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Ossifying fibroma of the temporal bone

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Abstract
A rare case of ossifying fibroma of the temporal bone is presented. Fibro-osseous lesions are benign neoplasms but may show an aggressive behaviour when invading important anatomical structures. The lack of experience in the treatment of those tumours is reflected in the small relative literature. The purpose of this paper is to contribute to the few cases already reported.

Key words: Fibroma, Ossifying; Temporal Bone

Introduction

Ossifying fibroma is a benign fibrous lesion in the head and neck region. Mandible and maxilla are the commonest sites. However, there are reports that the lesion develops in the nasal bones, the ethmoid cells, or the orbit.[1] Due to its rarity, the head and neck surgeon is not very familiar with the diagnosis and treatment of ossifying fibroma. In this paper we report a case of an ossifying fibroma of the temporal bone treated in our department, with a review of the current literature.

Case report

The patient, an 18-year-old female, was referred to our department after two unsuccessful surgical attempts to cure a stenosis of the right external auditory canal (EAC) the previous year.

The symptoms started two years before referral, when the patient suffered from an episode of acute right ear otalgia; she received antibiotics but the symptoms persisted. At the same time she noticed a decrease in her hearing acuity in the same ear. The family physician noticed the presence of a mass obstructing the right EAC and she was referred to an ENT specialist, who confirmed the presence of a mass filling the EAC. The audiological tests showed a conductive hearing loss of 45 dB. Computed tomography (CT) scan of the facial and petrous bones revealed a mass, lateral to the right temporomandibular joint and in close relation to the roof of the bony EAC. The mass was oval, with ring-like continuous peripheral and speckled central calcification and measured $1.7 \times 1.3 \times 1.2$ cm (Figure 1). It was connected to the EAC with a cord whose density was higher than that of soft tissue. The cord was extended into the EAC along the roof and the anterior wall reaching the tympanic membrane, without osteolysis. There was a narrowing of the EAC, while the middle ear cavity was normal as well as the ossicular chain. The other intracranial structures were normal.

The patient had already been operated twice for recurrence, first using an endaural and second a post-auricular approach. During the first operation, material was obtained from the lesion and sent for pathological examination. The diagnosis was of an ossifying fibroma. However, the patient relapsed again and she was then referred to us.

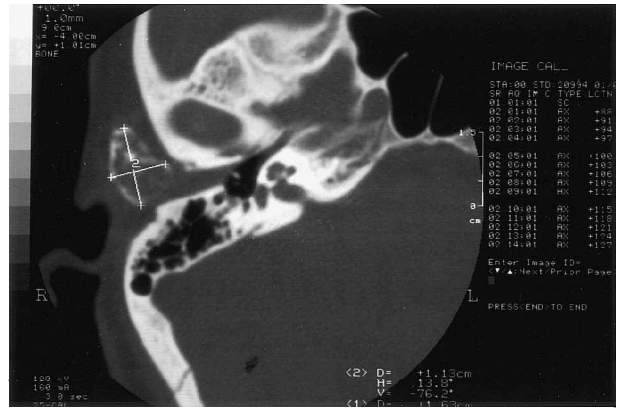

Fig. 1
Pre-operative CT scan of the temporal bones showing an oval mass with ring-like continuous peripheral and speckled central calcification, lateral to the right temporomandibular joint and in proximity to the roof of the EAC.

On referral, the patient had a completely obstructed EAC by a fibrous mass extending to the roof, the floor and the posterior wall of the canal, leaving the skin of the anterior wall intact. The mass was pale, spongy, vascular and irregular in shape with micronodular haemorrhagic surface. Mucopurulent secretion was noted in the EAC. The tympanic membrane could not be inspected as the mass spared only a very narrow fissure-like area in the lower part of the anterior wall of the EAC. A new CT scan showed deletion of the lateral part of the ossifying pathology of the first scan; however, its central, cord-like part, with density higher than soft tissue, still occupied the EAC in close relation to its roof and extended to the ear drum causing retraction (Figure 2).

