

Case Report

When Nothing Goes Right: An Unexpected Tongue Deviation in Internal Carotid Artery Dissection

Elisa Corazza^a Simone Lorenzut^b Mariarosaria Valente^a
Gian Luigi Gigli^a Giovanni Merlino^b

^aNeurology Clinic, Udine University Hospital, Udine, Italy; ^bStroke Unit, Udine University Hospital, Udine, Italy

Keywords

Cerebrovascular diseases · Internal carotid artery dissection · Hypoglossal nerve palsy · Neuroanatomy · Tongue

Abstract

Internal carotid artery dissection is a frequent cause of stroke in young people. The artery dissection and the formation of an intramural hematoma could also cause mass effect on surrounding structures, causing disorders such as cranial nerve palsies (about 12% of the cases), including XII cranial nerve. In the setting of an ischemic stroke, lower cranial nerve palsy could also be due to infratentorial ischemic lesions; however, there have been also rare reports of lower cranial nerve palsy due to supratentorial cerebral ischemic lesions. We describe a case of a 55-year-old man who presented with right internal carotid artery dissection and deviation to the left of the protruded tongue. The direction of the deviation of the protruded tongue was unexpected in this patient, because if the XII nerve palsy was due to mass effect related to the intramural hematoma of the dissected artery, a deviation to the right should have happened. Anyway, a subsequent magnetic resonance revealed also an acute ischemic lesion in the right tongue area in the primary motor cortex of the patient, providing a rare, but a fitting neuroanatomical explanation of the deviation and also providing clinical evidence of functional dominance of the crossed projections of the cortico-lingual tracts.

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Introduction

Internal carotid artery dissection represents an important cause of stroke in young and middle-aged patients [1]. During the dissection, a tear occurs in the artery, and blood enters the arterial wall under pressure, separating the wall layers. This process causes the formation of a false lumen, where blood leaks into the vessel wall, forming a hematoma and potentially leading to the occlusion of the vessel. The artery dissection could lead to cerebral thromboembolism and/or hypoperfusion, with subsequent ischemic stroke. It may also cause mass effect on surrounding structures, causing disorders such as Horner's syndrome and/or cranial nerve palsies.

Cranial nerve palsies occur in 12% of patients with carotid artery dissection, and in 5.2% of patients lower cranial nerve palsies happen (including the involvement of XII cranial nerve, with possible additional involvement of XI, X, and IX cranial nerves) [2].

In the setting of an ischemic stroke, lower cranial nerve palsy could also be caused by infratentorial ischemic lesions; however, there have been rare reports of lower cranial nerve palsy due to supratentorial cerebral ischemic lesions as well [3, 4].

Case Report

A 55-year-old man presented to the emergency department with sudden onset of dysarthria and inferior left facial palsy. His medical history was positive for dyslipidemia and arterial hypertension, treated with beta-blockers and statins.

On neurological examination, he had mild dysarthria, mild loss of strength in the left upper and inferior limb, left inferior (central) facial palsy, and deviation to the left of his tongue when protruded. The rest of the neurological and physical examination was normal.

An urgent head computed tomography (CT) showed no active parenchymal lesions; an angio-CT of the neck vessels and of the intracranial vessels showed reduction of caliber of the right internal carotid artery from its origin and then occlusion of the vessel from the C1–C2 level, involving the extracranial and intracranial tract (Fig. 1). The rest of the vessels were normal. These findings were suggestive of right internal carotid dissection, as suggested by the neuroradiologist.

The patient then underwent intravenous thrombolytic treatment, without complications. The right internal carotid artery dissection was also confirmed by an ultrasound examination, which showed flow occlusion in the right internal carotid artery, due to an intramural hematoma.

The patient underwent brain magnetic resonance imaging (MRI) 3 days after the symptom onset, which showed high signal intensity area in the territory of the right middle cerebral artery with restricted diffusion, involving also the right tongue area of the primary motor cortex (Fig. 2).

The neurological deficits, including tongue deviation, gradually improved, and after 5 months he only had mild inferior left facial palsy. The patient had no previous history of trauma, and there was no evidence of connective tissue disorders; no cause of the dissection was identified.

Discussion

The hypoglossal nerve innervates the ipsilateral muscles of the tongue, and its activation causes the protrusion of the tongue. Unilateral tongue weakness causes the tongue to deviate toward the weak side when protruded, as it happens in ipsilateral lesions of tongue muscles and in ipsilateral lesions of lower motor neurons or of the hypoglossal nerve. Lesions of the upper motor neurons (originating in the motor cortex) could cause contralateral tongue weakness, instead.

In our patient, the left deviation of the protruded tongue was not compatible with his carotid dissection side (if the weakness had been due to the right hypoglossal nerve compression the tongue should have been deviated to the right when protruded). The MRI finding of an ischemic lesion involving also the right tongue area of the primary motor cortex explained the left-sided deviation of the tongue.

Tongue palsy after a unilateral upper motor neuron lesion is uncommon and not completely understood. In fact, several studies with transcranial magnetic stimulation showed that the cortico-lingual projections are bilateral and both crossed and direct [5], but the crossed projections appear to dominate functionally, as shown by the amplitudes of the compound muscle action potentials [6].

In conclusion, our case provides a rare example of clinical evidence of functional dominance of the crossed projections of the cortico-lingual tracts and also provides evidence of inability of the unaffected hemisphere to compensate the deficit with the direct cortico-lingual projections.

We also would like to emphasize the importance of correlating every clinical finding with the neuroanatomy knowledge and to outline how the deficit of cranial nerves in an artery dissection should always be investigated before ascribing it to a mere compression mechanism.

Statement of Ethics

The authors have no ethical conflicts to disclose. The patient was informed and has given us his consent for the publication of the case.

Disclosure Statement

The authors have no conflicts of interest to declare.

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Author Contributions

E.C., G.M., and S.L. collected the data of the case, performed the literature search, wrote the initial manuscript drafts, selected the figures and references, and performed the first revision of the case. G.L.G. and M.V. supervised the work, performed the literature search, and completed the revision of the case.

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Fig. 1. Angio-CT showing the occlusion of the right internal carotid artery (arrow). The “flute beak” aspect is suggestive of carotid artery dissection.

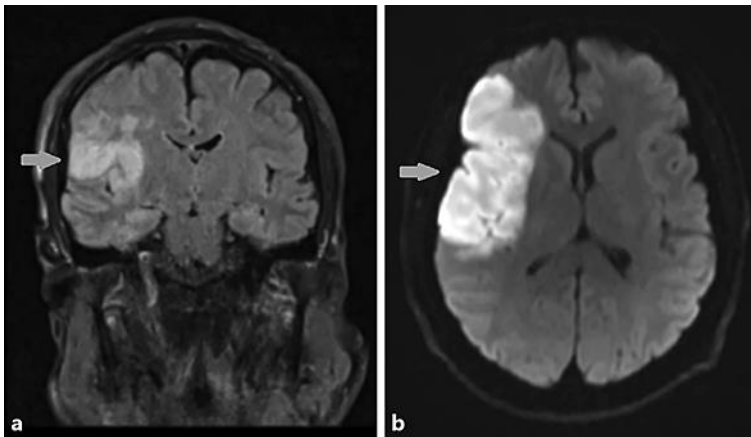


Fig. 2. **a** Brain MRI scan (coronal FLAIR sequence) shows a high signal intensity area on the right hemisphere, including also the tongue area of the primary motor cortex (arrow). **b** Brain MRI scan (axial diffusion-weighted image) shows restricted diffusion with high signal intensity in the same area (arrow).