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Metastatic carcinoma of the temporal bone presenting as glomus jugulare and glomus tympanicum tumours: a description of two cases

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Abstract
Two cases of metastatic carcinoma of the temporal bone, that simulated glomus tumours on thorough pre-operative evaluation are described. Although rare, metastatic spread to this area is recognized, but presentation in this way is unique.

Key words: Temporal bone; Neoplasm metastasis; Glomus jugulare tumour; Glomus tympanicum tumour; Prostatic neoplasms; Colorectal neoplasms

Introduction

Metastatic carcinoma involving the temporal bone that presents clinically is rare. A recent review of the literature revealed only 148 recorded cases (Nelson and Hinojosa, 1991). The actual incidence of involvement is probably higher, due to metastasis remaining clinically undetected and the temporal bone rarely being fully examined at post mortem. Paraganglionomas, or glomus tumours, of the temporal bone are more common, with a similar incidence to that of all malignant tumours of this region. Their presentation, clinically and radiologically, is usually considered to be different to malignant lesions. This report describes two cases of metastatic carcinoma, one presenting as a glomus jugulare and the other as a glomus tympanicum tumour. In the first case this was the primary presentation of the prostatic disease. We believe these to be the first such cases described in the English literature.

Case reports

Case 1

A 60-year-old man was referred to the neurosurgical unit as a petrous glomus tumour, with a two month history of a progressive loss of function of the left lower six cranial nerves VIIth–XIIth, pulsatile tinnitus and left-sided deafness. Magnetic resonance imaging (MRI) had already been performed and showed a vascular mass eroding the infralabyrinthine portion of the temporal bone, surrounding the carotid artery and extending into the petrous apex, bulging the dura of the posterior fossa (Figure 1).

Clinical examination confirmed the loss of function of the VIIth–XIIth cranial nerves, as well as reduced sensation in the distribution of the Vth nerve and a partial VIth nerve palsy. The otoscopic appearance of the middle ear was normal and audiometry confirmed a severe left-sided sensorineural hearing loss. Computed tomography imaging (CT) showed irregular enlargement of the left jugular foramen (Figure 2). Selective cerebral angiography was performed and demonstrated a large, vascular tumour with a pathological circulation derived from the ascending pharyngeal and occipital arteries (Figure 3). Embolization had previously been performed before referral with minor improvement of symptoms, and this was repeated. After embolization the patient went into retention and was catheterized. There was no past history of urological problems or prostatism.

The pre-operative diagnosis of a petrosal glomus tumour

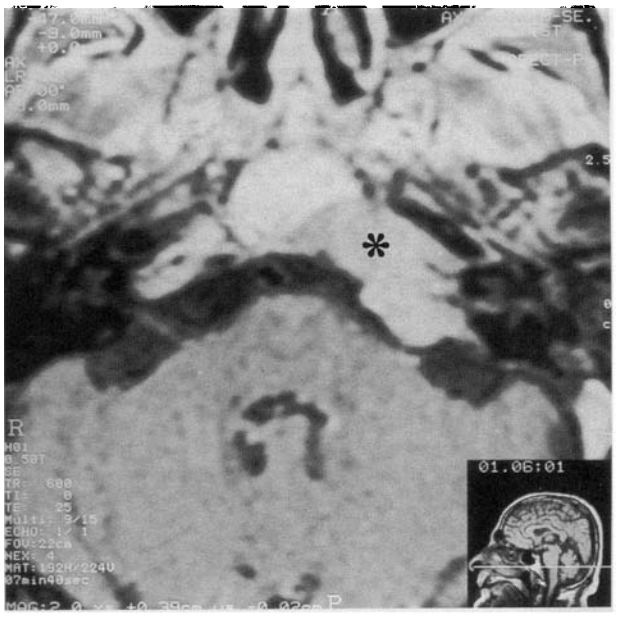

FIG. 1

Case 1. Axial MRI scan (T1 weighted) at the level of the cerebellopontine angle demonstrates the large tumour mass (star) extending towards the petrous apex, causing cerebellar displacement.

