Carotid Artery Dissection Induced Acute Tongue Swelling in a Cocaine User

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ABSTRACT ~ Carotid artery dissection represents a well-recognized cause of hypoglossal nerve paralysis even if it is less known the cause of acute tongue swelling. We report a 42-year-old man who presented to our observation with acute tongue swelling and atrophy of left side of tongue from a hypoglossal nerve injury. A magnetic resonance imaging revealed a denervation of the left half of the tongue from a hypoglossal nerve injury due to left extracranial internal carotid artery (ICA) dissection, without evidence of ischemic stroke. The urine toxicology screen test revealed a positivity for cocaine. This case report suggests to perform in young patients a toxicological drug screening test in presence of ICA dissection with hypoglossal nerve injury and acute tongue swelling. However clinical data must be performed to validate this observation and to analyze the negative effect of cocaine use.

INTRODUCTION

Carotid artery dissection, a well-recognized cause of ischemic stroke in both young and middle-age patients, may be traumatic or spontaneous, resulting in hemorrhage within the arterial wall. A traumatic dissection involves the common carotid resulting in a complete occlusion. Systemic hypertension and arteriopathy, such as fibromuscular dysplasia, Marfan’s syndrome, type 4 Ehlers-Danlos syndrome, and cystic medial necrosis, atheroma and pheochromocytoma are common causes of spontaneous carotid dissection. Previous clinical studies some authors reported the development of hypoglossal nerve paralysis with or without acute tongue swelling after internal carotid artery (ICA) dissection were reported. ICA dissection were observed in active cocaine users.

We describe the development of tongue swelling after spontaneous ICA dissection in a cocaine user.
CASE REPORT

A 42-year old men was admitted on June 29, 2018 to emergency room for painless tongue swelling. History excluded the presence of systemic disease, use or abuse of drugs and alcohol. No head movement with exertion of neck were reported. A blood pressure was 125/80 mmHg. Electrocardiography and echography excluded the presence of heart disease; biochemical tests were in normal range. A diagnosis of local (tongue) angioedema was postulated and a treatment with antihistamine and corticosteroid drugs was started, without clinical benefit. Three weeks later, the patient presented again to the hospital for tongue swelling and a subtle weakness and atrophy of left side of tongue was reported (Fig. 1). MRI of tongue, neck, and brain suggested a denervation of the left half of the tongue from a hypoglossal nerve injury due to left extracranial ICA dissection without ischemic lesions in the left ICA distribution. An urine toxicology screen test revealed a positivity for cocaine and a treatment with aspirin (100 mg/day) to prevent cerebral ischemia was started.

A follow-ups performed 1 and 3 months later documented an improvement of both tongue swelling and left-sided tongue weakness and on September 2018 brain images revealed a complete regression of the left ICA dissection.

FIGURE 1

LEFT SIDED TONGUE SWELLING AND ATROPHY OF LEFT SIDE OF TONGUE
DISCUSSION
The underlying arterial dissection was spontaneous and painless and this evidence combined with tongue swelling rather than more expected weakness and atrophy lead to the failure of recognition of neurogenic nature of this swelling.4

Even if hypertension could play a role in cocaine-mediated dissection,6 Dabbouseh et al.7 documented that cocaine exerts a direct damage of the vascular wall inducing an ICA dissection and in a cocaine-user with intracerebral hemorrhage a endothelial dysfunction of vascular wall was suggested.7 In agreement, in a cocaine-user with intracerebral hemorrhage and normal blood pressure a endothelial dysfunction of vascular wall was suggested.8

In the present case, clinical, radiological and biochemical findings excluded other causes involved in this clinical manifestation. Moreover, in our patient a normal blood pressure was reported and drug screen test revealed a positivity for cocaine use.

The use of cocaine could have induced either ICA dissection and acute tongue swelling due to a combination of exudative and trasudative edema in interstitium for an alteration of the endothelial bed. However it is possible that connective disease could play a role in this clinical manifestation. Unfortunately, we did not perform the genetic tests to excluded a possible involvement of the connective tissue disorders in vascular anomalies and this represent a limit for this case report. However, after the cessation of cocaine, the patient started a program for cocaine detoxification and now the patients is at the 5-month of this program e no other clinical manifestation of endothelial disease or connective disease was presented.

This case report suggest to perform in young patient with negative anamnesis for drug abuse at admission a toxicological drug screening test, in presence of acute tongue swelling with hypoglossal nerve injury induced by ipsilateral ICA dissection. ♦

Informed Consent
The patient provided written informed consent for publication of this report.

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