4	34.1	51.2
Repetition (15)	0	2.4	26.8	70.7
Naming (17)	12.2	0	14.6	73.2
ANELT (n=10)				
Comprehensibility (7)	10.0	0.0	40.0	50.0
Intelligibility (5)	0.0	33.3	33.3	33.3

SP = spontaneous speech, COM = communication, ART = articulation and prosody, AUT = automatic speech, SEM = semantic structure, PHON = phonological structure, SYN = syntactic structure, TT = Token Test in incorrect scores

Three factors did not meet the 25% condition (i.e., the cut-off criterion for inclusion of at least 10 patients in each sub-category) and were accordingly excluded from the regression analysis: aetiology, lesion location and diagnosis. The remaining eight factors were entered into the analysis and Table 7.4 shows the positive and negative associations between these predictors and the measurable changes in the AAT and ANELT scores.

Since the “other” subcategory of the factor ‘syndrome’ was too small for the Kruskal-Wallis test ($N = 6$), only 35 datasets were entered for the two subcategories, “global” and “Broca”, into the analysis. The number of months post-onset of the disease was unknown for six patients, also leaving 35 datasets.

The factors MPO, cognitive functioning and number of sessions SMTA were not significantly associated with recovery as assessed with the AAT and ANELT. Table 7.4 lists the nine significance values for the factors syndrome, type, severity, duration and number of sessions SLT for the different outcome variables.

Table 7.4 **Significance values of the prognostic factors for the AAT and ANELT subtest scores**

Factors (n = 41)	AAT SP COM	AAT SP ART	AAT SP AUT	AAT SP SEM	AAT SP PHON	AAT SP SYN	AAT TT	AAT REP	AAT NAM	ANELT COMP	ANELT INTEL
Syndrome (n=35)	0.15	0.36	0.80	0.09	0.14	0.50	0.06	0.79	**0.01**	0.69	**0.01**
Type	0.05	0.17	0.83	0.10	**0.01**	0.07	0.09	0.94	**0.01**	0.71	**0.01**
Severity	0.33	0.76	0.77	0.19	**0.02**	0.20	0.65	0.95	0.46	0.54	**0.02**
Cognitive functioning	0.73	0.29	0.82	0.33	0.95	0.68	0.18	0.49	0.78	0.12	0.67
Duration (weeks)	0.92	0.20	0.74	0.27	0.58	0.52	**0.01**	0.09	0.10	0.24	0.89
MPO (n=35)	0.36	0.94	0.65	0.94	0.84	0.43	0.43	0.86	0.95	0.64	0.79
Number of sessions SMTA	0.61	0.23	0.15	0.86	0.65	0.08	0.06	0.25	0.28	0.50	0.33
Number of sessions SLT	0.26	0.23	0.21	0.37	0.15	0.99	**0.01**	0.21	0.30	0.56	0.40

AAT: SP = spontaneous speech, COM = communication, ART = articulation and prosody, AUT = automatic speech, SEM = semantic structure, PHON = phonological structure, SYN = syntactic structure, TT = Token Tes in false scores, REP = repetition, WR = written language, NAM = naming, ANELT: COMP = comprehensibility INTEL = intelligibility, MPO = Months Post Onset, SLT = Speech Language Therapy, bold type = significant ($p<0.05$)

Four outcome measures revealed significant correlations with multiple factors: (1) the phonological structure of spontaneous speech, (2) the AAT Token Test, (3) the AAT naming test, and (4) the ANELT intelligibility measure. Two associations remained significant in the multiple regression analysis. The first significant association was between the AAT Token Test score and duration ($t = 4.42$, $p<0.05$): the longer the therapy period lasted, the more likely it was that the scores on the Token Test decreased. The second significant association concerned the scores on the AAT naming test with aphasia type ($t = 2.60$, $p<0.05$): non-fluent speakers showed most improvement on naming. The regression models of the significant and non-significant correlations are included in appendix A.3 - A.6.

7.5 | Discussion

The main research question of the current study was whether patients improved after therapy on language tests and which prognostic factors influence recovery after SLT and SMTA therapy in patients with non-fluent aphasia and AoS. The outcomes of the AAT and ANELT were studied in patients treated for non-fluent aphasia and AoS, in relation to factors that best predict the outcome.

