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A CASE OF FOIX-CHAVANY-MARIE SYNDROME WITH HISTORY OF GLIOBLASTOMA SHOWING PARTIAL RECOVERY

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ABSTRACT

Introduction: Foix-Chavany-Marie syndrome (FCMS) or bilateral opercular syndrome (OPS) is a rare pseudobulbar palsy characterized by facial, lingual, pharyngeal, and masticatory voluntary muscle paralysis resulting in anarthria with preservation of autonomic, involuntary, and reflexive functions. Damage to the posterior part of the inferior frontal gyrus and inferior part of precentral gyrus play a role in the pathogenesis of FCMS.

Case description: We report a rare case of bilateral OPS following an acute right middle cerebral artery (MCA) infarct in a patient with history of glioblastoma, and resection in the left MCA territory within the cingulate gyrus, showing recovery of speech and swallowing despite intensive bilateral opercular lesions owing to extensive multidisciplinary team support. However, the patient was discharged with a percutaneous endoscopic gastrostomy tube for long-term enteral feeding support due to partial recovery.

Discussion: The patient's history of glioblastoma, left MCA cingulate gyrus resection and right MCA infarction with automatic-voluntary dissociation led to the diagnosis of FCMS. Rehabilitation surprisingly showed mild improvement in speech and swallowing despite extensive bilateral opercular lesions proving that there are still chances of improvement in speech and swallowing in OPS with the right multidisciplinary approach.

Conclusion: Patients with a history of brain tumours like glioblastoma can develop bilateral OPS later in life in case of other vascular events that cause lesions in a previously unaffected operculum, triggering symptoms. Extensive involvement of the speech and language team is of significant in the management of FCMS cases as above where some recovery might still be seen.

KEYWORDS

Glioblastoma, bilateral opercular lesions, ischemic stroke, speech and language rehabilitation, automatic-voluntary dissociation

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LEARNING POINTS

- Foix-Chavany-Marie syndrome (FCMS) can be caused by bilateral opercular lesions (new or old or a combination of both) in patients with history of high-grade brain tumours.
- Patients with bilateral opercular syndrome experience full or partial or no recovery at all of speech and swallowing, hence proper rehabilitation with a Speech and Language Team is significant.
- Understanding automatic-voluntary dissociation in FCMS and being able to differentiate it from bulbar palsy and other similar phenomena is crucial in making a diagnosis of FCMS.

INTRODUCTION

Foix-Chavany-Marie syndrome (FCMS) or bilateral opercular syndrome (OPS) is a rare pseudobulbar palsy characterized by facial, lingual, pharyngeal, and masticatory voluntary muscle paralysis resulting in anarthria with preserved autonomic, involuntary, and reflexive functions[1]. Damage to the posterior part of the inferior frontal gyrus and inferior part of precentral gyrus plays an important role in OPS[2].

Voluntary movements, like speaking, chewing, and sticking out the tongue are primarily mediated by the corticobulbar tract, which originates from primary motor cortex (precentral gyrus), Premotor and supplementary motor areas and frontal operculum. These fibres descend contralaterally and bilaterally to the cranial nerve motor nuclei in the brainstem (e.g., facial, hypoglossal, trigeminal nuclei) and control precise, intentional movements of the face, tongue, pharynx, and jaw.

Involuntary/emotional movements (smiling, crying, yawning, laughing) are mediated by subcortical pathways: limbic system (amygdala, cingulate gyrus, hypothalamus), extrapyramidal tracts (reticulospinal, rubrospinal tracts), and brainstem centres that bypass the cortical motor areas. These pathways do not run through the operculum, so they remain intact in OPS. This is referred to as automatic-voluntary dissociation[3] hence affected patients are unable to voluntarily open the mouth, protrude the tongue, blowing out the cheeks, chew food, speak or swallow but can yawn, laugh, and cry[4].

Autonomic-voluntary dissociation is also seen in oral buccal apraxia, but patients are not entirely mute. Properties that make it different from bulbar palsy are preservation of jaw jerk, pharyngeal reflex, absence of fasciculation, atrophy and phenomenon of denervation and, unlike pseudobulbar palsy, the pathological laughter and emotional disturbances are lacking[5]. The most common cause of OPS is cerebrovascular disease, specifically ischemic stroke, involving the middle cerebral artery (MCA) territory[6]. Other causes are epilepsy, infections, neoplasia, vasculitis, traumatic brain injury, and degenerative diseases[3,4,6]. Unlike Broca's aphasia which can be associated with muteness OPS does not involve the total loss of voluntary movements.

We present a case of a man with paralysis of facial, lingual, pharyngeal, and masticatory voluntary muscles with preservation of autonomic, involuntary, and reflexive functions with history of glioblastoma and resection inn the left MCA territory within the cingulate gyrus territory and new acute right MCA ischemic stroke leading to the diagnosis of FCMS showing unexpected rapid partial recovery within 3 months despite severe bilateral opercular lesions.

