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The apraxia of speech rating scale: A tool for diagnosis and description of apraxia of speech

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1 INTRODUCTION

1.1 Background

The diagnostic classification of apraxia of speech (AOS) was first used by Darley in the late 1960's (Darley, 1968). He had observed that a subset of patients with aphasia following stroke also had difficulty with articulatory movements that could not be explained by phonologic errors or weakness. He suggested the use of this term to discriminate the language impairment associated with aphasia and the movement disorders associated with the dysarthrias from a third disorder which he believed resulted from impairment in planning and programming movement gestures for speech. Debate raged for the next 20 years (and to a small degree continues) as to whether there is indeed a category of impairment separate from either aphasia or dysarthria. Over the last 40 years, a large body of perceptual, acoustic, and physiologic research has demonstrated that there is support for such a category, and has provided copious descriptions of the characteristics associated with the disorder (see reviews in Duffy, 2013; McNeil, Robin, & Schmidt, 2009; McNeil, Doyle, & Wambaugh, 2000; Wertz, LaPointe, & Rosenbek, 1984).

More recently, AOS has been observed in progressive degenerative neurologic disease. In 2006, Duffy provided evidence that it may be the first or only sign of degenerative neurologic disease, and he posited the term primary progressive apraxia of speech (PPAOS)

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(Duffy, 2006). Recent research has further documented and described PPAOS as well as progressive nonverbal oral apraxia occurring in the context of progressive aphasia (Josephs, Duffy, Strand, Whitwell, Layton, & Parisi, 2006; Josephs & Duffy, 2008; Josephs, Duffy, Strand, Machulda, Senjem, Master, Lowe, Jack, & Whitwell, 2012). AOS, however, is less recognized in the neurology community, either as part of the phenotype of some categories of progressive aphasia (i.e. progressive non-fluent aphasia or PNFA), or as the earliest or only manifestation of neurodegenerative disease (PPAOS). This lack of recognition is likely due in part to the lack of biomarkers for apraxia of speech. Therefore, observations of specific speech characteristics remain the primary method for its diagnosis and classification. Although auditory perceptual assessment continues to be the primary method for differentiating among motor speech disorders (Duffy, 2013), we know that perceptual observations are vulnerable to a number of problems (Kent, 1996). One of the ways to reduce errors in perceptual judgments and improve consistency across clinicians, both for description and diagnosis, is to specify and quantify the presence and severity of the specific characteristics that have been accepted as consistent with the diagnostic label (Haley, Jacks, de Riesthal, Abou-Khalil & Roth, 2012).

In order to more specifically quantify the presence and severity of characteristics of AOS for a large research project designed to study individuals with progressive aphasia and/or apraxia of speech, a clinical rating scale, the Apraxia of Speech Rating Scale (ASRS), was developed. This scale was intended to be descriptive (versus diagnostic), primarily for the purposes of the research study rather than for broader clinical use. It became apparent, however, that with some revision the ASRS may be a useful clinical tool for differential diagnosis and estimates of severity, and a pertinent tool for subject description in research. Data regarding the reliability and validity of the scale in its current form will provide critical data informing future revisions.

The ASRS was initially designed to assist the description and quantification of characteristics that have been commonly accepted and reported as indicative of AOS. The rating scale uses a 5-point scale (Table 1), which describes not only the presence or absence of particular speech characteristics, but also their prominence and severity. The 16 items are organized according to whether they: a) are considered to be discriminative of AOS; b) can be apparent in patients with AOS but may also be exhibited by patients with aphasia; c) can be apparent in AOS but may also be seen in patients with dysarthria; or d) can be apparent in AOS but may also be present in aphasia or dysarthria (see Appendix). The ASRS is scored during and/or after listening to the individual's speech during conversational speech, picture description, word and sentence repetition and speech-like AMR and SMR tasks. Scoring procedures are summarized in the methods section.

1.2

The purpose of this report is to describe the 16-item ASRS, a rating scale designed to quantify the presence or absence, relative frequency, and severity of characteristics frequently associated with the diagnosis of AOS. We also report initial intra-judge and inter-judge reliability for the scale. Validity was examined through correlations of total ASRS scores with both the clinical diagnosis and clinical judgements of severity of AOS.

