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A Rare Presentation of Persistent Idiopathic Isolated Unilateral Hypoglossal Nerve Palsy

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Abstract

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Introduction

The hypoglossal nerve is the 12th cranial nerve and is composed purely of motor nerves.

Hypoglossal nerve palsy (HNP), often sustained as a complication of nerve injury in head and neck surgery, may also occur due to stroke, tumors, or neurological diseases. It frequently occurs in combination with glossopharyngeal, vagus, and accessory nerve palsy, (1)

Isolated unilateral hypoglossal nerve palsy (IUHNP) is a not so common condition which needs to be investigated extensively as it is usually a clinical manifestation of an underlying disease. Signs of hypoglossal nerve palsy (HNP) include, absence of power and movement on the same side, leading to deviation of the tongue to the affected side, fasciculations, and at a later stage lead on to atrophy. Dysarthria is most common, while a few patients complain of dysphagia [2]. Idiopathic IUNHP is indeed less frequent. In some cases, a diagnosis of idiopathic IUHNP was postulated as even after extensive investigations, the cause remained unknown, [2–7]. Most reported cases are self-limiting, and only a few are of persistent idiopathic IUNHP [5,6]

Case Presentation:

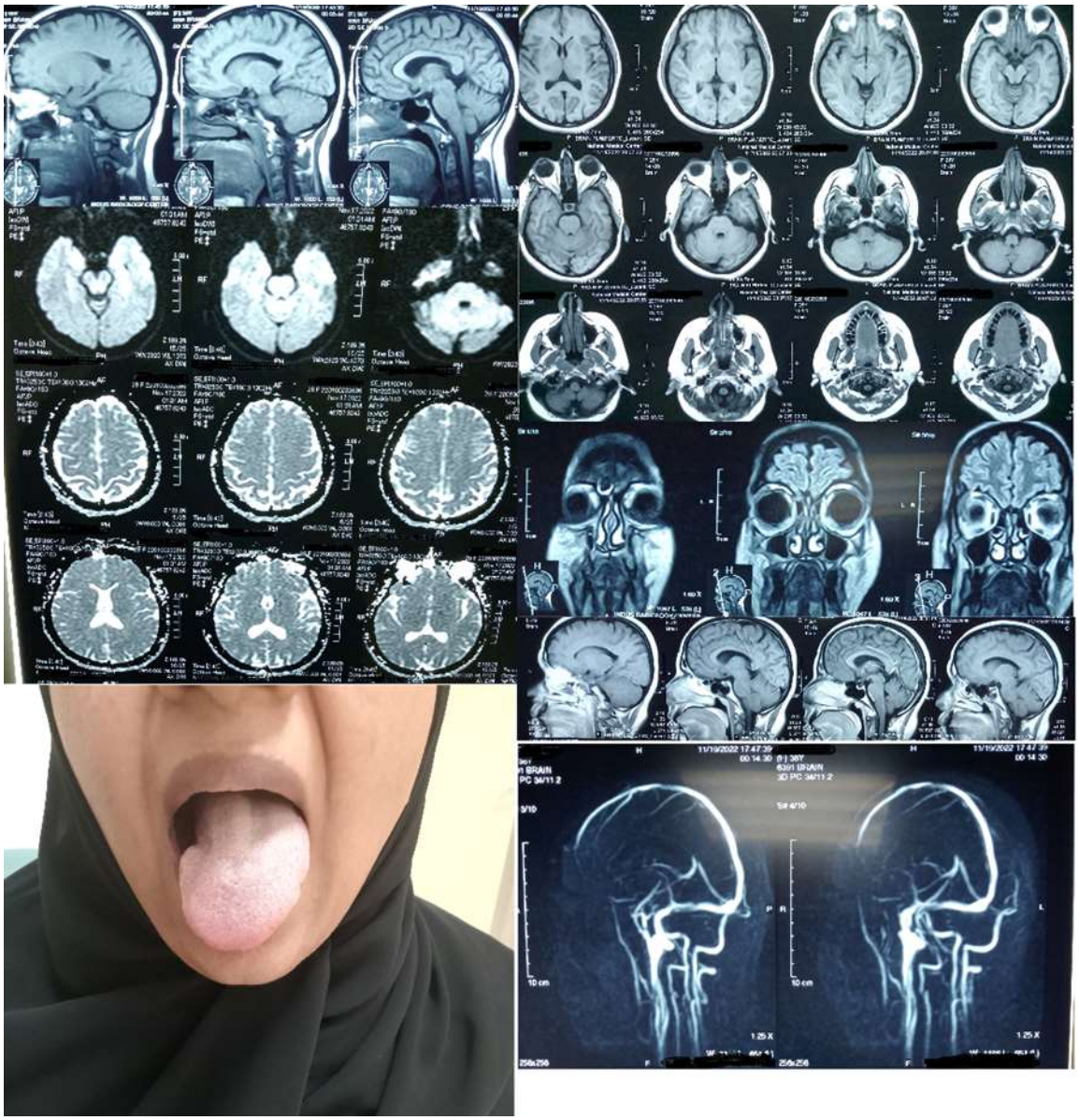
37 years old female, presented to our clinic with two months history of deviation of tongue. She was initially admitted in regional hospital with history of nausea, vomiting and epigastric pain for two weeks duration, with no other positive symptoms.

