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Clinical Records

A case of malignant otitis externa following mastoidectomy

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Abstract
We present a case of a 63-year-old diabetic male who developed malignant otitis externa following mastoidectomy. Extensive skull base osteomyelitis caused thrombosis of the jugular bulb and subsequent paralysis of cranial nerves VII, IX, X and XII. He was treated aggressively with intravenous antibiotics and debridement of granulation tissue in the mastoid bowl with full recovery of the cranial nerve palsies associated with recanalization of the jugular bulb. We believe this is the first reported case of malignant otitis externa to occur following mastoidectomy with complete recovery of the cranial nerve paresis.

Key words: Otitis externa; Jugular veins; Cranial nerves; Paralysis

Introduction

Malignant otitis externa (MOE) is a rare disease occurring principally in elderly diabetics and is usually caused by the organism *Pseudomonas aeruginosa* (Dawson, 1978). It is a necrotizing infection of the external ear canal which may spread to the mastoid and petrous parts of the temporal bone, leading to skull base osteomyelitis. The first case was described by Meltzer and Keleman in 1959 and was described as a progressive osteomyelitis of the skull base. There is still some debate over the most appropriate terminology and other terms in use include 'necrotizing' and 'invasive' otitis externa.

Cases have been reported in non-diabetics (John and Hopkins, 1978), immunocompromised patients (Hern *et al.*, 1996), and in children as young as five years old (Coser *et al.*, 1980). Although *Pseudomonas aeruginosa* is responsible for most of the cases of MOE, other pathogens have occasionally been isolated such as *Staphylococcus aureus* (Keay and Murray, 1988), *Proteus mirabilis* (Coser *et al.*, 1980) and *Aspergillus fumigatus* (Cunningham *et al.*, 1988).

As MOE spread to involve the adjacent temporal bone, the facial nerve may be involved causing ipsilateral facial nerve paralysis. Other cranial nerves (IX, X, XI and XII) may also be involved and this is a poor prognostic sign. The infection can prove fatal when there is intracranial extension or when the carotid artery or jugular vein are involved.

We report a case of MOE occurring following mastoidectomy in a 63-year-old diabetic Asian male. He had extensive skull base osteomyelitis with paralysis of cranial nerves VII, IX, X and XII and thrombosis of the jugular bulb. He was treated successfully and made a full recovery. We believe this is the first reported case of MOE to occur post-mastoidectomy with complete recovery of the cranial nerve paresis.

Case report

A 63-year-old Asian non-insulin dependent diabetic male presented with chronic otorrhoea secondary to a subtotal perforation. He underwent a right cortical mastoidectomy and tympanoplasty and granulation tissue was removed from the attic and mastoid. The tympanic membrane was grafted with temporalis fascia. He had an uneventful recovery and was discharged the following day. At five weeks post-operation, he developed severe right-sided otalgia with associated otitis externa and facial cellulitis. Examination revealed aural polyps deep in the ear canal. Both the ESR and CRP were raised at 90 and 103 respectively. The ear was swabbed and a tentative diagnosis of MOE made. He was commenced on i.v. ceftazidime and metronidazole and topical gentisone HC drops. He was monitored with serial CRP and ESR measurements and computed tomography (CT) scans of the skull base. This showed a large inflammatory soft tissue mass in the right ear canal, middle ear and skull base extending across the midline (Figure 1). There was some erosion of the temporal bone and temporomandibular joint and the internal jugular vein thrombosed. Soft tissue thickening was noted at the right side of the nasopharynx extending to the pterygoid muscles. The right internal carotid artery was patent. The ear swab culture confirmed a pseudomonal infection.

After one week of treatment, he developed a right facial palsy, had swallowing difficulties and started aspirating liquids. Neurological examination revealed weakness of the VIIth, IXth, Xth, XIIth cranial nerves on the right side. An EUA of the right ear and mastoid cavity was performed.

