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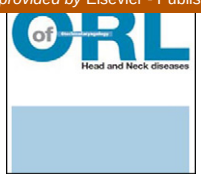

CASE REPORT

Isolated hypoglossal palsy due to cervical osteophyte

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KEYWORDS
Hypoglossal palsy;
Hypoglossal nerve;
Spondylosis;
Osteophyte

Summary
Introduction: Isolated hypoglossal nerve palsy is rare, and etiological diagnosis is difficult. We report a case of isolated hypoglossal compression by a cervical osteophyte in the hypoglossal canal exit.
Case study: An 86-year-old woman with history of cervical spondylotic myelopathy consulted for a lesion of the free edge of the tongue with impaired elocution. Clinical examination found a bite lesion on the right free edge of the tongue with right lingual amyotrophy and associated left deviation on retraction. Isolated right hypoglossal palsy was diagnosed. Skull base CT found a cervical osteophyte compressing the hypoglossal nerve at the exit from the right hypoglossal canal. Surgery was contra-indicated by the patient's general health status. No motor recovery was observed at 6 months' follow-up, but the elocution disorders regressed under speech therapy.
Conclusion: Hypoglossal palsy is infrequent, but generally a sign of skull base pathology. History-taking and careful examination guide rational selection of the radiological examinations required for etiological diagnosis.

Introduction

Cervical spondylosis is very widespread in the elderly. It is very often limited to neck pain, but may assume unexpected clinical forms according to its location along the cervical spine. We report a rare case of isolated hypoglossal compression by a cervical osteophyte in the hypoglossal canal.

Observation

An 86-year-old woman was referred for impaired elocution associated with a painful lesion of the free edge of the tongue of 2 weeks' evolution. She had no particular history except for cervical spondylotic myelopathy requiring C2—C4 laminectomy 2 years previously.

Examination of the mouth cavity found a bite lesion on the right free edge of the tongue with right lingual hemiatrophy and inability to move the tongue leftward, with rightward deviation on protraction (Fig. 1A), and leftward deviation on retraction (Fig. 1B). Neurological examination was otherwise normal. Isolated hypoglossal palsy (IHP) was diagnosed.

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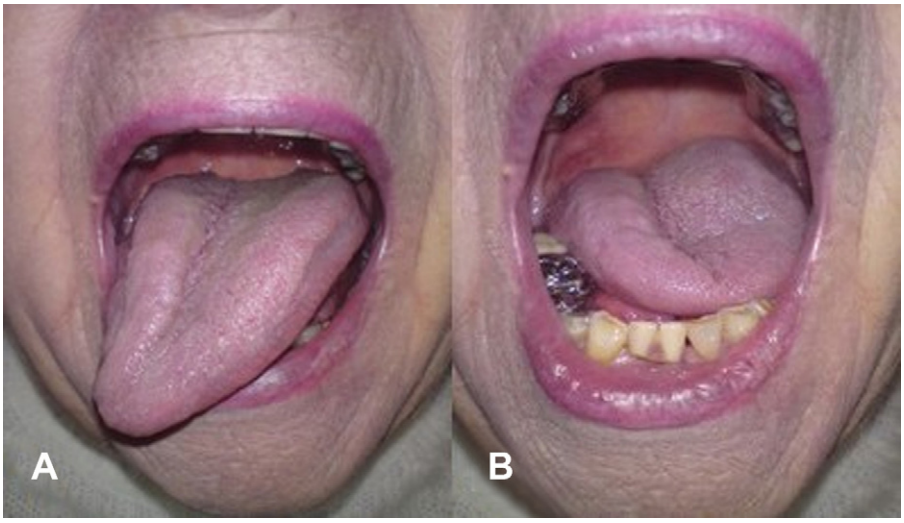

Figure 1 Right lingual palsy. (A) Right lingual deviation in protraction. (B) Left lingual deviation in retraction. Note right lingual amyotrophy, more visible in Fig 1B.