During the course of admission, she developed sudden onset weakness of the right side of tongue with marked deviation of the tongue, with intact sensation over the tongue. No other significant medical history. On examination, she had significant deviation of tongue to right, with minimal fasciculations and other intact cranial nerve functions.

Thorough otorhinological and ophthalmic examination were normal. Blood tests including CBC, renal, liver, lipids, electrolytes, coagulation profile, cardiac enzymes were within normal limits. TSH mildly reduced 0.142 with normal free T3/T4 levels, cortisol mildly elevated 1150. Viral markers, Antinuclear antibodies, anti-TTG were negative. Subsequently imaging studies performed, MRI brain plain and contrast – showed normal study and MRI DWI, MRV were normal. Ultrasound thyroid and color doppler study showed normal carotid and vertebral flow and a small nodule 0.8x0.9 nodule in right lobe thyroid, with no significant cervical lymphadenopathy. Echocardiography showed normal bilateral ventricular function.

Esophagogastroduodenoscopy revealed gastroduodenitis and Gr1 hiatus hernia, negative for H.Pylori infection. Patient was seen after 6 months, she had persistent deviation of tongue with marked atrophy over the right side of tongue. Normal swallowing and speech. She was reviewed at the end of 15 months, she had mild improvement with a residual atrophy over the affected side of tongue, otherwise no other focal deficit.

A diagnosis of Persistent Idiopathic Unilateral Hypoglossal Nerve Palsy was made., and since she presented late, oral steroids was not offered to her and speech therapy, general supportive measures provided.



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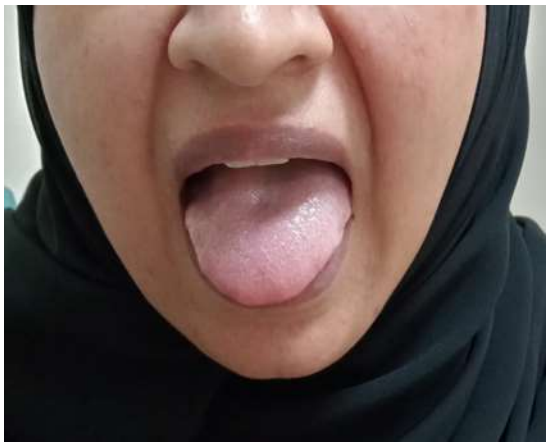

Discussion:

HNP predominantly appears together with other neurological abnormalities in a variety of diseases. A lesion in the brainstem generally involves other nuclei or tracts, and a lesion in base of skull most often affects other cranial nerves simultaneously , with tumors being the predominant underlying cause of HNP [9], Vertebral artery dissection, nasopharyngeal carcinoma, Arnold- Chiari malformation, and Dural arteriovenous fistula are other known causes to be considered [2]. Extensive clinical assessment is needed in view of varied etiological causes that could be expected with a case of HNP, specifically when assuming a diagnosis of idiopathic IUHNP.

Idiopathic IUHNP in spite of being a less reported condition , is more frequent than previously thought [1,7]. About 13 single case reports were documented by Ahmed and colleagues [4]. Ho and colleagues described a case of idiopathic IUHNP that lasted over 5 years and suggested a scheme of investigations to arrive the diagnosis of idiopathic IUHNP by way of exclusion [6]

It is relatively difficult to localize hypoglossal nerve lesions with neurological examination, hence imaging studies along the entire course of the nerve is necessary. Plain and contrast studies of CT and MRI are needed. Gadolinium enhanced MRI sequences are optimal in identifying most hypoglossal nerve pathologies. In case of CT imaging, contrast images greatly enhance the visualization of nearby vasculature and tumors. CT imaging is complementary to MRI in the skull base segment. [8,9]. Infrequently, this condition can be an early clinical manifestation of an underlying demyelinating disease, and hence, it is recommended to do a repeat brain MRI scan every 3 to 5 years [4, 6].

Most cases are reversible without any treatment and resolve within 2 to 4 weeks, however longer recovery periods between 2 to 5 months have been reported [2, 3, 7]. Two cases of idiopathic IUHNP reported persistence of symptoms for a period of 3 years and 2 years respectively [5, 10].

The patients with isolated IUHNP fall in a varied age group. [3].

Idiopathic IUHNP is a clinical entity similar to Bell's palsy (BP) as suggested by Lee and colleagues.[7]. The likelihood of recovery of the facial nerve increases with the use of oral steroids (OS) within 72 h of symptom onset in BP [11]. Oral Steroids have been used to treat idiopathic IUHNP in some case reports, but there are no clinical guidelines available to substantiate its use.[3]. As our patient was referred to us 2 months after symptoms onset, with already visible tongue atrophy and fasciculation, we did not initiate steroid therapy. Speech and language impairment are relatively common in HNP, hence ST is routinely used as a rehabilitation method [6]

Conclusion:

IUHNP is a rare diagnosis with a challenging clinical presentation with numerous differentials. We recommend that physicians should use a methodical approach to identify the cause and early appropriate referrals for targeted interventions.

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