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THE RELATIONSHIP BETWEEN APRAXIA OF SPEECH AND ORAL APRAXIA: ASSOCIATION OR DISSOCIATION?

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Running Head: Apraxia of speech and oral apraxia

THE RELATIONSHIP BETWEEN APRAXIA OF SPEECH AND ORAL APRAXIA: ASSOCIATION OR DISSOCIATION?

ABSTRACT

Acquired apraxia of speech (AOS) is a motor speech disorder which affects the implementation of articulatory gestures and the fluency and intelligibility of speech. Oral apraxia (OA) is an impairment of non-speech volitional movement. Although many speakers with AOS also display difficulties with volitional non-speech oral movements, the relationship between the two conditions is unclear. This study explored the relationship between speech and volitional non-speech oral movement impairment in a clinical sample of 50 participants with AOS. We examined levels of association and dissociation between speech and OA using a battery of non-speech oromotor, speech, and auditory/aphasia tasks. There was evidence of a moderate positive association between the two impairments across all participants. However, individual profiles revealed patterns of dissociation between the two in a few cases, with evidence of double dissociation of speech and oral apraxic impairment. We discuss the implications of these relationships for models of oral-motor and speech control.

Key words: speech apraxia, oral apraxia, stroke, dissociation, association

Introduction

Apraxia of speech (AOS) is a motor speech disorder where the movement plans which control speech production are impaired or inaccessible. As a consequence, speech output is characterized by a range of features which affect intelligibility. Speech often appears effortful and under conscious control (Lebrun, 1990; Lecours & Lhermitte, 1976), with a corresponding loss of automaticity in speech production. There is often evidence of initiation difficulties, and articulatory groping, which involves preparatory visible and sometimes audible speech movements and gestures. The temporal components of speech can be disrupted and features such as the voice onset time patterns of plosives can be disturbed (e.g., Van der Merwe, 2011); this impacts upon the robust signalling voiced/voiceless contrasts of plosives (Kent & Rosenbek, 1983; Ziegler & von Cramon, 1986a; Varley & Whiteside, 1998; Whiteside, Robson, Windsor & Varley, 2012). Other temporal dimensions of speech are also affected, with output displaying longer inter-syllabic pauses, prolonged segment and syllable durations (e.g., Haley & Overton, 2001) and disrupted prosody (Kent & Rosenbek, 1982, 1983; Whiteside & Varley, 1998a; Varley, Whiteside & Luff, 1999; Bartle-Meyer, Murdoch & Goozee, 2009). Furthermore, the spatiotemporal dimensions of speech are affected and substitutions and distortions of articulatory targets are perceived as a result of misdirected gestures (Bartle-Meyer et al., 2009). In addition, the overlap of articulatory gestures is reduced, resulting in lower levels of coarticulation (Ziegler & von Cramon, 1985, 1986b; Whiteside & Varley, 1998b; Whiteside, Grobler, Windsor & Varley, 2010).

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While the behavioural signs of AOS are well described, there remains considerable debate as to the underlying processing failures that are the source of the surface characteristics (Varley & Whiteside, 2001; Aichert & Ziegler, 2004). This debate stems from different theoretical perspectives regarding the mechanisms of speech control, and in particular with regard to whether all speech control requires segment-by-segment assembly of outputs (Shattuck-Hufnagel, 1979; Crompton, 1981; Keller, 1987; Levelt & Wheeldon, 1994). Controversies surrounding AOS also extend to its relationship with other forms of impairment resulting from left hemisphere (LH) injury such as aphasia and oral apraxia. In this report, we focus on the relationship between AOS and oral apraxia and explore the co-occurrence of speech and oral apraxia in a sample of 50 patients diagnosed with AOS.

AOS is recognized on the basis of spatiotemporal disruptions of speech gestures that impact upon fluency and intelligibility. By contrast, non-speech oral movements may be unimpaired in individuals with AOS (DeRenzi, Pieczuro & Vignolo, 1966; Galluzzi, Bureca, Guariglia & Romani, 2015). Both vegetative functions (respiration, laryngeal and palatal valving, chewing and swallowing) and volitional non-speech oral gestures, such as sticking out the tongue or performing lateral tongue movements to command, may be relatively normal. Oral apraxia (OA) is diagnosed when, despite intact sensory-motor function evident in vegetative use of the respiratory-oral tract, an individual is unable to use these effector systems under voluntary control. OA like AOS and other movement apraxias, typically occurs following LH damage which suggests that crucial movement control systems are lateralised. Typical clinical assessment tasks for OA involve imitating non-speech movements such as blowing, smiling

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or licking the lips (Bizzozero et al., 2000) or performing them to command (Dabul, 2000). A complicating factor in the identification of OA is that, as an impairment of volitional movement, evaluations require that a patient performs oral movements in response to spoken commands (Dabul, 2000). Patients with significant comprehension failure due to co-existing aphasia may fail to understand commands and may be slow to perform movements, or require a model before they can enact the movement. This attracts a scoring penalty in standard clinical evaluations. As a result, an individual might be classed as displaying OA deficits when the source of the impairment lies elsewhere and the presence and degree of OA may be over-determined by standard clinical evaluations. Laboratory investigations of non-speech oral movement employ tasks such as tracking a visual target with an articulator and as such are less prone to interference from extraneous factors such as auditory comprehension (Ballard, Granier & Robin, 2000; Ballard, Robin & Folkins, 2003; Bunton & Weismer, 1994). However, these tasks are not widely available.

