LETTER TO THE EDITOR

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A rare presentation of neuralgic amyotrophy: Hypoglossal neuropathy

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Sir,

In this letter, we highlight a rare presentation of neuralgic amyotrophy.

A 25-year-old man presented with difficulty in speaking of 3 months duration. Symptoms started in June 2021, with deviation of the tongue and difficulty in articulation. Over next few days, he noticed right shoulder pain and difficulty in overhead activities. He was evaluated and treated and had partially improved when he was seen at our center, 2 months into the illness. The patient reported significant improvement in pain and the right shoulder weakness, but speech had largely remained unchanged. On examination, he had severe wasting of the right half of tongue [Figure 1a], without fasciculations and right arm abduction weakness [Figure 1b] with mild winging of right scapula. Other cranial nerves were normal. Sensations in the affected dermatomes were normal. Sensory and motor nerve conduction study was normal. Needle examination did not reveal spontaneous activity (including the right genioglossus muscle). There was no evidence of active or chronic denervation. Magnetic resonance imaging brain was normal including brainstem and meninges. Based on the possibility of neuralgic amyotrophy, steroids were gradually tapered. On follow-up, the bulk of the tongue is better with no new complaints.

Our patient presented with tongue weakness and atrophy. This was followed rapidly by pain, and then weakness and wasting in right proximal arm musculature. Acute onset of pain followed by wasting and atrophy raised the possibility of neuralgic amyotrophy. Classic form of neuralgic amyotrophy involves the brachial plexus with frequent involvement of suprascapular, axillary, and long thoracic nerves.[1] Cranial nerve involvement at the onset is distinctly uncommon and has been described rarely.[2] Margaux Genevray and colleagues also described hypoglossal nerve involvement in neuralgic amyotrophy, like the present case.[3] The awareness of cranial neuropathies as a presenting feature of neuralgic amyotrophy is important for timely and effective management.

The normalcy of electrophysiology was unusual in the present case. We tested the patient in the 3rd month of illness when he had much recovered, which may explain the normal findings. Proximal electrical stimulation may have detected proximal conduction block, but the data were not available. Although neuralgic amyotrophy is considered predominantly an axonal process, literature shows evidence for focal proximal demyelination in the disease process.[4]

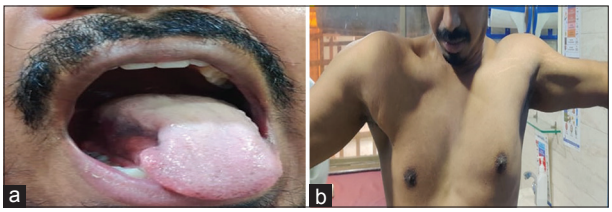

Figure 1: (a) Asymmetric tongue wasting (b) right arm abduction weakness

In conclusion, we report the unusual occurrence of hypoglossal nerve involvement in neuralgic amyotrophy, for awareness of this rare situation as early recognition and therapy is important in the overall prognosis.

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