The Journal of Laryngology and Otology
February 1998, Vol. **112,** pp. 182–185

Extra-articular pigmented villonodular synovitis of the temporomandibular joint

Louis T. C. Chow, F.R.C.Path., F.R.C.P.A.*, S. M. Kumta, M.S.(Orth.)†,
Walter W. K. King, M.D., F.R.C.S.‡

Abstract
Pigmented villonodular synovitis, a benign but locally destructive fibrohistiocytic proliferative lesion involving tendon sheaths, bursae and diarthrodial joints, is distinctly rare in the temporomandibular joint. We report one such case occurring in a 42-year-old housewife who presented with a progressively enlarging right zygomatic mass for six months. On exploration, an orange-brown firm mass, 5 × 3 × 2 cm, was seen adherent to the lateral aspect of the capsule of the right temporomandibular joint, and eroding into the inferior aspect of the right temporal bone and part of the mandibular condyle. The mass was completely excised. Pathological examination showed features typical of those of pigmented villonodular synovitis and the lesion was entirely extra-articular in location. The patient remained well with no evidence of local recurrence two years after operation. Review of the literature and careful analysis of the clinicopathological features showed that the vast majority of the reported cases of pigmented villonodular synovitis of the temporomandibular joint belonged to the extra-articular variant, which is associated with a more aggressive local infiltrative behaviour and higher rate of local recurrence than the localized type. The recommended treatment for this condition is therefore wide local excision, aiming to remove the lesion as completely as possible without producing severe disability for the patient.

Key words: Temporomandibular joint; Synovitis, pigmented villonodular

Introduction

Pigmented villonodular synovitis is a benign but locally destructive fibrohistiocyte proliferative lesion involving tendon sheaths, bursae and diarthrodal joints. More than 80 per cent of the cases involve the knee, and the hip joint accounts for about 15 per cent. It affects much less frequently the ankle, foot, hand, elbow and the shoulder (Dorwart *et al.*, 1984). Its occurrence in the temporomandibular joint has been distinctly rare. In this report, we describe the clinicopathological features of a patient with pigmented villonodular synovitis, the extra-articular type, involving the temporomandibular joint. The literature is reviewed with regard to the pathological typing and behaviour of the lesion in this particular location.

Case report

Clinical course

A 42-year-old housewife with good past health attended her private doctor because of a swelling in her right zygomatic region for six months. It was not painful but had been increasing gradually in size since its detection. A computerized tomogram (CT) revealed an expansile soft tissue mass in the region of the right temporomandibular joint, eroding into the temporal bone but not involving the dura (Figure 1). The patient was referred to our hospital for further management.

Physical examination revealed an ill-defined soft tissue swelling in the right zygomatic region, 5 × 3 cm across, situated deep to the subcutis. Fine-needle aspiration of the swelling showed a mixture of polygonal cells and osteoclast-like multinucleated giant cells, together with a few haemosiderin-laden macrophages. Cytologically, the features were those of a giant cell lesion, the differential diagnoses included giant cell tumour and chondroblastoma. On exploration, an orange-brown firm mass, 5 × 3 × 2 cm in maximal dimension, was seen adherent to the lateral aspect of the right temporomandibular joint. It eroded into the inferior aspect of the right temporal bone and also involved part of the mandibular condyle. The entire mass was totally resected, together with the right mandibular condyle, the joint capsule, and part of the zygomatic and temporal bone. The patient made an uneventful recovery and remained well with no evidence of local recurrence two years after operation.

Pathological findings

The main specimen consisted of fragments of orange-brown tissue, 5 × 3 × 1.5 cm across, encircling the capsule of the right temporomandibular joint, but without involving the joint itself or the head of the mandibular condyle (Figure 2). There were multiple similar brown tissue fragments, measuring 3 × 3 × 2 cm in aggregate, they

From the Departments of Anatomical and Cellular Pathology*, Orthopaedics and Traumatology†, and Surgery‡, Prince of Wales Hospital, Hong Kong.
Accepted for publication: 2 December 1997.

