Localizing unique and overlapping lesion locations in apraxia of speech and aphasia

Abstract

Since Darley's original description of apraxia of speech (AOS; 1968), controversy has centered around its diagnosis, treatment, and lesion location. Behaviors common to AOS are often shared among other communication disorders, complicating clinical management. The current study sought to identify crucial brain damage that causes apraxic speech, as well as errors common in both AOS and aphasia. Results revealed that damage to premotor and supplementary motor areas is unique to AOS, while involvement of temporal lobe areas predicts behaviors attributable to aphasia. These findings contribute to research regarding the neuroanatomical mechanism of AOS, and may ultimately improve differential diagnostic procedures.

Introduction

Apraxia of speech (AOS) is a disorder of planning and/or programming motor movements for speech, not related to a language deficit (aphasia) or neuromuscular involvement (dysarthria). The theoretical nature (Code, 1998; Mumby et al., 2007) and localization of crucial lesion locations (Dronkers, 1996; Hillis et al., 2004; Richardson et al., 2012) for AOS has been heavily debated in the literature. Characteristic behaviors include "effortful" visible and/or audible groping, off-target articulation, inconsistent production of speech targets, variable attempts at self-correction, difficulty with utterance initiation, and prosodic impairments (Wertz et al., 1984; Darley et al., 1975). Although these behaviors are commonly present in AOS, they are not unique to this disorder, as similar speech production difficulties may be evident in dysarthria and/or aphasia. To further complicate the controversies surrounding AOS, some have challenged perceptual evaluation of AOS and its validity in differentially diagnosing AOS from other neurologic communication disorders due to a lack of clear diagnostic guidelines (e.g., Haley et al., 2012; 2013).

In the current study, we delineated behaviors specific to AOS from those common amongst both AOS and aphasia (Josephs et al., 2012; 2013) and sought to determine sites of damage that corresponded to these behaviors. It was hypothesized that motor and premotor areas would be specifically involved in behaviors specific to AOS (Josephs et al., 2012; Whitwell et al., 2013), with the inferior frontal gyrus pars opercularis (IFGpo) (Hillis et al., 2004; Richardson et al., 2012) and posterior sensorimotor integration areas (Hickok & Poeppel, 2007) involved in both speech and language related deficits (i.e., AOS and aphasia). This project is novel in that we were able to more accurately separate behaviors related to two neurologic communication disorders with often similar behavioral features. In turn, this allowed us to identify brain areas that are both shared and unique across disorders.

Method

Participants. 30 participants (11 female; mean age = 59.73, range=37-80) were recruited as part of a larger stroke study, in which recruitment criteria were based on history of left hemisphere stroke. Participants did not need to present with aphasia or apraxia of speech to qualify for this study. All participants were tested at the chronic phase of stroke, at least six months post-onset. Participants varied in the presence or absence of aphasia type and severity, as follows: no aphasia =8, anomic aphasia =12, Broca's aphasia =8, Wernicke's aphasia =1, and conduction aphasia =1. Mean WAB score for all individuals with aphasia was 71.69 (range = 31.75 -93.2), and mean for individuals without aphasia was 97.5 (range = 94-99.2). See Table 1 for participant demographics. All participants agreed to study participation.

Procedure. Speech production was rated using the Apraxia of Speech Rating Scale (ASRS; Josephs et al., 2012; 2013). Speech samples were obtained from audiovisual recordings of three picture description tasks, diadochokinetic rates, and conversation. The speech characteristics included on this scale classify speech abnormalities into 4 categories: (a) features that occur in AOS, but not in dysarthria or aphasia; (b) features that can occur due to AOS and/or dysarthria; (c) features that can occur due to AOS and/or aphasia, and (d) features that can occur due to AOS/dysarthria/aphasia. The ASRS was completed for each participant by an ASHA-certified speech-language pathologist with experience using this scale for classification of speech production behaviors as related to AOS, aphasia and dysarthria. Each participant was rated on the presence/severity of all speech characteristics on the ASRS based on a 5-point scale (0=not present; 1 = detectable but not frequent; 2 = frequent but not pervasive; 3 = nearly always evident but not marked in severity; 4 = nearly always evident and marked in severity). An overall aphasia severity, AOS severity and dysarthria severity score was assigned to each participant as applicable, with the same aforementioned 5-point scale. For the purposes of the present study, only AOS and aphasia severities will be reported and discussed.

