

Unilateral pure trigeminal motor neuropathy and neurovascular conflict: More than a coincidence!

ABSTRACT

Sensory disturbances are usual manifestations of trigeminal neuropathy, but only a few cases of isolated unilateral pure trigeminal motor neuropathy were reported. We describe a rare case of a 65-year-old woman who presented with a 4-year history of progressive facial asymmetry and chewing weakness which was shown on imaging to be caused by unilateral atrophy of masticatory muscles, probably due to a neurovascular conflict between the third trigeminal branch and superior cerebellar vein. To the best of our knowledge, this probable association has never been described. We aimed through this article to describe the clinical presentation of this entity that clinicians might face in their daily practice and radiological aspect of it.

Keywords: Atrophy, conflict, motor, neuropathy, neurovascular, trigeminal

INTRODUCTION

The trigeminal nerve is the largest cranial nerve which serves both sensory and motor functions. Sensory disturbances are usual manifestations of trigeminal neuropathy, but only a few cases of isolated unilateral pure trigeminal motor neuropathy (UTMN) were reported since the first description by Chia in 1988.^[1] We present a patient with unilateral weakness and muscle atrophy which were not accompanied by trigeminal sensory involvement.

CASE REPORT

We report the case of a 65-year-old woman, with a history of type 2 diabetes and high blood pressure, who presented to the neurology consultation with facial asymmetry, left masticatory weakness, and muscle wasting in the left temporal area and cheek that has been progressing for 4 years without preceding pain, febrile illness, head trauma, stroke, or systemic symptoms. Neurological examination showed weakness and hypotrophy of the left temporalis and masseter muscle giving a sunken appearance in the cheek area, weakness of the jaw-closing muscles, and opening deviation [Figure 1a]. No abnormality was detected in the


functions of the temporomandibular joint. The patient's deglutition, tongue motion, corneal reflex, and facial sensation were normal. Electromyography (EMG) showed that the motor action potential was absent without any abnormal spontaneous activity and with a decreased recruitment pattern and chronic neurogenic motor unit potential changes in the affected masseter and temporalis muscles [Figure 1b], these remained normal in the frontalis, orbicularis oculi, orbicularis oris, sternocleidomastoid, and tongue muscles. Those results were in favor of trigeminal motor denervation.

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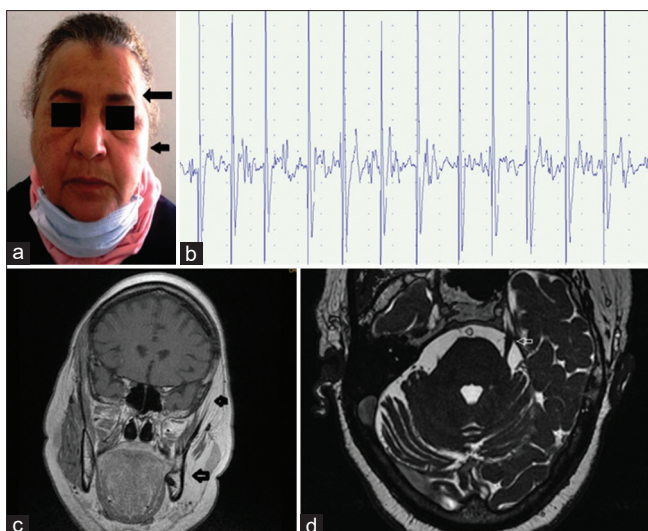


Figure 1: (a) Photograph of the face of the patient showing severe muscle wasting on the left side above and below the zygoma (black arrows), (b) Neuropathic trace in temporalis left muscle on electromyography(EMG), (c) Brain magnetic resonance imaging(MRI) of the face showing atrophy and fat degenerating of temporalis and masseter (black arrows), (d) Cerebral Brain magnetic resonance imaging(MRI), axial inframillimetric T2 weighted image showing neurovascular conflict between third trigeminal branch and superior cerebellar vein (white arrow)

Brain magnetic resonance imaging (MRI) of the face and head revealed severe atrophy and fat replacement of the left trigeminal motor nerve innervated muscles (temporalis, masseter, medial, and lateral pterygoid) [Figure 1c] and showed that the superior cerebellar vein crossed in a right angle of the trigeminal branch in the root entry/exit zone (REZ) with deformation of the nerve [Figure 1d]. Based on muscle denervation atrophy in the distribution of the third branch of the trigeminal in the absence of sensory symptoms or deficits, the patient's presentation was consistent with trigeminal motor neuropathy. Physiotherapy and regular follow-up were recommended; the patient refused the reconstructive surgery. However, there was no clinical improvement.

DISCUSSION

Pure UTMN is a very rare disease characterized by trigeminal motor paralysis without trigeminal sensory or any other cranial nerves disturbances. The clinical manifestation varies with the chronicity of the process, facial asymmetry, pain, weakness in chewing, and opening deviation.^[2] UTMN is due to a lesion anywhere along its course from the pons to distal peripheral nerve-innervating muscles. Diagnosis of this condition involves clinical neurological examination, radiological evaluation (MRI), and EMG. MRI typically shows a replacement of the muscle tissue by fat tissue, also reported in our case and guided the etiological diagnosis.^[3] There is no definite etiological factor associated with UTMN, it might have been idiopathic, although there are reports of its association with underlying disorders

such as a tumor, pontine infarction, viral infection, neoplastic, or head traumatic events.^[4] To the best of our knowledge, UTMN due to neurovascular conflict has never been described. Neurovascular contact is frequent imaging in asymptomatic patients, and the diagnosis of neurovascular contact can be retained as symptomatic when the following criteria are fulfilled: right angle crossing with direct vessel-nerve contact in the REZ, nerve displacement/deformation or atrophy, and concordant clinical and radiographic presentations, which was the case in our patient.^[5] The association between clinical and radiological presentation was strongly suggestive that the neurovascular conflict can be retained to be the cause of UTMN in our case report. There is no effective treatment for UTMN, and the management of underlying pathological causes such as a tumor may include surgical intervention. In patients where no etiology is confirmed, the treatment includes reassurance and physiotherapy, although some attempts have been made to surgically correct the disability to improve the mouth opening.^[2,3]

In conclusion, we have reported a rare case of left UTMN in a 65-year-old woman patient, in whom the muscles innervated by the left trigeminal motor nerve had undergone atrophy and wasting of the muscles. The MRI showed a neurovascular conflict between the superior cerebellar vein and trigeminal branch, meeting the criteria of symptomatic neurovascular conflict. This situation is unusual and leads us to think on the real relation between the neurovascular conflict and UTMN, is it a coincidence or more than that, and if the neurovascular conflict is present in some asymptomatic patients what are may be the risk of developing a UTMN. We believe that when a patient presents for a UTMN, that may be the only expression of some severe conditions such as strokes and tumors or inflammatory diseases, physicians must pay attention of the possibility of symptomatic neurovascular conflict between the superior cerebellar vein and trigeminal branch in their investigations before retaining the idiopathic origin.

Informed consent

Informed consent was obtained from the patient.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that name and initials will not be published and due efforts will be made to conceal the identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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