

## Bilateral skull base osteomyelitis in an immunocompetent patient

### Introduction

Skull base osteomyelitis is a relatively uncommon medical condition. The typical clinical scenario involves a diabetic or immunodeficient patient with otitis externa that was either inadequately treated or refractory to standard antibiotic therapy. Clinicians must have a high index of suspicion for osteomyelitis and begin the appropriate workup in a timely fashion. Treatment must be initiated without delay to prevent extension of the infection. Therapy consists of parenteral and topical anti-pseudomonal antibiotics coupled with debridement.

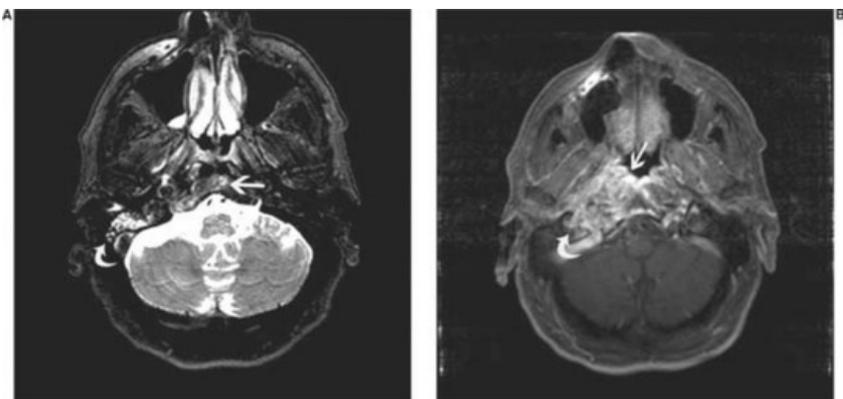
We describe an interesting case of bilateral temporal bone, skull base, and cervical spine osteomyelitis in an immunocompetent patient.

### Case report

A 75-year-old immunocompetent man with no history of ear disease presented to our outpatient office with intermittent right otalgia of 2 months' duration. During this period, he had been unsuccessfully treated with intermittent courses of oral and topical antibiotics for multiple episodes of otitis externa. Physical examination revealed exostoses in the ear canals, without granulation tissue or otorrhea. Findings of computed tomography (CT) of the temporal bones and skull base were consistent with chronic mastoiditis, and he was treated with a 3-week course of an oral fluoroquinolone antibiotic, hydrocortisone, and acetic acid ear drops.

The patient's otalgia worsened and the pain began to radiate to his right neck and shoulder. A three-phase technetium bone scan was done, which showed uptake within the right temporal bone and sphenoid bone extending into the skull base. The patient then began to experience tongue swelling, hoarseness, and dysphagia. Magnetic resonance imaging (MRI) of the brain revealed an extensive infiltrative process involving the right skull base, extending from the right lateral margin of the nasopharynx to the inferior aspect of the right temporal bone and jugular fossa ([figure 1](#)).

**Figure 1. A: T2-weighted MRI of the skull base demonstrates fluid signal within the right mastoid air cells (curved arrow). There is edema within the right nasopharyngeal soft tissues, as well as within the skull base. Bone marrow edema is seen within the clivus extending across the midline (straight arrow). B: T1 postcontrast fat-saturated MRI demonstrates enhancement of the skull base (curved arrow) with extension into the right nasopharynx (straight arrow).**



The patient was then admitted to the hospital and empirically started on intravenous vancomycin and ceftazidime. He had a normal complete blood count and chemistry panel on admission to the hospital, with a blood glucose level of 72 mg/dl and a hemoglobin A1c level of 6.1%. MRI of the cervical spine showed extension of a soft-tissue inflammatory process at the cranial base extending to the epidural and prevertebral spaces at the C1 to C2 level, with mass effect on the cervicomedullary junction.