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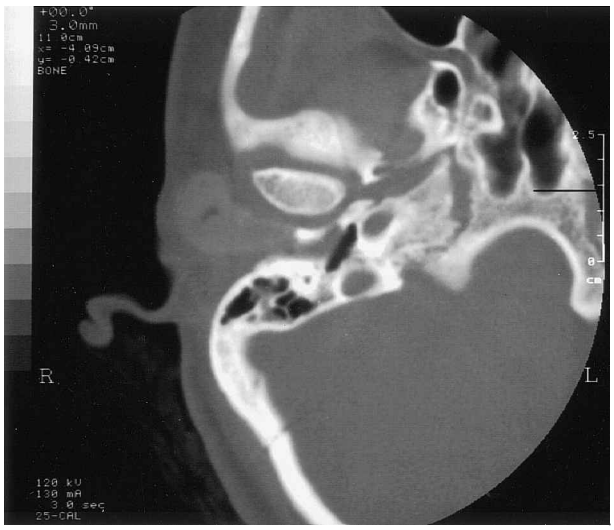

FIG. 2

CT scan of the temporal bones following the two initial operations. The oval ossified mass is not present but there remains its cord-like extension with density higher than that of soft tissues, occupying the EAC.

The patient was operated under general anaesthesia. The entire lesion was removed, along with the skin of the EAC, sparing only the skin of its anterior wall. Excision started from the region between the helix and the tragus to the temporal fascia, which was intact and continued to the bony EAC up to the tympanic membrane. Lateral to the ear drum and medial to the tumour, cholesteatoma of the EAC had been developed and was also excised. There was no need to enter the mastoid cavity or the middle ear space. Diamond burrs were used, as bleeding was remarkable. The cavity was plugged with antibiotic gauze that was left in place for 15 days post-operatively.

Histopathology

Several pieces of white homogenous elastic tissue were received, 0.7×1.0 cm in greatest diameter. Histological examination with haematoxylin-eosin showed a fibro-osseous lesion composed of broad anastomosing fascicles of loose vascular connective tissue with moderate to marked cellularity, and scattered bone spicules, characteristically lined by osteoblasts producing a rim of lamellar bone. With polarized light, bone was laid down in the centre of the spicules. At several areas, scattered osteoclasts and a mild chronic inflammatory infiltrate were seen. The features were those of an ossifying fibroma (Figure 3).

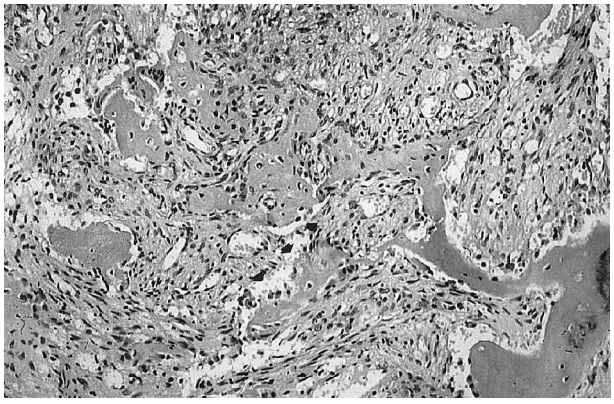

FIG. 3

Bone trabeculae with osteoblastic rim (arrows), in between loose cellular stroma (H&E; ×100).

After removal of the tamponade, recurrence of the tumour was noticed. For six months post-operatively, surgical debulking of the EAC under local anaesthesia was necessary, in order to remove relapsing tissues of the tumour. The procedure took place initially every two days, then twice a week and finally each week. At that time, the status of the EAC seemed to have stabilized in a normal condition and cleaning was discontinued.

The patient has been followed-up on a regular basis every three months since and has remained free of disease for two years.

Discussion

Ossifying fibroma is a benign tumour, easily excised, but occasionally shows an aggressive behaviour resulting in local destruction and recurrences.

