Short report

Hidradenoma with intracranial involvement

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SUMMARY A case of recurrent hidradenoma of the external ear with intracranial spread is described. The presentation, classification and management of this rare tumour are discussed and the importance of adequate long term review is stressed.

The hidradenoma is a rare tumour of ceruminous glands of the external auditory meatus, which as modified apocrine sweat glands normally produce a watery secretion as a constituent of cerumen. Differing opinions are expressed in the literature with regard to the classification and management of the tumour. In this case report, we describe a recurrent hidradenoma with intracranial spread, a feature only rarely described before.^{1,2}

Case report

The patient, a 79-year-old Caucasian male, gave a previous history of two operations to the left ear in 1969 and 1974. He had presented elsewhere in 1969 with a sub-acute painful swelling, 1 cm in diameter, located in the posterior aspect of the left external auditory meatus. Radiographs of the skull and internal auditory meati were normal. The lesion was excised without complication, the histological diagnosis being that of ceruminoma (hidradenoma).

The patient was lost to follow up until he presented himself to a different surgeon in 1974 with pain and deafness in the left ear. On examination, a large red hard mass was found, occluding the left external auditory canal and attached to the bony roof. Radiological examination was again normal. The mass was removed with a margin of apparently normal surrounding skin. The histological appearance of the lesion was identical to that of 1969. The patient was then lost to follow up.

He presented to this hospital as an emergency in July 1982. He was unable to give a clear history, but his family reported a marked emotional and intellectual change over the previous few months, in addition to a slowly progressive left facial weakness, pain in the left mastoid region, and weakness of the right arm and leg. On examination, he

was drowsy, irritable, disorientated and dysphasic, with a left lower motor facial weakness and a right hemiparesis. Examination of the external auditory meati was normal. He was deaf in the left ear. A chest radiograph showed no abnormality. A CT scan (fig 1) revealed a large tumour with an enhancing capsule emanating from the floor of the middle fossa and occupying the lower aspect of the temporal lobe. There was surrounding oedema with mass effect and two apparently cystic areas within the temporal lobe extension. Carotid angiography failed to show any tumour circulation.

At elective temporal craniotomy, the tumour was seen to arise from the petrous bone, pass through the dura, and extend into the upper surface of the temporal lobe invaginating to the tip of the lobe. Two secondary capsules

Fig 1 CT scan showing large tumour emanating from the floor of the middle fossa extending into temporal lobe on the left.

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were found deep within the temporal lobe extension, each containing altered blood. The intradural position of the lesion was removed completely. The patient made an excellent recovery from operation. One year later the only abnormalities were a mild nominal dysphasia, a dense left facial weakness and left sided deafness.

Pathology

The tumour measured approximately 3 cm × 4 cm, with a firm grey capsule consisting of fibrous tissue alone. Histologically the characteristic appearance was of papillary and tubular structures lined by cuboidal cells with small regular spheroidal nuclei arranged around central vascular cores. Frequent mitotic figures were seen (fig 2). The thickened dura mater contained islets of tumour infiltrating dural sinuses. The features were of a hidradenoma (ceruminoma).

Discussion

Tumours of the ceruminous glands of the external auditory meatus are rare. The first clear description of adenocarcinoma of the sweat glands of the ear, with myxomatous degeneration of the stroma, was in Case 10 of the series published by Haug³ in 1894. Unfortunately many subsequent reports contain insufficient clinical or pathological detail to allow definite classification. Even in acceptable reports there has been considerable discussion about the most appropriate name for the tumour.^{2,4-10} We agree with the view of Johnstone *et al*¹⁰ that the term *ceruminoma* is unjustifiable as the lesions are histologically similar to other sweat gland tumours, and therefore have used the term *hidradenoma*.

Previous authors have also differed with regard to the presenting features, management and prognosis of this condition.^{5,8,11,12} In their review of the litera-

ture in 1961, Peytz and Ohlsen¹¹ found 26 cases, 17 occurring in men and 9 in women, with an age range between 14 and 78 years. Patients may present with a mass, pain, bleeding or purulent discharge in the ear and rarely with apparent cranial nerve involvement and features suggestive of intracranial spread. Many such tumours are initially misdiagnosed as osteomata or exostoses associated with chronic otitis media, or rarely as glomus tumours. They may grow slowly and keep their histological identity, but may run a fulminating course with numerous recurrences. Whilst usually located in the external auditory meatus, spread to the tympanic cavity, extradural spread involving the 7th and 8th nerve, and facial weakness have been described.^{2,8,11,12} Pain is usually associated with local infiltration of the tumour, particularly along perineural lymphatics.

In addition to local spread, haematogenous dissemination can occur. The first report of pulmonary metastases came from Nussbaum¹³ in 1932. Pulec² reported a woman dying at the age of 45 years in whom necropsy revealed metastatic cylindroma in the brain, lungs, mediastinum, pleura, liver, left kidney and para-aortic lymph nodes. O'Neill and Parker⁴ reported a woman of 19 years who presented with a small lump in one ear which in spite of local resection and deep X-ray therapy, had spread 4 years later to involve the other ear and the deep cervical lymph nodes bilaterally.

With regard to management, whilst Juby⁸ proposed that local excision of the tumour seemed to be the treatment of choice he added that the patient should be kept under prolonged review. Johnstone *et al*¹⁰ commented on the high rate of local recurrence of these tumours (50%) in comparison with skin hidradenoma elsewhere (5%). They suggested that this might in part be due to the difficulty of adequate excision in the external auditory canal, but added that there seemed to be a preponderance of the less benign forms of hidradenoma at this site, and that even tumours without obvious malignant features histologically, seemed on occasion to spread widely despite radical surgical measures. Other authors also stressed the need for radical treatment in the light of a potentially bad prognosis, and for regular follow up.^{4,5,9,14} No clear view is expressed in the literature about the treatment of pulmonary metastases. Ramadass and Satuanarayana⁷ treated one patient with cyclophosphamide, but noted no decrease in the size of the chest secondaries. The role of chemotherapy would therefore seem to be unclear.

The aetiology of the intracranial involvement in our case is unclear, but it could be due to tumour penetration or be derived from residual ectodermal tissue in the tympanic cavity. In the light of our case,

Fig 2 Characteristic features of a hidradenoma (ceruminoma) with papillary and tubular structures orientated around central vascular cores. (Haematoxylin and eosin ×200.)

serial CT scanning would seem to be indicated in the follow up of cases, in addition to serial chest radiographs.

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