Diagnosing Apraxia of Speech on the Basis of Eight Distinctive Signs

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Diagnostiquer l'apraxie de la parole en se basant sur huit signes distinctifs

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Abstract

This paper reports the results of a study on the use of a fixed number of specific signs to differentially diagnose Apraxia of Speech (AoS) from aphasia or dysarthria. This was done with a diagnostic instrument for AoS that was developed in the Netherlands in 2012, the Diagnostic Instrument for Apraxia of Speech (DIAS; Feiken & Jonkers, 2012). There were 8 signs identified as specific to AoS, namely: inconsistency of errors, number of errors with consonants versus vowels, difference between sequencing and alternating diadochokinesis, groping, initiation problems, syllable segmentation, cluster segmentation, and articulatory complexity. The DIAS was administered to 30 individuals with AoS, 10 individuals with aphasia, 10 individuals with dysarthria, and 35 control individuals. Results showed that a differential diagnosis could be made in 88% of the cases using a minimum of 3 out of 8 specific signs of AoS as criteria. With the exception of 2 patients with aphasia, no other group exhibited the presence of 3 or more signs of AoS. It was concluded that the presence of 3 signs is sufficient to differentially diagnose AoS from aphasia and dysarthria, despite the fact that there is a large amount of variability in the presence of signs of AoS itself in the different individuals.

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Abrégé

Cet article présente les résultats d'une étude investiguant l'utilisation d'un nombre spécifique de signes pour distinguer l'apraxie de la parole de l'aphasie ou de la dysarthrie dans un processus de diagnostic différentiel. Pour ce faire, un test d'évaluation de l'apraxie de la parole ayant été développé aux Pays-Bas en 2012, soit le *Diagnostic Instrument for Apraxia of Speech* (DIAS; Feiken et Jonkers, 2012), a été utilisé. Huit signes ont été identifiés comme étant spécifiques à l'apraxie de la parole : inconstance des erreurs, nombre d'erreurs sur les consonnes versus les voyelles, différence entre les séries diadococinésiques en séquence et en alternance, tâtonnement, problèmes d'initiation, segmentation des syllabes, segmentation des groupes consonantiques et complexité articulatoire. Le *DIAS* a été administré à 30 participants ayant une apraxie de la parole, 10 participants ayant une aphasie, 10 participants ayant une dysarthrie et 35 participants formant un groupe contrôle. Les résultats ont montré qu'un diagnostic différentiel de l'apraxie de la parole peut être effectué dans 88% des cas en utilisant un minimum de trois critères sur huit. Aucun participant inclus dans les autres groupes expérimentaux n'a été identifié avec un minimum de trois signes spécifiques à l'apraxie de la parole, à l'exception de deux participants ayant une aphasie. La présence de trois signes spécifiques a ainsi été jugée suffisant pour distinguer l'apraxie de la parole de l'aphasie ou de la dysarthrie, et ce, malgré le fait qu'il existe une grande variabilité dans les signes observés au sein des individus ayant une apraxie de la parole.

The importance of standardizing the assessment of Apraxia of Speech (AoS) has been repeatedly emphasized in scientific literature (Knollman-Porter, 2008; Wambaugh, 2006; West, Hesketh, Vail, & Bowen, 2008; World Health Organization, 2005). AoS is generally defined as an impairment in programming the positioning of speech organs and the sequencing of articulations (Darley, 1968; Ziegler, 2008). There is, however, no consensus on how to diagnostically differentiate AoS from related communication disorders such as aphasia and dysarthria (Ziegler, Aichert, & Staiger, 2012). Also, there is still a debate in scientific circles regarding which particular signs lead to the diagnosis of AoS (Lowit, Miller, & Kuschmann, 2014; McNeil, Pratt, & Fossett, 2004; Ziegler, 2008).

To diagnose AoS, in the Netherlands, speech-language pathologists (S-LPs) usually administer general language tests or a dysarthria test (Feiken, Hofstede, & Jonkers, 2008; Jonkers, Terband, & Maassen, 2014), or base their diagnosis on clinical judgments. Internationally, there are a few standardized and normed instruments available, like the Apraxia Battery for Adults (ABA-2; Dabul, 2000) and the Motor Speech Examination (MSE; Ogar et al., 2006; Wertz, LaPointe, & Rosenbek, 1984) for English, as well as the *Hierarchische Wortlisten* (Liepold, Ziegler, & Brendel, 2002) for German. There are also criteria lists available to identify AoS, such as the Mayo Clinic Apraxia of Speech Battery (Darley, Aronson, & Brown, 1975; Duffy, 2005; Wertz et al., 1984); the checklist of McNeil, Robin, and Schmidt (2009); and the Academy of Neurologic Communication Disorders and Sciences (ANCDS) list (Knollman-Porter, 2008; Wambaugh, 2006). However, according to Knollman-Porter (2008) and West et al. (2008), there are no instruments or lists that provide reliable identification of AoS.