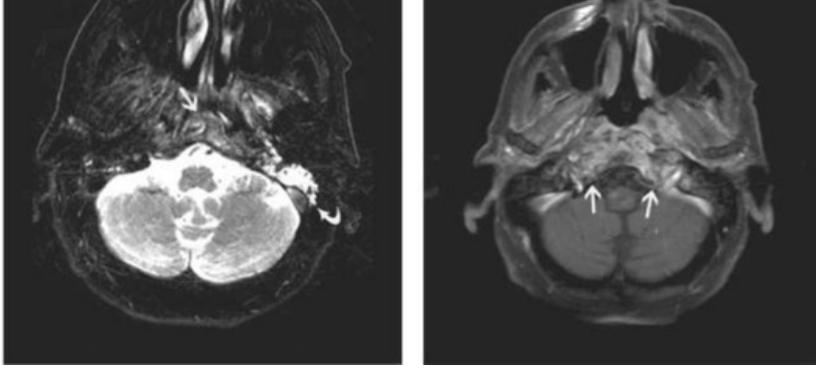
The patient also underwent an indium white blood cell scan to differentiate between infection and a possible neoplasm. There was no abnormal activity; however, this was thought to represent a false negative in the face of a chronic infection that had been partially treated with antibiotics.

The patient improved clinically and was discharged on a 6-week course of intravenous ceftazidime. He was maintained in a cervical collar for 3 months. He reported improvement of the right otalgia and neck pain after completing therapy, and serial bone scans showed improvement in the right temporal bone.

Four weeks after treatment, the patient began to experience left otalgia and left temporoparietal headache. Repeat imaging showed left otomastoiditis and left temporal bone and skull base osteomyelitis ([figure 2](#)). The patient had a left myringotomy tube placed. Cultures from the ear grew *Pseudomonas aeruginosa*, and he was treated with another 6-week course of intravenous ceftazidime, leading to clinical improvement.

**Figure 2. A: T2-weighted MRI of the skull base demonstrates fluid signal within the left mastoid air cells (curved arrow). Bone marrow edema is seen within the clivus extending across the midline (straight arrow). B: T1 postcontrast fat-saturated MRI demonstrates diffuse enhancement of the skull base (arrows) with diffuse extension throughout the nasopharynx.**





Three weeks after completion of antibiotic therapy, the patient developed significant neck pain, intermittent signs of upper extremity numbness, other sensory changes, and gait problems. CT and MRI revealed findings consistent with C1 to C2 instability with a large C1 to C2 pannus causing cervical medullary compression ([figure 3](#)).

**Figure 3. Presurgical sagittal reformatted noncontrast CT image of the skull base and upper cervical spine demonstrates a large pannus at the C1 to C2 level (straight arrow), resulting in severe narrowing of the spinal canal and cord compression (curved arrow).**

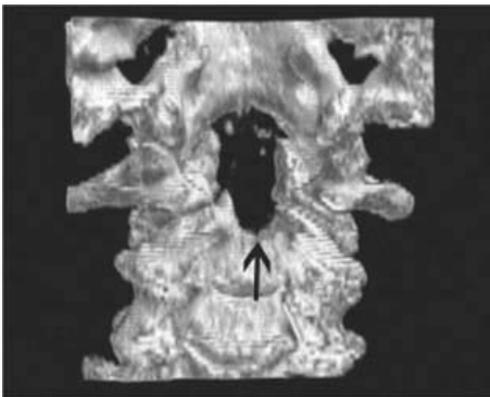


The patient underwent endoscopic transoral odontoidectomy and resection of the anterior aspect of the C1 vertebral ring and large ventral C1 to C2 pannus. The procedure began with a midline linear incision in the posterior pharynx from approximately the region of the inferior clivus to the lower aspect of the C2 vertebral body. Electrocautery was used to dissect through the mucosa and muscular layers to expose the fascia of the cervical spine. Subperiosteal dissection was then performed to expose the inferior aspect of the clivus, the C1 anterior tubercle, and the anterior aspect of the C2 body.

