A Case of Crossed Apraxia for Propositional Speech

In 1969 Darley and colleagues described[1] a disorder, apraxia of speech (AoS), of motor speech planning, distinct from aphasia, characterized by slow speech rate, segmentation of syllables, sound distortions, distorted substitutions, trial-and-error articulatory movements, and increased difficulty with increased length and complexity of utterances.

Localization of the lesion responsible for AoS had been more challenging than for the lesions causing aphasia but results of studies from a variety of disciplines converge on the left premotor cortex as one lesion site for AoS. Indeed, stroke cases,[2,3] detailed neuroimaging,[4] structural studies,[5] brain stimulation and a recent case of a patient who had AoS following a small focal surgical resection[6] all point to cortical area 55b as a key lesion site for AoS. Fascinatingly, Broca himself described a case[7] of what he called aphemia, and we now recognize as AoS in a patient with a lesion including area 55b!

One prediction of a lesion to area 55b being causative for AoS would be that a lesion disconnecting Broca's area from area 55b (lesion of the anterior-to-55b pathway) would cause severe AoS for propositional speech—rational use of language for specific communication goals—originating in Broca's area, but preserved speech with normal articulation for repetition and other speech originating in Wernicke's area (posterior-to-55b pathway). We here describe the first case of this "propositional apraxia of speech" in a right-handed patient with a lesion in the right prefrontal cortex including the right-sided homologue of area 55b. Functional MRI (fMRI) studies demonstrate 94–96% of right-handed subjects are left-hemisphere dominant for language function.[8] So our case is a rare case of crossed apraxia of speech.[9,10] Our patient also had severe dysphagia and aspiration requiring percutaneous endoscopic gastrostomy (PEG) tube placement. A recent paper[11] found a striking and statistically significant cooccurrence of severe dysphagia with aspiration risk and apraxia of speech (AoS) in patients with left hemisphere frontal cortical strokes, but not a statistically significant cooccurrence of Broca's (expressive) aphasia and aspiration risk. Interestingly, the causative lesion area in this paper responsible for AoS and severe dysphagia includes area 55b.

A 60-year-old right-handed male with a past medical history of hypertension and coronary artery disease presented (Day 1) to an outside hospital with altered mental status and difficulty speaking. He was found to have left side weakness. CT brain showed a large right middle cerebral artery infarct. Initial bedside dysphagia evaluation by the speech pathologist found the patient to have significant anterior spillage of liquid bolus with overt signs of aspiration and no initiation of the pharyngeal swallow. Trials of puree necessitated manual removal of the bolus from oral cavity and he was placed NPO. Eight days later (Day 9) the patient was admitted to acute inpatient rehabilitation. Physical examination showed left upper extremity strength 4/5, left lower extremity 4+/5 with intact sensation. His spontaneous speech was unintelligible, but the patient had normal receptive language. Speech therapy evaluation showed a severe oral dysphagia characterized by significant anterior spillage of bolus with overt signs of aspiration. A modified barium swallow (MBS) the next day (Day 10) showed severe oropharyngeal dysphagia. The patient agreed to have a percutaneous endoscopic gastrostomy (PEG) tube placed (Day 17, see Table 1 for timeline of Speech Pathology findings).

On admission to acute rehabilitation the patient's spontaneous speech was incomprehensible and was initially attributed by physicians to severe dysarthria. By contrast the patient's receptive language was intact. He also demonstrated reading comprehension significantly above chance—11/12 accuracy in matching read words to four different choices. As well, the patient was able to produce a normal-sized circular outline with symmetrically, well-placed features when given the clock drawing task.

Curiously, the patient was noted to be able to repeat phrases with adequate articulatory precision. An MRI was obtained [Figure 1], and this unexpected ability to repeat was the basis for further assessment of his speech articulation, which was probed using different cues such as auditory pattern super-sequences, and visual object, color and scene identification. In these tests, his speech production was markedly superior when utterances were automatic in nature (e.g., repeating, producing a numerical sequence) rather than volitional (e.g., naming visually presented objects and scenes).

