

Skull base osteomyelitis interpreted as malignancy

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Abstract

Lesions in the skull base may present difficulties in diagnosis primarily because the access needs to be invasive and one has to rely on imaging that may still be misleading. The case presented here illustrates this example in that the patient had abnormalities on computed tomography (CT) and magnetic resonance image (MRI) scans quite convincing of malignancy but which with time proved, essentially through clinical judgment, to be skull base osteomyelitis secondary to malignant external otitis (OME).

Key words: Osteomyelitis; Otitis externa; Radiography

Introduction

Skull base osteomyelitis is a known complication of malignant external otitis (MEO). Various nosological criteria have been designed to help in early diagnosis (Amorosa *et al.*, 1996). When such a condition presents in an elderly patient with facial nerve paralysis and a space-occupying lesion is detected on imaging, malignancy is suspected. Due to anatomical restraints, exact histological diagnosis may be difficult and a stereotactic biopsy may have to be precluded due to the patient's general condition. Described here is a case of malignant external otitis with skull base osteomyelitis illustrating this diagnostic dilemma that could only be resolved with conservative management and repeated imaging.

Case report

A 68-year-old insulin-dependent diabetic patient who also had chronic renal failure and cardiac decompensation, was being treated for relapsing external otitis of the right ear canal. The otorrhoea was slight, but she had a deeply-seated otalgia that was out of proportion to the degree of morbidity. This required her admission for treatment. Soon after admission she developed right-sided facial weakness that progressed to completion. Examination of the ear showed debris along the floor of the right ear canal and dullness of the tympanic membrane and tenderness in the mastoid region. All cranial nerves other than the facial nerve were intact. The oral cavity and the post-nasal space were clear. Swabs from the ear did not grow any organisms this time, but earlier *Pseudomonas aeruginosa* had been isolated. The ESR was raised and the leucocyte count was reduced. Biochemical readings were in keeping with diabetes and chronic renal failure. The patient had also developed profound sensorineural hearing loss in the right ear.

Imaging with MRI through axial T1 and T2 and coronal T1 images before, and after, gadolinium enhancement showed an abnormal soft tissue mass extending from the petrous ridge to the pterygopalatine fossa medially, where the fat planes were obliterated and the mass extended across the mid-line (Figure 1). The 'mass' was relatively

patchy without intense enhancement, and also featured as fullness in the post-nasal space. Axial CT scans of the skull base with two mm cuts showed a mass in the post-nasal space. There was loss of normal cortical bone pattern on the right margin of the foramen magnum extending into the clivus (Figure 2). The pterygopalatine fossa, mastoid, petrous ridge and attic appeared free of bone erosion. The ipsilateral eustachian tube and external ear canal were filled with soft tissue. The radiologist's opinion was that of a postnasal space tumour (probably of squamous cell carcinoma), extending outwards through the pharyngotympanic tube and involving the facial nerve laterally.

Biopsies of the post-nasal space were repeatedly undertaken but proved nothing more than chronic inflammation. A labelled white cell scan did not depict an area of

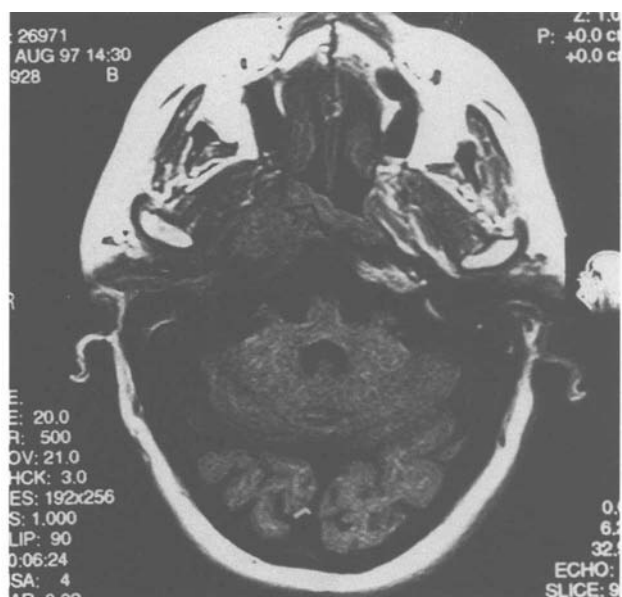


Fig. 1
MRI scan showing mass in the skull base.