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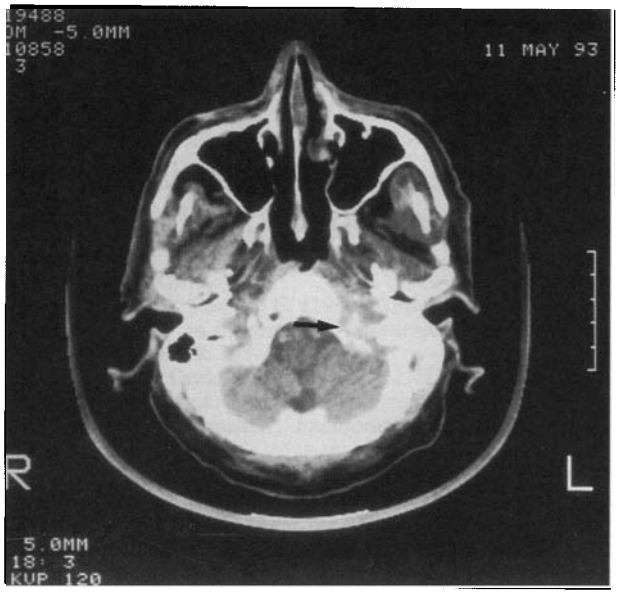

FIG. 2

Case 1. Axial CT scan demonstrating erosion of the left jugular foramen, indicated by the arrow.

was made on the basis of the angiography and the radiological destruction of the infra-labyrinthine petrous temporal bone, Fisch type C2/3 (Fisch, 1982).

Surgical exploration was performed using the postero-lateral approach (Cheesman and Symon, 1987). Tumour was found medial to the jugular bulb in the infra-labyrinthine portion of the temporal bone. It involved the facial nerve and carotid artery in the region of the processus cochlearformis, and the neural compartment of the jugular foramen. Frozen section was not performed and the per-operative diagnosis was that of a petrosal paraganglionoma. A radical excision was carried out. The

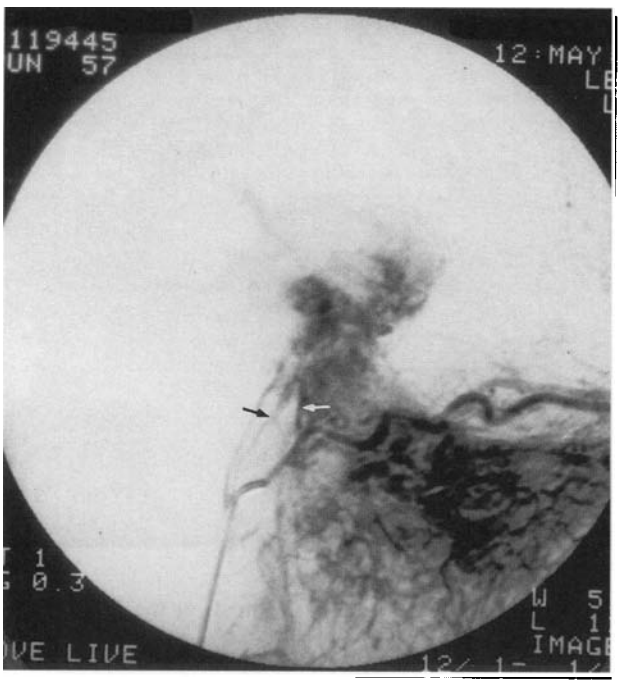

FIG. 3

Case 1. Selective cerebral angiogram showing the vascular mass with feeding vessels from occipital (white arrow) and ascending pharyngeal (black arrow) arteries.

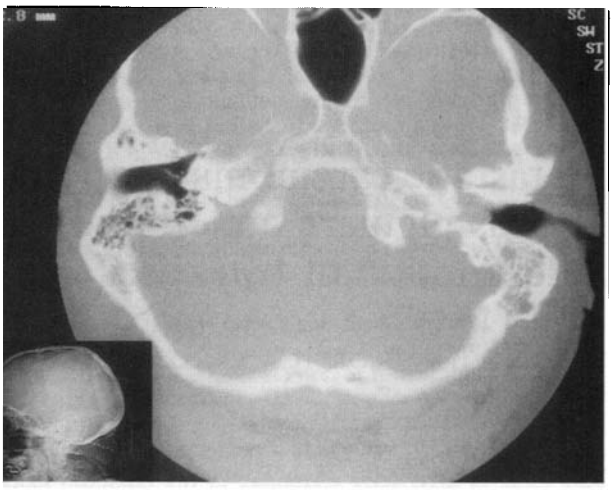

FIG. 4

Case 2. Axial CT scan at the level of the middle ear cleft. The soft tissue mass is shown in the left middle ear, without any evidence of bony erosion.

patient made a good post-operative recovery, but there was no recovery of cranial nerve function.

