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Case report

Unilateral vagus nerve lesion revealing clival meningocele

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ABSTRACT

Introduction: Cranial nerve lesions can be secondary to a space-occupying lesion of the skull base compressing adjacent nerves.
Case report: We report the case of an 84-year-old man, who presented with rapid and concomitant onset of dysphagia and ipsilateral recurrent laryngeal nerve paralysis, suggesting an isolated lesion of the vagus nerve. MRI revealed a diagnosis of previously unknown clival meningocele.
Discussion: Unilateral vagus nerve paralysis constitutes an exceptional mode of presentation of meningocele. Only a few isolated cases of clival meningocele have been reported to date, with no cranial nerve repercussions. The symptomatic management adopted in this case allowed rapid improvement of the patient's disorders.

1. Introduction

Nontraumatic meningoceles of the clivus are extremely rare and consist of prolapse of a pachymeningeal sac containing cerebrospinal fluid (CSF) through a bone defect in the clivus [1]. Clinical presentations are variable, but predominantly consist of rhinorrhoea, headache, and recurrent meningitis. To our knowledge, compression of the vagus nerve in this context has never been reported. We report a misleading initial presentation of this rare entity and highlight the value of non-interventional management in the absence of an infectious risk.

2. Case report

An 84-year-old man was urgently referred to the ENT outpatients department with progressive onset of dysphonia and dysphagia to solids over a period of 1 month, without any associated aspiration. He was an ex-smoker with a history of cardiac arrhythmia treated by anticoagulants, and myocardial infarction 1 year previously. He presented deterioration of his general state of health, with weight loss of 6 kilograms over 1 month.

Clinical and nasal endoscopic examination revealed a nasal voice, right hemipharyngeal paralysis with palatal droop, right recurrent laryngeal nerve paralysis in a paramedian position, and ipsilateral laryngeal sensory loss. The clinical features were in favour of complete vagus nerve paralysis with no lesion of the glossopharyngeal and accessory cranial nerves.

A nasogastric tube was inserted and the patient was admitted to the ENT department for enteral nutrition and aetiological work-up.

Contrast-enhanced neck CT scan demonstrated a meningocele in the right part of the clivus, associated with an incomplete appearance of the C1 vertebra with absence of the right posterior arch and hypotrophy of the right occipital condyle, with a malformative appearance (Fig. 1). Neck MRI confirmed the presence of a cerebrospinal fluid collection through the bone defect in the right part of the clivus, with a low-intensity T1 signal and a high-intensity T2 signal, with no gadolinium enhancement. This meningocele was crossed by the vagus nerve (Fig. 2). Two other meningoceles situated more inferiorly were also revealed, with no clinical repercussions: one between the odontoid process of C2 and the right hemibody of C1, and the other situated in the right C2-C3 intervertebral foramen.

In view of the negative staging assessment, symptomatic management was proposed in agreement with the neurosurgical team. Prophylactic pneumococcal vaccination was performed. Following weight gain, the right hemipharyngeal and laryngeal paralyses were gradually compensated by speech therapy. After 3 weeks of exclusive enteral feeding, the patient was able to gradually resume oral feeding and was weaned from the nasogastric tube at 5 weeks. Clinical follow-up at 1 month and three-monthly follow-up for 6 months showed stable weight gain and normal oral feeding.

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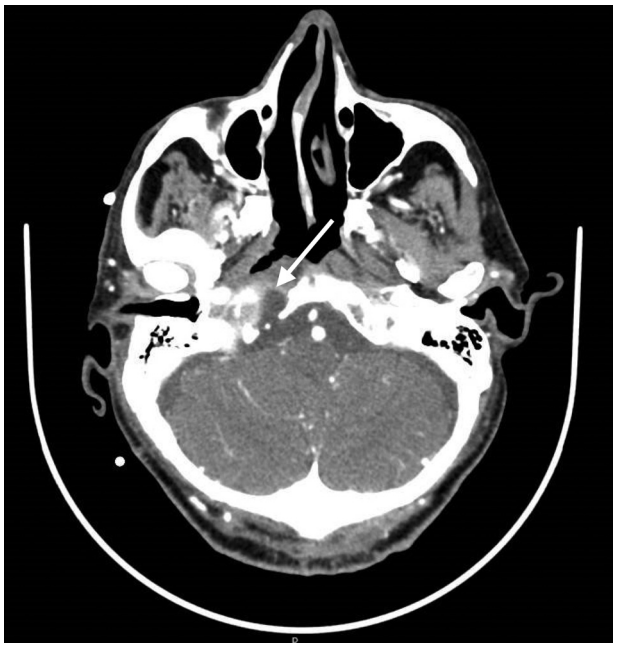

Fig. 1. Contrast-enhanced CT scan, axial section: low density area, 10 mm in diameter, corresponding to the meningocele (arrow). The overlying bone appears to be thinned, but with no signs of invasion.

