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Case report

Hypoglossal nerve paralysis in a child after a dental procedure



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ABSTRACT

Unilateral palsy of the hypoglossal nerve is a rare complication of orthodontic procedures. The main reported causes of HNP are: orthopedic and otorhinolaryngology surgical interventions, and in particular maneuvers involving compression or overstretching of the hypoglossal nerve, dental procedures and traumas, and also infections, motoneuron disorders, tumors, vascular diseases. Diagnosis is usually performed by electrophysiology studies (EMG-VCN), and brain magnetic resonance imaging (MRI) is useful to exclude other causes. The prognosis depends on the location and extension of the damage. Currently there is not a standardized treatment approach except the speech therapy, although, in some cases, the high-dose steroid treatment could be useful. We describe the case of a ten-year-old female, who was admitted in our Unit after a deviation of the tongue associated with dysarthria and dysphagia, occurred after the application of a mobile orthodontic device.

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1. Introduction

The hypoglossal nerve (Cranial Nerve XII) innervates the intrinsic and extrinsic muscles of the tongue, and is a pure motor nerve. It has both bulbar and spinal origin; nerve fibers originate from the medulla oblongata, and C1 fibers join the nerve as it exits the skull. The cervical root fibers innervate the strap muscles of the neck. Supranuclear fibers originate from the contra-lateral primary motor cortex. Both genioglossus muscles must be innervated and properly functioning to protrude the tongue in the midline [1] (Fig. 1).

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When monolateral, hypoglossal nerve palsy (HNP) is usually caused by intracranial or extracranial space occupying lesions, surgical or anesthesiology procedures, infections (dengue, poliomyelitis), motoneuron disorders, trauma, head and neck injuries, vascular anomalies, neuropathy or autoimmune diseases [2]. More rarely it can be idiopathic.

The anatomical location of the lesion is usually related to a different clinical presentation: (a) supranuclear lesions cause tongue deviation opposite to nerve lesion. There is not atrophy

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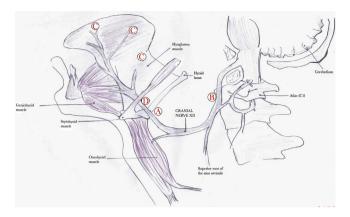


Fig. 1 – Anatomic locations of hypoglossal nerve injury: (A) Nerve compression or impingement (B) Nerve stretching (C) Pressure of the distal nerve fibers (D) Calcified stylohyoid ligament.

or fasciculation of the tongue and vascular disease is often the main cause; (b) nuclear or intranuclear lesions (unilateral or bilateral) cause a lower motor neuron dysfunction ipsilateral to the lesion, with fasciculation and atrophy: it is mainly caused by tumors and neurodegenerative diseases; (c) isolated lesions of nerve during its course cause ipsilateral deviation of tongue, and are sometime associated other cranial nerves (IX, X, XI) involvement. It is generally caused by trauma or expansive or compressive lesions [1,3].

2. Case report

We report a case of unilateral palsy of hypoglossal nerve in a 10-year-old aged female, admitted at our Unit. The patient, in full-health and not submitted to any treatment, presented a deviation of the tongue toward left (Fig. 2), associated with atypical swallowing and slurred speech, which had occurred for one year, after the insertion of a mobile orthodontic device. These symptoms still persisted during admission, after orthesis removal. At a phoniatric counseling, tongue deviation, fasciculation, atypical swallowing and dyspraxia were observed, without muscle atrophy. Speech therapy was therefore initiated but without benefit. At physical examination, adeno-tonsillar hypertrophy was observed, and, beside the tongue deviation, the neurological exam was normal. Routine laboratory exams (blood count, inflammatory markers, liver and kidney function, anti-streptolisine titer, thyroid hormones, EBV, CMV, Toxoplasma) were normal.

The electromyography (EMG) of the genioglossus muscles and the nerve conduction velocity (NCV) of hypoglossal nerves showed a motor axonal neuropathy of the left hypoglossal nerve. In particular, at EMG, a spontaneous activity and positive sharp waves were noticed only in the left genioglossus muscle. At a maximum contraction, the recruitment pattern of the same muscle was markedly reduced. The NCV of the right hypoglossal nerve showed normal distal latency (1.9 milliseconds), and a normal amplitude after mandibular-angle stimulation (2 mV). In the left nerve, latency was normal (1.7 ms), while the amplitude after mandibular-angle stimulation was reduced [0.1 mV (-95%)].



Fig. 2 – The present patient, at admission (10 years of age). The tongue is markedly deviated to the left as a consequence of left HNP.

At brain MRI, an asymmetry of the size of the vertebral arteries (left larger then right) was observed; the left hypoglossal nerve ran posteriorly to the left vertebral artery, and there were not any pathologic impregnation areas after CM. In order to exclude a neurovascular conflict, MRI was performed including cervical spine with vascular sequences, with normal results; moreover, a reduction of volume and a reduced enhancement after contrast of the left side of the tongue was observed.

After a 1-year speech therapy, the left-deviation of the tongue was reduced, even if the tongue, in its left side, appears to be still partially contracted (Fig. 3). The patient can speak properly and has not present anymore difficulties in swallowing. A new EMG of the muscle has showed a marked improvement of EMG (increase in recruitment pattern of the genioglossus muscle) and NCV of the left hypoglossal nerve, whose amplitude potential after mandibular angle stimulation had increased to 1.2 mV.

3. Discussion

The patient was admitted to our Unit for a left-deviation of the tongue, associated to atypical swallowing and slurred speech. Her medical history revealed an orthodontic appliance about 12 months before and her clinical symptoms had their onset few days after this procedure, without history of trauma nor signs of other neurological disorders. In the first months, the girl's symptoms had been overlooked, as it was thought that the fasciculation were a normal effect of the orthesis.

At admission, the general and neurological conditions and laboratory examinations were normal, thus an isolated lesion of the hypoglossal nerve was suspected. EMG – VCN confirmed a motor axonal neuropathy and a brain MRI with vascular sequences excluded intracranial masses, vascular disease or trauma.

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