Anterior Biopercular Syndrome Caused by Unilateral Infarction

Sindrome Biopercular Anterior Devido a Enfarte Unilateral

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ABSTRACT
The anterior biopercular syndrome is characterized by facio-pharyngo-glosso-masticatory diplegia, with automatic dissociation of movements. It generally translates bilateral opercular lesion, often of vascular etiology. There are very few cases described with unilateral lesions. We present the case of a patient with a bilateral anterior opercular syndrome caused by unilateral infarction.

Keywords: Deglutition Disorders; Cerebral Infarction.

RESUMO
A síndrome biopercular anterior caracteriza-se por diplegia facio-faringo-glosso-mastigatória, com dissociação dos movimentos automaticos. Traduz geralmente lesão opercular bilateral, frequentemente de etiologia vascular. Há casos raros descritos de lesão unilateral. Apresentamos o caso de uma doente com uma síndrome opercular anterior bilateral causada por um enfarte unilateral.

Palavras-chave: Disturbios da Deglutição; Enfarte Cerebral.

INTRODUCTION
Anterior opercular syndrome, or Foix-Chavany-Marie syndrome, was first reported by Magnus in 1837 and later detailed by Foix et al. in 1926. It is characterized by facio-pharyngo-glosso-masticatory diplegia, with preservation of reflex and automatic functions of these muscles. Ischemia is the most frequent cause and it is generally due to bilateral opercular lesions, but bilateral subcortical lesions have also been reported. Much less commonly, unilateral lesions can cause a biopercular syndrome.

We present the case of a patient with a bilateral anterior opercular syndrome caused by unilateral infarction.
CASE REPORT

A 47 years old female patient, right-handed, with a history of smoking and of two miscarriages, woke up one morning with speaking and swallowing difficulties. A week before she had felt paresthesia in the right hemiface and upper limb. On neurological examination the patient was awake, had good understanding of spoken and written language, but was unable to produce any sound. She had difficulty imitating simple gestures such as opening the mouth or protrude the tongue. Smile or yawn reflexes were preserved. She presented syntactic and praxis errors in her handwriting. She had a bilateral palate paralysis with anesthesia in this area and in the oropharynx, absence of palatal and gag reflexes, bilateral vocal cord palsy and dysphagia. She also had mild right hemiparesis, predominantly brachiofacial, with generalized osteotendinous hyperreflexia but without Babinski sign.

Magnetic resonance imaging (MRI) showed an acute left opercular infarction (Fig. 1). No other acute neither non-recent lesions were detected.

Routine blood tests, immunologic and pro-thrombotic studies, electrocardiogram and echocardiogram were normal. Doppler sonography of cervical arteries and the angiography confirmed a carotid dissection.

An anterior biopercular syndrome was diagnosed in the dominant hemisphere secondary to cerebral infarction caused by carotid dissection.

The patient was hypocoagulated with acenocumarol and recovered significantly. Four years later she had only slight language changes, and cervical Doppler sonography showed recanalization of the left internal carotid artery.

DISCUSSION

In the anterior opercular syndrome the connections between the motor cortex and the V, VII, IX, XII cranial nuclei and brainstem are interrupted bilaterally. Emotional and spontaneous movements, dependent on extrapyramidal connections, thalamus and hypothalamus, are preserved (automatic-voluntary dissociation).4

In this case the syndrome was incomplete because the V and VII nerves were not bilaterally affected, and curiously the lesion was unilateral. Although the classical neuroanatomical basis of the opercular syndrome involves bilateral lesions5, there are rare cases of unilateral lesions causing a bilateral palsy of the facial, chewing, tongue, pharynx and larynx muscles6,7 (Table 1). Several hypotheses have been suggested to explain this phenomenon; one possibility is the existence of an anatomical variant, so that the corticobulbar tract representation is predominantly unilateral.3

Regarding aetiology, cerebrovascular disease (mainly ischemic) is the most frequent cause.3,4 However other disorders were also described: encephalitis, brain tumours, cortical dysplasia, vasculitis, epilepsy, degenerative diseases and traumatic brain injury.

Several authors identified poor prognosis in the recovery of swallowing and especially of verbal language.4,6 In two reported cases of unilateral lesions with bilateral manifestations the prognosis was good3,7 and the same occurred in our patient.

This case illustrates a peculiar bilateral anterior opercular syndrome caused by unilateral brain infarction. Unlike the cases due to bilateral damage, functional outcome was good.

CONFLICT OF INTERESTS

None stated.

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Table 1 - Previously reported cases of biopercular syndrome caused by unilateral lesion.

<table>
<thead>
<tr>
<th>Reference</th>
<th>Age, years/sex</th>
<th>Handedness</th>
<th>Cerebral imaging</th>
<th>Lesion</th>
<th>Outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td>Starkenstein et al.7</td>
<td>55/M</td>
<td>Right</td>
<td>TC</td>
<td>Right operculum and insula</td>
<td>Marked improvement of jaw movement, muscle weakness and dysphagia; persistent anarthria</td>
</tr>
<tr>
<td>Moragas Garrido et al.3</td>
<td>61/M</td>
<td>ND</td>
<td>MRI</td>
<td>Left operculum</td>
<td>Absence of dysphagia and improvement of facial palsy and dysarthria</td>
</tr>
<tr>
<td>Moragas Garrido et al.3</td>
<td>36/M</td>
<td>ND</td>
<td>MRI</td>
<td>Right operculum</td>
<td>Improvement of dysarthria, dysphagia and brachial weakness; persistent facial diplegia</td>
</tr>
<tr>
<td>Giraldo-Chica et al.6</td>
<td>76/F</td>
<td>Right</td>
<td>MRI, SPECT</td>
<td>Right operculum</td>
<td>Improvement of dysphagia, persistent anarthria</td>
</tr>
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REFERENCES