A recently developed tool, published by Strand, Duffy, Clark, and Josephs (2014), could be valuable in diagnosing (progressive) AoS. Strand et al. presented a rating scale for the diagnosis and description of AoS and tested this in a group of participants with (progressive) AoS or aphasia, reporting high reliability scores as well as good validity of the tool. In the same vein, the current study investigates whether the identification of specific signs is useful for the differential diagnosis of AoS. These signs were measured with a recently developed Dutch diagnostic test, the Diagnostic Instrument for Apraxia of Speech (DIAS; Feiken & Jonkers, 2012). In contrast to the scale tool of Strand et al. (2014), the DIAS could be valuable in differentially diagnosing stroke-induced AoS from dysarthria and aphasia. The diagnosis is based on the presence of eight signs indicative of AoS, which were carefully selected based on theories regarding the characteristics and nature of AoS.

A literature review revealed 33 distinctive signs of AoS, which were categorized by the authors into primary and secondary signs. The primary signs were categorized into three subgroups: initiation errors, incorrectly articulated phonemes, and sequencing errors. Initiation errors include pausing before an utterance, visible or audible struggle to position the articulators (groping), and restarts (Duffy, 2005; Strand et al., 2014). Incorrectly articulated phonemes lead to signs like distortions or substitutions. In distortions, the target phoneme is still recognizable. If it is no longer possible to recognize the target phoneme a substitution occurs, where the change of one or more features leads to the production of another phoneme (den Ouden, 2002). Sequencing errors are exchange errors at the level of sound or syllable (Haynes, Pindzola, & Emerick, 1992; Square, Roy, & Martin, 1997; Ziegler, 2008). The number of initiation errors and distortions seems to be affected by articulatory complexity as well (Staiger & Ziegler, 2008). This is reflected at the phoneme level in a larger number of errors with consonants as compared to vowels (Wertz et al., 1984), and at the word level in a larger number of errors with syllables containing consonant clusters as compared to simple syllables (Staiger & Ziegler, 2008).

The secondary signs are signs that can be assumed to be reactions to the underlying disorder. Speakers with AoS may pause more often (Aichert & Ziegler, 2004; Duffy, 2005) between the consonants of a cluster (cluster segmentation; McNeil, 2002) or the syllables of a word (syllable segmentation; Staiger & Ziegler, 2008), and lengthen vowels (Van der Merwe, 2009). In so doing, individuals with AoS create more time for articulatory motor programming, to lower the number of articulation errors.

The categorized primary and secondary signs were compared to the signs seen in other neurologic speech disorders, like aphasia and dysarthria. Overlapping signs were omitted. Examples of these signs are a word-length effect or the presence of substitutions, which are signs that can be found in both AoS and aphasia (Romani & Galluzzi, 2005; Ziegler, 2005). A sign that is found in both individuals with AoS and dysarthria is slow speech, but also problems with diadochokinesis in general (Duffy, 2005; Ziegler, 2002). However, as alternating diadochokinesis (/pa-ta-ka/) is specifically more difficult for individuals with AoS than sequencing diadochokinesis (/pa-pa-pa/; Ziegler, 2002), this characteristic can be considered as a specific sign of AoS.

The resulting eight signs were considered to be critical signs useful for the differential diagnosis of AoS. The scored signs are: 1) inconsistency in the pronunciation of repeated phonemes, 2) more errors with consonants than with vowels, 3) more difficulty in alternating diadochokinetic rate (/pa-ta-ka/) than sequencing diadochokenitic rate (/pa-pa-pa/), 4) visual or audible groping, 5) initiation problems (restarts), 6) syllable segmentation, 7) segmentation of consonant combinations, and 8) effect of articulatory complexity. These signs are assessed using the DIAS, which is described in the Methods section.

In this study, it will first be investigated whether the eight signs of AoS can be scored reliably. The diagnosis of AoS will be based on the presence of a number of these signs. For the differential diagnosis of AoS with aphasia and dysarthria not all signs need to be present, as the same underlying deficit may lead to different primary and secondary signs. How many signs need to be present in order to come to a differential diagnosis will be investigated. The outcomes of a study with 50 brain-damaged speakers and 35 non-brain-damaged control speakers will be presented.

Methods

Participants

Participants were selected as possibly having AoS by the treating S-LP based on the most recent criteria, i.e., the ANCDS list (Wambaugh, 2006). Another S-LP then independently confirmed this judgment. This S-LP was blinded to the diagnosis of the first S-LP. Both S-LPs were independent in the sense that they were not co-authors of the article. This study only considered those cases where both S-LPs agreed on the clinical diagnosis of AoS.

