

## CASE REPORT

## Dysphagia management in bilateral frontal opercular syndrome (Foix-Chavany-Marie syndrome)

Thomas Richards,<sup>1</sup> Thomas Tudor Hughes,<sup>2</sup> Jake Moss,<sup>3</sup> Tessa Lomax<sup>4</sup>

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<sup>1</sup>Adult Speech and Language Therapy Department, Cardiff and Vale University Healthboard, Cardiff, UK

<sup>2</sup>Department of Neurology, Cardiff and Vale University Health Board, Cardiff, UK

<sup>3</sup>Cardiff and Vale University Healthboard, Cardiff, UK

<sup>4</sup>Pennine Acute Hospitals NHS Trust, Oldham, UK

**Correspondence to**

Mr Thomas Richards,  
Thomas.Richards@wales.nhs.uk

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**SUMMARY**

An 80-year-old woman was admitted to hospital with acute anarthria and severe dysphagia, characterised by loss of voluntary control of the muscles of facial expression, mastication, bolus manipulation (tongue) and pharyngeal constriction, with relative preservation of involuntary movements. Brain imaging revealed an acute infarct in the left frontal operculum and an area of gliosis consistent with an old infarct in the right frontal operculum. A clinical and radiological diagnosis of bilateral anterior frontal opercular syndrome (Foix-Chavany-Marie-syndrome) was made. Deglutition consists of a voluntary oral stage and an involuntary pharyngeal and oesophageal phase. As involuntary movements were relatively preserved and no sensory loss was suspected, it was hypothesised that, once initiated or triggered, pharyngeal swallowing could be adequate. This was trialled at the bedside without adverse effects and subsequently videofluoroscopy demonstrated timely laryngeal elevation and closure, adequate opening of the upper oesophageal sphincter and smooth transit of the bolus from mouth to oesophagus. Oral intake was restored within days of admission and a percutaneous endoscopic gastrostomy was avoided. This is the first time to the authors' knowledge that this management approach has been described for this condition.

**BACKGROUND**

Previous reported management of this condition has involved a period of long-term enteral nutrition using either nasogastric (NG) or percutaneous endoscopic gastrostomy (PEG) feeding (figure 1).<sup>1–6</sup> This report makes the case that patients with bilateral frontal opercular syndrome may be able to return to oral intake rapidly with timely referral to speech and language therapy (SALT), avoiding a PEG and the associated risks. In common with other conditions in which the lower motor neuron circuitry of cranial nerves V, VII and IX–XII is preserved, pure reflex swallowing (laryngeal closure and elevation in response to the presence of a bolus) may facilitate oral feeding. In this case, this was associated with a reduced length of hospital stay and satisfactory nutrition and hydration solely from oral intake. The case illustrates the importance of a multidisciplinary approach to the management of oral feeding failure and the value of a detailed understanding of the mechanism of oral feeding failure in every patient.

**CASE PRESENTATION**

An independent 80-year-old woman presented with acute onset of difficulty moving her face, tongue and jaw, with associated loss of speech and an inability to swallow. Her medical history included a previous right opercular infarct from which she had made a full recovery, hypertension, left ventricular hypertrophy, coronary heart disease and gastro-oesophageal reflux disease. Medication on admission included losartan 50 mg once daily, atenolol 50 mg once daily and esomeprazole 20 mg once daily.

During the admission, esomeprazole was changed to lansoprazole 15 mg once daily and the patient was started on clopidogrel 75 mg 14 days poststroke, rosuvastatin 10 mg at night, biotene oral balance dry mouth moisturising liquid and chlorhexidine 1% dental gel taken as directed four times a day.

General medical examination revealed an irregular pulse and a BP of 170/89 mm Hg. Neurological examination revealed a bilateral eyelid apraxia, slow and broken vertical saccades, and a complete absence of voluntary facial, lingual, jaw and palatal movements with associated anarthria and aphagia. During laughter, spontaneous facial movements were observed. There was no emotional lability and no primitive reflexes. Comprehension of the spoken and written word was preserved and the patient was able to communicate her story using a pen and paper. Neurological examination of the limbs was normal. CT was suggestive of acute infarct in the left frontal operculum and old infarct of the right frontal operculum (figure 1).

**TREATMENT**

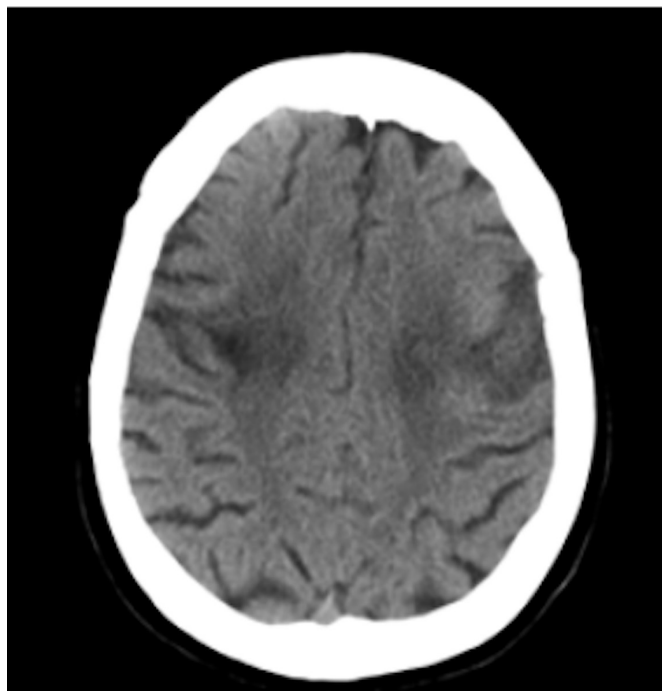
Medical management followed the local stroke protocol. The patient was prescribed 300 mg of aspirin and as she did not meet the inclusion criteria, she was not thrombolysed. Having failed the nurse-led swallow screen, an NG tube was inserted and the patient was referred to SALT for dysphagia evaluation and management. Ten days following admission, she was anticoagulated with the direct oral anticoagulant apixaban, as her stroke was attributed to atrial fibrillation.

