



Case Reports & Case Series

The neck-tongue syndrome following cervical whiplash injury[☆]

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ABSTRACT

Background: Neck-tongue syndrome is a disorder causing unilateral neck pain, accompanied by simultaneous ipsilateral numbness of the tongue. It is explicable by compression of the second cervical root in the atlantoaxial space on sharp rotation of the neck. It may be secondary, idiopathic, familial, more frequent in females, in young people and the pain lasts from a few seconds to a few minutes. Treatment is symptomatic but also prophylactic, benefit is made by physiotherapy treatment. International Headache Society (IHS) places it between “Painful lesions of the cranial nerves and other facial pain”, describing as sudden onset of pain in the occiput or upper neck associated with abnormal sensation in the same side of the tongue.

Methods: A 42-year-old female, who a month after reported a whiplash injury, following a car accident, manifested the symptoms of neck-tongue syndrome. The rotational movements of the head to the right side provoked intense pain in the neck and along the right half of the tongue. The pain also appeared spontaneously, duration varied from a few seconds to few minutes, associated with burning, frequency of about two episodes per day. Radiography and cervical spine MRI showed signs of spondylosis.

Results: Topiramate combined with non-steroidal anti-inflammatory drugs, improved pain both in the neck and the tongue, reducing the episodes in frequency and intensity.

Conclusions: Neck-tongue syndrome is a rare disorder, which manifests with pain in the neck and pain associated with burning of half of the tongue, by unknown etiology. Secondary forms following whiplash injury have not been described, Treatment of these secondary forms concerns use of symptomatic drugs for pain, both also anticonvulsants as prophylaxis.

1. Introduction

Neck-tongue syndrome is a rare headache disorder provoked by sudden rotation of the neck, causing unilateral neck and/or occipital pain and ipsilateral tongue sensory disturbance. Described by Lance in 1980, NTS was found in young subjects and associated with Chiari I-type malformation [1]. International Headache Society (IHS) has placed it between “Painful lesions of the cranial nerves and other facial pain”, describing as “sudden onset of pain in the occiput or upper neck associated with abnormal sensation in the same side of the tongue” (2). The rapid rotation of the neck causes compression of the second cervical root in the atlantoaxial space. Numbness of half the tongue is caused by compression of the second cervical root that receives afferents fibers from the lingual nerve, travelling through the hypoglossal nerve [1,2]. Treatment of pain is usually with either non-steroidal anti-inflammatory drugs (NSAIDS) like indomethacin or anti-convulsants like gabapentin. Blockage of C2 nerve root can be considered useful in

severe cases. We describe a rare case of neck-tongue syndrome, following cervical whiplash in a female, describing clinical symptomatology and treatment. We found no similar cases in literature.

2. Case Report

We describe the case of a 42-year-old female, who a month after reported a whiplash injury, following a car accident, manifested the symptoms of neck-tongue syndrome. The rotational movements of the head to the right side provoked intense neck pain and along the right half of the tongue, where it was associated with burning. The pain also appeared spontaneously, the duration varied from a few seconds to few minutes, frequency of about two episodes per day. Neurological examination was normal except for neck pain during head movements. Radiography and cervical spine MRI showed signs of spondylosis (Figs. 1–2). Oral-dental examination showed no alterations, except for a flush of the right half of the tongue (Fig. 3). The patient brought a cervical collar for about two

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Fig. 1. Cervical radiography in lateral projection showing signs of spondylosis prevalent on the first three cervical vertebrae.



Fig. 2. Sagittal cervical MRI in weighted T1 images showing signs of a dominant spondyloarthrosis on the second, third and fourth cervical vertebrae.

weeks. The administration of topiramate at the daily dose of 100 mg, combined with non-steroidal anti-inflammatory drugs (NSAIDs), improved pain both in the neck and the tongue, and at present the episodes have been reduced in frequency and intensity.

3. Discussion

The neck-tongue syndrome (NTS), consists of neck pain and altered sensation in the ipsilateral half of the tongue, caused by rapid neck movements. It has been attributed to damage to lingual afferent fibers going through the hypoglossal nerve to C2 spinal roots. Proprioceptive



Fig. 3. Slight reddening of the lateral margin of the right half of the tongue.

fibers from the tongue enter the central nervous system through the second cervical dorsal root by lingual-hypoglossal nerves connections and between the latter and the second cervical root [1]. Therefore, for this reason, C2 root is compromised by sudden rotation of the neck, and compression is more severe when subluxation of the atlantoaxial joint occurs [1,9]. The abnormal sensation in the ipsilateral side of the tongue can be numbness, paraesthesias or the sensation of involuntary movements. In neck-tongue syndrome, pain from the upper part of the neck sends signals towards the brain, interpreted as coming from the tongue. NTS may be defined as secondary, for example related to trauma, idiopathic, or hereditary. Some studies show the prevalence of NTS in female (56%) and onset at a young age (16%) [2]. Regarding the appearance at a young age, it may be that the existence of ligamentous laxity, during growth and development, may facilitate transient subluxation of the atlantoaxial joint, leading to sudden head turning. As secondary form, a case of neck-tongue syndrome with lingual pseudoarthrosis, related to an atlantoaxial osteoarthritis of tuberculous origin, is reported [3]. Familial cases suggest a genetic predisposition in some individuals [4]. Lewis studied eight patients, five teenagers and three adults, affected by neck-tongue syndrome. Each of the five adolescents had normal examinations and normal neuroimaging. The three adults were parents of the affected children and had experienced transient symptoms during their adolescence suggesting an autosomal dominant inheritance pattern [5]. NTS can be treated with chiropractic procedures even if the opinions are controversial (worsening of symptoms?). Roberts treated a female patient with spinal manipulation, myofascial release, and home exercises. After 2 weeks, she was symptom free. At the 2-year follow-up, the patient remained free of symptoms [6].

It seems paradoxical but NTS responded favorably to a course of chiropractic care. Obviously, other authors affirm the importance of immobilization with cervical collars, or physical therapy [7,10].

Pharmacological-prophylactic therapy with anticonvulsants, such as arthrosic, seems to be the most effective [8]. With regard to the case described, we hypothesize that the cervical whiplash, in an arthritic spine, may have triggered the syndrome. However, in our case the prognosis was good, having produced the pharmacological therapy a definite improvement of the symptoms.

4. Conclusions

Neck-tongue syndrome is a rare disorder, which manifests with pain in the neck and pain associated with burning of half of the tongue, by unknown etiology. Secondary forms related mainly to cervical spine traumas have been described, but also idiopathic and familial forms. Treatment is pharmacological-prophylactic. In this work we describe a case of NTS secondary to cervical spine whiplash injury, making a brief dissertation on the few statistics about the neck-tongue syndrome, its physiopathology and treatment.

Statement

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