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Clinical Records

Osteomyelitis of the temporomandibular joint in patients with malignant otitis externa

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Abstract
Malignant (invasive) otitis externa is an infection involving the external ear canal, often in elderly diabetic patients, which carries a high morbidity and mortality. It may involve widespread areas of soft tissue around the skull base, and in more advanced cases, may give rise to osteomyelitis and cranial neuropathy. We describe two patients who were treated for malignant otitis externa complicated by destructive osteomyelitis of the temporomandibular joint (TMJ). For both patients, diagnosis was made using magnetic resonance imaging (MRI), and repeat scans were employed during follow-up. Improved scan appearances mirrored improvements in clinical condition in both cases.

Key words: Otitis externa; Osteomyelitis; Temporomandibular joint; Magnetic resonance imaging

Case reports

Case 1

An 81-year-old insulin-dependent diabetic woman was referred to the ENT department complaining of right-sided otalgia for the past month. She had an ear canal full of copious purulent discharge, skin debris and granulation tissue. A pus swab grew *Pseudomonas aeruginosa*. A working diagnosis of malignant otitis externa was made, and she was commenced on gentisone HC drops (gentamicin 0.3 per cent, hydrocortisone acetate one per cent) and oral ciprofloxacin 750 mg b.d.

Over the next few weeks she continued to complain of increasing otalgia and TMJ pain. She was admitted to hospital and an MRI scan performed which demonstrated features consistent with a progressive malignant otitis externa and involvement of the TMJ: a T1-weighted coronal image (Figure 1) demonstrated extensive low signal soft tissue surrounding the right mandibular condyle (short arrows) in keeping with advanced arthropathy. At this stage there was preservation of the normal high marrow signal in the head of the mandible (long arrow). There was thickened, low signal intensity soft tissue inferior to the right temporal lobe (curved arrow) representing dural involvement; compared with the normal opposite side.

She was given intravenous ciprofloxacin 750 mg b.d. and her symptoms improved over a few days. She was discharged home with a six-week course of oral ciprofloxacin 750 mg b.d. Regular follow-up showed satisfactory control of symptoms. Her diabetes was proving difficult to control, however, and a repeat MRI scan (Figure 2) performed four months after initial diagnosis now showed

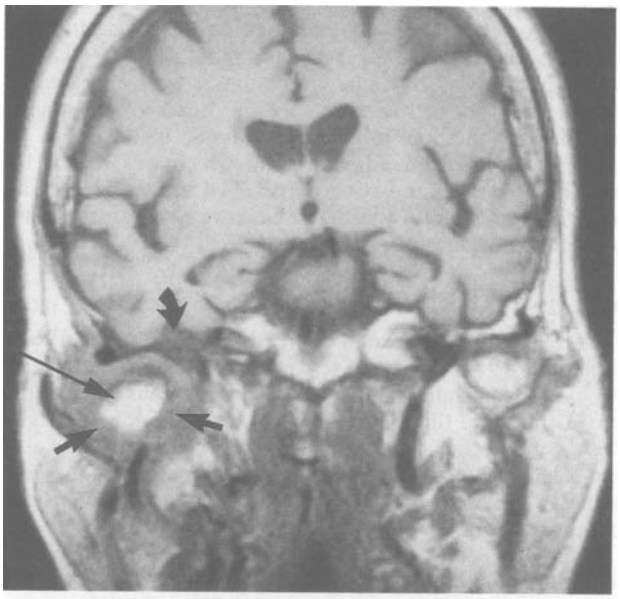

FIG. 1 *Case 1.*
SE T1-weighted (600/15) coronal image performed a few weeks following first presentation, showing inflammation surrounding the TMJ (see text for detailed description).

features consistent with chronic osteomyelitis of the right TMJ: a T1-weighted coronal image showed extensive destruction of the right mandibular condyle, with low signal throughout its marrow.