2. METHODS

2.1 Participants

The data reported in this paper were collected in the context of a larger project to study individuals with progressive aphasia and/or progressive AOS. As part of this work, a comprehensive speech-language test battery is administered in addition to a neurologic examination, a neuropsychological test battery, and neuroimaging.

The ASRS was completed for 133 participants who met study criteria for progressive aphasia and/or progressive AOS (Table 2). Participants ranged in age from 42 to 84 years. Methods for initial clinical classification are described first, followed by methods for administration of the ASRS, then methods for examining reliability and validity.

2.2 Apraxia of Speech Clinical Diagnosis

The examining clinician (authors EAS or JRD) made clinical judgements about the presence versus absence of AOS based on the spoken language tasks of the Western Aphasia Battery (WAB) (Kertesz, 2007) (i.e., conversational questions; picture description; repetition; naming; word fluency and sentence completion), as well as conversation, word and sentence repetition, letter and action word fluency tasks, and alternating movement rate (AMR) and sequential motion rate (SMR) tasks. These clinical judgments were made independent of scoring of the ASRS, and only one of the two judges scored and had access to the ASRS ratings. After the entire speech-language protocol was completed, a judgment about the broad clinical diagnosis was made (e.g. progressive aphasia [logopenic, semantic dementia, agrammatic, or fluent]; PPAOS; or progressive AOS with agrammatic aphasia). A five point scale, common to our clinical practice (0= normal – no evidence of AOS; 1= mild AOS; 2= moderate AOS; 3= marked AOS; and 4=severe AOS), was used to judge the presence and severity of AOS (clinical diagnosis), also independent of the ASRS score.

The final clinical diagnosis regarding presence or absence of AOS was confirmed through consensus by the examining clinician and the second speech-language pathologist on the research team (EAS or JRD). The consensus decision involved joint review of video recordings of the patient's speech (conversational speech, picture description, word and sentence repetition, and diadochokinetic tasks). Based on these samples, and without knowledge of the ASRS scores, the second clinician gave an opinion regarding the presence or absence as well as the severity of AOS. The clinician who had tested the patient then reported his/her initial clinical diagnosis, which had also been made prior to scoring the ASRS. When there was disagreement, the clinicians came to consensus via discussion and further review of the recordings. Immediate consensus was reached for all but two of 133 patients, both of whom were ultimately judged by consensus to have very mild AOS.

2.3 ASRS Scoring

After the initial clinical diagnosis of AOS was determined by the examining clinician, but prior to the consensus meeting, the ASRS was scored by the examining clinician. For each item, a score was given based on the 5-point scale (Table 1). A score of 0 was assigned if the characteristic was not noted in any task. The score of 1 was given if the item was observed,

but very infrequently. The score of 2 was given if the item was observed fairly frequently, but not necessarily across all tasks and not on most utterances. A score of 3 was given if the characteristic was pervasive (nearly always evident) but not marked in severity; that is, the feature was mild enough that intelligibility was not significantly affected. A rating of 4 was given if the feature was noted across tasks on most utterances and was severe enough that targets were difficult to recognize. The total number of items noted, as well as the total score over all items, was then calculated.

2.4 Reliability

Ten patients (13% of the sample) were quasi-randomly selected for reliability judgments based on diagnostic classification and AOS severity. The two examining clinicians had each originally assessed five of the ten patients. Four individuals had been classified as having progressive aphasia without AOS(1 PFA; 1 SD; 2 LPA); three were classified as having PPAOS; and three were classified as having with AOS plus agrammatic aphasia. A range of clinical severity was represented. The ASRS had been scored initially immediately following testing, either from notes made during the session and/or from review of video recordings immediately following testing, but only after a clinical judgement of presence or absence and severity of AOS had been made.

To assess intra-judge reliability, the two clinicians (JRD and EAS) then each independently rescored, using video recordings, the ASRS for the five patients they had initially tested. Six months or longer had passed between the original assessment and the rescoring, and no reference was made to the original scores until rescoring was complete.

Interjudge reliability measures were derived from ASRS ratings made by a third clinician (HC) for each of the ten patients. Total scores as well as the item scores for each of the 16 characteristics were used in the analyses.

3. RESULTS

3.1 Reliability

Reliability was computed through intra-class correlations (ICC) (Table 3). The overall inter-judge ICC among the three clinicians was 0.94 for the total score and 0.91 for the number of items present. Inter-judge ICCs ranged from 0.87 to 0.91. Intra-judge ICC measures were also high, ranging from 0.91 to 0.98.