To our knowledge, there are only three reports in the English literature of ossifying fibroma of the temporal bone. The first one, by Stecker,[2] was misdiagnosed pre-operatively as a glomus jugulare and the author reported a six-month disease-free period; the second one, by Levine *et al.*,[3] presented as exostoses of the external ear and recurred twice in a six-month period. Finally, the most recent report, by Zappia *et al.*,[4] presented as an asymptomatic mass of the mastoid area, and was complicated after 18 years by an epidural abscess and a cholesteatoma. Two other reports in the literature of ossifying fibromas of the temporal bone, turned out to be cases of monostotic fibrous dysplasia.[5,6]

The facial bones are sites for developing of several fibro-osseous lesions, such as monostotic and polyostotic fibrous dysplasia, cementifying fibroma, benign cementoblastoma, periapical fibrous dysplasia and ossifying fibroma. Confusion regarding their terminology and classification began in 1927, when Montgomery first used the term ossifying fibroma to describe a lesion that was eventually diagnosed as fibrous dysplasia.[7] In 1938, Lichnenstein[8] considered as fibrous dysplasia a tumour that might have been an ossifying fibroma and, in 1958, Jaffe[9] suggested that all types of fibro-osseous lesions of the facial bones should be regarded as types of fibrous dysplasia.

Today, ossifying fibroma is considered a distinct pathological entity that can be accurately diagnosed using specific clinical, radiological and pathological criteria. The greater incidence of occurrence is in the third to fourth decade of life, without sex predilection. However, the younger the patient, the more aggressive the behaviour of the tumour. Some authors claim that the proliferative activity of the tumour subsides with time.

Its clinical presentation varies according to the bones that are involved. However, swelling, sensory disturbances and asymmetry are the most common symptoms when the maxilla or the mandible are involved.[10] When the temporal bone is involved, conductive hearing loss, otorrhoea and pain are the most common complaints. Our patient experienced all the above-mentioned symptoms, as did the patient reported by Levine *et al.*[3] On the other hand, the case reported by Stecker[2] presented only with hearing loss and tinnitus, mimicking a glomus jugulare, and the one reported by Zappia *et al.*[4] presented as an asymptomatic mass in the mastoid area.

Imaging of the facial bones shows a round or oval mass, well demarcated and radiolucent, which appears like an egg shell. Usually, the lesion has lower density than the surrounding structures and older areas tend to be calcified. On the contrary, the margins of fibrous dysplasia are not

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well defined, and there is no osseous peripheral response but a ground glass appearance.[11] In the present report, radiological findings were consistent with an ossifying tumour as it was oval, with distinct boundaries of thin bone and calcified areas. In Stecker's[2] report, simple X-rays used to assess the temporal bone were normal, while in Levine's[3] report temporal bone tomography revealed obliteration of the EAC with a dense mastoid bone and normal middle ear space. Finally, in the most recent report, by Zappia *et al.*,[4] a computed tomography (CT) scan was performed only when the tumour relapsed and showed hyperostosis of the temporal, parietal and occipital bones, along with a cholesteatoma and an epidural abscess which complicated that case.

Pathologically, ossifying fibroma presents as evenly spaced spicules of bone rimmed with osteoblasts and osteoclasts, randomly distributed within a fibrous stroma. Most of the spicules are centrally composed of woven bone but there is evidence of lamellar transformation at the periphery. The lesion resembles fibrous dysplasia but the finding of woven bone with lamellar bone producing peripheral osteoblasts distinguishes ossifying fibroma from the latter. Fibrous dysplasia is characterized by bony trabeculae composed of woven bone without lamellar transformation and no apposition of the spicules by osteoblasts and osteoclasts.[12]

The biochemical profile of the patients is usually normal, with an occasional rise in alkaline phosphatase, if there is chronic osteolysis.

The differential diagnosis should include fibrous dysplasia and osteogenic sarcoma. The lack of literature concerning these tumours reflects their rarity and the limited experience for the proper surgical treatment of each individual case.

Levine *et al.*[3] stressed the fact that, although the tumour is biologically benign, its anatomic position may involve vital structures that should not be removed, because of danger to the patient. They suggested that periodical surgical removal of pathological tissue may be necessary for definitive treatment of the lesion, respecting these structures.

We report this rare case of ossifying fibroma of the temporal bone as a contribution to the small number of cases that have been reported in literature. In our opinion, persistent and careful surgical debulking is the answer to the problem caused to the ear surgeon by this rare entity, which may have an aggressive behaviour. In our case, this strategy seemed to have a therapeutic result for the patient. However, any experience relative to the behaviour and treatment of these tumours would be welcomed, in order to enhance our knowledge.

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