The patient was re-admitted to hospital, and an open exploration of her right temporomandibular joint was carried out. At operation a thickened joint capsule was

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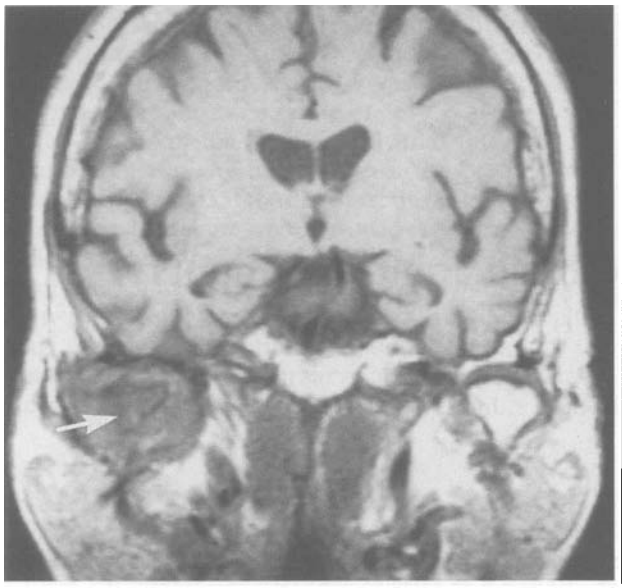


FIG. 2 *Case 1.*

Coronal SE T1-weighted (600/15) image four months after initial diagnosis. Disease progression to TMJ osteomyelitis is seen (see text).

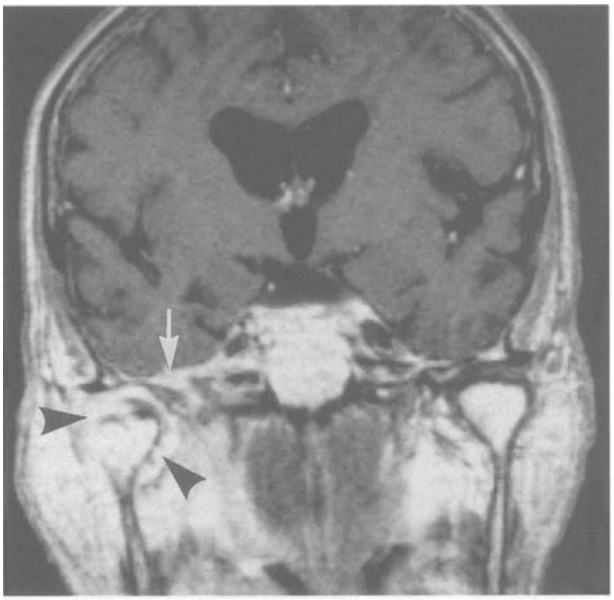


FIG. 3 *Case 2.*

Coronal SE T1-weighted (450/15) gadolinium enhanced images at the time of first presentation, demonstrating features consistent with inflammation around the right TMJ and middle cranial fossa (see text).

exposed through a pre-auricular incision. It was opened to reveal an eroded, avascular mandibular head, which was excised. It was noted that the bone of the glenoid fossa was soft, and that there was no articular cartilage.

Post-operatively, the patient made a good recovery. There was no growth from the culture of the excised tissue, and histology reported appearances consistent with a healing osteomyelitis. She was discharged on oral ciprofloxacin 400 mg b.d. for a further month. An MRI scan (not shown) was repeated four months post-operatively, which showed a resection of the head of the right mandible, and some inflammatory tissue in the space of the right temporomandibular joint which was felt to be consistent with granulation tissue. The mastoid area was clear. Overall there was much improvement, and far less oedema and inflammatory change.

Case 2

An 82-year-old insulin dependent diabetic man was admitted with a painful, discharging right ear, having been treated for unstable diabetes over the previous few weeks. Over the same time period he had also developed a right lower motor facial nerve palsy. Examination of the ear revealed the presence of granulation tissue in the ear canal, and copious pus, culture of which grew *Pseudomonas aeruginosa*.