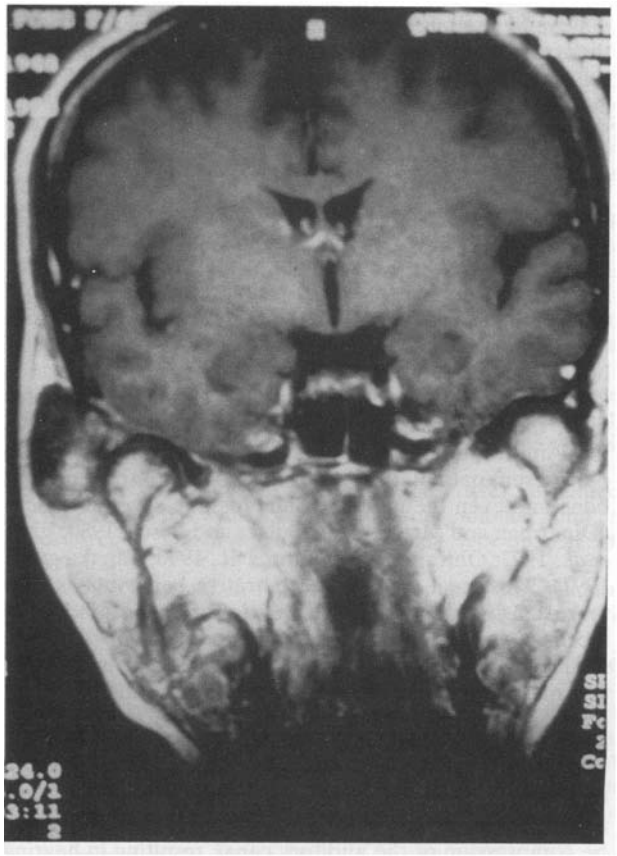

FIG. 1

Computerized tomogram of the lesion, showing its location in the region of the right temporomandibular joint, eroding into the inferior aspect of the temporal bone, but not involving the dura.

showed a firm spongy cut surface. A piece of bone, 1 × 1 × 1 cm, labelled zygomatic arch, was also included.

The lesion showed a uniform histological appearance. It consisted of a mixture of mainly polygonal stromal cells, a number of interposed haemosiderin-laden macrophages, and evenly distributed osteoclast-like multinucleated giant cells (Figure 3). Histological examination confirmed that the lesion affected only the external aspect of the capsule with no involvement of the synovium or interior of the temporomandibular joint. However, it infiltrated extensively into the surrounding soft tissue and skeletal muscle, with involvement of the cortex of the included portion of temporal bone as well. In summary, the features were those of the extra-articular type of pigmented villonodular synovitis of the temporomandibular joint, with infiltration into the temporal bone, and presenting as a progressively enlarging mass in the right zygomatic region of a 42-year-old housewife.

Discussion

Pigmented villonodular synovitis is a benign fibrohistiocytic proliferative lesion affecting tendon sheaths, bursae and diarthrodal joints. Its gross appearance is fairly characteristic, consisting of brownish villous and nodular spongy growths with variable foci of bright orange areas. Histologically, it consists of a mixture of mainly polygonal stromal cells, and varying proportions of haemosiderin-laden macrophages, foamy histiocytes and evenly distributed osteoclast-like giant cells. The gross features thus correspond well to the microscopic picture in that the haemosiderin-laden macrophages impart a brown discolouration to the lesion, whereas the foamy histiocytes are responsible for the bright orange areas. Despite its benign nature, the lesion is locally destructive, in that it may extend into adjacent bone, simulating a primary or metastatic bone tumour. Accordingly, local recurrence may develop, especially if the lesion is incompletely excised.

Since the first description of pigmented villonodular synovitis by Jaffe *et al.* in 1941, it had been recognized that some of these lesions were mainly extra-articular in location. Accordingly, Enzinger and Weiss (1995) employed the term extra-articular pigmented villonodular synovitis, or tenosynovial giant cell tumour of the diffuse type for this group of lesions, when the involvement was largely confined to soft tissue with, or without, affecting the adjacent joint. The clinical importance of recognizing this extra-articular variant of pigmented villonodular synovitis lies in their distinct clinical behaviour. Compared with the localized type, for which the recurrence rates have been reported as 25 per cent by Schwartz *et al.* (1989), the extra-articular type of pigmented villonodular synovitis is more aggressive locally, and in the experience of Enzinger and Weiss (1995), the recurrence rates have been in the range of 40 to 50 per cent. The recommended treatment was therefore wide local excision, aiming to remove the lesion as completely as possible without producing severe disability for the patient.