As is recorded on Audio 1 (Example 1 in Transcripts) and Example 2 (see Transcripts), sequencing and "super-sequencing" tasks were completed with little error in either speech or arithmetic. Namely, the patient was able to count by 2s and 5s up to 12 and 30, respectively, after being prompted by the first two numbers only (i.e., with minimal

Figure 1: T2 FLAIR Brain MRI showing lesion in the right dorsal premotor cortex

Table 1: Dysphagia timeline

Day Number	Speech Pathology Findings
Day 1	Significant anterior spillage of liquid bolus with no initiation of the pharyngeal swallow. Trials of puree results in manual removal of bolus from oral cavity
	REC: NPO
Day 2	Ice chip trials- limited oral manipulation and posterior transport of bolus. Severely limited laryngeal elevation. Oral suctioning performed
	REC: NPO
Day 4	Improved toleration of pureed bolus and thin liquids, with mild anterior spillage. Patient placed on pureed diet and thin liquids.
Day 9	Severe oral stage dysphagia characterized by significant anterior spillage of bolus. Left buccal pocketing, poor bolus manipulation, and delayed swallow initiation. Reduced hyolaryngeal elevation noted on palpation. Majority of bolus spilled anteriorly from oral cavity.
	REC: MBSS and alternative means of nutrition
Day 10	Modified Barium Swallow Study
	Severe oral dysphagia secondary to limited lingual range of motion and poor lip seal- significant anterior spillage and lingual pumping.
	Severe oropharyngeal dysphagia- delayed swallow initiation, reduced hyolaryngeal elevation, incomplete laryngeal vesitbular closure. Partial anterior hyoid excursion and diminished pharyngeal stripping wave. Mild valleculae, pyriform sinuses, and posterior pharyngeal wall residue.
	Penetration of puree and aspiration of all liquid consistencies.
	REC: NPO, alternative means of nutrition. Dysphagia therapy. Supplemental oral feeds of puree 2 x 4 oz. container post extensive oral care.
Day 13	Mildly improved lingual and labial range of motion and strength. Significantly reduced anterior spillage and improved bolus control. Continued to present with overt signs and symptoms of aspiration.
	Recommendation: NPO with NG tube
Day 14	Improved secretion management and strength of swallow mechanism. Continued to present with overt signs and symptoms of aspiration. Speech unintelligible.
Day 15	Repeat Modified Barium Swallow Study
	Reduced anterior spillage of bolus, disorganized lingual motion for bolus formation, repetitive mastication, and moderate collection of residue in left buccal cavity.
	Delayed initiation of the pharyngeal swallow with bolus head at the pyriform sinuses. Significantly reduced superior movement of the thyroid cartilage with minimal approximation of arytenoids to epiglottic petiole. Incomplete laryngeal vestibular closure with aspiration of pureed boluses (1/3 times). Silent aspiration and study was terminated.
Day 17	PEG tube placed

cues). Interestingly after the patient had counted by 2s to 14, he next said "thirteen," (Audio 1/Transcripts Example 1). In a similar cuing paradigm, he successfully counted backwards from 10 and correctly recited the days of the week and months of the year. Throughout super-sequencing tasks, the patient consistently substituted/t/for/d/in 'two' but pronounced other words starting with consonants fluently (Examples 1-3).

The patient's ability to verbalize visual observations was assessed next. As captured in Example 4, the patient mumbled unintelligibly when asked to identify a photograph of pizza. However, as illustrated by Example 5, he read the word "pizza" aloud with clarity. As demonstrated in the remaining examples, his ability to initiate articulation of visibly salient objects was markedly improved once he had heard the phrases aloud; i.e., when he was prompted to repeat. Per Examples 6 and 7, the patient softly uttered the color he was presented. However, he easily vocalized the word "yellow" after he had been prompted to repeat the instructor's utterance (Example 7). Finally, as is captured on Audio 2 (transcribed in Example 8), while the patient could correctly identify and point to the most salient elements of the Cookie Theft Picture, he could not do so comprehensibly verbally, despite being able to repeat them aloud (e.g., "water is running over") without any articulatory imprecision.

We think that our patient has a *forme fruste* of AoS—"propositional apraxia of speech"—which we have not seen previously reported. His deficit is differentiated from transcortical motor aphasia (and expressive aphasia) by facile production of significant propositional speech output, albeit not comprehensible speech, as for example during his description of the Cookie Theft Picture (Audio 2/Example 8). A parsimonious explanation for our patient's speech deficit would be damage from the stroke to the connection between Broca's area and motor articulation hubs, but an intact pathway from Wernicke's area to the speech articulation area [Figure 2]. As demonstrated by Examples 1–4, 7 and the end of Example 8 (Audio 2), the patient has also at most only a mild dysarthria when performing automatic speech tasks.