Skull base and neck CT found an osteophyte involving the occipito-atloid joint facing the external orifice of the right hypoglossal canal (Figs. 2 and 3). On neurosurgical advice, surgery was contra-indicated by the patient's age and the iatrogenic risk.

At 6 months' follow-up, following speech therapy, the patient no longer showed problems of elocution, but amyotrophy and impaired tongue mobility remained unchanged.

Discussion

The XIIth cranial or hypoglossal nerve is a purely motor nerve assuring lingual musculature innervation. It may suffer damage at any point along its trajectory from the myelencephalon to the lingual muscles. Its trajectory can be divided into five segments: medullary (nuclear), cisternal (extramedullary and intracranial), basicranial (hypoglossal canal), retro- and prestyloid (near to the medial and lateral carotids and Xth and XIth nerves), and submandibular (near to the lingual muscles). Hypoglossal palsy is thus seldom isolated but rather associated with palsy of other cranial nerves [1].

The literature reports multiple IHP etiologies. Skull base tumor (metastasis, or primitive malignant or benign tumor) is the most frequent [2]. Trauma is the second, comprising cranial trauma (occipital condyle fracture) sustained in road accidents and surgical trauma (neck surgery, prolonged cervical hyperextension during anesthesia). Thirdly, vascular etiologies notably comprise medial carotid artery dissection. Joint and auto-immune pathologies (cervical rheumatoid arthritis, occipito-atloid cyst) and occipital hinge deformity (Arnold-Chiari malformation) have also been implicated. In children and adolescents, infectious mononucleosis should be investigated [3]. Finally, in certain cases no etiology can be determined.

Clinical examination should systematically concern the paired cranial nerves, to rule out associated nerve damage. The Claude Bernard-Horner sign should be systematically looked for, being frequently associated with hypoglossal

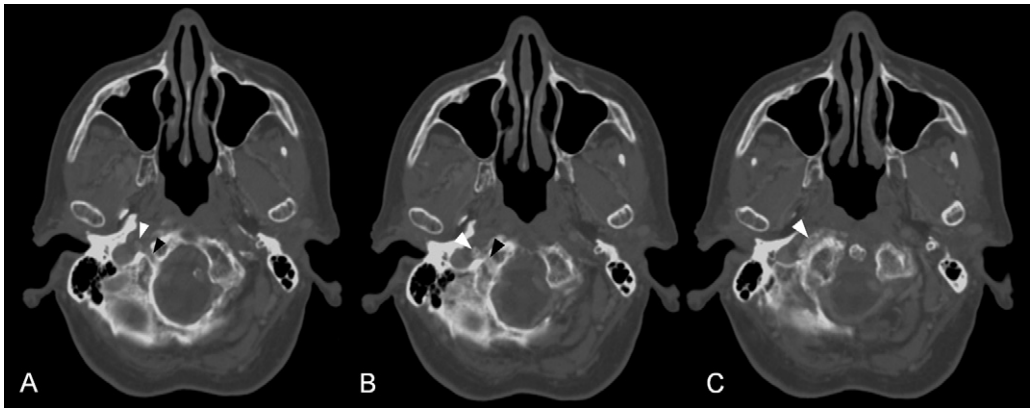

Figure 2 Basicranial CT, axial slices. (A) Black arrow: hypoglossal canal. White arrow: osteophyte. (B) Slightly lower slice, showing the osteophyte (white arrow) in the lower hypoglossal canal (black arrow). (C) Slice through hypoglossal canal. Note osteophyte protrusion (white arrow) into jugular foramen.

