Characteristics of Patients with Brain Damage and Suspected Severe Swallowing Apraxia

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Abstract: The detailed mechanisms and neural networks underlying swallowing apraxia (SwAp) are unknown. We retrospectively examined the clinical symptoms and computed tomography (CT) brain images of 8 patients with suspected SwAp. We showed that SwAp is associated with the left side of the brain because the dominant hemisphere was affected in each case. Three cases presented with rippling SwAp and had damage to the precentral gyrus in the dominant hemisphere. In contrast, there was no common anatomical feature associated with stasis SwAp. We hypothesized that SwAp is a subtype of buccofacial apraxia because the brain areas involved in buccofacial apraxia were in close proximity to the areas affected in SwAp (observed in 3 of 8 cases). However, from a semiology viewpoint we consider the 2 types of apraxia independent because they did not always occur together.

Key words: swallowing apraxia, dysphagia, precentral gyrus, supramarginal gyrus, apathy

Introduction

Apraxia is a disorder of voluntary movement observed in patients with brain damage. Individuals with apraxia cannot initiate a willed or planned purposeful movement or activity despite the presence of adequate strength, sensation, coordination, and comprehension. There are several types of apraxia, including oral-verbal, constructional, dressing, ideational, ideomotor, limb-kinetic, and swallowing apraxia (SwAp). SwAp is a disturbance in the ability to perform a continuous swallowing movement.

Initially, Logemann proposed that SwAp resulted from a delay in initiating the swallow without tongue motion after presentation of a bolus in the mouth, or by mild to severe searching motions of the tongue before initiating the swallow. SwAp has also been described as a “lack of coordination of labial, lingual, and mandibular movement during the oral stage.” In the left hemisphere, the precentral gyrus (PrCG), Broca’s area (BA), and the supramarginal gyrus (SMG) have been associated with the praxis system. A recent review by Tsubahara noted that SwAp could be divided into 2 pathological conditions. The first is a disorder associated with buccofacial apraxia (BFAp), which correlates with lesions in the opercular part (POp) of...
the PrCG to the anterior insula in the dominant hemisphere. The second is a serious disorder in the preparatory and oral phases of the swallow, which develops in association with severe apathy, the lack of the will to move. This correlates with lesions in the area from the medial aspect of the frontal lobe to the cingulate gyrus (CG).

Despite knowledge of the brain regions involved in SwAp, the details of the underlying mechanisms and neural networks are unknown. Moreover, unlike other traditional forms of apraxia that are recognized based on this exclusion definition, SwAp has not readily been accepted as a true form of apraxia.

In this study, we retrospectively analyzed the clinical symptoms and computed tomography (CT) brain images of 8 subjects with suspected SwAp. By reconsidering these diagnostic images and additional neuropsychological symptoms we aimed to gain further insight into the mechanisms of SwAp.

**Subjects and methods**

We selected 8 patients with suspected severe SwAp in accordance with Logemann’s definition: patients who are unable to intake necessary nutrition from the mouth, yet present a normal swallowing reflex at the pharyngeal stage. The 8 patients were admitted to our hospital between 2013 and 2016. Nutrition was provided by an intravenous drip or nasogastric tube because the patients were unable to intake the necessary nutrition by mouth. This study was performed with the approval of the medical research review committee at Nishi-Hiroshima Rehabilitation Hospital.

Clinical records, CT scans, and videofluoroscopic swallowing studies (VF) were retrospectively examined. The patients were divided into 2 groups according to VF results. The first group was characterized by a repetitive rippling tongue movement in the oral phase and classified as the rippling SwAp group. The second group was characterized by a lack of movement of the tongue, which led to stagnation of water or jelly on the floor of the cavity, and classified as the stasis SwAp group.

**Results**

Patient demographics and clinical characteristics are shown in Table 1 and anatomical references are shown in Fig. 1 and 2. Cases 1 and 2 were assigned to the rippling SwAp group and Cases 4-8 were placed in the stasis SwAp group. Case 3 showed characteristics of both rippling and stasis SwAp. The cases in the rippling SwAp group all had PrCG damage in the dominant hemisphere. However, we found no common anatomical features in the stasis SwAp group. Case reports representative of rippling SwAp and stasis SwAp are presented below.