Item level agreement was also examined (Figure 1). For most judgments there were no substantial differences between ratings. For example, for item 1 (distorted substitutions), 15 of the 20 judgments (across both intra-judge and inter-judge) were in perfect agreement; the remaining five judgments were within one point on the 0–4 scale. For item 14, however, 10 of the 20 judgments were in perfect agreement, while 7 were within one point and 3 differed by two points. No items were discrepant by 3 or 4 points. Ninety seven percent of the judgments across the 16 items were in perfect agreement or differed by only 1 point.

Agreement on whether each particular item was present or absent was also examined (Table 4). Five items were judged as being present or absent with at least 90% agreement. Nine

items were judged as being present or absent with 70–89% agreement. Two items yielded less than 70% agreement across all three judges.

3.2 Validity

Participants with AOS had higher total ASRS scores than those without AOS [$t(54.68) = 14.83, p < 0.001$ (Figure 2)]. Specificity, which is the proportion of individuals without the disorder (according to a gold standard which in this case was the independent clinical diagnosis) who are shown to not have the disorder on the test) was calculated at 100% with a cutoff score on the ASRS set at 8 (Figure 3). Sensitivity (the proportion of individuals with the disorder who are shown on the test to have the disorder) ranged from 75% with a cutoff at 14, to 90.5% with a cutoff at 10, and 96% with a cutoff at 8 (Figure 3).

Clinical judgement of AOS severity was strongly correlated with ASRS scores [$r(132) = .88, p < .001$]. As shown on Figure 3, there was overlap on two participants who had mild AOS with those who were judged to have no AOS. There was expected overlap among patients who exhibited mild-to-moderate or moderate-to-marked AOS. There was significant overlap between marked to severe levels; however, the strong correlation shows that the scale adequately represents differences in clinically judged severity.

4. DISCUSSION

The ASRS was originally developed for a large research study of progressive aphasia and progressive AOS, with intent to help quantify the presence, frequency, and prominence or severity of characteristics commonly associated with AOS. Although the original intended purpose of the scale did not focus on its utility for general clinical use, its potential for usefulness in clinical settings became apparent to us during our research, especially given the paucity of clinically valid and reliable measures for quantifying the disorder in general practice. This led us to pursue further development of the measure and this initial report regarding the scale's reliability and validity.

The results presented here show demonstrate good initial validity on the basis of strong correlations with clinical diagnosis, as well as strong correlations of ASRS scores with the clinical judgement of AOS severity. Sensitivity and specificity measures suggest that the ASRS is not likely to identify someone as having AOS if they would not be given that clinical diagnosis.

Reliability was fairly good but needs more extensive examination, especially across a larger number of clinicians who have different levels of experience. The clinicians administering or scoring the ASRS in this study have many years of clinical experience in motor speech disorders and two were involved in the scale's development (which, admittedly may have inflated our measures of reliability). It is likely that reliability would not be as good with clinicians who have less experience or training in the identification of specific AOS speech characteristics. Providing video examples that illustrate each of the characteristics rated on the scale, and the ratings that reflect their prominence/severity, may be very helpful to assuring reliability of ratings among clinicians and researchers.

A number of changes in the ASRS items may improve its broader clinical and research use. Our experience in using it as well as the data from this study suggest that discarding or modifying some items may improve reliability and perhaps validity. Although our examination of item-level agreement indicate that most rating disagreements were only 1 point off, it is quite important that items are reliably measured as present or absent. For example, two items (increased sound distortions or distorted sound substitutions with increased speech rate, and sound or syllable repetitions) had less than 60% agreement on whether the item was present or absent. This may partly reflect the fact that many of our participants with AOS were not able to produce utterances at a faster rate, causing uncertainty about the appropriate rating of this item across clinicians. There was also some uncertainty about the distinction between “sound/syllable repetition” and “false starts and/or groping.” Omitting or more adequately defining some items will likely improve reliability.

Some of the items are not easily scored given the 0–4 scale. For example, “deliberate, slowly sequenced, segmented, &/or distorted (including distorted substitutions) speech SMRs in comparison to speech AMRs” could be rated as present or absent, rather than as more or less pervasive or severe. As a result, there may be some items for which we switch to a binary scoring system (i.e., 0 = not present; 1 = present). There is also potential redundancy across items. For example, positive identification of either “syllable segmentation within word” and “syllable segmentation across words” may result in obligatory identification of “lengthened intersegment durations.” Each of these issues will be addressed as we complete a more detailed item analysis and work to improve reliability without compromising the validity and descriptive value of the scale.