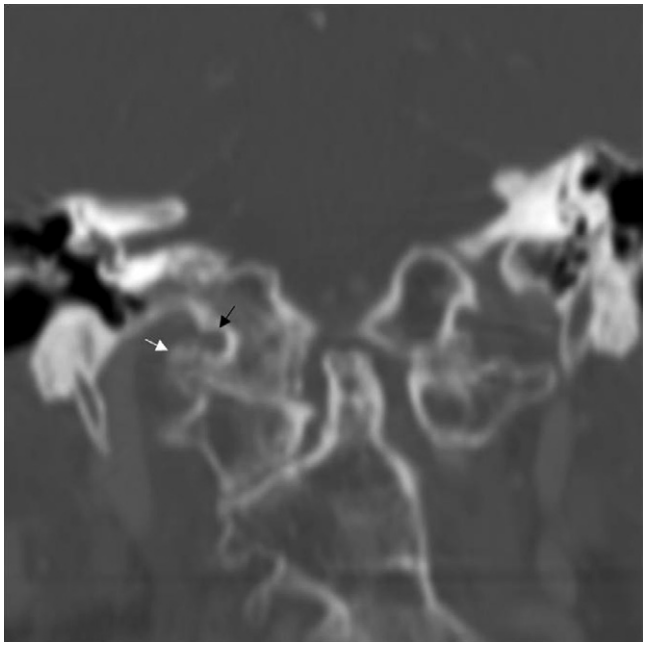

Figure 3 CT reconstruction of the cervico-occipital hinge, coronal slices. Note proximity of the osteophyte (white arrow) to the external orifice of the right hypoglossal canal (black arrow).

palsy in carotid dissection [4]. The other most frequently associated cranial pairs are X, VII and V [5]. Once isolated involvement has established a diagnosis of IHP, etiology is to be investigated.

Meticulous history taking is the key to etiological diagnosis in IHP. As the present case illustrates, the history of the disorder is primordial. Onset circumstances are to be examined for trigger factors (cervical trauma, recent surgery) and predisposition (cardiovascular history, arthropathy, systemic illness, malignancy), to guide paraclinical assessment.

Imaging should be systematic. Brain and skull-base MRI centered on the hypoglossal trajectory is the most frequent examination. In case of suspected carotid dissection, angio-MRI should be associated to explore for flame shaped arterial occlusion, double arterial lumen, or aneurysmal dilation of the medial carotid. Basicranial CT may be preferred, as in the present case, in case of suspected degenerative osteoarticular or cervico-occipital hinge trauma etiology [6]. It provides better evidence of osseous pathology of the hypoglossal canal or of condylar fracture. Serology for infectious mononucleosis should be prescribed in pediatric cases when radiological assessment is negative [3].

Prognostic hypoglossal electromyography may be used to assess neural recovery, which is directly related to the pathological process: most cases of idiopathic IHP or IHP secondary to mononucleosis show spontaneous regression within a few weeks or months [2], but tumoral, vascular and traumatic etiologies show variable and incomplete recovery, especially when amyotrophy is already established.

IHP management has two objectives: to treat the lesion mechanism, and to restore neurologic function. The latter often takes second place, partly because the lesional process (metastasis, basicranial tumor) has priority, and also because the impairment of swallowing and phonation induced by the paralysis is not severely disabling, even when it fails to regress. In many cases, treatment of the lesion (carotid dissection, cervical trauma) will also be effective in restoring neurologic function. In all cases, speech therapy focusing on swallowing and phonation should be considered; this was the treatment prescribed in the present case, in view of the patient's general health status and of the negative risk/benefit trade-off for neurosurgical intervention on the craniocervical junction. Such surgery is currently restricted to benign and malignant basicranial tumor, and is often complicated by chronic posterior headache or cranial palsy [7]; it may also threaten the vertebral artery.

Conclusion

IHP is infrequent and usually symptomatic of skull base pathology. To the best of our knowledge, the present case is the second to be published concerning isolated hypoglossal compression by osteophyte. History-taking and meticulous examination guide rational selection of the radiological examinations required for etiological diagnosis.

Disclosure of interest

The authors declare that they have no conflicts of interest concerning this article.

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