Definite histology of the lesion was that of an adenocarcinoma of metastatic origin. Immunofluorescent staining confirmed it as arising from a prostatic primary tumour. Rectal examination revealed a rock hard smooth prostate compatible with prostatic carcinoma, and serum alkaline phosphatase and prostate specific antigen were both highly raised. A bone scan showed multiple bony metastases. The patient was therefore commenced on a course of post-operative radiotherapy plus anti-androgen treatment.

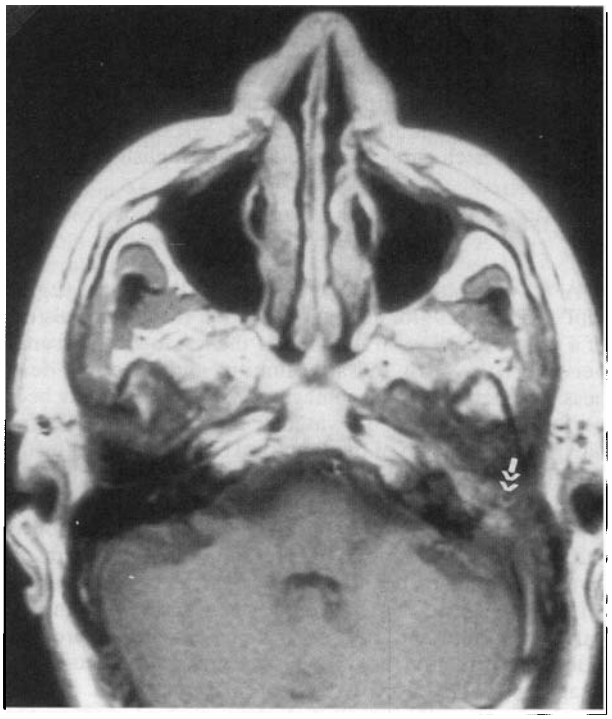

FIG. 5

Case 2. Axial MRI scan (T1 weighed) post-gadolinium contrast demonstrating enhancement of the mass in the middle ear cleft (indicated by the arrow).

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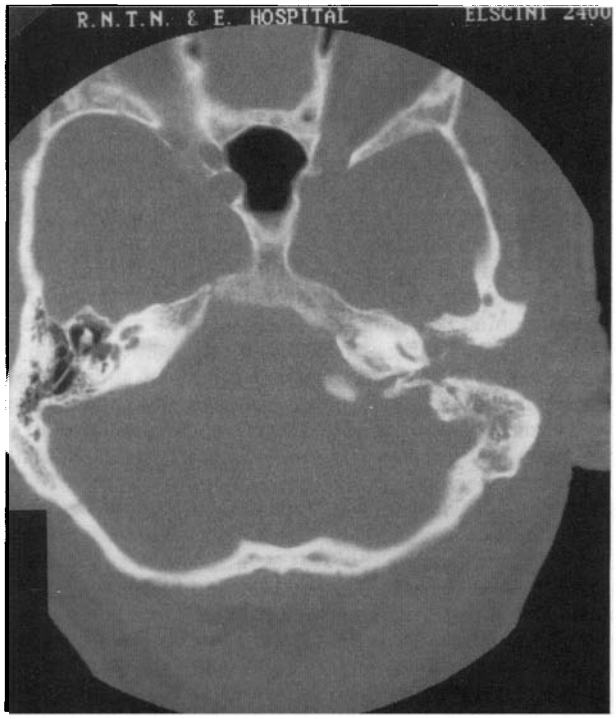

FIG. 6

Case 2. Axial CT scan illustrates the expansion of the soft tissue mass with bony erosion of the anterior mesotympanum and external meatus.