Thirty participants (15 male, 15 female; mean age 58.4 years, range 34–78) clinically diagnosed with AoS were assessed with the DIAS. To study the potential of differentially diagnosing between patients with AoS, aphasia, and dysarthria on the basis of clinical signs, 20 participants without AoS but with aphasia (n = 10; eight male, two female; mean age 62.7 years, range 45–77) or dysarthria (n = 10; nine male, one female; mean age 55.8 years, range 18–77) were also tested with the DIAS. All participants with AoS suffered from a single stroke. The same holds for eight of the participants with dysarthria and eight of the participants with aphasia. One individual with aphasia and one with dysarthria suffered from a traumatic brain injury. One other individual with dysarthria suffered from a subarachnoid bleed, and for one participant with aphasia the specific etiology was unknown. Aphasia was diagnosed with the standard Dutch diagnostic test, the Aachen Aphasia Test (Graetz, De Bleser, & Willmes, 1992). Only participants having aphasia with phonological deficits, reflected in low scores for repetition and phonological errors in spontaneous speech, were included. Dysarthria was diagnosed with the Dutch *Radboud Dysartrie Onderzoek* [Radboud Dysarthria Investigation] (RDO; Knuijt & de Swart, 2007). The 20 participants without AoS were selected on the basis of their entry in the rehabilitation centre where this study was performed. The first 10 participants with aphasia and dysarthria—irrespective of the type of aphasia or dysarthria—that fit the inclusion criteria were tested. Therefore, this group was less balanced with respect to sex than the group with AoS. A group of 35 control speakers that matched the participants with AoS in age, sex, and education was also tested to determine the cut-off points for the different signs. This group consisted of 14 male and 21 female participants, mean age 52.3 years (range 23–64). A chi-square test revealed no difference between the AoS group and the control group with respect to sex ($\chi(1) = 0.754$, $p > .05$). However, the AoS group turned out to be significantly older than the control group ($t(63) = 2.489$, $p < .05$). Nevertheless, the mean age of both groups was below 60, and in the AoS group only four of the 35 participants were older than 70. Therefore, age is not assumed to be of influence on the outcomes.

All participants gave their informed consent. Testing was done with permission of the Medical Ethics Committee of the University Hospital Groningen (UMCG). All participants were native speakers of Dutch. Participants had a normal intellect (IQ > 70) and vision, and their hearing and neurocognitive abilities did not interfere with an acceptable assessment. All patient group data are presented in Table 1.

All individuals with AoS also suffered from aphasia. In order to determine if the results of this study could be explained by a difference in the severity of aphasia between the group with AoS and aphasia, their scores on the Token Test of the AAT were compared. Originally, the Token Test was developed to be a test for the reception of language, but currently the Token Test is used as a selective instrument to detect the presence of aphasia and as an indicator of its severity (El Hachioui et al., 2013; Orgass & Poeck, 1966). The maximum score on this test is 50, which reflects a negative score. Individuals without aphasia had a mean score of 2.4 (SD = 2.5) on this test (Graetz et al., 1992).

Table 1. Participants by Speech Category

Group	Age in years (Mean and *SD*)	Sex	TPO in months (Mean and *SD*)
Apraxia of speech (*n* = 30)	58.4 (11.6)	15 m, 15 f	32.0 (25.4)
Aphasia (*n* = 10)	62.7 (9.8)	8 m, 2 f	29.7 (53.9)
Dysarthria (*n* = 10)	55.8 (16.3)	9 m, 1 f	10.5 (4.4)
Control speakers (*n* = 35)	52.3 (11.3)	14 m, 21 f	-

Note. m = male, f = female, TPO = time post onset

Materials

All individuals were tested with the DIAS (Feiken & Jonkers, 2012). The DIAS contains four tests, of which three were used in this study.[1] The test for orofacial apraxia will not be discussed here, as it is only part of the instrument to diagnose orofacial apraxia. Three tests were administered to assess the presence of the eight aforementioned signs: articulation of phonemes, diadochokinesis, and articulation of words. All items can be found in Appendix A. In Table 2, an overview is given of the three tasks that were used for differential diagnosis, mentioning the different signs that were studied. Not all signs were investigated in every subtest, but the three subtests were indicated for specific signs. Below, the tests are described including descriptions of the specific signs per test.

In the test for the articulation of phonemes, participants are instructed to repeat vowels and consonants three times consecutively. This test evaluates the conscious production of individual consonants. In AoS, inconsistent distortions and substitutions of phonemes often occur (Sign 1; Darley et al., 1975; den Ouden, 2002; Varley & Whiteside, 2001;Wertz et al., 1984). Inconsistent errors in this study are assumed to be different pronunciations during the repetition of three phonemes. Wambaugh (2006) states that errors of speakers with AoS are consistent. However, this is a different kind of consistency, as it refers to the consistency of error types across different tests.

With respect to the number of errors made with consonants or vowels (Sign 2), more errors with

Table 2. Subtests of the DIAS

Test	Differential diagnostic criteria	Control score mean (*SD*)	Cut-off score
Articulation of phonemes (15 consonants; 15 vowels)	- Inconsistency of errors (1) - Number of errors with consonants vs. vowels (2)	0.09 (0.51) 0.09 (0.74)	2 2
Diadochokinesis (6 series of sequencing and alternating syllables or words)	- Difference between sequencing and alternating diadochokinesis (3) - Groping (4)	0.94 (0.11) 0	0.74 2*
Articulation of words (6 blocks of 11 words)	- Initiation problems (5) - Syllable segmentation (6) - Cluster segmentation (7) - Articulatory complexity (8)	0.003 (0.02) 0 0 0.10 (0.39)	1 out of 11 blocks > 0 > 0 0.88

Note. *Groping was not seen in the control group, thus every occurrence could be considered deviant. However, as clinicians questioned this symptom during the pilot phase on certain occasions, the cut-off was set to 2.