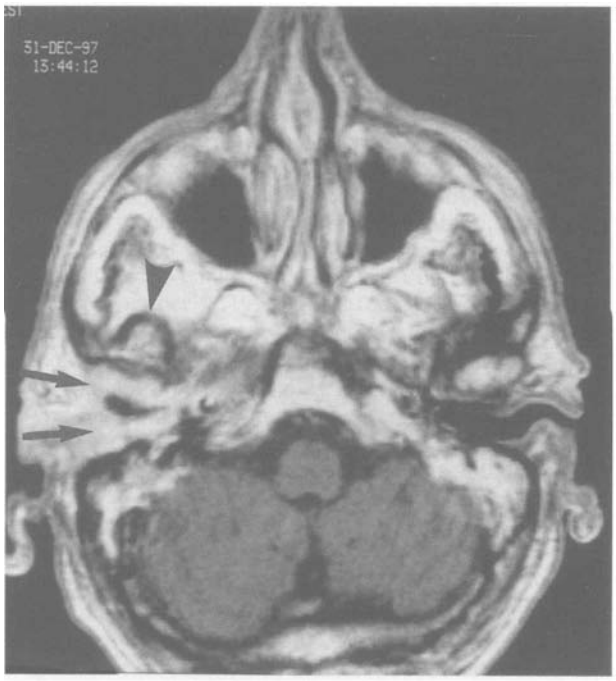
An MRI scan of the skull base was performed, which demonstrated appearances consistent with malignant otitis externa and TMJ involvement. On a T1-weighted coronal image with intravenous gadolinium (Figure 3), there was intense enhancement of the thickened joint capsule around the right TMJ (arrowheads) consistent with an active inflammatory process in these tissues. There was extension of enhancing tissue to involve the dura of the middle cranial fossa (white arrow). Note the loss of clarity of the cortical bone, which appears black, on the right mandibular condyle compared with the left, consistent with cortical destruction. On an axial T1-weighted gadolinium-enhanced image (Figure 4a), there was near obliteration of the external ear canal by enhancing circumferential soft tissue thickening (arrows). The right mandibular condyle (arrowhead) was displaced forwards by the swelling. A further image from the same series (Figure 4b), showed markedly asymmetrical appearances of the infratemporal fossae: on the right side there was intense enhancement around the pterygoid muscles (arrows) indicating involvement in the inflammatory process.

A diagnosis of malignant otitis externa was made. The patient was commenced on intravenous ciprofloxacin 400 mg b.d. and Otomize ear spray (neomycin 325 units per ml, dexamethasone 0.1 per cent, acetic acid two per cent). He improved clinically over the next few days, with a reduction in pain, and improved diabetic control. He was discharged home on oral ciprofloxacin 400 mg b.d. Regular review in clinic saw the patient continue to improve on this medication, and eight months after his initial referral, a repeat MRI scan (not shown) was performed. This showed an improvement in the appearance of the inflammatory tissue surrounding the right TMJ, and the dural enhancement was less evident than it had been previously. The improved appearances on the scan mirrored the clinical improvement seen in the patient.