Some patients with AOS appear to display a movement disorder that is restricted to speech (e.g., Galluzzi et al., 2015). However, many patients with AOS are identified as also having OA (e.g., Dronkers, 1996; New et al., 2015). The frequent co-occurrence of conditions might indicate that the control mechanisms for oro-motor and speech motor control depend to some degree on shared substrates and resources (Ballard et al., 2003). Instances of dissociation between the two conditions in the direction of speech impairment with retained non-speech oro-motor control (AOS>OA) may be the result of partial damage to an essentially integrated mechanism responsible for both forms of movement. While gross oral movements such as moving the tongue from side-to-side remain

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relatively unimpaired in instances of limited damage, the finer and faster movements of speech, requiring narrowly targeted and tightly integrated gestures, are disrupted. Consistent with the proposal that co-occurrence of AOS and OA stem from greater severity of impairment of motor control, Botha et al. (2014) examined the presence of OA and AOS in patients with progressive apraxia of speech and reported that patients with OA tended to display more severe AOS. Evidence from functional neuroimaging also supports the proposal that neural networks underpinning speech and non-speech behavior overlap to some degree (New et al., 2015).

However, there are also reports of cases of OA in the absence of AOS (e.g., Kwon, Lee, Oh & Koh, 2013). Botha et al., (2014) also report instances of double dissociation between OA and AOS within patients with progressive apraxia of speech. This reverse dissociation (OA>AOS) constrains the extent of commonality or overlap between the control mechanisms for the two forms of movement control, particularly if difficulty in producing oral movements to command could not be attributed to auditory comprehension failure. The evidence of a double dissociation between speech and oro-motor control (Shallice, 1988).

Proposals of independence between components of control systems for speech and non-speech oral movement are consistent with approaches that view complex behaviours as being mediated by multiple assemblies of processing systems. Various sub-systems are recruited to meet the demands of a particular task (Hickok & Poeppel, 2007; Golfinopoulos, Tourville & Guenther, 2010). In the case of speech and non-speech oral motor control, the total assemblies that mediate each form of behaviour are likely to be rather different (Weismer, 2006).

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The movement control system for speech will be closely interconnected with auditory and more general linguistic processing mechanisms. Furthermore, speech movements in adults are entrained actions that have been executed many times. By contrast, a request to produce an action such as moving the tongue alternately between the top and bottom lips is a novel action sequence. Different neural systems are recruited in executing novel as opposed to overlearned movements. In particular, cortico-cerebellar activation is evident in sequence learning, while activation shifts to cortico-striatal regions when reproducing overlearned movements (Hikosaka, Nakamura, Sakai & Nakahara, 2002; Doyon, Penhune & Ungerleider, 2003; Dayan & Cohen, 2011). Nonspeech oral movement is also likely to place greater dependence upon occipitoparietal somatosensory and visuo-spatial systems in targeting movements and determining whether visual targets have been reached. Functional brain imaging studies support the proposal that the neural networks for speech motor control can be differentiated from those employed in non-speech oral movement. For example, while non-speech oral movements elicit a bilateral network of neural activations, speech movements evoke left-lateralized activity, reflecting a close inter-relation with language processing systems in healthy participants (Bonilha, Moser, Rorden, Baylis & Fridriksson, 2006).

In addition to difference in the total neuronal assembly for speech and nonspeech oral movements, there are also differences in the movement parameters of the two forms of action. Differences in force, speed and spatial targeting have been described (Ziegler, 2003; Weismer, 2006; Bunton, 2008), suggesting that even within dedicated motor control mechanisms there is potential for separation between the two forms of movement. However, it is likely that movements of the

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vocal tract, whether they result in speech or non-speech gestures, involve some common units, for example at the level of primary motor cortex. Therefore, the degree of autonomy between speech and oromotor control will be constrained (Ballard et al., 2003) and a partial autonomy model would represent the most plausible characterization of the architecture of speech and non-speech oromotor control.

In terms of patterns of association and dissociation that might occur between AOS and OA, a partial autonomy model is able to account for all possible patterns. This includes the instance of OA>AOS which is more problematic for a model proposing close overlap of substrates of speech and oromotor control. Both models are able to account for instances of association between AOS and OA.

In this report, we evaluate the severity of speech and volitional non-speech oral movement impairment in a large clinical sample of fifty participants with AOS. In addition to exploring relationships at group-level, we conducted individual case profiling to determine if there was evidence of double dissociation of speech and oral apraxic impairment. Given the theoretical importance of cases where OA impairment was disproportionate to degree of AOS, we examined the behavioural profiles of such cases to determine if their difficulty in performing oral movements to command could be attributed to impaired auditory comprehension.

Methods

Study design

Fifty one participants with AOS were recruited to the study. The study was granted ethics approval under the National Health Service Local Research Ethics

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Committee (NHS LREC) procedures. All participants gave their informed consent, but one participant later withdrew from the study. The remaining fifty participants were recruited to an intervention study (Whiteside et al., 2012) and prior to intervention, a series of speech and language assessments were undertaken in order to profile non-speech, speech and language performance. These baseline profiles are the focus of the current paper.

Participants

The participants were 21 females and 29 males with a mean age of 65 years (SD = 15 years; age range = 28 to 91 years). All were at least 5 months post-onset of a cerebrovascular accident (CVA), with a mean time post-onset time of 21 months (SD = 20, range = 5 to 105 months). Brain imaging was not available for all participants. There was attested LH pathology for 38 participants. There was no information regarding lesion location for 11 participants, and one participant, who was right handed, showed signs of right hemisphere pathology. All participants were assessed for handedness by asking them to report their hand preference in opening a jar, brushing teeth, throwing a ball, and writing, and footedness (kicking a ball). All participants except for six were either right, or predominantly right-lateralised for handedness and footedness. Of the six nonright handers, three displayed mixed laterality, and three were predominantly lefthanded. Two speech and language therapists independently identified the participants as having AOS using standard diagnostic criteria such as longer syllable durations, speech errors (sound substitutions and sound (phonetic) distortions), reduced speech fluency, and dysprosody (e.g., Wambaugh, Duffy,

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McNeil, Robin & Rogers, 2006; Haley, Jacks, de Riesthal, Abou-Khalil & Roth, 2012). All participants had some degree of coexisting aphasia.