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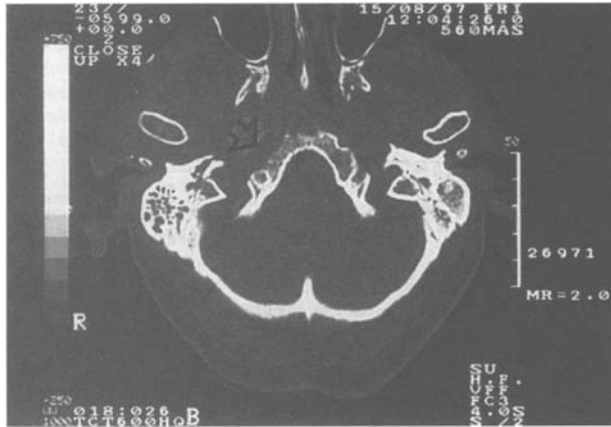


FIG. 2

CT scan showing bony erosion of anterior margin of the foramen magnum.

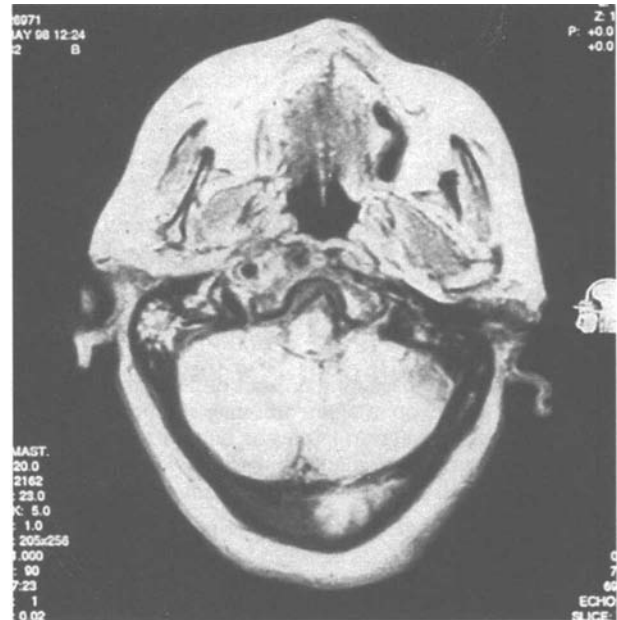


FIG. 4

Follow-up MRI scan showing complete disappearance of the mass from the skull base region.

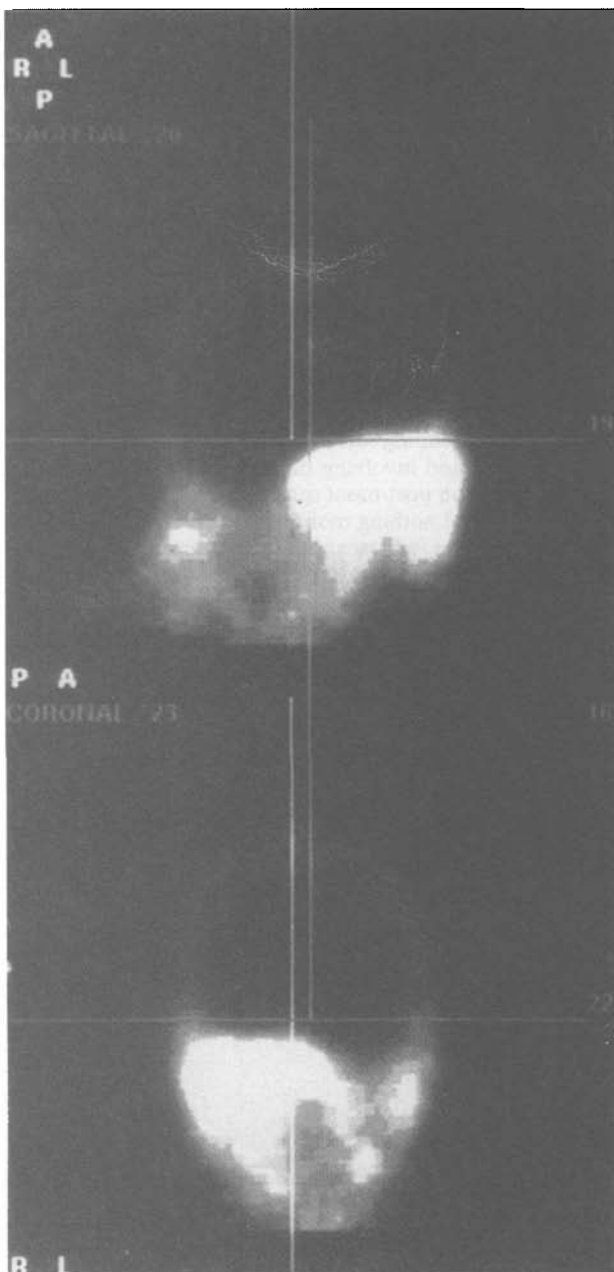


FIG. 3

Gallium scan showing increased uptake in the skull base.

increased accretion but Gallium scan (Figure 3) showed massive uptake along the base of the skull suggestive of skull base osteomyelitis. Electromyography (EMG) of the facial nerve indicated poor prospects of recovery. The granulations in the ear canal were debrided and the patient was put on long-term ciprofloxacin. Deep stereotactic biopsy of the skull base was considered in view of the seemingly indeterminate nasopharyngeal biopsies, but the poor general condition of the patient precluded this.