Extensive granulations were encountered in the ear canal extending through the flap onto the malleus and obliterating the mastoid cavity. The mastoid bone was noted to be congested. The canal wall was drilled down and the granulation and scar tissue excised and submitted

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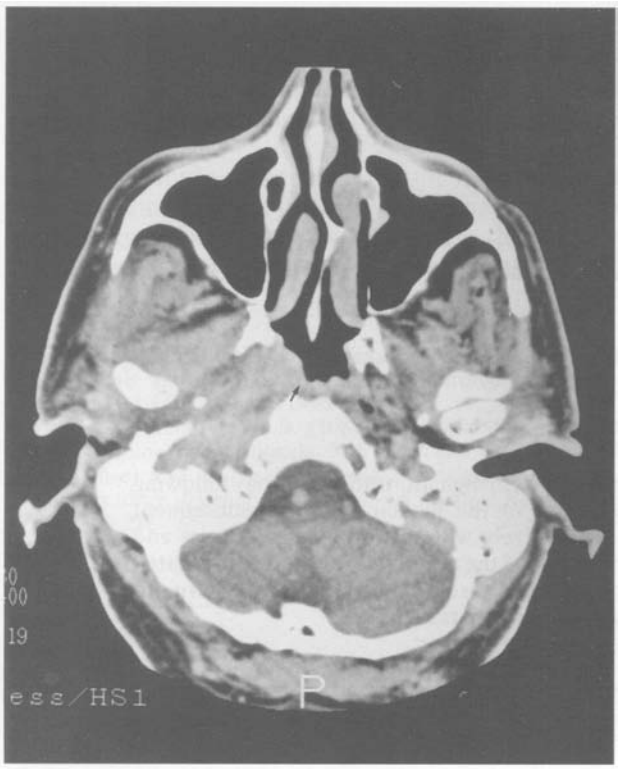

FIG. 1
Axial CT scan showing extensive soft tissue swelling at the base of skull extending across the midline to the nasopharynx (arrowed). A right mastoid cavity is present.

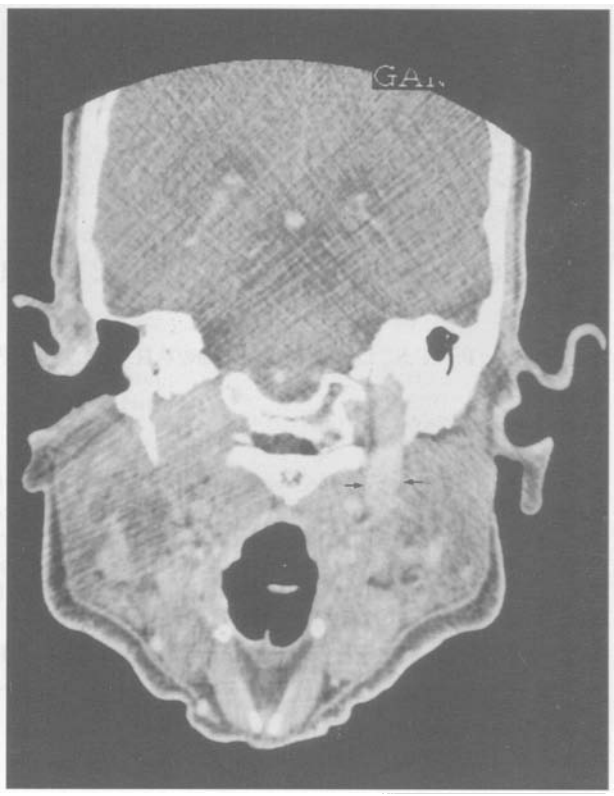

FIG. 2
Coronal CT scan showing occlusion of the right internal jugular vein and a patent left internal jugular vein.

for histology. This revealed chronic granulation tissue only with no evidence of malignancy. Blood tests for autoimmune and chronic specific granulomatous conditions were negative. A technetium bone scan showed increased uptake in the right temporal bone and sphenoid sinus. A repeat CT scan showed the skull base osteomyelitis remained the same and there was no sequestrum present. Antibiotic therapy was continued with full recovery of cranial nerve function over the next two weeks. A follow-up CT scan showed improvement in the inflammatory swelling at the skull base and demonstrated recanalization of the jugular bulb. He was discharged from the ward after six weeks of i.v. antibiotics and continued oral ciprofloxacin for a further three months as an out-patient. At present, he is pain free, has normal swallowing and has a dry mastoid cavity.