Although the ASRS was designed primarily as a descriptive tool, the validity results show potential for differential diagnosis. At this point we recommend using the cut-off of 8 for the scale’s index of presence versus absence of AOS, recognizing that further analysis and revision of the scale may modify the recommended cut-off score. And, as with any test or rating scale used for diagnostic purposes, its interpretation regarding the diagnosis of AOS must be tempered by clinical judgment independent of the scale, including relevant variables that may not be reflected in the rating scale.”

The ASRS may provide an additional tool for quantifying the presence and severity of AOS during spontaneous conversation, narrative picture description, and repetition tasks. One strength of the ASRS is the grouping of perceptual features according to their relationship to dysarthria and aphasia. For example, if all of the identified features can be associated with disorders other than AOS, the clinician can direct their observations to behaviors that support an alternative diagnosis.

Additional research is planned to develop the scale more completely. Studying the ASRS with individuals diagnosed with AOS due to stroke may further establish its validity and clinical applicability. In addition, we have not yet examined the degree to which the ASRS distinguishes AOS from dysarthria. Some revision to the current version, and direct comparison to a cohort of individuals with dysarthria and no AOS, may further strengthen its potential for broader clinical use.

In summary, this study has shown that the ASRS meets some of the reliability and validity criteria for a rating scale that documents the presence and prevalence of specific AOS characteristics. Further development of the scale will hopefully permit its broader use in clinical practice and as a tool for better subject description in AOS research.

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Appendix

Apraxia of Speech Rating Scale (ASRS- V1)

Name: _____ #: _____ Date: _____

Examiner: _____

Total of Ratings: _____ # of items rated present: _____

Aphasia present Y N Severity (0–4) _____ AOS present Y N Severity (0–4) _____
 Dysarthria present Y N (type= _____); Severity (0–4) _____

1	AOS - primary distinguishing features^a (no overlap with dysarthria or aphasia). One or more must be present for diagnosis of AOS.	Score (0–4)
1.1	Distorted sound substitutions	
1.2	Distorted sound additions (not including intrusive schwa)	
1.3	Increased sound distortions or distorted sound substitutions with increased utterance length or increased syllable/word articulatory complexity	
1.4	Increased sound distortions or distorted sound substitutions with increased speech rate	
1.5	Inaccurate (off-target in place or manner) speech AMR's (alternating motion rates, as in rapid repetition of "puhpuhpuh")	
1.6	Reduced words per breath group relative to maximum vowel duration	
2	Distinguishing features unless dysarthria present^b	Score (0–4)
2.1	Syllable segmentation within words > 1 syllable	
2.2	Syllable segmentation across words in phrases/sentences	
2.3	Sound distortions	
2.4	Slow overall speech rate	
2.5	Lengthened vowel &/or consonant segments	
2.6	Lengthened intersegment durations (between sounds, syllables, words, or phrases; possibly filled, including intrusive schwa)	
3	Distinguishing features unless aphasia present^c	Score (0–4)
3.1	Deliberate, slowly sequenced, segmented, &/or distorted (including distorted substitutions) speech SMRs in comparison to speech AMRs	
3.2	Audible or visible articulatory groping; speech initiation difficulty; false starts/restarts	
4	Distinguishing features unless dysarthria &/or aphasia present^d	Score (0–4)
4.1	Sound or syllable repetitions	
4.2	Sound prolongations (beyond lengthened segments)	

Apraxia of Speech Rating Scale Scoring Instructions

- 0 = not present
- 1 = detectable but infrequent
- 2 = frequent but not pervasive
- 3 = nearly always evident but not marked in severity
- 4 = nearly always evident and marked in severity

^a Present in AOS and not a feature of aphasia or dysarthria.

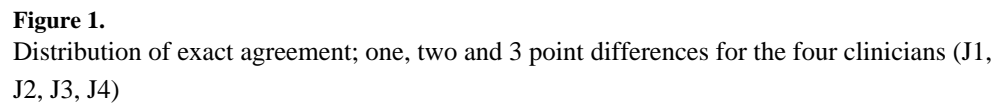
^b Associated with AOS but can also occur in dysarthria. Consider as characteristic of dysarthria if primary distinguishing characteristics of AOS not present, and other features of dysarthria are present. If dysarthria present with AOS, score with the modifier "d" (e.g. 2d).