CASE DESCRIPTION

A 58-year-old male, active smoker presented to the emergency department with occipital headache, unable to swallow and unable to speak, with a sensation of something stuck in his throat, and with left facial droop. He was admitted to the stroke ward. The patient had a background of epilepsy and glioblastoma multiforme in the left temporal lobe for which he underwent sequential operations including resection in the left MCA in cingulate gyrus territory plus radical chemotherapy.

On examination, he was unable to voluntarily open his mouth, protrude his tongue, show his teeth, whistle, chew,

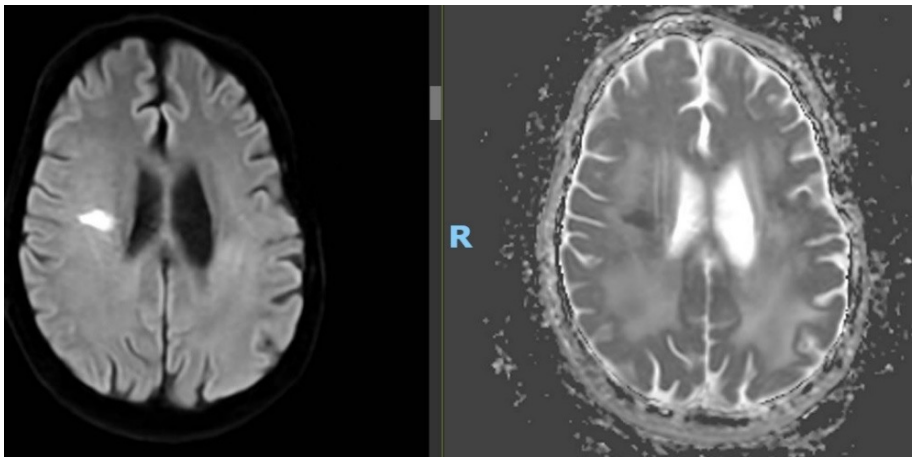


Figure 1. Diffusion-weighted magnetic imaging of the head with contrast (with apparent diffusion coefficient mapping) showing acute right middle cerebral artery territory infarction.

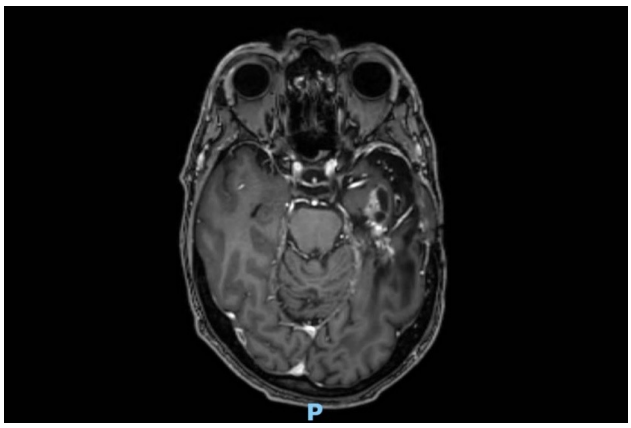


Figure 2. Magnetic resonance imaging of the head with gadolinium showing an increase in the volume of the enhancement, along with more noticeable nodularity along the medial aspect of the temporal lobe resection, extending superiorly to the ventricular margin.

or swallow. The tongue was midline with no tongue atrophy, fibrillation, and deviation observed. The patient scored 1 for facial droop and 3 for best language on the National Institutes of Health Stroke Scale, with a total score of 4. No limb weakness was observed, and reflexes were intact. Blood tests and vital signs were within normal limits.

As part of the stroke workup, the patient underwent an initial computed tomography (CT) scan of the head which did not show any acute stroke. CT angiogram did not show any significant stenosis of the large neck vessels. Magnetic resonance imaging (MRI) of the head with gadolinium was done later to identify tumour progression and identify any stroke due to ongoing symptoms and a negative CT scan of the head. MRI of the head showed acute infarct in the central right cerebral hemisphere *(Fig. 1)* along with slight glioblastoma progression *(Fig. 2)*. Oncology was consulted and suggested conservative management of the slight glioblastoma progression.

Blood tests for acute ischemic stroke including autoimmune antibodies and antiphospholipid syndrome antibodies were done to look for the cause of stroke and were found to be negative. A transthoracic echocardiogram showed a left ventricular ejection fraction of >55% without vegetation, thrombus, valve, or wall motion abnormalities.

Due to the above presenting symptoms – aphasia, anarthria, drooling, inability to voluntarily open the mouth, protrude the tongue protrusion, puff out the cheeks, chew food , speak or swallow but preserved reflex-motor and automatic functions and the ability to smile along with a new right MCA ischemic infarct *(Fig. 1)*, a history of glioblastoma and resection in the left MCA territory within the cingulate gyrus *(Fig. 3 and 4)* territory with history of epilepsy, the patient was diagnosed with FCMS. The patient did not receive thrombolysis and did not undergo thrombectomy due to not meeting the criteria. The patient communicated by writing on a board and by using hand signs.