MRI data for each participant were acquired using a Siemens 3T system with a 12-element head coil. All participants underwent a high-resolution T1 and T2 MRI sequences.

Data Analysis. For the purpose of lesion-symptom mapping, a region of interest (ROI) analysis with aphasia and AOS severity as dependent variables (as measured by the ASRS) was conducted to localize cortical damage related to these disorders. Univariate and multivariate linear regression was completed using an in-house code written in MatLab (The MathWorks, Natick, MA) and corrections for multiple comparisons completed using permutation thresholding with 2,000 (univariate) and 3,000 (multivariate) permutations (Rorden et al., 2009).

Results

Twelve participants received a classification of AOS (mean ASRS score for those with AOS =2.75). Only one of these individuals did not have an additional classification of aphasia. Of the 22 individuals with aphasia, mean ASRS aphasia severity was 1.55. In the univariate analysis, seven ROIs survived thresholding for ASRS aphasia severity, and five ROIs survived for ASRS AOS severity. Significant ROIs were unique to each disorder's severity, except for BA 48, which was shared by both AOS and aphasia. Interestingly, areas found to be significant for AOS severity were localized in primary somatosensory, premotor/supplementary motor areas, *pars opercularis* of the inferior frontal gyrus (IFGpo), and the operculum. Areas significant for ASRS Aphasia Severity were localized along the temporal gyrus. See Table 2 for z-scores for each significant ROI, and Figures 1 and 2 for localization of significant ROIs for each disorder, based on the univariate analysis. Once lesion variability between AOS and aphasia is accounted for in multivariate analysis, 10 ROIs survived permutation thresholding for aphasia severity only, as reported in Table 3. Once again, regions along the temporal lobe emerged as significant ROIs. No significant ROIs emerged in the multivariate analysis for AOS severity. See Figure 3 for a comparison between ROIs for aphasia (corrected) and AOS (uncorrected).

Discussion

This study is novel in that we aimed to localize ROIs unique to, and shared between, AOS and aphasia. Because diagnostic metrics in AOS are plagued by poor inter- and intra-rater reliability (Haley et al., 2012; Haley et al., 2013), localization of lesion locations critical for AOS and aphasia may help to improve diagnostic capabilities for post-stroke disorders through the use of neuroimaging. It is recognized that the ASRS itself is based on perceptual ratings of speech behaviors; however, it may reduce some of the confound in perceptual assessment of AOS and aphasia, as it delineates behaviors into categories that are unique to AOS, or that could be shared between disorders. Additionally, with regard to the localization of speech production, our results add to the growing body of work that supports the role of premotor and supplementary motor areas in AOS (Josephs et al., 2012; Josephs et al., 2013; Whitwell et al., 2013). Overall, not only do our findings have implications in refining clinical diagnostic procedures for AOS, but they also contribute to localization of AOS, which has been heavily contested in the literature.