Case 2

A 53-year-old man presented to the ENT department with a four-month history of left-sided hearing loss, associated with tinnitus which was occasionally pulsatile. Over the preceding three weeks he had developed a left-sided facial weakness, which seemed to be improving, and some otalgia. Examination revealed an almost complete left VII palsy but no other cranial nerve palsies. Otoscopy showed a pink, fleshy, tender mass bulging behind an intact tympanic membrane. Physical examination was otherwise unremarkable. Audiometry revealed a predominantly conductive hearing loss of 50 dB on the left side. He had undergone a large bowel resection for a Duke's stage B adenocarcinoma of the colon four years previously, but there had been no sign of any recurrence on close follow-up.

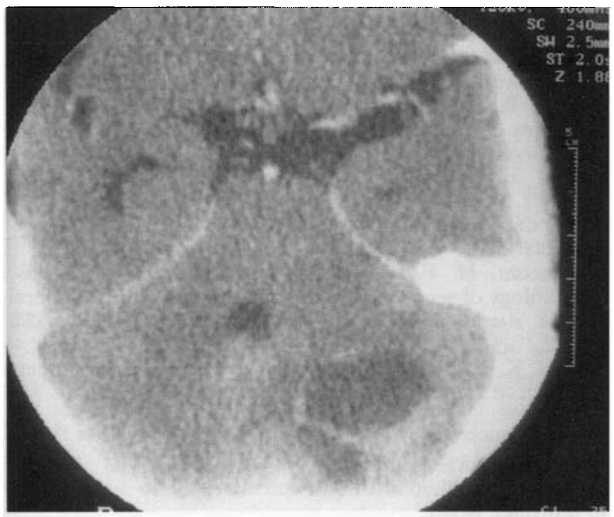

FIG. 7

Case 2. Axial CT scan at the level of the posterior cranial fossa demonstrates two areas of low attenuation consistent with cerebellar metastases.

A provisional diagnosis of a possible glomus tumour was made. A CT scan showed soft tissue opacification of the left middle ear, without any bone erosion and an intact, normal, jugular fossa (Figure 4). A MRI scan revealed similar features with some enhancement of the tumour with gadolinium (Figure 5). Blood haematology and biochemistry were both normal and by this time the facial weakness had completely resolved. This all supported the diagnosis of a glomus tympanicum tumour. He was therefore added to the waiting list to be admitted for excision.

The patient returned as an emergency three months later, before his operation date, with increased pain, and bleeding from the left ear. Examination revealed the tumour extending into the external auditory meatus. The facial nerve function was almost entirely intact. A biopsy was performed and a CT scan carried out. The histology of the biopsy was reported as invasive papillary adenocarcinoma, with appearances suggestive of a large bowel metastasis. The CT scan showed that the lesion had expanded, now with bony erosion (Figure 6), and the appearance of two areas of low attenuation in the left posterior cranial fossa consistent with metastases (Figure 7).

In view of the diagnosis and cranial metastases, the patient was referred to the oncology department, where he was treated palliatively with radiotherapy.

Discussion

Metastases in the temporal bone are rare but are being reported with greater frequency. In 1962 a review of the literature revealed only 25 such cases Maddox (1967), Hill and Kohut in 1976 found a total of 103 and more recently Nelson and Hinojosa (1991) discuss the 148 published reports. Since then there have been several more reports. A number of different groups have studied the temporal bones of patients with a diagnosed malignancy at a different anatomical site. Jung *et al.* (1986) found approximately 24 per cent of a series of patients with a proven distant carcinoma had temporal bone metastases. Nelson and Hinojosa (1991) studied 1,200 bones in the temporal bone collection of the University of Chicago. Sixty bones from 33 patients were noted to contain secondary lesions from a distant primary malignancy. In 27 patients the temporal bone metastases were bilateral, however seven cases were included where direct or meningeal spread had occurred from either a tumour originating in a nasal sinus or from intracranially. In the other 20 cases, the mode of spread of the malignancy was described as being haematogenous, and the petrous apex was the most common site for the secondary deposit (involved in all of the bones). This may explain the high incidence of bilateral metastases, as the petrous apex contains vascular channels and may filter out neoplastic cells from the circulation (Proctor and Lindsay, 1947). Lesions in this area may remain asymptomatic and clinically undetected for a significant period, allowing time for bilateral temporal bone seeding to occur. Interestingly the middle ear in nine of these bones was described as being the site of metastatic tumour, as in the second case described above, but whether these were isolated lesions or as a result of spread from the deposit in the petrous apex was not indicated.