In the following months, the patient generally stabilized, but the facial weakness persisted. Other cranial nerves remained uninvolved. Serial ESR showed a declining trend. Interestingly enough, a MRI performed nine months after the first imaging did not show to the same extent the intensity of the 'masses' previously reported (Figure 4). The gallium scan was also repeated and this time showed no inflammation. It was therefore deemed that the patient actually had skull base osteomyelitis complicating malignant external otitis (MEO).

Discussion

The possibility of MEO arises in an elderly diabetic patient with an inflamed ear canal and granulations with or without facial weakness. Chandler first presented a series of 13 patients in whom MEO was diagnosed (1968). With the advent of anti-pseudomonal antibiotics and third generation macrolides, this condition is more manageable (Lucente and Parisier, 1996). However due to facial nerve involvement and occasional radiological findings of abnormal masses the condition can mimic malignancy as did our case.

Several nosological criteria have emerged in the literature to confirm the diagnosis of malignant external otitis (Corey *et al.*, 1985; Babiatzki and Sade, 1987; Cohen *et al.*, 1987; Sade *et al.*, 1989; Levenson *et al.*, 1991). These comprise a group of clinical features such as severe otalgia that is out of proportion to the signs, granulation in the external canal, isolation of *Pseudomonas aeruginosa*, and positive temporal bone scanning with Tc99. Interestingly enough, positive MRI findings do not appear in any of

these criteria. Gherini *et al.* (1986) reported on the relatively little experience with MRI in MEO. Bath *et al.* (1998) consider MRI to be better than CT scan in delineating soft tissue although the latter excel in defining the extent of disease and detecting bony destruction. CT is also useful in diagnosing extradural involvement and occlusion of venous sinuses (Grandis *et al.*, 1995). However other temporal bone disease such as carcinoma cannot be distinguished from necrotizing external otitis on the basis of CT findings (Curtin *et al.*, 1982). Because the bone does not remineralize with cure (Rubin *et al.*, 1990), CT scans are not helpful in evaluating recovery.

In MEO with skull base osteomyelitis, the infection preferentially travels in vascular and fascial planes rather than in pneumatized tracts (Nadol, 1980). This accounts for the fact that the mastoid air cell system in the petrous bone is spared and cancellous parts are involved facilitating spread of infection along the skull base as it did in this case. Bernheim and Sade (1989), also noted dermal and osseous inflammation of the ear canal in MEO, but stated that although one can histologically diagnose carcinoma, histiocytosis and Wegeners granulomatosis, the features of MEO are not specific. This was the case in repeated biopsies in this patient. This case did fulfil some of the criteria for MEO, although *Pseudomonas* sp. was isolated much earlier than the onset of facial paralysis. This may well have been due to use of topical gentisone. Perhaps tissue culture may have isolated the organism.

The age of the patient, facial nerve involvement and presence of soft tissue masses on MRI and CT scan evoked suspicion of malignancy but repeated nasopharyngeal biopsies were negative and stereotactic skull base biopsy of these masses appeared unsafe in this rather frail elderly patient. Further efforts to establish the diagnosis of skull base osteomyelitis complicating MEO were not helped by negative results obtained from a labelled white cell scan which may have been due to a low white cell count and reduced immune response. However a gallium scan did establish skull base infection and on this was based our decision to treat this patient for MEO with skull base osteomyelitis. Uri *et al.* (1991) reported the usefulness of a gallium scan in establishing the diagnosis. Gallium scanning is useful for monitoring the results of treatment because its uptake reduces as osteomyelitis is controlled with therapy (Garty *et al.*, 1985). Its uptake reduces totally with control of infection as happened in this patient after some months of therapy. Scanning with technetium on the other hand can still show positive results after infection has subsided.

The effectiveness of ciprofloxacin in MEO is well documented (Levenson *et al.*, 1991), but the therapy should be long term (Uri *et al.*, 1984), although caution has been raised against build up of resistant organisms (Pickard, 1990). The facial nerve function did not recover in this patient. In MEO this does not necessarily signify a grave prognosis (Chandler, 1972). Surgery was not undertaken in our patient. The facial nerve is not merely compressed by granulations but also gets inflamed (Chandler, 1972; Nadol, 1980). It is also affected by neurotoxins made by *Pseudomonas aeruginosa* (O'Sullivan *et al.*, 1978). The role of decompression is, therefore, not very clear (Krause *et al.*, 1988; Benecke Jr, 1989).