Behavioural Profiling

The extent of aphasic difficulties, the severity of AOS, and the presence of nonspeech oral impairment were assessed by raters who were blind to the purpose of the analysis. All raters were qualified speech and language therapists who went through a process of consensus training. The details of the assessments are provided below under three headings (*Severity of Oromotor impairment*; *Severity of AOS*; and *Aphasia severity*). All vocal responses were recorded to a Marantz PMD660 Portable Solid State Recorder which was attached to a Beyerdynamic M58 Omnidirectional Dynamic Microphone mounted on a table top microphone stand; the microphone was placed within 0.5 metres of the participant's seated position.

Severity of oromotor impairment

Volitional non-speech movements to command

The non-speech oromotor assessments consisted of three tasks which involved sub-components of the speech production system (laryngeal, lingual and labial). In the laryngeal task, participants were instructed: "Can you make a cough". The lingual task required movement of the tongue from side-to-side. The labial task involved alternate lip spreading and rounding. In all three sub-tasks, if no response was made after 10 seconds, a demonstration was provided.

The scoring of the non-speech oromotor tasks was based on the established system and assessment devised by Dabul (2000), with a score of 5

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for correct responses, and 4.5 if a participant displayed initiation difficulties, followed by an accurate response. A score of 4 indicated a correct response after an initially errorful response. Scores of 3 corresponded to responses which roughly approximated to the target (e.g., reduced speed or amplitude). A correct response after a demonstration gained a score of 2, while a rough approximation after demonstration was awarded a score of 1. A score of 0 was given where responses were incorrect following a demonstration.

Severity of AOS

Repetition of words of increasing numbers of syllables

Speakers with AOS show durational abnormalities with increasing word length (e.g., Haley & Overton, 2001). A word repetition task was designed in which a monosyllabic word subsequently had various affixes attached in order to create two and three-syllable word forms. Importantly, word frequency was closely controlled within these triads. Repetition performance across words of increasing length was used to assess the severity of AOS. Participants were presented with five sets of words of increasing syllable length (1, 2 and 3 syllables). All three words in each series were low frequency items (The British National Corpus, 2001). Participants repeated each word after the experimenter. The words shared the same onset syllable (i.e., bung, bungle, bungalow; puck, pucker, puckering; buff, buffer, buffalo; pill, pillar, pillory; orb, orbit, orbital). Correct responses without struggle and which did not require any prompts were awarded scores of 2. Responses which included self-corrections, or those which displayed visible or audible searching were given scores of 1. No responses or

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responses with uncorrected speech errors were scored as 0, with a maximum possible score of 30

Non-word repetition accuracy

The non-word repetition task investigated participants' auditory-to-phonetic transcoding abilities. The assessment also explored the effects of syllable frequency and length of utterance on non-word production. Stimuli were 10 monosyllabic non-words and 10 trisyllabic non-words. There were five non-words comprised of high frequency syllables, and five non-words comprised of low frequency syllables. Frequency values were based on the Celex database (Baayen et al., 1993). Participants were instructed that all items in the test were nonsense words and they were required to repeat them after the experimenter. A second repetition of the word was provided if requested and without scoring penalty. Three practice non-words were presented. If the participant produced a real word, they were reminded that all the words were non-words and they were encouraged to repeat the form exactly as presented by the experimenter. Only first responses were scored. Correct items were given a score of 1 and incorrect items with phonetic errors were assigned the value 0, with a maximum possible score of 20.

Word repetition task and assessment of accuracy

Participants repeated a list of 105 real words (ABC list) after a single presentation. Responses were coded on a 0-7 scoring protocol by an independent assessor who had not participated in the data collection phase. This rater was also blind to the purpose of the analysis. A brief summary of examples

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in the scoring system is provided here. A score of 7 indicated rapid and accurate responses, without struggle or articulatory groping. A score of 6 was given if the response was slow (response latency greater than 2 seconds) but accurate. A score of 5 represented those responses which were accurate following a false start. Scores of 4 were given for slow responses with false starts and responses displaying phonetic errors and distortions. Scores of 3 included responses which contained two phonetic errors, while 2 indicated a response which contained two phonetic errors/distortions following a false start. Scores of 1 were assigned to repetitions which had two phonetic errors/distortions and were slow following a false start. Scores of 0 included those repetitions which were completely off target, or if a participant took longer than 10 seconds to make a response. Median scores on the 105 items were then calculated for each participant for subsequent analysis (see "Whole Group Profiles and Data Reduction").

Accuracy scoring was repeated for a subset of the speech repetition data by a second rater who was blind to the first rater's scores. This was done for 593 speech samples drawn from 17 participants. The samples represented individuals with different levels of AOS severity (mild: n=3, moderate: n=4; and severe: n=10). Spearman's Rank Correlation indicated a high level of inter-rater reliability for the entire data set (n=593, rho=.898, p<.0001). Robust-to-high levels of reliability were also found for the subgroups representing different levels of AOS severity (mild: n=104, rho=.806, p<.0001; moderate: n=139, rho=.708, p<.0001; severe: n=350, rho=.896, p<.0001). A further level of analysis was conducted to assess inter-rater reliability: absolute differences between the first and second raters were calculated for the 593 samples representing the 17 participants, and the samples for the three AOS severity subgroups. All four sets

of absolute differences indicated a median absolute difference of 0 between the two raters.

