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Unilateral isolated hypoglossal nerve palsy: A rare complication of Diabetes Mellitus

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Abstract

We report a middle-aged male who presented with sudden onset slurring of speech with polyuria and polydipsia of three weeks duration. Evaluation suggested unilateral hypoglossal nerve palsy. The patient was detected to have uncontrolled hyperglycemia and exhaustive workup for possible alternative etiology including connective tissue disorders, retropharyngeal infection, hypothyroidism, thrombosis of median branches of vertebral artery, lacunar infarct over the hypoglossal nerve nucleus, multiple sclerosis, epilepsy, malignancy etc. was unrewarding. Unilateral isolated hypoglossal nerve palsy not only unmasked his diabetes, but also got corrected following glycemic control. The case report exemplifies a relatively infrequent clinical manifestation of diabetes, thereby reiterating that although unilateral isolated hypoglossal nerve palsy is rare, nevertheless warrants thorough evaluation before being labelled as "idiopathic" and diabetes remains one amongst various potentially reversible medical causes.

Keywords: unilateral isolated hypoglossal nerve palsy, diabetes mellitus

Introduction

A forty-five years old male was admitted with sudden onset slurring of speech, lasting for about an hour. This was preceded by three weeks history of polyuria and polydipsia along with significant and unintentional weight loss of six kilograms. There was no history of preceding head injury, nausea, vomiting, headache, blurred vision, diplopia, dysphagia or altered sensorium. The patient denied history of any limb, truncal or neck muscle weakness, rash over the body or fever. On further questioning, patient denied difficulty in mastication or swallowing. Family history was not significant. Clinically the patient was afebrile, normotensive and had dysarthria, with a score of two on the NIH stroke scale. He could not repeat "Fifty-Fifty," but was able to repeat "Mama" and "Tip-Top", suggesting dysarthria secondary to difficulty in tongue movement. He had a boggy tongue, which on protrusion deviated to right side (Figure 1), due to unopposed action of the unaffected left side. There was no wasting or fasciculations on the right side of his tongue and rest of the neurological examination was unremarkable. During laboratory workup, he was diagnosed with uncontrolled diabetes mellitus (HbA1C-9.5%). Rest hematological and biochemical profile was normal. Viral markers (HIV, HBsAg and HCV) were negative by Enzyme linked immunosorbent assay and work up for target organ damage revealed bilateral nonproliferative retinopathy. Diffusion-weighted magnetic resonance imaging (MRI) brain & MR angiography (intracranial as well as extracranial neck vessels) was unremarkable without any parenchymal signal abnormality in either the brainstem or the cerebellar hemispheres. Further exhaustive workup for possible alternative etiology antibody, anti-neutrophil cytoplasmic (anti-nuclear factor, C-reactive antibody, rheumatoid protein, complements, thyroid function test, ACE level, vitamin B12, folate, serum electrophoresis, cerebrospinal fluid analysis and PET-CT) was unrewarding. The patient was

managed with insulin (basal – bolus regime) and other supportive measures. His tongue weakness and dysarthria improved, concurrent to glycemic control and subsequent follow up for a year didn't reveal recurrence of his symptoms thereafter.

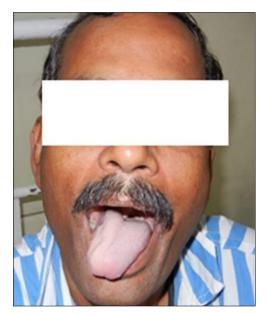


Fig 1: Boggy tongue deviated to right side on protrusion

Discussion

Unilateral isolated hypoglossal nerve palsy (HNP) remains relatively uncommon diagnostic challenge ^[1], with postoperative cases being the most common etiology followed by neoplastic and idiopathic causes ^[2].

Medical causes although rare, nevertheless include connective tissue disorders, secondary vasculitis, retropharyngeal infection, acute poliomyelitis, thrombosis of median branches of vertebral artery, vertebral artery dissection ^[3], lacunar infarct over the hypoglossal nerve nucleus, multiple sclerosis, epilepsy ^[4].

Although isolated palsy of peripheral or upper cranial nerves in diabetic patients is well documented, but isolated unilateral HNP^[5] is rare and tongue weakness unfolding diabetes further adds to the novelty. Pathophysiology appears multifactorial and include duration of hyperglycemic exposure, elevated lipids, blood pressure, smoking and concurrent exposure to other potentially neurotoxic agents such as ethanol^[6].

In absence of plausible alternative aetiologias, transient idiopathic ^[7] HNP comparable to Bell's palsy, may be a convenient differential in index case, nevertheless retrospectively in background of evident uncontrolled diabetes, subsequent improvement following glycaemic control and applying principle of Occam's razor, it may be conceivable to attribute isolated unilateral HNP in index case to uncontrolled diabetes.

Learning points

- Unilateral isolated hypoglossal nerve palsy is a rare entity and requires thorough evaluation before it is labelled as "idiopathic."
- Tenable etiology includes "3Ts"; trauma, tumors and thrombosis of vertebral artery.
- Diabetes is one of the reversible medical causes of isolated unilateral hypoglossal nerve paralysis, besides connective tissue disorders, secondary vasculitis and retropharyngeal infection.

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