Our patient had a right brain stroke. Lateralization of language is a well-recognized feature of brain function, with fMRI studies demonstrating[8] 94-96% of right-handed subjects to be left hemisphere dominant for language function. Though rare, several cases of crossed apraxia of speech have been reported, and in particular, two well-described cases of crossed apraxia of speech following stroke[9,10] had a lesion that would appear to include the right homologue of area 55b. These cases show that the area controlling speech articulation, in addition to the classical speech areas (Broca's and Wernicke's), can be in the right brain even in right-handed individuals. Further, our case shows that in a patient

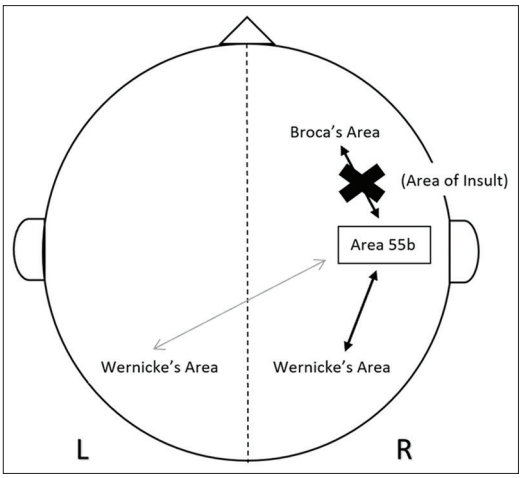

Figure 2: Diagram showing (black arrows) intact pathway from Wernicke's area (in the right brain) to the speech articulation area (55b) in the right prefrontal cortex, but a damaged pathway from Broca's area (also presumed to be on the right side in this patient) to the speech articulation area. The grey arrow shows the possibility of a pathway from Wernicke's area in the left brain to the speech articulation on the right side of the brain

with AoS for propositional speech, preserved normal articulation is a marker of tasks that can be subserved by Wernicke's area without assistance from Broca's area.

Our case is the first instance where AoS emerged solely during propositional speech. In addition to AoS, our patient also had severe swallowing deficits with aspiration of all food consistencies that were resistant to standard dysphagia exercises and requiring percutaneous endoscopic gastrostomy (PEG) placement. So our case may be an instance of a right middle cerebral artery stroke causing the recently described syndrome[11] following left middle cerebral artery stroke of co-occurrence of dysphagia with aspiration and AoS. As such, our case and subsequent reports of patients with similar presentations may be helpful in further localizing the brain areas responsible for speech articulation and swallowing, thereby improving rehabilitation methods for speech and swallowing and minimizing aspiration risks. In particular, our case may motivate the search for methods to engage, recruit and utilize potentially useful latent right brain homologues of left-brain speech areas damaged after a stroke or other brain injury.

Declaration of patient consent

The patient gave written consent for publication of the case and use of the audio tapes and MRI image.

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Conflicts of interest

There are no conflicts of interest.

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Supplementary Material

Transcripts:

AUDIO 1/EXAMPE 1

Patient: do, three, four, five, six, seven, eight, nine, ten

Examiner: Okay, two, four, six

Patient: do, four, six, eight, ten, twelve, fourteen, thirteen

Examiner: Very good.

EXAMPLE 2

Examiner: Count backwards, ten, nine

Patient: ten, nine, eight, seven, six, five, four, three, do, one

Examiner: Days of the week, Monday

Patient: Monday, Tuesday, Wednesday, Thursday, Friday, Saturday, Sunday

Examiner: by fives, five, TEN

Patient: five, ten, fifteen, twenty, twenty-five, thirty

Examiner: Very good.

EXAMPLE 3

Examiner: How many fingers (showing two fingers)

Patient: do

Examiner: How many fingers (showing three fingers)

Patient: Three

Examiner: It's good.

EXAMPLE 4

Examiner: Can you read that word, sir?

Patient: Pizza.

Examiner: One more time.

Patient: Pizza.

EXAMPLE 5

Examiner: What do you see? (showing a picture of pizza)

Patient: incomprehensible sounds

EXAMPLE 6

Examiner: What color is that? (pointing to blue color on tablet computer)

Patient: Blue (impaired articulation but comprehensible)

EXAMPLE 7

Examiner: What color is that? (pointing to yellow color on tablet computer)

Patient: Yellow (impaired articulation but comprehensible)

Examiner: Can you say yellow?

Patient: Yellow

AUDIO 2/EXAMPLE 8

Examiner: See this picture. Tell me what you see in this picture. (Showing the Cookie Theft Picture)

Patient: Patient makes incomprehensible sounds with pointing salient activities in the picture such as flowing sink, children stealing cookies

Examiner: Okay. Can you say, “Water is running over”?

Patient: Water is running over.

Examiner: Very good.