Aphasia severity: (1) Auditory perception/comprehension tests

Auditory minimal pairs and spoken lexical decision

Both tasks were presented via a CD recording of the stimuli. A subset of real word minimal pairs (same speaker, test P3) from the ADA Comprehension Battery (Franklin, Turner & Ellis, 1992) was used to assess auditory processing ability. Twenty four trials were completed, with half the items being the same and the other half, different. Participants listened to the pairs and judged if they were the same or different.

The auditory lexical decision task was again derived from the ADA Comprehension Battery and was composed of a subset of 20 items. Participants listened to each word/non-word and judged if the item was a real word or not. One participant was unable to complete the auditory assessments. The scores of both auditory tasks were summed to give an 'Auditory' score, with a maximum possible score of 44.

Spoken sentence comprehension

The experimenter said 16 spoken sentences from the Comprehensive Aphasia Test (CAT) Comprehension of Spoken Sentences sub-test and participants matched each sentence to one of four pictures which best depicted its meaning (Swinburn, Porter & Howard, 2004). Standard CAT scoring procedures were used, with a maximum score of 32.

Aphasia severity: (2) Spoken Picture Naming

Ten low frequency and 10 medium frequency items were selected from the PALPA 54 sub-test (Picture Naming x Frequency) (Kay, Lesser & Coltheart, 1992). Participants named pictures of the 20 target words. No prompts were provided. The item was marked as correct if the participant retrieved the correct or synonymous lexical item, with no penalty for apraxic errors.

Statistical Analysis I: Whole Group Profiles and Data Reduction

The median, minimum and maximum values representing all 50 participants were first calculated for each task. Subsequently, the individual scores for each participant on all tasks were correlated using a series of Spearman's rank order correlation tests (Spearman's rho). Groups of correlations were systematically conducted to investigate the associations between the scores on tasks and subtasks within and across the full range of non-speech, speech, and auditory/aphasia battery assessments. In addition, the internal reliability and consistency of the scores and scales for selected sub-tasks were analysed using Cronbach's alpha (α) coefficient of reliability (Cronbach, 1951; Cronbach, Schönemann, & McKie, 1965; Cronbach & Shavelson, 2004). Cronbach's alpha (α) values which are between .7 and .9 are interpreted as reflecting good levels of internal consistency (Nunnally & Bernstein, 1994). The behavioural tasks were grouped as follows for the correlation tests (Spearman's rho) and Cronbach's alpha (α) analysis. The first set of analyses explored the laryngeal, lingual, labial sub-tasks and the total oromotor scores. The second group of analyses explored speech production measures and included each participant's median values for word repetition accuracy (ABC list), non-word repetition accuracy, one-syllable

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word accuracy, two-syllable word accuracy and three-syllable word accuracy. The third set of analyses represented a consolidation of the speech production measures from the second set of analyses: the total scores for the increasing word length accuracy tasks were explored with the values for word repetition accuracy (ABC list) and non-word repetition accuracy. The fourth grouping of analyses explored consistency of scores on the aphasia measures and included the total auditory scores, the CAT sentence comprehension scores and the picture naming tasks scores.

Statistical Analysis II: Individual profiles and the characterisation of associations and dissociations between speech and oral apraxia impairment

This phase of analysis investigated the patterns of association and dissociation between the non-speech oromotor task scores and word production scores. The scores for the sub-tasks of the non-speech oromotor tasks (laryngeal, lingual and labial) were summed to give a total oromotor score. A total word score was calculated by summing the values for each participant for the three word production tasks: non-word repetition total, increasing word length total, and word repetition accuracy (ABC list) median. Total word scores were examined as a function of the total oromotor scores for each participant in the sample using Spearman's rho to measure the patterns and strength of association between oromotor and speech scores.

Statistical Analysis III: Can accuracy levels in speech production accurately predict levels of oromotor performance?

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Patterns of dissociation between oromotor and speech scores were examined statistically using discriminant function analysis. First, total oromotor scores were categorised into levels of performance (low (scores of 0, 1, 2, 3), low to moderate (scores of 4, 5, 6), moderate (scores of 6.5, 7, 8, 9), moderate to high (scores of 10, 10.5, 11, 12), high scores (scores of 13, 14, 14.5, 15)). Subsequently, speech production score (total word score) was used as the predictor variable to predict group membership of the 5 categories of oromotor performance. All 50 cases were used in the analysis. Prior probabilities of group classification based on the oromotor scores were computed from group size. Box's test of equality of covariance matrices yielded log determinants of 5.424, 5.389, 4.351, 4.407, 4.918 and 5.065 for the five oromotor groups (from the low to the high score groups, respectively). In addition, a Box's M value of 3.876 was not significant (p=.448), therefore suggesting that the covariance matrices did not differ and that group variance was equal.

Results I - Whole group profiles

The median, minimum and maximum values representing all 50 participants for the non-speech oromotor tasks are given in Table 1. Table 2 provides the median, median absolute deviation (MAD), minimum and maximum values for the following tasks: increasing word length (one-, two- and three-syllables, and total increasing word length), non-word repetition, word repetition (ABC list), total auditory score, the CAT spoken sentence comprehension, and spoken picture naming (PALPA 54).