Cases 1, 3, and 4 also had BFAp, which may be associated with SwAp. Cases 3, 5, 6, and 7 displayed apathy; cases 5 and 6 also had left-side CG damage. Of the 8 cases, 7 had left hemisphere brain damage. It is possible that the dominant hemisphere in Case 2 was the right hemisphere even though the patient was right-handed, as this case showed motor aphasia. We found no other relationships between the type of apraxia and specific brain damage.
### Table 1. Patient demographics and clinical characteristics

| Case | Age (y) | Sex | Diagnosis                        | Brain region damaged          | SwAp type       | BFAp | Other apraxia | Apathy |
|------|---------|-----|----------------------------------|-------------------------------|-----------------|------|---------------|--------|
| 1    | 85      | F   | Subcortical hemorrhagic infarction | L.PrCG, L.SMG, L.POp, L.BA    | rippling        | +    | + (LKA)       | -      |
| 2    | 69      | M   | ICH + SAH                        | R.MCA, R.PrCG, R.IG, R.BA     | rippling        | -    | -             | -      |
| 3    | 82      | F   | ICH                              | L.Pu, L.PrCG, L.IG             | rippling plus stasis | +    | -             | +      |
| 4    | 58      | F   | SAH                              | A-com, L.SMG                   | stasis          | +    | + (IA)        | -      |
| 5    | 42      | F   | SAH + Cerebral infarction        | A-com, L.CG, R.CG, R.PrCG, R.IG, R.POp | stasis        | -    | -             | +      |
| 6    | 81      | F   | ICH + SAH + CSDH (traffic accident) | L.CG                           | stasis          | -    | -             | +      |
| 7    | 82      | F   | ICH                              | L.PrCG, L.SMG                  | stasis          | -    | -             | +      |
| 8    | 89      | F   | Cerebral infarction              | L.IG                           | stasis          | -    | + (IMA)       | -      |

**Abbreviations:** ICH, intracerebral hemorrhage; SAH, subarachnoid hemorrhage; CSDH, chronic subdural hematoma; PrCG, precentral gyrus; SMG, supramarginal gyrus; POp, part of opercularis; BA, Broca’s area; Pu, Putamen; IG, insular gyrus; CG, cingulate gyrus; MCA, (ruptured aneurysm of) middle cerebral artery; A-com, (ruptured aneurysm of) anterior communicating artery; SwAp, swallowing apraxia; BFAp, Buccofacial apraxia; LKA, limb-kinetic apraxia; IA, ideational apraxia; IMA, ideomotor apraxia.

**Fig. 1.** View of the left hemisphere of the brain. The insular gyrus (IG) is in the back of the Sylvian fissure. IG, insular gyrus; POp, part of the opercularis; PrCG, precentral gyrus; SMG, supramarginal gyrus.

**Fig. 2.** Axial plane at the level of the semioval center. CG, cingulate gyrus; PrCG, precentral gyrus.
Representative case of rippling SwAp

Case 1 was an 85-year-old female patient with a subcortical hemorrhagic infarction in the left cerebral hemisphere who had previously received conservative treatment. After 35 days from onset, the patient was transferred to our hospital for rehabilitation. She received a small amount of food by mouth, but received most nutrition through a nasogastric tube. After 112 days from admission (148 days from onset), she had gained the ability to receive nutrition from the mouth only in the form of a pureed meal. The CT scan revealed a high-density lesion due to a subcortical hemorrhagic infarction that had affected parts of the PrCG and SMG. There was also some mass effect to the POp and BA. VF studies showed that the patient could chew and attempted to transfer the bolus to the pharynx repeatedly; however, she was unable to do so smoothly and took a long time to eat. She could swallow without any aspiration when the bolus finally reached the pharynx. This case represented rippling SwAp. This patient also presented with BFAp, limb-kinetic apraxia, sensory aphagia, memory disturbance, disorientation, right-side unilateral spatial neglect, and inattention but did not exhibit apathy.