Therefore, in presenting this case of MEO complicated by skull base osteomyelitis, the authors wish to indicate how a clinical decision although necessitated by circumstances, appeared feasible despite sophisticated imaging suggestive of malignancy.

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References

- Amorosa, L., Mopdugno, G. C., Pirodda, A. (1996) Malignant external otitis: Review and personal experience. *Acta Otolaryngologica (Stockh)* (Suppl 521): 3–16.
- Babiatzki, A., Sade, J. (1987) Malignant external otitis. *Journal of Laryngology and Otology* 101: 205–210.
- Bath, A. P., Rowe, J. R., Innes, A. J. (1998) Malignant otitis externa with optic neuritis. *Journal of Laryngology and Otology* 121: 274–277.
- Benecke, J. E. Jr. (1989) Management osteomyelitis of skull base. *Laryngoscope* 99: 1220–1223.
- Bernheim, J., Sade, J. (1989) Histopathology of the soft parts in 50 patients with malignant external otitis. *Journal of Laryngology and Otology* 103: 366–368.
- Chandler, J. R. (1968) Malignant external otitis. *Laryngoscope* 78: 1257–1294.
- Chandler, J. R. (1972) Pathogenesis and treatment of facial paralysis due to malignant external otitis. *Annals of Otology, Rhinology and Laryngology* 81: 648–658.
- Cohen, D., Friedman, P. (1987) The diagnostic criteria of malignant external otitis. *Journal of Laryngology and Otology* 101: 216–221.
- Cohen, D., Friedman, P., Eilon, A. (1987) Malignant external otitis versus acute external otitis. *Journal of Laryngology and Otology* 101: 211–215.
- Corey, J. P., Levandowski, R. A., Panwalker, A. P. (1985) Prognostic implications of therapy for necrotizing external otitis. *American Journal of Otology* 6: 353–358.
- Curtin, H. D., Wolf, P., May, M. (1982) Malignant external otitis: CT evaluation. *Radiology* 145: 383–388.
- Garty, I., Rosen, G., Holdstein, Y. (1985) The radionuclide diagnosis, evaluation and follow-up of malignant external otitis (MEO). The value of immediate blood pooling. *Journal of Laryngology and Otology* 99: 109–115.
- Gherini, S. G., Brackmann, D. E., Bradley, W. G. (1986) Magnetic resonance imaging and computerized tomography in malignant external otitis. *Laryngoscope* 96: 542–548.
- Grandis, J. R., Curtin, H. D., Yu, V. L. (1995) Necrotizing (malignant) external otitis: prospective comparison of CT and MR imaging in diagnosis and follow-up. *Radiology* 196: 499–504.
- Krause, D. H., Rehm, S. J., Kinney, S. E. (1988) The evolving treatment of necrotizing external otitis. *Laryngoscope* 98: 934–939.
- Levenson, M. J., Parisier, S. C., Dolitsky, J., Bindra, G. (1991) Ciprofloxacin: drug of choice in the treatment of malignant external otitis. *Laryngoscope* 101: 821–824.
- Lucente, F. E., Parisier, S. C. (1996) Classics from *The Laryngoscope*: James R. Chandler: 'Malignant External Otitis'. (*Laryngoscope* 1968; 78: 1257–1294). *Laryngoscope* 106: 805–807.
- Nadol, J. (1980) Histopathology of pseudomonas osteomyelitis of the temporal bone starting as malignant external otitis. *American Journal of Otolaryngology* 1: 359–371.
- Pickard, R. E. (1990) Caution indicated in prescribing ciprofloxacin [letter; comment]. *Archives of Otolaryngology – Head and Neck Surgery* 116: 742.
- O'Sullivan, T. J., Dickson, R. I., Blockmanis, A., Roberts, F. J., Kaan, K. (1978) The pathogenesis, differential diagnosis and treatment of malignant otitis externa. *Journal of Otolaryngology* 7: 297–303.
- Rubin, J., Curtin, H. D., Yu, V. L., Kamerer, D. B. (1990) Malignant external otitis: utility of CT in diagnosis and follow-up. *Radiology* 174: 391–394.
- Sade, J., Lang, R., Goshen, S., Cohen, K. R. (1989) Ciprofloxacin treatment of malignant external otitis. *American Journal of Medicine* 87 (Suppl 5A): 138S–141S.

- Uri, N., Gips, S., Front, A., Meyer, S. W., Hardoff, R. (1991) Quantitative bone and Ga-67 scintigraphy in the differentiation of necrotizing external otitis from severe external otitis. *Archives of Otolaryngology – Head and Neck Surgery* **117**: 623–626.
- Uri, N., Kitzes, R., Meyer, S. W., Schuchman, G. (1984) Necrotizing external otitis. The importance of prolonged drug therapy. *Journal of Laryngology and Otology* **98**: 1083–1985.

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