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TABLES 1 & 2 ABOUT HERE

Spearman's rho correlations for all participants and Cronbach's alpha (α) -

Oromotor tasks

Table 3 displays the correlation coefficients (Spearman's rho) for the associations between the sub-tasks of the non-speech oromotor assessment (the volitional cough, the lingual task, the labial task), and the total score for the oromotor tasks. Due to the number of multiple comparisons conducted (n=6), the alpha level (p=.05) was adjusted to avoid type-1 errors. The adjusted alpha level (.05/6=.00833) was used to assess the significance in the associations between the tasks. There were significant correlations between the total oromotor scores and all the oromotor sub-tasks which ranged from rho=.768 (p<.0001) for the laryngeal sub-task, to rho=.798 (p<.0001) for the lingual sub-task. In addition, there was a significant correlation between the lingual and labial sub-tasks (rho=.568, p<.0001). Cronbach's alpha (α) was subsequently applied to the laryngeal, labial and lingual scores of the oromotor assessment for the 50 participants. This gave a Cronbach's alpha (α) value of .710 which suggested that there was internal consistency amongst the scores for the sub-tasks within the non-speech oromotor battery. On this basis, the oromotor total score was used in subsequent analyses, and in the characterisation of dissociations between non-speech and speech tasks (see sections on Statistical Analysis II and Results II below).

TABLE 3 ABOUT HERE

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Word repetition and non-word repetition accuracy

Table 4 displays the coefficients (Spearman's rho) for the correlations between word repetition accuracy (ABC list), non-word repetition accuracy, onesyllable word accuracy, two-syllable word accuracy, and three-syllable word accuracy. Due to the number of multiple comparisons conducted for this set of data (n=10), the alpha level (p=.05) was adjusted to avoid type-1 errors. The adjusted alpha level (.05/10=.005) was used to assess the significance of the associations between the word repetition and non-word repetition scores. The accuracy scores for the word repetition task (ABC list) displayed significant correlations with non-word repetition (rho=.834, p<.0001), one-syllable word repetition (rho=.757, p<.0001), two-syllable word repetition (rho=.831, p<.0001), and three-syllable word repetition (rho=.808, p<.0001). Non-word repetition scores were also significantly correlated with all the sub-tests in the increasing word length task: rho=.750, p<.0001 for one-syllable word repetition; rho=.782, p<.0001 for two-syllable word repetition; and rho=.768, p<.0001 for three-syllable word repetition. The correlations between the different sub-tasks of the increasing word length task were all significant and ranged from rho=.583 (p<.0001) for the one-syllable and two-syllable words, to rho=.858 (p<.0001) for the two-syllable and three-syllable words. Cronbach's alpha (α) was applied to the one-, two- and three-syllable word accuracy scores for the 50 participants. A value of .925 value suggested that there was a high level of internal consistency amongst the scores for the increasing word length sub-tasks. On the basis of the correlation and Cronbach's alpha (α) analyses, the increasing word length total which represented the sum of the one-syllable, two-syllable and three-syllable accuracy scores was used in subsequent analyses.

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TABLE 4 ABOUT HERE

Table 5 displays the coefficients (Spearman's rho) for the correlations between word repetition accuracy (ABC list), non-word repetition accuracy, and the increasing word length total. Due to the number of multiple comparisons conducted for this set of data (n=3), the alpha level (p=.05) was adjusted to avoid type-1 errors. The adjusted alpha level (.05/3=.017) was used to assess the significance in the associations between the word repetition and non-word repetition scores. As above, word repetition and non-word repetition accuracy scores were all significantly correlated: rho=.834 (p<.0001) for word repetition accuracy (ABC list) and non-word repetition accuracy; rho=.895 (p<.0001) for word repetition accuracy (ABC list) and increasing word length total accuracy; and rho=.863 (p<.0001) for non-word repetition accuracy and increasing word length total accuracy (see Table 5). Cronbach's alpha (α) was applied to the scores for three repetition accuracy scores: word repetition accuracy (ABC list), non-word repetition accuracy and increasing word length total accuracy. The resulting value was .758 suggesting that the scores were internally consistent for the three repetition tasks. On the basis of the correlation and Cronbach's alpha (α) analyses, a total word score based on the sum of the word repetition accuracy (ABC list) scores, non-word repetition and increasing syllable word length total was computed and used in the characterization of associations and dissociations between performance on non-speech and speech tasks (see Results II and III below).

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Auditory tasks, sentence comprehension and picture naming

Table 6 provides the correlation coefficients (Spearman's rho) for associations between the scores for the following auditory/aphasia battery assessments: auditory, CAT sentence comprehension, and spoken picture naming (PALPA 54). Due to the number of multiple comparisons conducted for this set of data (n=3), the alpha level (p=.05) was adjusted to avoid type-1 errors. The adjusted alpha level (.05/3=.017) was used to assess the significance in the associations between the scores. The total auditory scores were significantly correlated with the CAT sentence comprehension task scores (rho=.494, p<.0001), and spoken picture naming (PALPA 54) scores (rho=.435, p<.003). In addition, the CAT sentence comprehension task scores were significantly correlated with spoken picture naming (PALPA 54) (rho=.687, p<.0001). Cronbach's alpha (α) was applied to the three sets of scores. A value of .780 value suggested that there was a good level of internal consistency amongst the scores for the auditory/aphasia battery assessments.