Representative case of stasis SwAp

Case 4 was a 58-year-old female patient with subarachnoid hemorrhage caused by rupture of an anterior communicating (A-com) artery aneurysm, who had received coil embolization by intravascular surgery. At 32 days post-surgery, a ventriculoperitoneal (VP) shunt was inserted for secondary hydrocephalus. At 67 days post-surgery, the patient was transferred to our hospital for rehabilitation and received an intravenous drip because she could intake only one-tenth of a meal. At 110 days from onset, she was able to intake a whole meal. CT images revealed that coil embolization at the A-com caused halation and that the VP shunt had been inserted in the posterior horn of the left lateral ventricle, passing around the SMG. VF studies showed that the bolus stopped in the oral cavity between the velum and the tongue for a long period. The patient was unable to transfer the bolus to the pharynx but could swallow well, rarely when the bolus arrived at the pharynx. This case was an example of stasis SwAp. This patient also presented with ideational apraxia, BFAp, motor impersistence, memory disturbance, disorientation, inattention, and apathy.

Discussion

Apraxia is defined as a symptom that cannot be explained by other neurological signs, such as motor palsy, ataxia, or high brain function. Logemann proposed that SwAp could be defined in 2 ways: as a delay in the swallow at the oral stage or as involving abnormal tongue searching motions. Tsubahara also reported 2 types of SwAp: one associated with BFAp and one including symptoms of severe apathy, which correlate with lesions in the CG associated with SwAp. However, Tsubahara was unable to discern whether it is correct or not to include the state of severe apathy into SwAp. A recent study by Saito et al substituted “swallowing hesitation” for SwAp to prevent misunderstanding of
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definition, and they classified the “swallowing hesitation” into 2 categories depending on tongue movements: rippling and stasis\textsuperscript{14}. Their stasis type of “swallowing hesitation” included apathy. No one could have been sure about relationship between SwAp and apathy yet. In short, depending on the definition of SwAp used, cases diagnosed as SwAp may or may not include apathy. Additional discussion is necessary to establish a solid definition of SwAp. For the purpose of this study, we suggest that SwAp is associated with left hemisphere function because all 8 cases had brain damage in the dominant hemisphere. There is evidence that damage to the left hemisphere is also associated with SwAp\textsuperscript{17-19}.

We attempted to identify the regions of the brain responsible for SwAp and to correlate them with swallowing patterns in the cases without apathy (Cases 1, 2, 4, and 8). Cases 3, 5, 6, and 7 displayed apathy; therefore, they were maybe excluded from receiving a SwAp diagnosis. We found that cases with the rippling pattern only (Cases 1 and 2) had damage in the PrCG in the dominant hemisphere. Case 3, which had the rippling and stasis patterns, also had damage in the left PrCG. It is possible that brain damage in the left PrCG is associated with rippling SwAp, a finding that is consistent with previous reports\textsuperscript{6,14}. However, we were unable to associate a specific part of the brain with the stasis pattern. According to Saito et al, stasis SwAp tended to be associated with broad lesions in the left hemisphere\textsuperscript{14}. However, since 4 of the stasis SwAp cases in this study also displayed apathy, we were unable to identify a connection between SwAp and specific brain regions in the remaining 2 stasis SwAp cases without apathy.

Robbins and Levine found that 50% of patients with damage to the left cerebrovascular region have difficulty in initiating coordinated swallow movement and also show oral and verbal apraxia\textsuperscript{5}. In contrast, the neural mechanisms that mediate lingual discoordination during swallowing might be partly independent of the neural systems that mediate BFAp functions\textsuperscript{19}. The critical areas for BFAp include the frontal and central opercula, anterior insula, and a small area of the first temporal gyrus that is adjacent to the frontal and central opercula\textsuperscript{20}. Since these areas were in close proximity to the areas affected by SwAp in cases 1, 3, and 4, which also had BFAp, we hypothesize that SwAp is a subtype of BFAp\textsuperscript{21}. However, we consider BFAp and SwAp to be independent from a semiological viewpoint because they did not always occur together.

Although there are many reports about the brain areas that might be responsible for SwAp\textsuperscript{6,8,11,14,18}, no report describes the size of the area involved or the extent of damage. It is rare that a limited singular part of the brain is damaged by brain vascular disease or a traffic injury, usually plural parts are damaged at once. Hence, it is difficult to guarantee that a precise anatomical region is responsible for the observed neuropsychological symptom. Although Cases 4 and 8 had sustained damage in a relatively narrower cerebral area than seen in the other cases, this sample size was too small for us to declare a new hypothesis. Further research with a larger sample size is required to determine an association between SwAp and the size of the brain area that has sustained damage and the extent of damage involved.
Conflict of interest disclosure

The author has no conflicts of interest directly relevant to the content of this article.

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[Received January 11, 2017 : Accepted February 2, 2017]