[1]It is not intended to provide an elaborate description of the subtests and the theoretical background of the DIAS. Feiken and Jonkers (2012) and Jonkers et al. (2014) provide more information on construct and item validity, specificity, and sensitivity of the test.

consonants than with vowels are expected (Duffy, 2005; Wertz et al., 1984).

The test for the articulation of phonemes consists of 30 items: 15 consonants (C) and 15 vowels (V). This composition allows one to assess whether there is a difference in the number of errors between consonants and vowels. Consonants differed in place or manner of articulation. Vowels were chosen on the basis of their position in the vowel triangle (Kooij & van Oostendorp, 2003). Place of articulation of the consonants was varied to circumvent perseveration. After, for example, the consonant /m/, an alveolar sound like /d/ followed. The internal consistency of this test is .96 (Cronbach's alpha). To account for a possible effect of consistency, participants were asked to repeat every phoneme three times in a row.

The second test in the DIAS that plays a role in differential diagnosis is a diadochokinesis task. Oral diadochokinesis is seen as a sensitive measure for neuromotoric speech capacities (Ziegler, 2002), as it demands maximum performance of a participant. Deger and Ziegler (2002), Ogar et al. (2006), and Wertz et al. (1984) note that individuals with AoS will have more difficulties in alternating different syllables (alternating diadochokinesis) than repeating the same syllables (sequential diadochokinesis), which is defined as Sign 3.[2] Initiation problems, substitutions, omissions, slow speech rate, segmentation of syllables or clusters, and repeated attempts to produce an item are possible consequences of difficulties with alternating diadochokinesis. In accordance with Duffy (2005), the diadochokinesis test was also specifically used to observe the symptom of groping (Sign 4).

The diadochokinesis test contains 12 items: six sequencing and six alternating items. This subtest is set up according to the level of complexity, starting with simple CV structures, like the sequencing item /pa-pa-pa/ versus the alternating item /pa-ta-ka/, and ending with CCVCC structures, like /stank-stank-stank/ versus /stank-blank-drank/. In some of the alternating items the consonant in initial or final position changes, whereas in others the consonants within a cluster change. Most of the words used in these structures were meaningful words. The words were controlled for frequency of occurrence using the CELEX frequency list for Dutch (Baayen, Piepenbrock, & Gulikers, 1995). The sequential items always had the lowest frequency, to prevent any poor performance on the alternate version of the item, which could be explained by a word frequency effect. The internal consistency of this test is .97 (Cronbach's alpha).

With the test for the articulation of words, the presence of the final four signs of AoS is studied, among which are initiation problems (Sign 5). Problems with the initiation of speech are often seen in individuals with AoS (Haynes et al., 1992; LaPointe, 1990). They can appear in different forms. LaPointe (1990) describes false starts and repetition of sounds or syllables as instances of initiation problems. As mentioned in the introduction, as a reaction to articulation problems speakers with AoS may also pause more often, leading to cluster segmentation (McNeil, 2002; Sign 6) or syllable segmentation (Staiger & Ziegler, 2008; Sign 7). Finally, individuals with AoS make more repetition errors with consonant clusters (Staiger & Ziegler, 2008) and with longer words (Ziegler, 2005), and this is reflected in the *articulatory complexity* sign (Sign 8).

The test for the articulation of words (word repetition) contains 66 items with increasing length and articulatory complexity. The test consists of 11 blocks of six words, where every block differed in complexity, with respect to the number of syllables, number of phonemes and articulatory complexity (CV structures, CC clusters within a syllable, CCC clusters within a syllable, and CC clusters at the syllable boundary). Every block of six items focused on a specific structure. The words in the test do not differ with respect to word frequency. The internal consistency of this test is .99 (Cronbach's alpha). Kuschmann, Miller, and Lowit (2014) provide requirements for intelligibility tests used in speakers with AoS, considering, among others, adequacy, completeness, levels of difficulty, number of items, and frequency of items. The list of items in this test fits with the requirements mentioned here.

Procedure

All tests were administered in one session in a fixed order. All assessments were videotaped and scored later. The administration of the subtests was multimodal, meaning that the items were presented both visually and auditorily to circumvent influences of visual or auditory problems. Participant and tester sat face-to-face in a quiet room. To prevent lip reading, the participant was asked not to look at the tester during the assessment. In cases where this was not possible, the mouth of the tester was covered. Testing (including the test for orofacial apraxia) lasted about 45 minutes. After instruction, all subtests

[2]There is some confusion in the literature about what should be seen as sequential diadochokinesis and what should be seen as alternating diadochokinesis. Duffy (2005), for example, uses the terms with the inverse meaning. However, there is agreement on the fact that the repetition of different syllables, like /pa-ta-ka/, is more difficult for individuals with AoS than the repetition of the same syllable (/pa-pa-pa/).

started with two examples. In the case of an inadequate response to (one of the) examples, the participants were corrected. During assessment no help or feedback was provided, except for one repetition of the target if the participant requested it. There was no time pressure to answer, except for in the diadochokinesis test. In this test participants were first asked to repeat every sequence of three syllables once, and if this was possible, they were asked to repeat every sequence as often and correctly as possible within 8 seconds. The tester told the participant when to start and stop.