Evaluating the pre- and post-treatment scores on the primary outcome measures, significant improvement was found on speech production (i.e., AAT spontaneous speech, repetition and naming) and the comprehensibility of verbal communication (ANELT); this means that patients improved after therapy on language tests.

Contrary to our expectations, no improvement was found in the intelligibility of verbal communication as measured with the ANELT. The most plausible explanation is a lack of generalisation from test measures to functional measures since significant improvement was found for various phonemic aspects (e.g., repetition) on the AAT that were not reflected in the functional communication (i.e., intelligibility) measure of the ANELT. Furthermore, only ten patients were evaluated with ANELT, and, therefore, the results of the ANELT are based on a small group of patients.

As to the clinical implications of the findings, there was an obvious difference between the statistically significant (i.e., group) and the clinically significant (i.e., individual) changes as described in the test manuals. For the group, changes on the AAT's spontaneous speech variables were all significant, whereas at the individual level significant positive changes ranged between 19% and 34% of the patients. For all other AAT subtests, the changes in the raw group scores were clinically significant, with individual positive changes ranging between 51% and 73%.

Apparently, repetition and naming improve, but generalisation to spontaneous speech is not consistently achieved.

To identify prognostic factors, eleven factors were related to the patients' AAT and ANELT outcome scores. Four outcome measures revealed significant correlations with multiple factors and two associations remained significant in the multiple regression analysis. First, the regression model showed a significant association between *duration* and the AAT's Token Test, which is considered a severity outcome measure. The longer the therapy period, the more likely it was that the scores on the Token Test decreased as the Token Test counts the numbers of errors; this means that the severity of aphasia decreased more with longer treatment duration. These results are in line with the findings of Pederson et al. (1995), who suggested that in severe-aphasia language recovery is a long and slow process. The patients tested post-treatment had, on average, received 30 weeks of therapy, which is quite long for this type of treatment. These findings are not in line with the findings of Bhogal et al. (2003) who concluded that effects are strongest with intense treatment (eight hours/week within eleven weeks). The duration data of the current study are in line with Sage et al. (2011) who observed a superior effect in non-intensive learning. Finally, these findings do not correspond with the results of Maas et al. (2008) who found no effect on practice distribution (i.e., duration) and practice amount (i.e., number of sessions).

The regression analysis also yielded a significant association between aphasia *type* (non-speaking versus non-fluently speaking) and the AAT naming test. Non-fluent speakers showed most improvement on naming. The model, thus, suggests that non-fluent aphasic speakers improve more than non-speaking aphasic patients.

It was concluded that subjects in this study (i.e., patients treated with SMTA and SLT) form a homogeneous group. From the 41 patients evaluated, more than 80% were right-handed, had a lesion in the left hemisphere in the area of the middle cerebral artery and were diagnosed with aphasia, AoS and impaired cognitive functions. However, no outcome measures related to AoS were included. Therefore, to study the effectiveness of SMTA in a prospective study a sensitive test to measure improvement on accuracy, consistency and fluency of articulation is needed and must be added to the general language test (AAT) and the verbal communication measure (ANELT). A general articulation test is available with the Diagnostic Instrument for Apraxia of Speech (DIAS), recently developed by Feiken and Jonkers (2012). However, a test that measures improvement on speech motor programming for weekly testing in a case-series design with multiple measurements, was missing. The development of such an instrument will be described in Chapter 8.

7.6 | Conclusion

In this retrospective study patients received SMTA in parallel with SLT. The results revealed significant changes on all spontaneous-speech measures, repetition and naming of the AAT. Also, the comprehensibility measure of the ANELT improved significantly. No significant change was found on the intelligibility measure of the ANELT.

Duration and aphasia type were identified as prognostic factors of speech recovery after SMTA and SLT. Duration was associated with severity of aphasia: the longer the therapy period, the more likely it was that the scores on the Token Test decreased. Finally, aphasia type was associated with naming: non-fluent speakers showed most improvement on naming.