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CASE REPORT

Isolated right hypoglossal nerve palsy- A case report

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ABSTRACT:
We report a 46 year old woman who developed isolated right hypoglossal nerve palsy two weeks after traditional uvulectomy. Radiological and other general haematological investigations were normal. Isolated unilateral hypoglossal nerve palsy is rare. Thorough physical examination and meticulous investigation is advocated where available.

Key words: isolated, hypoglossal nerve palsy, unidentified cause

Introduction

The nuclei and fascicles of the hypoglossal nerve (12th cranial nerve) are located in the medulla. The nerve exits the cranium via the hypoglossal foramen to innervate the tongue musculature. Although hypoglossal nerve palsy received inconspicuous attention in most standard text books,[1] several aetiological factors have been attributed to it. This include injuries to the head and neck, vascular injuries, space occupying lesions, autoimmune diseases or idiopathic.[2] Clinically, patient may present with lingual dysarthria, atrophy and fasciculation of tongue musculature and deviation of the tongue to ipsilateral side on attempt at protrusion. A patient with isolated right hypoglossal nerve palsy is presented in this report with highlights on challenges of thorough investigation in a developing economy.

CASE SUMMARY

A 46-year old woman was enjoying apparent good health until a year prior to presentation when she developed dysarthria and deviation of the tongue to the right side on protrusion. She had initial difficulty with swallowing which she overcame within short time. Her symptoms were preceded by traditional uvulectomy two weeks earlier, indication for which was not clear but claimed to be due to swollen uvula. There was no history of trauma, stroke or surgery. Examination revealed an otherwise fit woman, with an amputated uvula and deviation of the tongue to the right side (fig 1) with some degree of muscle atrophy.

Fig. 1. Atrophy of the right side of the tongue and deviation to the right on protrusion.

General haematological investigations (full blood count & erythrocytes sedimentation rate) were normal. Retroviral screening was negative and chest radiograph was within normal. Cranial Computerized tomographic (CT) scan did not reveal any ischaemic or space occupying lesion.

Fasting blood glucose (FBG) and electrolyte,

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urea & creatinine (e/u/c) were normal. At the time of writing this report, we do not have facilities for virological studies like anti cytomegalovirus (CMV), Epstein-Barr virus etc. These serological tests would have given a clue for possible viral infection or exposure. Electrophysiological evaluation like electromyography (EMG) was also not done due to non availability of equipment; EMG will give objective assessment of return or otherwise of muscular electrical activity which is useful in prognostication.

She was placed on multivitamins and has been receiving physiotherapy. As at last visit, there was clinically appreciable improvement in the tongue muscle bulk and deviation. She also said there has been significant improvement as regards the dysarthria.

Fig.3. Six months of physiotherapy: Note the improvement in muscle bulk and tongue deviation on protrusion.

DISCUSSION

The hypoglossal nerve (XII) takes its origin from a nuclear column located in the floor of the IVth ventricle and is derived from the same cell group as the nuclei of nerves III, IV and VI. Its fascicular fibres traverses the full sagittal diameter of the brainstem exiting from the ventral surface of the medulla between the pyramid and olive.[3] Although the nucleus consist of four distinct subnuclear columns, its motor composition is not clearly understood owning to its complexity.[4] Isolated unilateral hypoglossal nerve palsy is rare. As an entity, its occurrence therefore is more often than not attributable to lesions at the supranuclear, nuclear or infranuclear levels. Blomquist and co-workers found ultrasonic assessment unremarkable but MR imaging/MR angiography useful in hypoglossal nerve palsy due to Dural Arteriovenous fistula.[5] In our patient cranial CT scan did not reveal any lesion. MRI would have been considered for better soft tissue delineation if there has been the slightest suspicion of soft tissue lesion on the CT.

This rare condition has varied causes which ranges from trauma through cervical vertebra dislocation, carotid aneurysm, intracranial neurolemoma and infectious mononucleosis to vaccination.[6,7,8] Multiple sclerosis, hysteria, Guillain-Barre neuropathy, stroke, surgery and infection have been reported as other causes in a review of 26years experience in which up to 3% of cases were idiopathic.[9] In our patient, although no virological investigation was carried out, we couldn't establish a cause. Careful clinical history and thorough systematic physical examination is mandatory in evaluating a patient with isolated hypoglossal nerve palsy. In this index case, absence of positive history and negative loco-regional features ruled out neck surgical procedure, parapharyngeal/ retropharyngeal infections and skull base metastatic disease. Ho[10] and co-workers developed Potential pathway of investigations in a patient presenting with isolated hypoglossal nerve palsy depending on clinical assessment as follows: Basic haematological: Full blood count, Erythrocyte sedimentation rate and C-reactive protein. Special haematological (immunology tests): rheumatoid factor, complement. Serology for infectious agents: Herpes simplex, Epstein-Barr virus, Cytomegalovirus. Radiology: CT, MRI, CXR (to exclude tuberculosis). CSF analysis may also be helpful.

Uvulectomy is the rarest throat surgery mentioned in the early medical literature, it receives only brief attention in most modern standard text. This throat surgery is unusual in present day otorhino-laryngological practice. In some African countries like Kenya, Nigeria, Sierra Leone and Tanzania, traditional uvulectomy is carried out due to varied throat lesions by the traditional barbers. Children and adults are victims and the procedure is fraught with several health hazards including infections.[11] In a study of 385 children, Isa and co-workers reported that, the commonest influencing factor for traditional uvulectomy was prevention or cure of frequent throat infections.[12] In this case report, the patient had traditional uvulectomy one year prior to presentation, indication was not confirmed but she said the uvula was swollen. It is possible that it was throat infection which could be viral or bacterial that influenced her decision to undergo

the traditional procedure. On the other hand the procedure might have been complicated by infection which may have lead to the isolated nerve palsy. The reality remained unascertained since neither coherent history nor equipment for serological (virology) was available at the time of this report. There is paucity of literature on traditional uvulectomy globally, further research on this procedure especially in countries where this is still a common practice is essential.

Treatment of hypoglossal nerve palsy is essentially treatment of the cause, in idiopathic, isolated cases however a minority may resolve without specific treatment[13]. In our patient there has been significant improvement clinically although basic electrophysiologic assessment was not possible at the time of this write up. It has been reported that only about 15% of cases of hypoglossal nerve palsies recover fully[4].

In conclusion, isolated hypoglossal nerve palsy requires careful clinical evaluation in order to establish its aetiology. Practitioners in developing countries may be challenged by inadequate medical equipment.

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