Next, under intraoperative neuronavigation guidance and direct endoscopic visualization, a cutting burr was used to remove the anterior aspect of the C1 vertebra, the odontoid process, the superior half of the C2 vertebral body, and the most inferior aspect of the clivus. The cruciate ligament was opened, allowing exposure to the underlying large C1 to C2 pannus, which was then resected and decompressed.

Postoperative imaging revealed decompression of the central canal ([figure 4](#)). The patient had no evidence of a cerebrospinal fluid leak postoperatively, and the following week he underwent a posterior occipital to cervical instrumented fusion with autograft. He was discharged 10 days later on oral clindamycin 300 mg daily for 4 weeks. The patient had a full neurologic recovery following surgery, and resolution of neck pain at his last follow-up visit 18 months postoperatively.

**Figure 4. Postsurgical 3-D reformatted noncontrast coronal CT image of the skull base and upper cervical spine shows interval resection of the anterior arch of C1 and the dens (arrow).**



## Discussion

Osteomyelitis of the central cranial base is an uncommon condition that has a high morbidity and mortality rate if not promptly diagnosed and adequately treated. Conditions that

adversely affect blood flow through bone such as radiation, malignancy, osteoporosis, osteopetrosis, and Paget disease predispose to osteomyelitis. Furthermore, systemic conditions such as diabetes, anemia, and malnutrition increase the likelihood of osteomyelitis.<sup>1</sup>

Patients with diabetes mellitus or an underlying immunodeficiency disorder seem to be predisposed to malignant otitis externa which, if improperly treated, can lead to the development of skull base osteomyelitis. Once the infection invades the bone of the external auditory canal, it then progresses to the vasculature of the pneumatic spaces. The infectious process then spreads submucosally, causing localized bone destruction at the adjacent sites.<sup>2</sup> Thus the infection can extend to contiguous structures, such as the cervical spine, inoculating the site with bacteria.

Our case was unique for many reasons. First of all, the patient had none of the conditions that predispose patients to developing osteomyelitis. He had a normal complete blood count, and his blood glucose and hemoglobin A1c levels were within normal limits. Therefore, there was no known microangiopathy or compromised immunologic response to explain the fulminant disease course.

In a review of 84 cases of osteomyelitis in the head and neck, Prasad et al found that all 4 patients with temporal bone osteomyelitis had diabetes mellitus with malignant external otitis.<sup>1</sup> Our patient, although he did not have diabetes, had experienced multiple episodes of otitis externa that might not have been adequately treated or might have been resistant to therapy. Therefore, there might have been persistence of the infection in the underlying bone.

Infections in the external ear may spread through the fissures of Santorini to the surrounding temporal bone. From there, the infection can spread through the Haversian system of compact bone to involve the entire skull base and cervical vertebrae. In our patient, the infection began in his right temporal bone and then spread posterolaterally to involve the jugular foramen and lower cranial nerves. Osteomyelitis then manifested in his left temporal bone and ultimately involved the craniovertebral junction, leading to vertebral instability.

Bilateral skull base osteomyelitis is very rare.<sup>3</sup> It is thought to occur with partial treatment of the initial condition, whereby the primary site of infection is healed but the central infectious process in the bone marrow remains active.<sup>2</sup> Our patient was treated with parenteral antibiotics for 6 weeks on two occasions and had local improvement each time. Theoretically, a deeper infective process might have persisted and provided a nidus for spread. The infection might have progressed along the prevertebral soft-tissue planes, thus leading to involvement of the cervical spine.

Only 3 cases of cervical spine invasion from skull base osteomyelitis have been reported in the American literature, and in one case, the patient succumbed to the disease.<sup>3-5</sup>

In conclusion, skull base osteomyelitis must be included in the differential diagnosis of patients with otitis externa refractory to standard therapy, even if they have no predisposing medical conditions. Prompt initiation of treatment is of utmost importance to prevent the potential osseous spread of the infection.

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*Ear Nose Throat J.* 2011 December;90(12):E23

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