Scoring

For each test, the presence of the specific signs was evaluated. Cut-off points for the presence of signs were determined based on scores of the control speakers. A symptom was considered to be present if a score differed more than two standard deviations from the mean score of the control speakers (adjusted upwards if necessary). These cut-off points are presented in Table 2. In Appendix B, how the specific signs were scored per test is described. Scoring and interpreting of the errors could be done in 45 minutes.

The number of signs was counted for every participant and it was evaluated whether it was possible to distinguish individuals with AoS from individuals with dysarthria or aphasia based on the number of signs present. Severity is not considered in the current study. This means that the presence of a sign is important but the frequency with which a sign is noted is not.

Reliability

Intra-rater reliability was obtained by comparing the scoring of sign presence on the basis of video recordings of the DIAS of 30 participants twice, with an intermediate period of six months, by the same experienced clinical linguist. Inter-rater reliability was based on the scores of three experienced S-LPs not involved in the intra-rater reliability, who scored the video recordings of the DIAS administration independently. Test-retest reliability was obtained by testing 10 participants with the DIAS twice, with an intermediate period between two and six weeks. Again, video recordings were scored.

Results

Reliability

Intra-rater and inter-rater reliability correlations (intra-class correlations (ICC) or Kappa scores) for scoring the eight signs were significant and showed overall good reliability. In Table 3, the ICC values and Kappa scores for all reliability measures are presented. The lowest inter-rater agreement was seen for *cluster segmentation*, although this agreement is still acceptable. There was a strong agreement for *more errors with consonants than with vowels*. All other intra-class correlations showed very high agreement. The Kappa values for groping indicated good to excellent agreement. With respect to the ICC values for the intra-rater reliability, all correlations were significant at the level of .001 and indicated a very high agreement. Not all correlations were significant for the test-retest reliability. A non-significant and poor agreement was found for *articulatory complexity*. Ratings for the other signs were again significant, and agreement varied from good (groping) or strong (cluster segmentation) to very high (all other signs).

Number of signs

In order to find out what the necessary number of signs would be for the diagnosis of AoS, the number of signs in the three groups was calculated and afterwards it was decided what the ideal number needed for a reliable diagnosis would be. In comparing the presence of signs in participants with AoS with those noted in individuals with dysarthria and aphasia, it was found that the presence of at least three signs was needed to diagnose AoS in most of the individuals with AoS. In 26 of the 30 individuals with AoS, three or more signs were determined. Three of the four individuals with fewer signs were individuals with very severe speech problems. In these individuals only the first two subtests could be administered, and therefore most of the signs could not be determined. Only in one case a participant was able to do all the subtests and still had fewer than three signs. Three individuals with AoS, however, showed only three signs, which means that when using four signs as diagnostic criteria, a smaller number of individuals with AoS would be diagnosed properly.

In the group of individuals with dysarthria (n = 10), none of the individuals had three or more signs of AoS. In the aphasia group (n = 10), two individuals had three signs of AoS, while the other eight individuals showed fewer signs. Seven individuals not assumed to have AoS showed two signs, which would lead to a larger number of misdiagnoses if these were to be used as diagnostic criteria. This means that the presence of three signs was the best way to divide the groups into individuals with and without AoS.

In Table 4, an overview is provided with the number of individuals in the AoS group that displayed a specific sign. Every sign was found in almost half of the speakers

Table 3. Reliability Measures for the Different Signs

	Inter-rater reliability	Intra-rater reliability	Test-retest reliability
Inconsistent realization of phonemes	.84 ($p < .01$)	.98 ($p < .001$)	.93 ($p < .001$)
More errors with consonants than with vowels	.76 ($p < .01$)	.95 ($p < .001$)	.81 ($p < .05$)
More problems with alternating than with sequencing syllables	.92 ($p < .001$)	.98 ($p < .001$)	.98 ($p < .001$)
Initiation problems	.81 ($p < .001$)	.95 ($p < .001$)	.92 ($p < .001$)
Syllable segmentation	.81 ($p < .001$)	.98 ($p < .001$)	.99 ($p < .001$)
Cluster segmentation	.62 ($p < .001$)	.90 ($p < .001$)	.73 ($p < .05$)
Articulatory complexity	.80 ($p < .001$)	.95 ($p < .001$)	.32 ($p > .05$)
Groping	Kappa		
Rater 1-2	.73 ($p < .05$)	.86 ($p < .001$)	.74 ($p < .05$)
Rater 1-3	.73 ($p < .05$)		
Rater 2-3	1.00 ($p < .001$)		

Note. All comparisons: intra-class reliability, except for the *groping* sign, for which Kappa-scores were used.