TABLE 6 ABOUT HERE

<u>Results II - Individual profiles and the characterisation of associations and</u> <u>dissociations between speech and oral apraxia impairment</u>

Figure 1 displays the total word score (Non-word total + Increasing word length total + word repetition accuracy (ABC List) median) for all 50 AOS participants plotted against the total oromotor score (Laryngeal + Lingual + Labial). The correlation between total word score and total oromotor score was significant (rho=.395, p<.01) at the level of the whole group. The data points in

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Figure 1 indicate patterns of heterogeneity in the performance across the 50 participants; this explains the moderate level of association between the total oromotor score and the total word score in this sample. Figure 1 also displays the total oromotor scores for each participant as a function of performance (low to high scores), and shows evidence of dissociations between the total oromotor scores and total words scores of three participants; participant 51 has a very low total word score suggesting severe speech impairment, but a high oromotor score. In contrast, participants 20 and 31 are amongst the highest scorers for total word score, but display severe oromotor impairment. When the oromotor scores for participants 20 and 31 were examined further, the following profiles were found for the three sub-tasks: i) Laryngeal: participant 20 was unable to perform the laryngeal task, even after a demonstration, which resulted in a score of 0, and participant 31 produced a sneeze which was also received a score of 0.; ii) Lingual: participant 20 moved their tongue up and down initially (as opposed to the requested lateral movement), but was able to produce a correct response after demonstration which received a score of 2. Participant 31 only moved their mouth and received a score of 0; and iii) Labial: Participant 20 was unable to round and spread their lips even after demonstration, and received a score of 0, while participant 31 partially rounded and spread their lips (reduced amplitude) which received a score of 3.

The auditory processing and sentence comprehension scores for Participants 20 and 31 were then examined to determine if their performance on the oromotor tasks was likely to have been influenced by auditory processing and comprehension failures, accounting for their failure to produce volitional movements to command. Participant 20 scored 39/44 on the auditory battery,

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which is within \pm 1 MAD of the group median, and 14/32 on the CAT sentence comprehension test which was more than 1 MAD below the group median of 21.5 (see Table 2). The pattern of errors for Participant 20 on the CAT comprehension test suggested difficulties with decoding syntactic/structural information with errors on reversible and gapped sentences, accounting for 7 out of the 9 errorful responses. Participant 31 achieved a score of 28/44 on the auditory battery which was more than 1 MAD below the group median of 37, indicating some auditory perceptual difficulties. However, the score on the CAT spoken sentence comprehension test (21/32) was within \pm 1 MAD of the group median (see Table 2). Errors were again on reversible and gapped sentences (3 errors and 1 delayed response). Overall, the failures of sentence comprehension suggested specific difficulties with processing structural/syntactic information.

The results presented so far suggest that for the current clinical sample, the presence of AOS may not be an accurate predictor of accompanying OA. This is the focus of the next section.

FIGURE 1 ABOUT HERE

Results III: Discriminant function analysis

Table 7 presents the group statistics for the discriminant function analysis, and provides the mean and standard deviation values of total word score by oromotor performance category. The number of cases in each category of oromotor performance is also given. The mean and standard deviation values for each category in Table 7 suggest some degree of variation in speech performance as a function of the oromotor performance category, with the lowest

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variation being observed for the moderate group. This category also included the smallest number of cases (n=7).

There was a moderate association between speech performance and oromotor score category (Wilks Λ = .792, χ^2 (4) = 10.71, p=.03), which supports the results of the Spearman's rank correlation test (rho=.395) (see Results II). The discriminant function accounted for 100% of the between-group variability, and the canonical correlation value of .456 was low (effect size = .208), therefore indicating that total word score was not a good predictor of group membership of the oromotor score categories. This is corroborated by the data in Table 8 which provide the cross-validated classification results of the discriminant function analysis; only 34.0% of cases were correctly classified across all groups. The most frequent directional trend for misclassification was from a lower actual group classification into a higher predicted group. The highest rates of correct classification occurred when patients' actual oromotor performance measures were in the low (60%) or high (69.2%) scoring groups. Cases for both the moderate and moderate to high groups were misclassified 100% of the time (see Figure 2). Moreover, not a single individual was predicted to fall within the moderate or moderate to high categories. The classification results also indicate that two participants belonging to the low oromotor category were classified as belonging to the high oromotor category. Casewise statistics showed these to be participants 20 and 31 (see Table 8 and Figure 2). Conversely, one participant belonging to the high oromotor category was classified as belonging to the low oromotor category. Casewise statistics showed this to be participant 51 (see Table 8 and Figure 2).

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TABLES 7 & 8 ABOUT HERE FIGURE 2 ABOUT HERE

Discussion

We evaluated the severity of speech and volitional non-speech oral movement impairment in a sample of fifty participants with AOS using a broad battery of assessments. The battery included evaluation of non-speech gestures, word repetition, and a range of measures to determine aphasic impairment, particularly in the domain of auditory processing and sentence comprehension. The associations among sub-tasks of each battery were investigated and the internal consistency of the scores was determined using Cronbach's alpha (α) coefficient of reliability. In addition to assessing these relationships at a grouplevel, individual performance was examined to identify any participants whose profiles displayed evidence of a dissociation of speech and oral apraxic impairment (i.e., AOS>OA; OA>AOS). Given the theoretical importance of cases who show the pattern OA>AOS for discriminating between different accounts of mechanisms governing speech and non-speech oral movement control, we examined the auditory perceptual and sentence comprehension abilities of individuals who showed this pattern of dissociation. In particular, we sought to determine whether the apparent disproportionate oral apraxic impairment in these cases was due to poor auditory processing and comprehension. The evaluation of volitional oral movement in clinical settings requires the capacity to rapidly respond to spoken commands.

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The results revealed that at the group level, there was moderate positive association between the severity of AOS and the degree of OA impairment (see Figure 1). Evidence of associations is important in informing the process of clinical diagnosis. The clinician who is aware of co-occurrence of impairments might sensibly seek evidence of a second impairment on the basis of the presence of the first. However, as is the case in the interpretation of all correlations, the co-occurrence of the two forms of apraxic impairment does not entail that they both stem from the failure of a common mechanism. Other possibilities that might account for the coincidence of conditions include distinct neural mechanisms that are in close anatomical proximity, or that are dependent on blood supply from the same artery.

