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Pigmented villonodular synovitis of the temporomandibular joint: report of a case

ALESSANDRO FRANCHI*, PAOLO FROSINI†, ROBERTO SANTORO†

Abstract
Pigmented villonodular synovitis is a benign reactive process of the synovial tissue, that usually involves the knee or other major joints. Reports of this entity in the temporomandibular joint are rare. The authors describe a case involving this joint, initially clinically diagnosed as a parotid tumour.

Key words: Synovitis, pigmented villonodular; Temporomandibular joint

Introduction

The term pigmented villonodular synovitis was introduced by Jaffe *et al.* (1941) to describe an idiopathic proliferative lesion involving tendon sheaths, bursae and diarthrodal joints. It more often affects the knee and the fingers, and less frequently the hip, ankle or wrist joints. The purpose of this report is to describe the clinico-pathological features of a case involving the temporomandibular joint, which is an extremely rare location for this entity.

Case report

A 59-year-old woman was admitted to the Oto-Rhino-Laryngology Clinic of the University of Florence with a complaint of painful swelling in the left parotid area of six months' duration. There was no history of trauma. On examination, there was a 3 × 2 cm firm mass anteriorly to the left tragus, fixed on deep planes. Ecography showed an ill-defined echogenic area, containing an echo-free area of 1.9 × 0.6 cm, considered to be an abscess. An orthopantomonogram revealed no alteration of the mandibular condyles and a parotid sialography was unremarkable. The patient underwent surgery, with a clinical pre-operative diagnosis of parotid tumour. The left parotid gland was dissected and a 1.5 × 1 cm, reddish-brown mass adherent to the capsule of the temporomandibular joint was found (Figure 1). A frozen section resulted in the diagnosis of a benign giant cell-containing lesion. The mass was completely excised, together with part of the capsule but with preservation of the meniscus. A sternocleidomastoid muscle flap was used to cover the exposed mandibular condyle. At one year follow-up, there was no evidence of recurrence or functional impairment of the joint.

Pathological findings

On histological examination, the specimen consisted of synovial tissue with marked villous hypertrophy. This was infiltrated by polygonal or spindle hystiocytes, arranged in a vague nodular pattern. Numerous multinucleated giant cells were scattered throughout the lesion (Figure 2). Interstitial haemorrhagic areas and abundant intra- and extracellular deposits of haemosiderin were present. The central area of the growth was marked by dense fibrosis with hyalinization. The synovial lining was hyperplastic, with one or two layers of tall synoviocytes.

Discussion

The occurrence of pigmented villonodular synovitis (PVNS) in the temporomandibular joint is an extremely rare event. Eisig

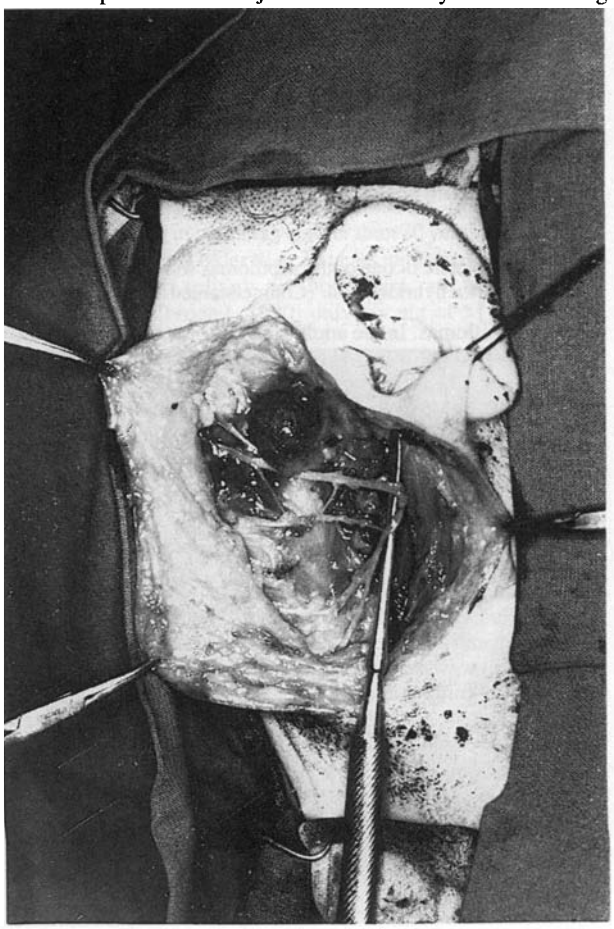

FIG. 1
After removal of the parotid gland, a reddish-brown nodule tightly adherent to the temporomandibular joint is visualized.

