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Pigmented villonodular synovitis of the temporomandibular joint

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Abstract
The first case of pigmented villonodular synovitis of the temporomandibular joint in a Chinese patient is reported. The clinicopathological features are described and the presentation as a parotid mass is emphasized. This rare tumour requires a high clinical suspicion for diagnosis. For removal, meticulous dissection of tumour and facial nerve is necessary.

Key words: Synovitis, pigmented villonodular; Temporomandibular joint diseases; Parotid gland

Case report

A 10-year-old girl with a left preauricular swelling present for one year was seen in the Division of Otorhinolaryngology at the Prince of Wales Hospital in June 1992. The mass had not changed significantly in size during this time. Physical examination showed a left preauricular swelling which was thought to originate in the parotid. It measured approximately 1.5 × 1.5 cm. The facial nerve function was normal. An ultrasonogram of the area showed a hypoechoic lesion within the parotid gland which measured 10 × 10 mm. Fine needle aspiration biopsy was inconclusive.

A left superficial parotidectomy was performed but no tumour mass was identified in the specimen. However, a 2 × 2 cm yellowish mass, firm in consistency was found lying deep to the facial nerve. During dissection it was found to be arising from the left temporomandibular joint extending medial to the medial surface of the neck of the ramus of the mandible. Facial nerve dissection was extremely difficult because the nerve was stretched over the tumour. The tumour was removed together with part of the left temporomandibular joint capsule. The patient had a complete facial nerve paralysis immediately after surgery but there were no problems with jaw movement. The facial nerve function recovery was complete after eight weeks.

Pathology

The parotid gland specimen was histologically normal. Macroscopically, the tumoral tissue measured 2.5 × 2 × 2 cm and was soft and yellowish in colour, lobulated, well circumscribed and no necrosis or haemorrhage was identified. Microscopically, sections of the tumour showed cleft-like spaces lined by a proliferative synovial lining (Figure 1). The underlying stroma showed multinucleated giant cells scattered among mononucleated cells (Figure 2). Focal irregular distribution of osteoclasts were noted. No frank nuclear anaplasia was noted. Tissue from the temporomandibular joint showed the same morphological spectrum as the tumour. This histological picture confirmed the diagnosis of pigmented villonodular synovitis on both the tumour and the material from the temporomandibular joint.

Discussion

Pigmented villonodular synovitis (PVS) is a benign lesion first described by Jaffe *et al.* (1941). The condition develops in the joint lining and is characterized by an exuberant granulomatous inflammation. PVS may be either localized or diffuse (Jaffe, 1958). In localized PVS, the synovial membrane may give rise to one or more yellow-brown nodular growths. Diffuse PVS is characterized by numerous fine brownish, villous, nodular lesions, containing variable foci of bright orange areas (lipid laden histiocytes) involving the entire synovial membrane of a single joint. Rarely, a lesion may extend into the bone and simulate a primary or metastatic bone tumour. The importance of this behaviour is that it may suggest malignancy, both radiologically and histologically to both clinician and pathologist. Although the lesion is benign, it is often composed of exuberant masses of plump fibrohistiocytic cells, which may be mistaken for a sarcomatous process. It may be confused wth lesions such as fibrosarcoma, undifferentiated sarcoma and chondroblastoma. The infiltrative nature of this lesion increases the risk of local recurrence following surgical excision.

Most patients with PVS are young to middle-aged adults. In a review of the twelve previously reported cases affecting the temporomandibular joint, there were eight female patients and four male patients (Eisig *et al.*, 1992). Most of these cases presented

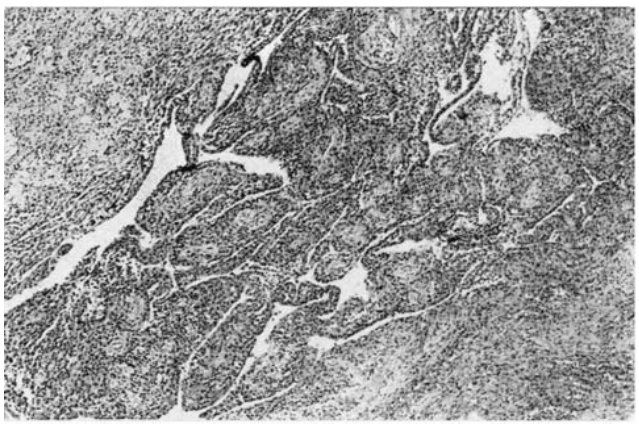

FIG. 1
Typical cleft-like spaces lined by active synovial cells. (× 60).

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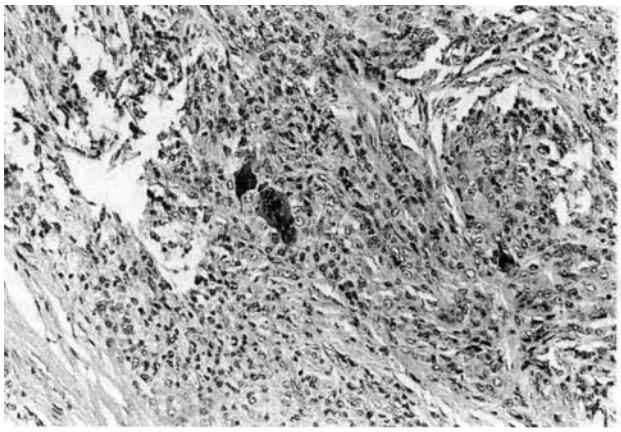

FIG. 2

Multinucleated giant cells are scattered among mononucleated cells (× 100).

as a preauricular swelling or a parotid mass. One patient presented with hearing loss from a mass in the ear canal which was a direct extension of a lesion arising from the right temporomandibular joint. Symptoms were referable to the temporomandibular joint which included pain, decreased joint movement and clicking. The radiological features of invasive PVS are not pathognomonic, but may indicate a synovial lesion which has invaded bone. A plain radiograph is adequate to detect erosion of bone. In our patient the ultrasonogram failed to detect the PVS mass possibly because the consistency of PVS tissue is similar to that of the parotid gland.

Summary

Pigmented villonodular synovitis of the temporomandibular joint is a rare cause of an apparent parotid mass and appears to predominate in females. PVS should always be included in the differential diagnosis of any solitary joint process with erosive and cyst-like bone changes. Bone erosion can be recognized in a radiograph of the temporomandibular joint which should be performed if there are any joint symptoms. A CT scan of the base of skull should be performed pre-operatively if a diagnosis of PVS is considered in order to show deep lobe extension. During removal of the tumour, the dissection must be performed with great care to avoid damage to the facial nerve as it is situated deep to the nerve.

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