Table 4. Number of Individuals With AoS Showing Specific Symptoms of AoS

Signs	Individuals with AoS Symptoms ($n = 30$)
Inconsistency of errors	17/30
Number of errors with consonants vs. vowels	13/30
Difference between sequencing and alternating diadochokinesis	18/30
Groping	23/30
Initiation problems	28/30
Syllable segmentation	25/30
Cluster segmentation	14/30
Articulatory complexity	18/30

with AoS. The sign *more errors with consonants than with vowels* was found in the lowest number of speakers with AoS. Only 13 of the 30 speakers showed this sign. In almost all speakers with AoS (28/30), initiation errors occurred. No specific pattern was seen with respect to the number of primary or secondary signs.

Severity of aphasia

The mean Token Test score of the individuals with AoS was 24.9 (*SD* = 13.5) and of the individuals with only aphasia 27.0 (*SD* = 16.6). An unpaired *t*-test did not show a significant difference between these scores (t(38) = 0.4, $p > .05$). This indicates that differences between the groups with respect to the presence of signs do not relate to the severity of the aphasia.

Discussion

The current study investigated whether it is possible to differentially diagnose AoS from dysarthria or aphasia on the basis of the presence of signs of AoS. With the Dutch DIAS (Feiken & Jonkers, 2012), the presence of eight specific signs of AoS was studied in a group of individuals with AoS, dysarthria, and aphasia, as well as a control group. The individuals with AoS were selected on the basis of clinical judgment by an S-LP using the most recent selection criteria for AoS, i.e., the ANCDS list (Wambaugh, 2006) and this judgment was independently confirmed by the judgment of a second blinded S-LP. The individuals with dysarthria and aphasia were diagnosed with the RDO (Knuijt & de Swart, 2007) and the Aachen Aphasia Test (Graetz et al., 1992), respectively.

Haley, Jacks, De Riesthal, Abou-Khalil, and Roth (2012) showed that clinicians are reliably able to list and interpret the signs of AoS, but show poor agreement in differentially diagnosing AoS. This is because clinicians observe and prioritize the signs differently, and consequently reach different conclusions. In this study, we showed that the eight signs can be scored reliably by experienced S-LPs. Both the inter- and intra-rater reliability showed significant and sufficiently high correlations. This also holds for the test-retest variability, except for the *articulatory complexity* sign. Although this sign was found in 18 of the 30 speakers with AoS, it seems that the presence of this sign is not as clear to interpret as the other signs. This might have to do with the fact that the calculation of this sign is more complex than the other signs, although the inter- and intra-rater reliability were good. It could also be that the presence of this sign is subtler to detect than the others, which means that in some cases raters might miss its presence. For future studies it is recommended to detect the presence of the influence of articulatory complexity with a simpler measure.

In the individuals with AoS, the signs were present, but with a large amount of variation. This is consistent with the assumption that the same underlying disorder can manifest itself in different primary or secondary signs. However, the differential diagnosis could be determined with the presence of three of eight signs. In 26 of 30 tested individuals with AoS, three or more signs were present. Three of the four remaining individuals could not be diagnosed properly as they were severely impaired patients who could not complete all the subtests. These individuals were for example unable to do the diadochokinesis test at all, or could only repeat one or two words of the repetition test. Therefore, in these individuals not all signs could be counted. This leads to a restriction on a valid diagnosis on the basis of signs, namely that individuals should be assessed with the entire diagnostic test and that all signs can at least be scored properly. Only one individual with a clinical diagnosis of AoS scored with fewer than three signs. For this individual it is difficult to decide whether he/she was incorrectly diagnosed with AoS by the S-LP or incorrectly diagnosed as not having AoS using the DIAS.

The presence of three of more signs was not seen in any of the ten individuals with dysarthria; however, three signs were present in two of the 10 individuals with aphasia. This result can be interpreted in two different ways. One could conclude that it is not always possible to make a differential diagnosis between aphasia and AoS in some cases. Another possible interpretation is that diagnosing on the basis of the presence of symptoms is preferable to clinical judgment, because of the possibility that these aphasic speakers also suffer from AoS. Control speakers and the individuals in the other patient groups rarely showed these signs. The fact that more signs were present in the group of speakers with AoS than in the group of speakers with aphasia appeared to be unrelated with severity of aphasia, because Token Test scores for both groups were comparable. However, there is some debate about the role of the Token Test as a measure for severity of aphasia. Although authors use the Token Test in such a way (e.g., El Hachioui et al., 2013), the developers of the Token Test originally presented it as an instrument to diagnose language comprehension impairments only (see also De Renzi & Faglioni, 1978). In that case, the only justified conclusion is that the presence of three or more distinctive signs in participants with AoS in the current study seems unrelated to the presence of an aphasic comprehension disorder.