High dose atorvastatin was started to control cholesterol which is a risk factor for stroke. The patient was reviewed by the tobacco team for smoking and started on nicotine replacement therapy. The patient also developed spontaneous jaw swelling and pain. CT scan of the face showed bilateral temporomandibular joint dislocation which spontaneously reduced. The patient also had an inpatient fall during his stay and CT scan of the head showed acute haemorrhage within the left temporal resection cavity *(Fig. 5)* due to which antiplatelet therapy was stopped. The patient was initially started on antiplatelet therapy on identification of stroke but this was later stopped due to fragility of blood vessels post radiotherapy received earlier. Due to difficulties



Figure 3. Non-contrast computed tomography scan of the head showing left temporal craniotomy.

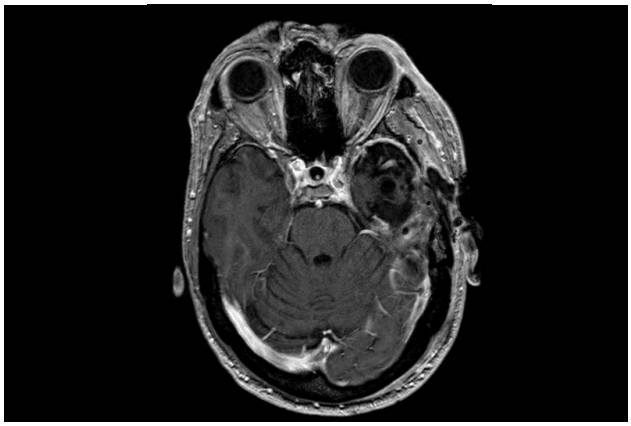


Figure 4. Gadolinium-enhanced T1-weighted magnetic resonance imaging of the head showing the resection cavity and pneumocephalus in the left temporal lobe post craniotomy.



Figure 5. Non-contrast computed tomography scan of the head showing left temporal haemorrhage.

with swallowing and speech, the patient was reviewed by the speech and language team and dietitians and underwent nasogastric tube insertion for support with feeds and medications initially. Surprisingly the patient made some rapid recovery in his speech and swallowing with continuous rehabilitation within 3 months (he was able to tolerate International Dysphagia Diet Standardisation Initiative level 3 diet and level 0 fluid despite severe bilateral opercular lesions). However, he still had a percutaneous endoscopic gastrostomy tube inserted for long-term nutritional rehabilitation due to only partial recovery.

DISCUSSION

The part of the brain cortex covering the insula, inferior frontal, pre- and postcentral, supramarginal, angular (inferior parietal), and superior temporal gyrus is referred to as the operculum. OPS is defined as anarthria and facial, pharyngeal, masticatory, tongue, laryngeal, and brachial voluntary muscles paralysis[1,2]. As described, typical automatic-voluntary dissociation was observed in our patient. Both bilateral and unilateral cases of OPS have been observed previously[3].

Often new and older lesions together in combination can also produce bilateral OPS which was observed in our patient[6,7]. FCMS due to epilepsy, as in our patient, has been reported[8]. Tumours are an established but rare cause of FCMS, however no cases of FCMS associated with glioblastoma have been reported previously. Our patient with history of progressive glioblastoma with left MCA cingulate gyrus resection and new right MCA infarction qualified for the diagnosis of FCMS making this case extremely rare.

Also, previous research has shown that very rarely patients with bilateral OPS have demonstrated only partial recovery but the majority have not shown any improvement at all with either speech or swallowing[5,7,9]. On the other hand, almost all the patients with unilateral opercular lesions have relatively good recovery[3,5]. Hence, the prognosis for bilateral OPS tends to be worse than that for unilateral OPS[10].

Despite aggressive bilateral opercular lesions, it was surprising that our patient showed rapid partial recovery with his speech and swallowing within months since it is rare to observe any improvement with both speech and swallowing in such cases. This can possibly be attributed to regular and aggressive rehabilitation measures by the speech and language therapy team. This emphasizes that rehabilitation therapy is of utmost significance in any patient with bilateral OPS as there might be scope for some degree of recovery.

A history of high-grade progressive glioblastoma and rapid partial recovery of both speech and swallowing despite intense bilateral opercular lesions made this case of FCMS rare and notable.

CONCLUSION

Early recognition and involvement of multidisciplinary team is necessary to effectively treat FCMS patients. The present case shows that some amount of recovery of speech and swallowing is possible in these patients with the right therapy assessments. Also, in patients with old high-grade brain tumours new infarcts or lesions in the brain opercula can contribute to OPS.

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