In addition to exploring performance at the level of the group, we examined patterns of dissociation or disparity in non-speech oral movement and speech production ability. Discriminant function analysis was conducted to investigate whether accuracy levels in speech production could accurately predict levels of oromotor performance. This revealed that 66% of cases were misclassified, and that in general, the total word score over predicted oromotor ability. Furthermore, of the misclassifications, three cases gave evidence of double dissociation of speech and non-speech oral movement impairment. Participant 51 displayed a high degree of speech apraxic impairment in comparison to the extent of disruption of non-speech oral movement (AOS>OA) (see Figure 1), and was incorrectly classified as having a low oromotor score on the basis of their speech production score (see Figure 2). This pattern of impairment (AOS>OA) is consistent with patients documented in other reports (e.g. Galluzzi et al., 2015).

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Models which propose considerable overlap in speech and oromotor control mechanisms can account for this pattern of dissociation. However, the reverse dissociation (OA>AOS) was observed in participants 20 and 31 who were classified as having a high oromotor score on the basis of their speech production scores (see Figures 1 and 2). This reverse pattern of dissociation has also been observed elsewhere (e.g. Kwon et al., 2013), and is problematic for claims of considerable overlap in control systems. The extent of auditory perceptual and sentence comprehension did not appear to fully account for the disproportionate impairment of volitional oromotor control. Although both Participant 20 and 31 displayed some difficulties decoding structural/syntactic information in sentence comprehension, the spoken commands for the volitional oral movement task had relatively simple syntactic structure and could largely be decoded on the basis of lexical information alone (e.g., "Can you make a cough."). Furthermore, the extent of OA for Participant 31 in particular appeared not to be due to failure to comprehend spoken commands. This participant showed relatively preserved performance on the spoken sentence comprehension test. The finding of a double dissociation between the two forms of oral movement control provides support for autonomy/partial autonomy models and the proposal that the control mechanisms for speech and non-speech oral movements are either entirely or partially distinct (Ziegler, 2003; Weismer, 2006; Bunton, 2008). This result is consistent with the findings of neuroimaging investigations (e.g., Bonilha et al., 2006) that reveal different patterns of activation during speech and non-speech oral movement tasks in healthy participants. Tremblay and Gracco (2009), however, using repetitive transcranial magnetic stimulation (rTMS), report overlapping substrates in the pre-

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supplementary motor association area (pre-SMA) for volitional speech and oromotor gestures involving the lips (whistling, raspberry, kiss). Furthermore, although TMS of the pre-SMA disrupted production of both words and oromotor gestures, a stronger interference of oromotor gestures was observed. Tremblay and Gracco (2009) posit a number of possible explanations for the latter finding, including higher levels of motoric demand for oromotor gestures which might be more susceptible to interference by TMS, or the novelty and less familiar nature of the oromotor gestures employed in their study. Overall, current evidence supports partially autonomous models of speech and oro-motor control.

Given the evidence for partial autonomy, it is possible that OA can occur without AOS. This pattern of dissociation may not always be identified within the speech pathology clinic as only patients with speech impairment may routinely have performance probed on non-speech volitional movements. In the evaluation of ideomotor apraxia, the ability to perform gestures to command in oral and nonoral effector systems is impaired (e.g., pantomime of blowing out a match and hammering a nail; Lezak, 1995). The presence of OA in some individuals displaying ideomotor apraxia without AOS in a recent study confirms the dissociation between OA and AOS (Botha et al., 2014). Furthermore, the report of association between OA and ideomotor apraxia suggests the recruitment of common resources in the production of oromotor gestures and limb movements (Botha et al., 2014).

In summary, in this investigation of a large sample of speakers with AOS we observed patterns of association and double dissociation between impairment of speech and non-speech oral movement. While associations might result from

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shared processing components, or close neural substrates, dissociations may be indicative of independent mechanisms controlling components of the two forms of movement. Key features that differentiate speech from non-speech oral movement include their patterns of connectivity to sensory-perceptual and linguistic processing systems and the novel versus entrained nature of the action plans.

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Table 1. Oromotor evaluation results for all 50 participants with AOS.

Task	Median	Median Absolute Deviation (MAD)	Minimum	Maximum
Non Speech Laryngeal (volitional cough) Max score 5	1.00	1.00	.00	5.00
Non Speech Lingual (lateral: side to side) Max score 5	5.00	4.00	.00	5.00
Non Speech Labial (alternate rounding ('oo') and spreading ('ee') of lips) Max score 5	3.00	2.00	.00	5.00
Total Non Speech Max score 15	8.00	3.50	.00	15.00

Table 2. Speech tasks, and auditory/aphasia assessments for all 50 participants with AOS.

Task	Median	Median Absolute Deviation (MAD)	Minimum	Maximum
Increasing word length 1 syllable Max Score 10	8.00	2.00	.00	10.00
Increasing word length 2 syllables Max Score 10	8.00	1.00	.00	10.00
Increasing word length 3 syllables Max Score 10	6.00	3.00	.00	10.00
Increasing word length total Max Score 30	23.00	4.00	.00	30.00
Non-word repetition Max Score 20	3.5	3.00	.00	11.00
Word Repetition (ABC List) Max Score 7 on each word item	5.50	1.50	.00	7.00
Total Auditory Score* Max Score 44	37.00	4.00	21.00	41.00
CAT Comprehension Task Max Score 32	21.50	5.00	4.00	31.00
Spoken Picture Naming (Subset of PALPA 54) Max Score 20	15.50	4.50	.00	20.00

*n=49; 1 participant failed to complete the auditory assessments

Table 3. Oromotor tasks: correlations (Spearman's rho (n=50)) for all 50 participants with AOS.