No specific signs seem to favour the diagnosis of AoS. All signs were found regularly in the different individuals, with a minimum of 13 out of 30 speakers with AoS showing the sign of more errors with consonants than with vowels. This reveals that it does not seem to be possible to further restrict the number of symptoms to be present. It is also clear that not all signs are found in all individuals with AoS. Only the symptom of initiation problems was seen in almost all speakers.

As mentioned in the introduction, there is a lively debate on the diagnosis of AoS and on which type of tasks to use for diagnosis. There is a discussion in the literature as to what importance non-speech tasks, such as repetition of phonemes and diadochokinesis, could contribute to the diagnosis of AoS. Ziegler (2003) doubts the role of such tasks due to their unrelatedness to natural speech. It is indeed impossible to diagnose AoS on the sole basis of such tasks, but in line with Kuschmann et al. (2014), it is assumed that these non-speech tasks provide information on the underlying impairment, whereas it is also necessary to focus on real words, as is done in the repetition task, for a closer correlation with natural speech. Both types of tasks, therefore, have merit in the assessment of AoS. The importance of a diadochokinesis test is also reflected in the fact that 18 of the individuals with AoS showed greater problems with alternating diadochokinesis as compared to sequential diadochokinesis.

The discussion on diagnosis partly has to do with the lack of consensus on the exact underlying deficit(s) and the differential diagnosis with respect to aphasia and dysarthria. There seems to be agreement on some of the signs of AoS, but even with respect to these signs there is discussion regarding whether they should really be seen as purely signs of AoS. All of the eight signs that are evaluated in the DIAS were mentioned as signs of AoS in the literature. The assertion that not all eight signs need to be present in all individuals with AoS has been shown in this study and was also confirmed by Strand et al. (2014). Strand and colleagues recently showed that it is possible to reliably score the presence of signs of AoS and to validly diagnose (progressive) AoS on the basis of the presence of these signs, also without the necessity of all signs being present for a group of individuals with (progressive) AoS (Strand et al., 2014).

In line with the findings of Strand et al. (2014), the current study indicates that the discussion about the differential diagnosis with respect to aphasia and dysarthria should not be about finding signs that are present in all AoS patients. When the division of the signs of AoS into primary signs (like initiation errors and distortions) and secondary signs (like segmentation of consonant clusters or intersyllabic pauses) is taken into account, it is likely that individuals differ in how they express AoS. Therefore, different specific signs could lead to the diagnosis of AoS. The present study showed that, nevertheless, only three signs need to be present to result in a valid differential diagnosis between speakers with and without AoS.

This current study is limited by the fact that, although a significant number of individuals with AoS participated, the groups of individuals with dysarthria and aphasia were rather small. Accordingly, no specific distribution was made in the different types of individuals with dysarthria (e.g., ataxic dysarthria or flaccid dysarthria) or aphasia (e.g., conduction aphasia or Wernicke's aphasia). In future studies, the authors intend to account for the type of dysarthria or aphasia by testing a larger number of participants.

A second limitation is the fact that this study was conducted with Dutch participants using a Dutch instrument. It is assumed, however, that the specific symptoms that were considered with this instrument might be considered in other languages as well. The fact that Strand et al. (2014) were able to use signs for the diagnosis of AoS shows that a diagnosis on the basis of the presence of signs does not have to be test-specific.

Finally, the fixed order of the subtests could have influenced the outcomes. Participants might have had more speech problems at the beginning of the administration of the test due to starting problems, or at the end due to, for example, fatigue, which could lead to a bias in the presence of specific symptoms. However, given the fact that no specific sign was the most common in the participants with AoS, it seems unlikely that more symptoms would be shown in the first or final test for the group of participants with AoS.

In this study, it was shown that, by assessing the specific signs of AoS, AoS can be distinguished from aphasia and dysarthria. The possibility of differentially diagnosing AoS from aphasia and dysarthria is important in clinical practice. S-LPs will be able to connect their treatment properly to the actual deficit(s), creating a better basis for treatment. In addition, by knowing which signs are present in a specific patient, better choices can be made in setting priorities for therapy. Administration of the test and scoring of the responses can be done in roughly 90 minutes. S-LPs are able to do the scoring

and interpretation on the basis of the description in the manual. One-day courses are also offered, however, to acquaint S-LPs with the procedures. Haley et al. (2012) already showed that S-LPs often have different opinions on the presence of a sign, but that training on the basis of a systematic protocol clearly reduces these differences.

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Two of the authors were involved in the development of the Diagnostic Instrument for Apraxia of Speech (DIAS; Feiken & Jonkers, 2012).