To feed, the patient used a semireclined posture with her chin up to prevent anterior loss of fluids and to direct the bolus towards the pharynx. The patient commenced intake with 1–2 mL bolus of fluid via a syringe. The fluid was placed sublingually and allowed to trickle around the lateral ventral tongue and floor of mouth, towards the faucial arches thus providing the required sensory



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**Figure 1** CT head demonstrating bilateral frontal opercular infarcts.

information for appropriate calibration of a pharyngeal swallow. The patient tolerated this without adverse clinical signs. As she became familiar with this approach, she quickly progressed from sublingual syringe to a 5 mL bolus via a spouted measuring cup placed on the lips with the spout directed to the ventral tongue/floor of mouth. Delivery was then controlled by the patient and volume increased with practice. Once a safe, efficient pharyngeal response was confirmed with video fluoroscopy (figure 2; see online supplementary video 1), the patient was introduced to puréed meals via spoon as this did not require chewing. Oral intake, with addition of high-calorie supplement drinks, was continued using sublingual administration with spouted cup and chin-up posture. This allowed her to meet her nutritional requirements.

#### OUTCOME AND FOLLOW-UP

Over the course of the admission, eyelid apraxia improved with some improved jaw function. Nutritional requirements were

satisfied orally with a combination of puréed meals and supplement drinks under supervision of a dietitian. PEG feeding was not required. The NG tube was removed 48 hours post-commencing puréed meals, and the patient was discharged home. On 2-month follow-up, voluntary control of musculature for feeding and speech was improving, and she was able to chew soft foods and articulate single words with adequate intelligibility.

#### DISCUSSION

There are limited published case reports describing anterior bilateral frontal opercular syndrome as a consequence of serial cerebral vascular events, and few provide an account of the considerations around diagnosing and managing dysphagia. Those that do advocate for enteral feeding and early PEG placement has also been suggested<sup>6</sup>. PEG placement is associated with infrequent but known risks of haemorrhage, infection and blockage.<sup>7</sup> We have described a successful case of restoring safe and sufficient oral intake in a condition where early PEG feeding may superficially appear to be the most effective, low-risk management approach.

The neuroanatomical basis of autonomic voluntary dissociation, whereby involuntary control is preserved despite loss of voluntary control, has been described in previous case reports of bilateral anterior opercular syndrome.<sup>8</sup> The operculum houses neurons that send supranuclear fibres to the cranial nuclei responsible for voluntary control of the facial, pharyngeal, lingual and masticatory muscles, whereas emotional and autonomic control of these muscles come from pathways running from the amygdala and lateral hypothalamus to the brainstem.<sup>8</sup> This may explain why in this syndrome reflex and behavioural movements are relatively preserved. Disruption of these muscles results in oral stage dysphagia, which has a detrimental effect on the ability to meet nutritional and hydration needs orally.

To remediate oral stage deficits, it would not be effective to simply bypass the oral stage via a syringe or similar. To understand why this is the case, it is necessary to understand the underlying mechanisms. The medullary swallow centres are responsible for the crude pharyngeal swallow,<sup>9</sup> including the sensory nuclei of cranial nerves IX and X which, in part, form the 'Nucleus Tractus Solitarius' (NTS). The NTS relies on sensory information obtained during the oral stage. The supra nuclear sensory afferents and motor efferents for swallowing project into the NTS and augment the 'central pattern generator' of the crude swallow. This is hypothesised to appropriately calibrate the onset, velocity and



**Figure 2** Video fluoroscopy using fluids of normal consistency showing minimal post-swallow residue (typical for age) without laryngeal penetration or aspiration.

duration of the pharyngeal swallow response; appropriate to the volume, texture and consistency of the bolus.<sup>10</sup>

For this reason, syringing fluids towards the posterior pharynx in a reclined position was hypothesised to be unsuccessful due to the absence of crucial sensory data acquired during the oral stage. Without these data, it can be predicted that the pharyngeal response would be mistimed. The consequence may be laryngeal penetration of fluids while the vestibule is open as supraglottic approximation (false cords, arytenoids) does not typically occur until the pharyngeal swallow is initiated. Thus, increasing the likelihood of preswallow laryngeal penetration of fluids and increasing aspiration risk. Aspiration is associated with pneumonia and is a leading cause of mortality in the stroke population.<sup>11</sup> This knowledge informed the successful reintroduction of oral feeding in this case; without it, one may solve

the problem of oral stage dysphagia while inadvertently causing pulmonary aspiration.

**Contributors** TR, TL, JM: Literature search and initial drafts. TR and TL sourced radiology imaging. TTH conducted review and contributed critical revisions to article draft before submission. TTH and TR act as joint guarantor.

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**Patient consent** Obtained.

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### Learning points

- ▶ This report contributes to an evolving understanding of the supratentorial lesions that cause dysphagia and how this manifests clinically.
- ▶ This report makes the case that it may be possible to avoid PEG feeding and the associated risks in patients with preserved lower motor neuron innervation of the larynx, pharynx and tongue.
- ▶ Multidisciplinary management of oral feeding failure in bilateral frontal opercular syndrome is likely to yield superior outcomes.
- ▶ This case illustrates one instance of a successful therapeutic progression of oral intake. Video-fluoroscopy is considered the gold standard for assessment of swallowing however therapists without access to this may want to consider a cautious trial similar to that described in this report.

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