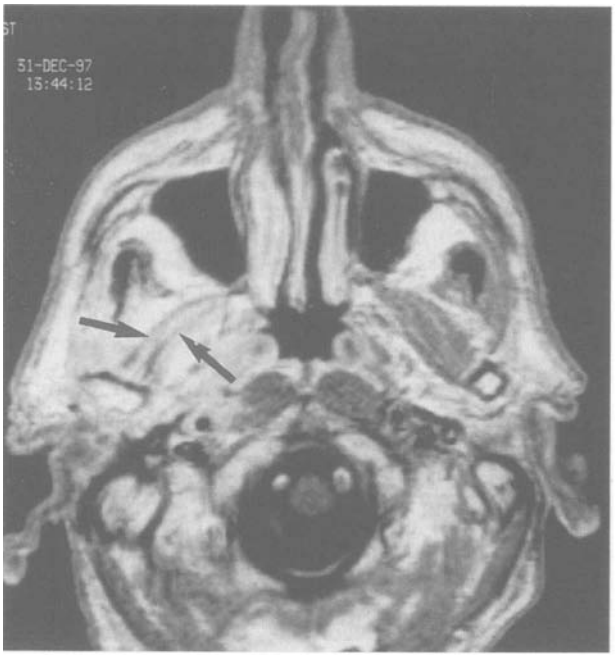
Discussion

Malignant otitis externa is a serious infection of the external ear canal, most often affecting elderly patients with diabetes mellitus. The pathogen typically found is *Pseudomonas aeruginosa*. The infection causes a high level of morbidity since in advanced cases it may progress to involve the surrounding structures including the skull base, where it causes an osteomyelitis and can give rise to cranial neuropathy (Slattery and Brackmann, 1996). Rarely, the dura may become involved and result in meningitis, or sigmoid sinus thrombosis, and so can be fatal.

The diagnosis of malignant otitis externa may be difficult to distinguish from that of chronic otitis externa. Attempts have been made to draw up diagnostic criteria since the disease was first characterized in Chandler's original series (Chandler, 1968). Most authors agree that the essential factor is the presence of a persistent otitis externa which is refractory to treatment, severe otalgia, and the presence of granulation tissue in the external ear canal (Levenson *et al.*, 1991; Amorosa *et al.*, 1996). Patients are usually, but not exclusively, elderly diabetics. The pathogen is most commonly *Pseudomonas aeruginosa*, but other organisms



(a)



(b)

FIG. 4 (a and b) *Case 2.*

Axial SE T1-weighted (500/15) gadolinium enhanced images at presentation. (a) External ear canal involvement. (b) Infratemporal fossa involvement in the inflammatory process (see text).

such as *Aspergillus flavus* are occasionally isolated, therefore these factors can only be relative criteria. The development of a cranial neuropathy represents a more advanced disease state, and if present, distinguishes the disease from chronic otitis externa. Likewise, the presence of skull base osteomyelitis is diagnostic, but quite unusual these days, since malignant otitis externa is usually diagnosed before this occurs.

Increasingly, a diagnosis of malignant otitis externa is made on clinical criteria together with positive diagnostic imaging. Standard investigation has usually been with CT scan or Tc-99 bone scan, both of which are capable of showing bony involvement and erosion (Amorosa *et al.*, 1996). There have been a few reports of MR imaging being employed in the diagnosis and follow-up monitoring of these patients (Gherini *et al.*, 1986; Rubin *et al.*, 1990). The two patients reported here underwent MR imaging during the investigation of their disease. This is employed in our department as a standard imaging technique in the diagnosis and follow-up of patients with malignant otitis externa. It has been found to be particularly useful in evaluating the soft tissue involvement, including the meninges, and parotid area. We have found that MR imaging allows demonstration of both cortical bone erosions as well as narrow infiltration (Figures 2 and 3) and in the latter evaluation is superior to CT. It also seems that improvements in the clinical condition of the patient are mirrored by the MR imaging appearances.

The two patients described in this report both developed an extremely unusual complication of malignant otitis externa, in that the ipsilateral TMJ was destroyed. As mentioned above, the development of osteomyelitis in malignant otitis externa is an advanced event, and not often encountered now. It is especially rare for the TMJ to become directly involved. A review of the literature found one report of osteomyelitis (Drew *et al.*, 1993), and very few reports of septic arthritis of the TMJ associated with malignant otitis externa (Dingle, 1992). Of our two patients, one underwent surgical debridement of her TMJ, the other was treated conservatively, and both improved clinically. It is uncertain whether a secondary osteomyelitis should be managed conservatively or operatively: the current trend is towards conservative treatment. It may be that the first patient would eventually have responded to medical therapy, particularly as the dead bone removed was showing signs of healing and contained no pathogens. It must be noted, however, that of the two, this patient showed the more rapid improvement in her symptoms, which may provide an argument for operative treatment.

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