From the Institute of Anatomic Pathology* and Otolaryngology Clinic†, University of Florence, Florence, Italy.
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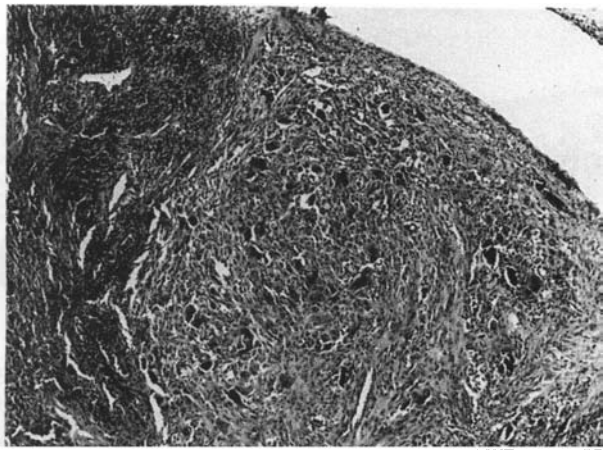

FIG. 2

The synovial tissue is densely infiltrated by histiocytes and multinucleated giant cells. Large deposits of haemosiderin are present. (H&E; ×100).

et al. (1992) recently reviewed the clinicopathological features of 12 cases; two further instances were described by Drommer (1978) and by Dawiskiba *et al.* (1989). Patients were chiefly young adults, mostly in the third and fourth decades, ranging between 22 and 62 years, without sex predilection. The most common complaint was swelling of the preauricular region. Other clinical signs were limitation of mouth opening, pain, trismus and clicking of the joint. Compression of the auditory canal resulted in hearing loss and/or tinnitus in three patients (Lapayowker *et al.*, 1973; Dinerman and Myers, 1977; Eisig *et al.*, 1992). The initial clinical diagnosis was parotid tumour in a third of the cases (Eisig *et al.*, 1992).

On radiological examination, the temporomandibular joint was usually preserved, but erosion of the juxta-articular bone could be detected in the more advanced cases (Lapayowker *et al.*, 1973; Miyamoto *et al.*, 1977). CT scan may be useful in defining the extent of the lesion (Eisig *et al.*, 1992).

Histologically PVNS must be distinguished from other hyperplastic synovitis with haemosiderin deposition, like haemosiderotic synovitis, which is due to chronic intra-articular bleeding. Multinucleated giant cells and a diffuse or nodular histiocytic infiltrate are typical features of PVNS and are not found in haemosiderotic synovitis.

In the case described by Dawiskiba *et al.* (1989), fine needle aspiration cytology showed the typical diagnostic features, including haemosiderin containing histiocytes and numerous multinucleated giant cells.

A combination of PVNS and synovial chondromatosis was found in the lesions described by Raibley (1977) and by Takagi and Ishikawa (1981), suggesting a possible relationship between the two entities.

The aetiology of PVNS remains unknown. Trauma with intra-articular haemorrhage has been proposed as a possible cause, but a clear history of trauma is absent in most patients (Eisig *et al.*, 1992). PVNS is regarded as a reactive, benign lesion, but in some cases it may behave aggressively, with erosion and invasion of the adjacent bone. In three of the cases with temporomandibular joint localization, the lesion extended into the temporal bone and the middle cranial fossa (Dinerman and Myers, 1977; Geiger and Pesch, 1980; Eisig *et al.*, 1992), requiring an aggressive treatment with craniotomy.

If incompletely removed, PVNS frequently recurs. The case described by Takagi and Ishigawa (1981) recurred five years after treatment and the one reported by O'Sullivan *et al.* (1984) recurred three times in a seven-year period, diffusely involving the neighbouring tissues, and requiring radiation therapy. Thus, a wide excision and long-term follow-up are recommended (Eisig *et al.*, 1992).

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Address for correspondence:
Dr Roberto Santoro,
Istituto di Clinica Otorinolaringoiatrica,
Università degli Studi di Firenze,
Viale Morgagni 85,
50134 Firenze,
Italia.