There is no doubt of the significant incidence of temporal bone metastases in malignancy. Previously these may not have been recognized due to the temporal

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bone not being fully examined at post mortem or deposits remaining clinically silent. The apparent increase in the incidence of clinical cases presenting may be because of the improved survival of patients with carcinoma and therefore more chance of signs developing, or a reflection of greater suspicion and reporting of more minor otological symptoms. The most common primary tumours leading to secondary temporal bone deposits are breast, lung, kidney and prostate (Nelson and Hinojosa, 1991). There are only four reported cases of metastases from adenocarcinoma of the colon (Schuknecht *et al.*, 1968; Kobayashi *et al.*, 1986; Ruah *et al.*, 1987), and this represents the fifth such case.

Glomus tumours or non-chromaffin paraganglionomas of the temporal bone, originate from the paraganglion cells of either the jugular bulb or from along the course of Jacobsen's nerve. They are the most common tumour of the middle ear (Stewart, 1993). Greer *et al.* (1976) reviewed 99 cases of glomus tumour and 110 malignant tumours (comprising only nine metastatic cases) of the temporal bone seen at the Mayo Clinic. Their incidence was similar at approximately eight to nine cases per 100 000 population. The commonest symptoms were hearing loss (70 per cent), tinnitus (64 per cent) and otalgia (24 per cent) for glomus tumous, and pain (70 per cent), otorrhoea (49 per cent) and hearing loss (41 per cent) for malignant lesions. Facial weakness was seen in 11 per cent of glomus tumours and 23 per cent of malignancies, but lower cranial nerve palsies were more common in the glomus series (13–18 per cent compared with two to seven per cent).

The radiological appearances of both glomus tumours and temporal bone metastases although considered different may exhibit similar features. CT scanning shows erosion of the temporal bone with each tumour, but if this is centered on the jugular foramen, it is logical to assume that it is a glomus jugulare tumour. Middle ear involvement is usually found with paraganglionomas, however with the increased use of MR scanning as part of a general neurological investigation, medially placed glomus tumours are being detected more frequently, and these may have a paucity of otological symptoms. Magnetic resonance imaging shows an enhanced image on T2 weighting and with gadolinium contrast in both cases, often with flow voids indicating vascular spaces. Angiography is an aid to diagnosis, but rather than truly differentiating the two tumour types it merely confirms the presence of a vascular tumour, and its main role is for pre-operative embolization. The diagnosis is made from the clinical presentation as well as an evaluation of the CT and MR images. This paper demonstrates that the clinical and radiological findings normally associated with glomus tumours are not necessarily diagnostic.

In the second case presented, where a red mass was seen behind the drum, there could be no doubt about the clinical diagnosis of a glomus tumour. The subsequent rapid bone erosion on repeat CT imaging was the pointer to the correct diagnosis. Goebel *et al.* (1987) described three cases of primary adenocarcinoma of the temporal bone presenting as glomus tumours. In these cases, clinical presentation, CT scanning and angiography were thought, as in our paper, to be diagnostic of a paraganglionoma. The correct diagnosis was only made on histological examination of the operative specimen. In one of their patients, more aggressive bony erosion than expected from a glomus tumour was found on repeat CT scanning, and this resembles the second report in our paper. Aggressive, progressive bony erosion does seem to be more characteristic of malignant disease than of a paraganglioma.

There is a degree of overlap in symptomatology of glomus and malignant tumours of the temporal bone. In the cases reported here, it can be seen that all the accumulated clinical and radiological evidence pointed to a glomus lesion in both instances, even though some features were common to both tumour types (pain, VIIth nerve palsy, and in retrospect some radiological findings). Although rare, there is a definite incidence of temporal bone metastases from a distant primary malignancy. The recognition and diagnosis of these deposits may increase with the lengthening survival times for patients with a carcinoma and as more detailed means of radiological examination becomes available. These cases emphasize the importance of both a past history of carcinoma, plus a thorough physical examination, including per rectal, in all cases of suspected temporal bone tumours.

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