	Volitional Cough (Laryngeal)	Lingual	Labial	Total Oromotor
Volitional Cough (Laryngeal)		.385	.399	.768***
Lingual			.568***	.798***
Labial				.786***

***Significant at p<.0001 (two-tailed; using adjusted alpha level of .00833 for multiple comparisons)

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Table 4. Correlations (Spearman's rho (n=50)) for speech tasks for all 50 participants with AOS.

	Word Repetition (ABC List)	Non-word repetition	One syllable word repetition	Two syllable word repetition	Three syllable word repetition
Word Repetition (ABC List)		.834***	.757***	.831***	.808***
Non-word repetition			.750***	.782***	.768***
One syllable word repetition				.634***	.583***
Two syllable word				-	.858***
repetition ***Significant	at p< 0001 (ítwo-tailed: u	sing adjuste	d alpha level	of 005 for

***Significant at p<.0001 (two-tailed; using adjusted alpha level of .005 for multiple comparisons)

Table 5. Word repetition accuracy tasks: correlations for all 50 participants with AOS.

	Word repetition (ABC List) accuracy	Non- word repetition accuracy	Increasing word length total
Word repetition		.834***	.895***
(ABC List)			
accuracy			002***
Non-word			.863***
repetition			
accuracy			

***Significant at p<.0001 (two-tailed; using adjusted alpha level of .017 for multiple comparisons)

Table 6. Auditory, comprehension and picture naming tasks: correlations for participants with AOS.

	CAT Comprehension Score	Picture naming task (PALPA 54)
Total Auditory Score (n=49)*	.494***	.435**
CAT		.687***
Comprehension (n=50)		

*1 participant failed to complete the auditory assessments

**Significant at p<.003 (two-tailed; using adjusted alpha level of .017 for multiple comparisons)

***Significant at p<.0001 (two-tailed; using adjusted alpha level of .017 for multiple comparisons)

Table 7. Mean and standard deviation values of total word scores as a function of category of oromotor performance. The number of participants in each category of oromotor performance is also indicated.

Mean	Standard Deviation
Total Word Score	Total Word Score
17.6	15.1
26.0	14.8
34.7	8.8
31.4	9.1
33.1	11.7
	Total Word Score 17.6 26.0 34.7 31.4

Table 8. Cross-validated classification results for the discriminant analysis function using total word score as the predictor of oromotor performance.

Actual	Predicted Oromotor Group Membership ^a				
Oromotor			(n,		
Group (n)			%)		
	Low	Low to	Mod	Mod to	High
		Mod		High	
Low	6	2	0	0	2*
Scores n=10	60	20	0	0	20
Low to	3	2	0	0	7
Moderate	25	16.7	0	0	, 58.3
Scores					
n=12					
Moderate	0	2	0	0	5
Scores ^b n=7	0	28.6	0	0	71.4
Moderate	0	3	0	0	5
to High ^b	0	37.5	0	0	62.5
Scores	Ũ	01.0	Ũ	Ū	02.0
n=8					
High	1**	3	0	0	9
Scores n=13		23.1	0	0	69.2

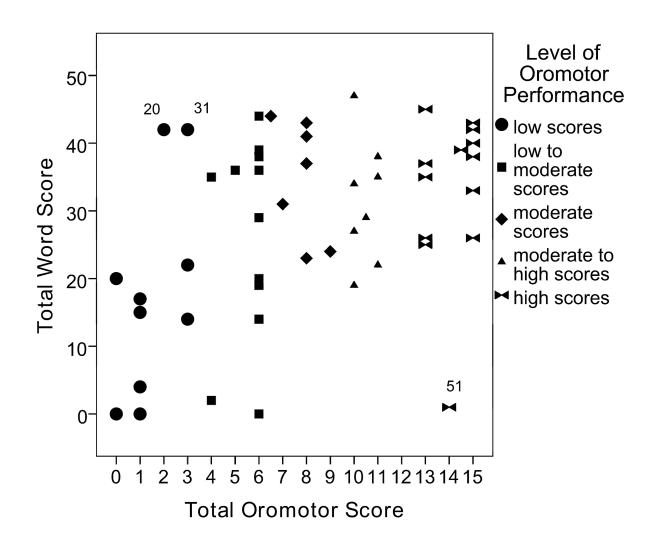
^a34.0% of original grouped cases were correctly classified.

^bCategories which were misclassified 100% of the time

*Represents participants 20 and 31

**Represents participant 51

Figure 1. Total word score (Non-word total score + Increasing word length total score + word repetition accuracy (ABC List) median score) for all 50 AOS participants plotted against the total Oromotor score (Laryngeal + Lingual + Labial) (rho=.395, p<.01). The oromotor scores in the scatterplot are also coded by level of performance (see text for explanation). Three participants displaying patterns of dissociation in this sample are also highlighted (20, 31 & 51 – see text for explanation).



http://acn.oxfordjournals.org/content/30/7/670.full?keytype=ref&ijkey=zl9hW4LyB5SrGzd

Figure 2. Total word score (Non-word total score + Increasing word length total score + word repetition accuracy (ABC List) median score) for all 50 AOS participants plotted against the total Oromotor score (Laryngeal + Lingual + Labial) by level of oromotor performance (actual group). Also depicted in the graphs is the predicted group membership for each participant using total word score as the predictor variable in the discriminant function analysis (see text for explanation). The top panel (low scores) indicates that 2 participants (20 & 31) with low oromotor scores were classified as belonging to the 'high scores' group. The bottom panel (high scores) indicates that 1 participant (51) was classified as belonging to the 'low scores' group. Moderate and moderate to high score groups were misclassified 100% of the time and therefore do not appear as a predicted group in the graph (see Table 8).

