CASE REPORT

Skull base osteomyelitis presenting with an isolated hypoglossal nerve palsy

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SUMMARY

This is the first case of skull base osteomyelitis presenting with isolated bilateral hypoglossal nerve palsy reported in the literature. A 75-year-old man presented with tongue paralysis without any other cranial nerve palsy. He was otherwise well apart from recently having a high prostate-specific antigen level recorded. Investigations for malignancy or cerebrovascular insult were negative with the diagnosis of skull base osteomyelitis confirmed using CT. Following treatment with intravenous antibiotics for 6 weeks, symptoms resolved.

BACKGROUND

This is the first case of skull base osteomyelitis presenting with isolated bilateral hypoglossal nerve palsy presented to the literature. It presents a diagnostically challenging case which adds to the body of information concerned with skull base infection.

CASE PRESENTATION

We present the interesting case of a previously well 75-year-old Caucasian man who presented with a 6-week history of dysphagia to solids, dysarthria and headache on a background of weight loss for 6 months with no other constitutional symptoms. He underwent a temporal artery biopsy which was normal. Three months prior to the admission, he had complained of otalgia, diagnosed as otitis externa and treated with antibiotic ear drops. On follow-up in the ENT clinic 6 weeks later he complained of poor tongue movement and dysphagia and was investigated with an MRI of his brain which was reported normal. He also had an episode of urinary retention and was found to have a raised prostate-specific antigen (PSA), while awaiting prostate surgery.

There was no history of cerebrovascular disease or arthritis.

On clinical examination, vital signs were within normal limits and the patient had no fever. Neurological examination on presentation revealed bilateral tongue wasting without fasciculations. There was no evidence of other cranial nerve palsies, and funduscopy and peripheral neurological examination was normal. Examination was otherwise unremarkable, notably with no lymphadenopathy found.

INVESTIGATIONS

Initial blood tests showed a haemoglobin of 9.9 g/dL (mean corpuscular volume (MCV) 90.1 fL) with a previous value of 11.2 g/dL 2 weeks previously,

white cell count (WCC) of $16.5 \times 10^9/L$ and platelet count of $440 \times 10^9/L$. Biochemical analysis showed acute kidney injury with a sodium of 133, potassium of 4.4, urea of 17.5 and a creatine of 121. Vasculitic screen, Lyme serology and Bence-Jones protein was negative. Fasting glucose, glycated haemoglobin, complement level, antiGQ1b, serum electrophoresis, ACE, lactate dehydrogenase Antistreptolysin-O test Epstein-Barr virus serology, immunoglobulins and haematinics were all normal. C reactive protein was mildly raised at 54 mg/L. Lumbar puncture was performed showed slightly raised protein (0.73 units).

Electromyography revealed a generalised axonal peripheral neuropathy.

Examination and biopsy of the postnasal space revealed oedematous tissue with pus present so representative biopsies and microbiological specimens were sent for analysis.

Following multidisciplinary discussion with radiology, neurology, rheumatology and haematology colleagues a further MRI with gadolinium contrast was carried out which confirmed that the brain was structurally normal.

Subsequent CT scan of the skull base demonstrated bony destruction of the skull base with bilateral involvement of the inferior margins of the petrous temporal bone.

The patient proceeded to have an endoscopic biopsy of the postnasal space, which revealed a heavy growth of *Staphylococcus* and *Pseudomonas* species.

TREATMENT

A diagnosis of skull base osteomyelitis was made and the patient was started on a long-term course of intravenous tazobactam/piperacillin and he rapidly improved clinically and biochemically, while in hospital.

OUTCOME AND FOLLOW-UP

The patient was discharged within a month from diagnosis and a routine follow-up 3 months post-treatment has shown significant improvement of his hypoglossal palsy.

DISCUSSION

Isolated hypoglossal palsy, especially bilateral, is very rare and has a broad differential diagnosis. Most cases of hypoglossal nerve palsy occur with other neurological or systemic signs. Reported cases of truly isolated palsy are due to cancer¹—which is the most common cause²—carotid artery aneurysms,³ vertebral artery dissection,⁴ cervical

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osteophytosis,⁵ calcified persistent hypoglossal artery,⁶ synovial cysts,⁷ vasculitides/rheumatological diseases,² dental disease,⁸ trauma⁹ and following transoral intubation¹⁰ among others.

The hypoglossal nerve originates from the hypoglossal nucleus and appears from the medulla oblongata from where it passes into the hypoglossal canal, between the internal carotid artery and the internal jugular vein. It then passes through the digastric muscle and courses anteriorly to provide efferent motor innervation to all the tongue muscles apart from palatoglossus. It is responsible for the voluntary movements of the tongue. This course means that it may be liable to compression or injury at a number of different sites, particularly in its journey through and out of the skull base. We believe that this is the first case of isolated hypoglossal nerve palsy caused by skull base osteomyelitis reported in the literature. It is likely that the point of entry for the infection was the episode of otitis externa which occurred 3 months prior to admission but there were no signs of necrotising otitis externa on presentation.

Learning points

- ▶ Hypoglossal palsy is a rare clinical presentation.
- ▶ Most common causes are vascular pathology, cervical spondylosis, vasculitides and trauma.
- ▶ The diagnostic yield of a scan is not 100%, if clinical presentation is not explained by the results of a scan, the scan should be revised or repeated.
- ▶ Common diseases can present with uncommon presentations.

Competing interests None.

Patient consent Obtained.

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