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Appendix A
Items of the Subtests

Articulation of phonemes

	Consonants	Vowels
1.	/t/	/oo/
2.	/f/	/ee/
3.	/s/	/u/
4.	/h/	/a/
5.	/p/	/ie/
6.	/k/	/o/
7.	/r/	/uu/
8.	/l/	/i/
9.	/j/	/eu/
10.	/b/	/ei/
11.	/n/	/oe/
12.	/g/	/aa/
13.	/m/	/ui/
14.	/d/	/e/
15.	/w/	/ou/

Diadochokinesis

1. Pa pa pa

2. Pa ta ka

3. Mok mok mok

4. Mok sok hok

5. Dam dam dam

6. Dam das dak

7. Schel schel schel

8. Schel stel spel

9. Vlok vlok vlok

10. Vlok stok brok

11. Stank stank stank

12. Stank blank drank

Articulation of words

a. One syllable, not complex

1. sok (sock)

2. web (web)

3. kat (cat)

4. noot (nut)

5. veer (feather)

6. tas (bag)

b. Two syllables, not complex

1. kanon (canon)

2. minuut (minute)

3. banaan (banana)

4. debuut (début)

5. zadel (saddle)

6. gebak (cake)

c. One syllable, CC, 3 phonemes

1. knie (knee)

2. vlo (flea)

3. trui (sweater)

4. sla (salad)

5. prei (leek)

6. twee (two)

d. One syllable, CC, 4 phonemes

1. tand (tooth)

2. wesp (wasp)

3. punt (point)

4. gans (goose)

5. koord (cord)

6. bank (bank)

e. One syllable, CCC, 4 phonemes

1. arts (doctor)

2. sprei (bedspread)

3. angst (fear)

4. stro (straw)

5. oogst (harvest)

6. eerst (first)

f. One syllable, CCC, 5 phonemes

1. spraak (speech)

2. schrik (fright)

3. dorst (thirst)

4. schroef (screw)

5. kunst (art)

6. streep (line)

g. Two syllables, C-C, 5 phonemes

1. oksel (armpit)

2. pasta (pasta)

3. advies (advice)

4. omdat (because)

5. asbak (ashtray)

6. afweer (defense)

h. Three syllables, C-C, 8 phonemes

1. impulsief (impulsive)

2. abnormaal (abnormal)

3. aantasten (affect)

4. verwonden (wound)

5. onwaarheid (untruth)

6. inpalmen (to charm)

i. 4 syllables, not complex, 8 phonemes

1. televisie (television)

2. limonade (lemonade)

3. vitamine (vitamin)

4. politica (politician; fem.)

5. mayonnaise (mayonnaise)

6. apparatuur (apparatus)

j. not complex, 8\9-11 phonemes

1. fotocamera (photo camera)

2. kilometer (kilometre)

3. honorarium (fee)

4. figureren (figure; verb)

5. papegaaien (parrot; verb)

6. telefoneren (telephone; verb)

h. complex, 9-11 phonemes

1. invloedrijk (influential)

2. handtastelijk (palpable)

3. fietstassen (cycle-bags)

4. gras groeit (grass grows)

5. herfstblad (autumnal leaf)

6. eerstejaars (first-year student)

Appendix B
Determination of the Cut-off Scores

The *articulation of phonemes* subtest was used to assess two signs. To detect whether or not participants produce inconsistent realizations of phonemes, the number of inconsistencies within a three-time repetition was calculated (range: 0–30). To detect whether participants produced more errors in consonants than in vowels, the scores for correctly produced consonants and vowels were subtracted from each other (range: 0–15).

The *diadochokinesis* test was used to assess two signs. First, it was evaluated whether participants experience more difficulties in alternating than sequencing syllables and words, and secondly, it was observed whether participants show visible or auditory groping. For the assessment of the first symptom, the number of correct realizations in the repeating sequence (/pa-pa-pa/) was compared to those in the alternating sequence (/pa-ta-ka/). If the participants were able to perform the single repetition, they were asked to produce as many repetitions as possible in eight seconds. The number of correct realizations for the alternating sequence was then divided by the correct realizations for the repeated sequence, where a run of three syllables constituted a sequence. The obtained scores were increased by 1 in order to circumvent nil scores (range: unlimited). A score below 1 indicates a poorer performance on the alternating sequences.

The diadochokinesis test was also used to score the symptom of *groping*. This was done by scoring the presence of this symptom during the repetition of the alternating sequences.

The four remaining signs (initiation problems, syllable segmentation, segmentation of consonant combinations, and effect of articulatory complexity) were captured in the *articulation of words* subtest. Sixty-six words were divided into 11 blocks of increasing complexity. To prevent reliance on one single instance of a symptom, but also to keep scoring time within proportional limits, the presence of the signs was scored per block of six words. Initiation problems were scored in all blocks, so the highest score is the presence of 11 signs in 11 blocks (score 11/11 = 1). The other signs were only observed in a selected group of blocks. Syllable segmentation can only be observed in the polysyllabic words. There were 36 polysyllabic words, used in six blocks, so the highest score is 6/6 (score = 1). Segmentation of consonant clusters can only be observed in words including a consonant cluster. This was the case for 30 words, used in five blocks. The highest score is 5/5 (score = 1).

The effect of articulatory complexity was determined by comparing words of similar length but different articulatory complexity; two blocks contained non-complex words, two blocks contained a two-consonant cluster, and two blocks contained words with a three-consonant cluster. To account for an effect of articulatory complexity, the score of the third block was subtracted from the mean score of the three blocks.