^c Associated with AOS but can also occur in aphasia. Consider as characteristic of aphasia if primary distinguishing characteristics of AOS not present, and other features of aphasia are present. If aphasia present with AOS, score with the modifier "a" (e.g. 2a).

^d Associated with AOS but can also occur in aphasia and dysarthria. Consider as characteristic of dysarthria or aphasia if primary distinguishing characteristics of AOS not present. If aphasia or dysarthria also present with AOS, score with the modifier “d,” “a,” or “ad” (e.g. 2ad).

Highlights

- The ASRS quantifies the frequency/severity of characteristics associated with AOS
- ASRS correlates with clinical diagnosis and severity of AOS
- Overall intra judge and interjudge reliability is acceptable
- Sensitivity and specificity of the ASRS are acceptable



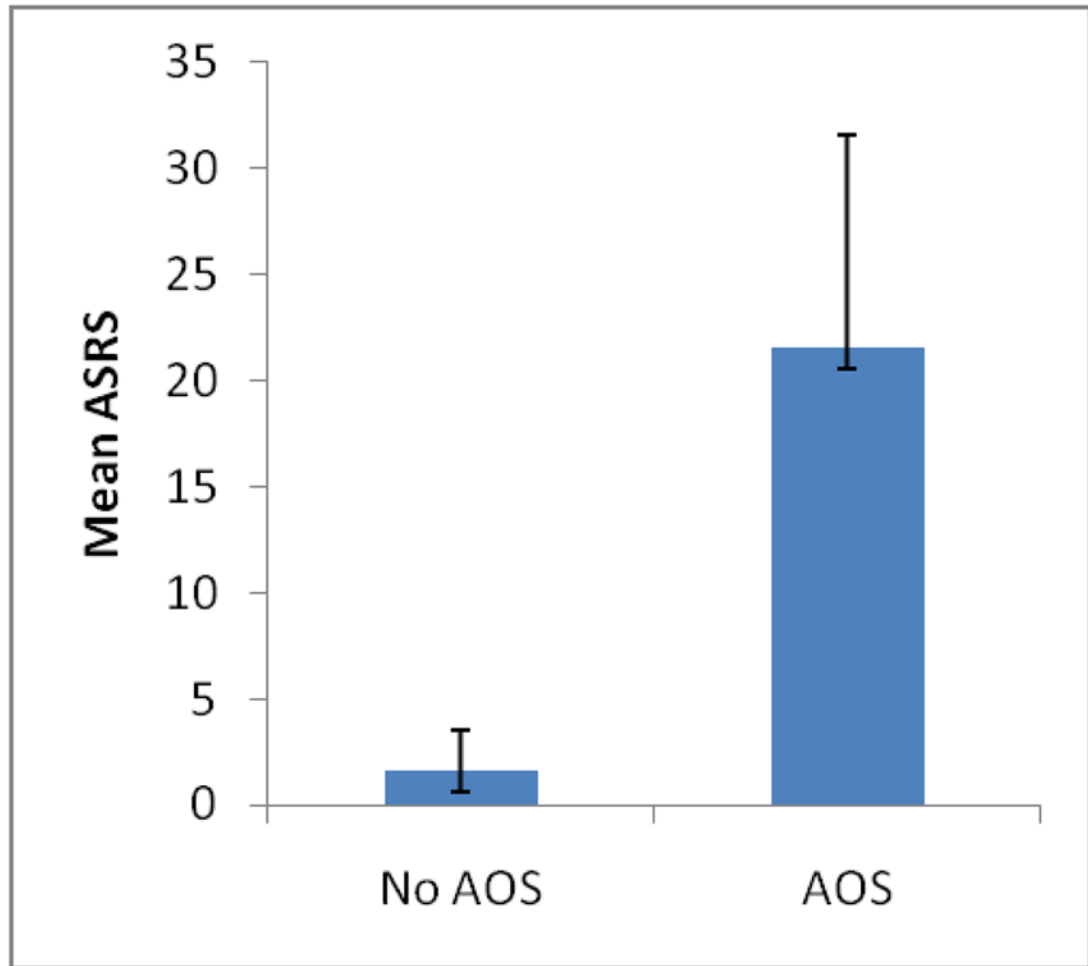


Figure 2.
Total ASRS Scores for Individuals with AOS and those without AOS

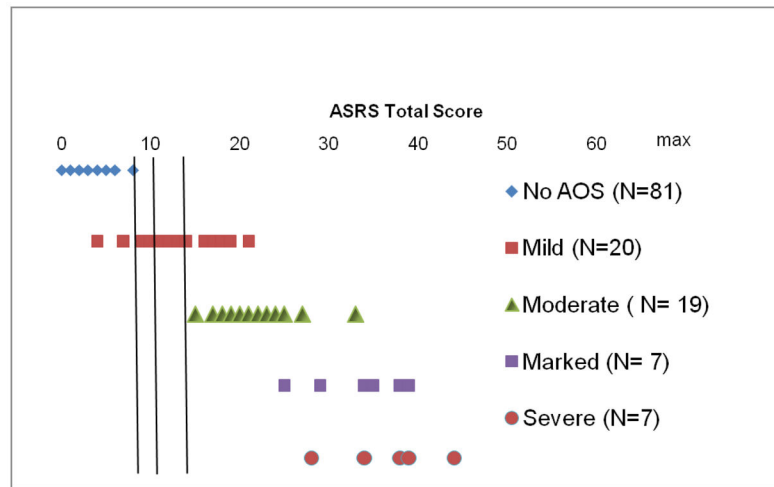


Figure 3.
ASRS scores for each level of clinical judgment of severity

Table 1

Scoring for each of the 16 items on the ASRS

Score	Description
0	not present
1	detectable but infrequent
2	frequent but not pervasive
3	nearly always evident – not severe
4	nearly always evident - severe

Table 2

Participant characteristics

Classification	N	Mean Age (years) (range)	Mean WAB AQ ^I (SD)
Progressive Aphasia			
Logopenic	49	66.2 (47 – 85)	77.1 (16.3)
Semantic Dementia	8	66.0 (57 – 75)	78.9 (18.0)
Primary Agrammatic Aphasia	2	75.5 (73 – 78)	92.6 (3.11)
Primary Fluent Aphasia	10	66.8 (54 – 80)	93.8 (4.0)
Unclassified	9	68.1 (57 – 76)	83.5 (13.0)
Apraxia of Speech + Agrammatic Aphasia	33	68.2 (42 – 83)	81.7 (13.3)
Primary Progressive Apraxia of Speech	23	72.0 (53 – 84)	96.6 (3.2)