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Table 1

Demographic information for all participants

Participant	Gender	Age	Months Post Onset	Aphasia Type	WAB AQ	ASRS: Aphasia Severity	ASRS: Apraxia Severity
Stroke1	F	80	83	None	99.1	0	0
Stroke2	M	60	42	None	99.2	0	0
Stroke3	F	70	94	None	99.2	0	0
Stroke4	F	42	19	None	94.2	0	3
Stroke5	F	62	19	None	97.3	0	0
Stroke6	M	50	47	None	95.9	0	0
Stroke7	F	64	22	None	98.6	0	0
Stroke8	M	62	59	None	96.9	0	0
Average	M:F (3:5)	61.25	43.14		97.55	0	0.375
Min		42	19		94.2	0	0
Max		80	94		99.2	0	3
Aphasia1	M	57	29	Broca's	64.6	2	2
Aphasia2	M	70	54	Broca's	63.6	2	4
Aphasia3	F	39	84	Broca's	55.2	2	3
Aphasia4	M	55	47	Broca's	76.2	1	0
Aphasia5	M	57	129	Anomic	83.2	1	3
Aphasia6	M	65	113	Anomic	80.4	1	3
Aphasia7	F	78	10	Anomic	90.5	1	0
Aphasia8	F	60	185	Anomic	86.2	1	1
Aphasia9	F	57	80	Conduction	51.5	2	0
Aphasia10	M	63	65	Anomic	94	1	0
Aphasia11	M	56	73	Broca's	59.4	2	3
Aphasia12	M	66	18	Wernicke's	52.7	2	0
Aphasia13	M	55	65	Anomic	88.8	1	0
Aphasia14	M	43	27	Broca's	31.75	4	3
Aphasia15	M	66	33	Anomic	93.2	1	0
Aphasia16	F	37	85	Anomic	98.5	1	0
Aphasia17	M	62	54	Broca's	58.2	2	2
Aphasia18	M	52	36	Broca's	57.5	2	4
Aphasia19	M	76	10	Anomic	72.1	2	2
Aphasia20	M	55	26	Anomic	91.1	1	0
Aphasia21	F	72	21	Anomic	84.6	1	0
Aphasia22	M	61	24	Anomic	93.1	1	0
Average	M:F (16:6)	59.18	57.63		73.93	1.54	1.36
Min		37	10		31.75	1	0
Max		78	185		98.5	4	4

Table 2

Significant regions for ASRS Aphasia Severity and AOS Severity

Univariate Analysis					
ASRS Aphasia Severity			ASRS AOS Severity		
	ROI	Z-value		ROI	Z-value
BA 21	Middle Temporal Gyrus	4.16	BA 3	Primary Somatosensory Cortex	3.27
BA 22	Superior Temporal Gyrus	4.22	BA 6	Premotor/Supplementary Motor Cortex	2.88
BA 34	Anterior Entorhinal Cortex	4.1	BA 43	Subcentral Area	3.17
BA 38	Temporopolar Area	3.94	BA 44	Pars Opercularis	2.99
BA 41	Primary and Auditory Association Cortex	4.29	BA 48	Retrosubicular Area	3.87
BA 42	Primary and Auditory Association Cortex	4.01			
BA 48	Retrosubicular area	4.04			

Table 3

Regions associated with ASRS Aphasia Severity

Multivariate Analysis - ASRS Aphasia Severity		
	ROI	Z-value
BA 20	Inferior Temporal Gyrus	2.61
BA 21	Middle Temporal Gyrus	3.29
BA 22	Superior Temporal Gyrus	2.67
BA 28	Anterior Entorhinal Cortex	2.4
BA 34	Temporopolar Area	2.43
BA 35	Perirhinal Cortex	2.45
BA 36	Parahippocampal Cortex	2.27
BA 38	Primary and Auditory Association Cortex	2.14
BA 41	Primary and Auditory Association Cortex	2.51
BA 42	Retrosubicular area	2.03

Figure Captions

Figure 1. Aphasia severity. This figure displays significant ROIs from the univariate analysis of ASRS aphasia severity scores.

Figure 2. AOS severity. This figure displays significant ROIs from the univariate analysis of ASRS AOS severity scores.

Figure 3. Aphasia and AOS severity. This figure displays significant ROIs from the multivariate analysis for ASRS aphasia severity (in red). ROIs for AOS severity are displayed in green, although analyses for AOS severity did not survive thresholding in the multivariate analysis.