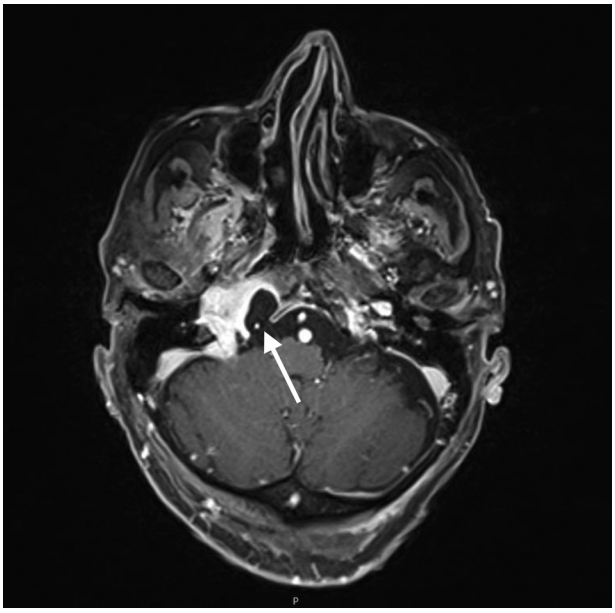

Fig. 2. Gadolinium-enhanced MRI, T1-weighted sequence, axial section: bone defect in the right part of the clivus (arrow), containing a cerebrospinal fluid collection, continuous with the perimedullary subarachnoid spaces. The vagus nerve passes through this meningocele.

3. Discussion

Cranial meningoceles correspond to a CSF collection in a meningeal hernia through a zone of fragility of the bones of the skull base. Meningoceles can be either congenital or acquired (Fig. 3). Acquired meningoceles may be traumatic (80 to 90% of cases [1]), neoplastic, infectious, or spontaneous, which represent 3 to 4% of all causes of CSF collection [2].

An idiopathic meningocele can form through a congenital dehiscence or through a bone defect due to bone remodelling during the patient's life. However, it is difficult to formally exclude a long-standing, minimal traumatic cause.

It is now widely accepted that spontaneous skull base CSF collections are the result of a multifactorial process involving both raised intracranial pressure and an anatomical predisposition. Intracranial pressure varies during the cardiac cycle, determined by arterial and venous blood flow, cerebral compliance and CSF flow. CSF movements can progressively induce meningeal herniation through the skull.

Meningoceles are rare with an estimated prevalence of 0.05%. The case reported here is unusual in terms of the clinical presentation and the exceptional site of the bone defect. One case of recurrent bacterial meningitis, secondary to a meningocele through the occipital part of the clivus, has been reported [3] and a review of the literature on this subject revealed 8 cases of transclival CSF collection, and 2 cases of clival meningocele [4].

The clinical features of acquired meningoceles are dominated by rhinorrhoea, headache, meningitis and signs of intracranial hypertension [5], possibly associated with local swelling [6].

Compressive lesions of the glossopharyngeal, vagus and accessory cranial nerves have been reported in two cases, in a context of tumour of the jugular foramen [7] or retroclival haematoma [8].

The diagnosis is based on MRI visualizing the meningeal hernia through the bone defect. In particular, T2-weighted

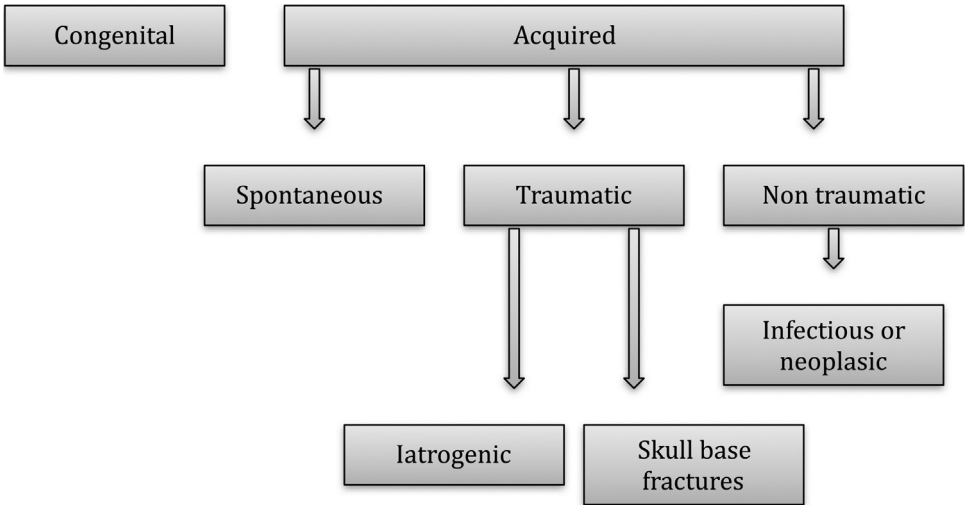

Fig. 3. Classification of meningoceles according to their aetiology.

sequences demonstrate a high-intensity signal corresponding to CSF with gadolinium enhancement of the herniated meninx. In the present case, the gadolinium-enhanced T1-weighted sequence more clearly visualized the nerve passing through the meningocele. CT scan with bone window setting demonstrated possible erosion of the clivus and the meningeal hernia.

Surgical management of these meningoceles is usually recommended in order to prevent meningitis, intracranial abscess and pneumocephalus. Some cases may resolve spontaneously, but with a persistent risk of repeated meningitis. Surgery was not proposed in our patient, in view of his poor general state, and management based on speech therapy appeared to be sufficient to relieve his symptoms.

4. Conclusion

To our knowledge, this is the first reported case of meningocele of the clivus inducing complete and isolated vagus nerve compression. The misleading clinical presentation of this lesion and its exceptional nature should be emphasized. Symptomatic management was appropriate in this particular case.

Disclosure of interest

The authors declare that they have no competing interest.

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