^I Western Aphasia Battery Aphasia Quotient

Table 3

Intraclass correlations (ICC) for the ASRS total score and number of items observed to be present.

Reliability Measures	Total Score	Item Count
Overall ICC based on three raters	0.94	0.91
Intra-judge #1	0.98	0.97
Intra-judge #2	0.95	0.91
Inter-judge: #1 vs. #3	0.91	0.87
Inter-judge: #2 vs. #3	0.88	0.91

Table 4

Agreement across all judgments as to the presence or absence of individual items

Agreement	Item
100%	<ul style="list-style-type: none"> Reduced words per breath group relative to maximum vowel duration Lengthened intersegment durations (between sounds, syllables, words, or phrases; possibly filled, including intrusive schwa)
90%	<ul style="list-style-type: none"> Syllable segmentation within words > 1 syllable Sound distortions Lengthened vowel and/or consonant segment
80%	<ul style="list-style-type: none"> Increased sound distortions or distorted sound substitutions with increased utterance length or increased syllable/word articulatory complexity Inaccurate (off-target in place or manner) speech AMRs Syllable segmentation across words in phrases/sentences Deliberate, slowly sequenced, segmented, and/or distorted (including distorted substitutions) speech SMRs in comparison to speech AMRs
70%	<ul style="list-style-type: none"> Distorted sound substitutions Distorted sound additions (not including intrusive schwa) Slow overall speech rate Audible or visible articulatory groping; speech initiation difficulty; false starts/restarts Sound prolongations (beyond the patient's likely lengthened vowel and consonant segments)
40–60%	<ul style="list-style-type: none"> Increased sound distortions or distorted sound substitutions with increased speech rate Sound or syllable repetitions