Figure 1

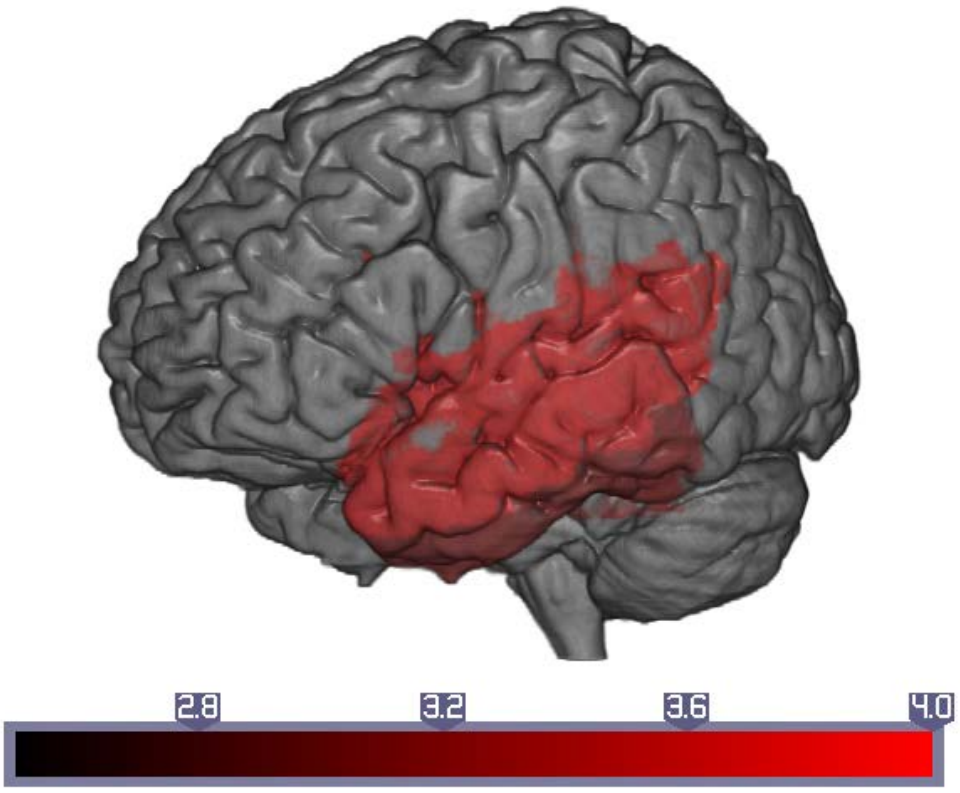

Figure 2

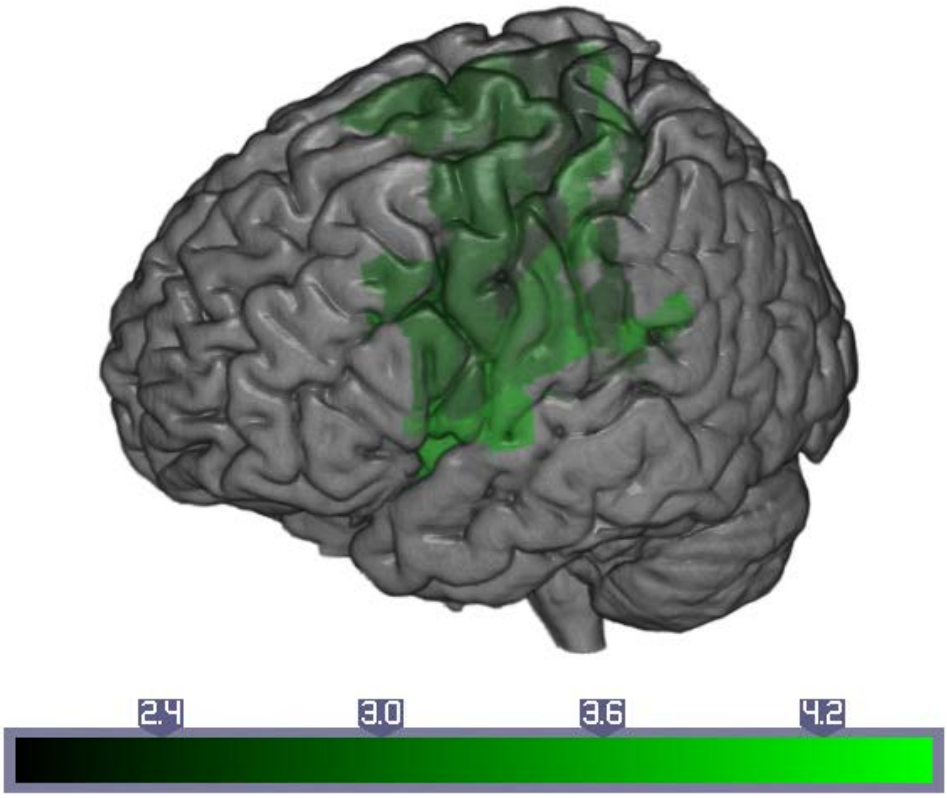

Figure 3