Discussion

MOE is a serious condition with a significant morbidity and mortality. It develops after an initial dermatitis of the external ear canal, which compromises the natural barrier of the skin. *Pseudomonas* spp. frequently colonize the moist environment of the external ear canal, especially if previous systemic or local antibiotic therapy has been administered. It can cause a localized external otitis that may progress to a more invasive disease process, particularly if the host is immunosuppressed. MOE therefore represents a spectrum of diseases. When it is limited to the soft tissues and cartilage, it is called necrotizing otitis externa (or stage I disease), but as it progresses to involve the temporal bone and other parts of the skull base, it is called skull-base osteomyelitis. Skull-base osteomyelitis can be further subdivided on the basis of radionucleide scans. Early forms of the disease show restriction of inflammation to the ear and mastoid (stage II) while later the osteomyelitis spreads across the skull base and can involve the facial bones (stage III) (Benecke, 1989). The occipital bone and sphenoid sinus is frequently involved at this stage (Murray and Britton, 1994). It has been proposed that most cases of skull-base osteomyelitis are due to inadequately treated localized MOE (Grobman *et al.*, 1989).

Our case developed advanced skull-base osteomyelitis (stage III) post-mastoid surgery with inflammation in the parapharyngeal area and the bone marrow in the sphenoid sinus. He also developed lower cranial nerve palsies which were associated with thrombosis of the jugular bulb. This is associated with a poor prognosis (Dawson, 1978) but with early and aggressive treatment, he made a full recovery. His cranial nerve palsies resolved as the jugular bulb recanalized and currently he is symptom free.

Early diagnosis and treatment is essential if a favourable outcome is to be achieved. Maintaining a high index of suspicion is important in making a proper diagnosis of MOE Cohen and Friedman (1987) have proposed the following criteria: a severe otitis externa with either a positive bone scan or that fails to respond to local treatment for at least one week, accompanied by pain, oedema, exudate, granulations and microabscesses. Arguably, *Pseudomonas* spp. should be cultured from the ear but it is now recognized that a small proportion of cases are caused by other pathogens.

Autoimmune conditions, chronic granulomatous causes and carcinoma should be excluded (Al-Shihabi, 1992). CT scanning can accurately delineate the amount of soft tissue involvement and bone erosion caused by the disease (Fritz *et al.*, 1989; Murray and Britton, 1994). It can show subtle

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foci of skull-base involvement that may be clinically unsuspected and demonstrates soft-tissue thickening of the parapharyngeal space and roof of the nasopharynx implying advanced disease (Gold *et al.*, 1984). Radionuclide bone and gallium scan images accurately depict the biological activity of the disease process and permit accurate treatment evaluation and patient monitoring (Noyek *et al.*, 1984).

The mainstay of treatment is prolonged antibiotic therapy. Usually a third generation cephalosporin e.g. ceftazidime, or a quinolone e.g. ciprofloxacin, is used, the latter having the advantage that oral preparations are available. These have now replaced aminoglycosides in the treatment of MOE that have the unacceptable risk of ototoxicity if used for prolonged periods especially in elderly patients with impaired renal function. High levels of antibiotics are required to achieve a minimal inhibitory concentration in the infected tissue. The tissue uptake is affected by vascularity and the presence of concomitant disease. Small vessel disease, especially in diabetics, further compromises antibiotic uptake in affected tissues. Because chronic osteomyelitis is associated with necrotic bone that may serve to sequester bacteria and the organisms are not rapidly dividing, it is necessary to attain high levels of antibiotics over a prolonged period of time in affected tissues (Grobman *et al.*, 1989). Antibiotics must be continued until sequential gallium scans reveal resolution of the osteomyelitis, so that recurrence can be prevented. Surgery has only a limited role in MOE. Biopsy must be performed to exclude other conditions mimicking the disease. Abscesses and sequestra must be drained and frequently a cortical mastoidectomy is performed for this reason. The extent of the disease determines the degree of surgical exenteration.

In summary, MOE is a potentially life threatening condition which is diagnosed with a high degree of clinical suspicion and appropriate investigations. Early treatment with high dose, long-term antibiotics and timely surgery can reduce the morbidity and mortality of this serious condition.

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