The pathological features of the lesion in our patient are typical of those of the extra-articular variant of pigmented villonodular synovitis, consisting of an expansile orange-brown mass with a firm spongy cut surface, adherent to the

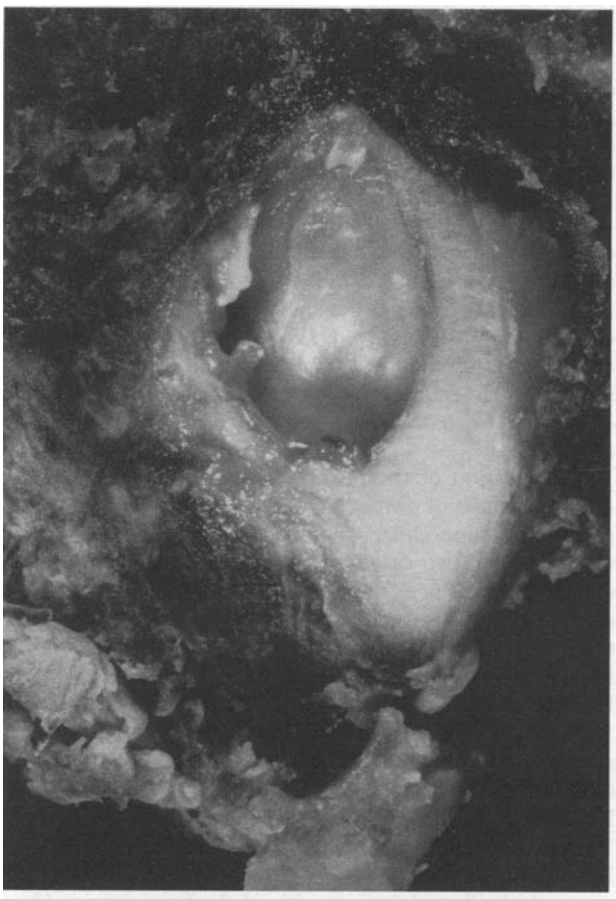

FIG. 2

Gross appearance of the resected specimen. The lesion consists of brown spongy firm tissue encircling the capsule of the temporomandibular joint, but both the joint itself and the mandibular condyle are uninvolved.

https://doi.org/10.1017/S0022215100140265 Published online by Cambridge University Press

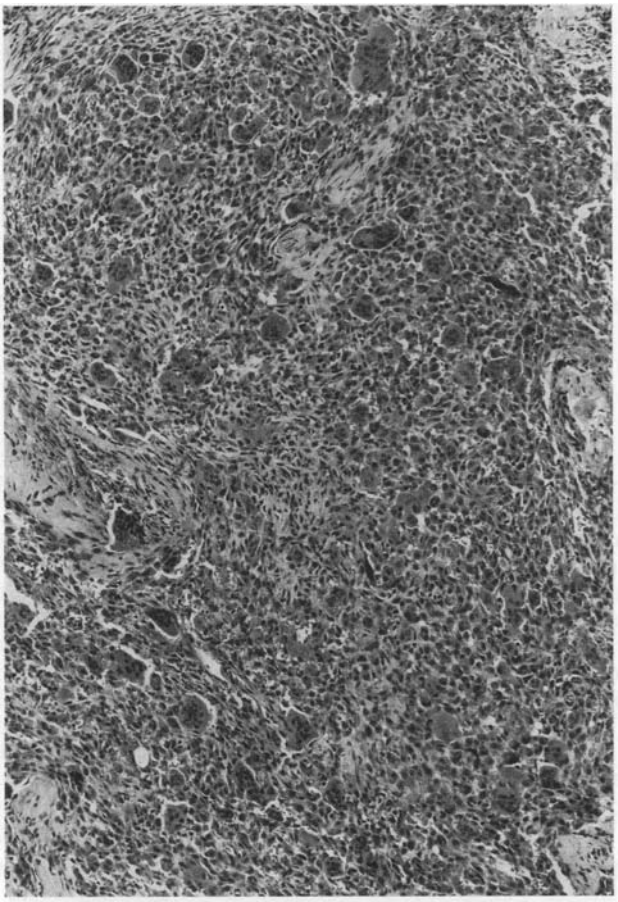

FIG. 3

The lesion shows a uniform histological appearance. It consists of a uniform mixture of mainly polygonal stromal cells, a number of interposed haemosiderin-laden macrophages, and evenly distributed osteoclast-like multinucleated giant cells. (H & E; × 40).

external aspect of the capsule of the right temporomandibular joint. It infiltrates in the surrounding soft tissue and temporal bone, but neither the synovium nor the interior of the joint are involved (Figure 2). Histologically, it consists of a mixture of mainly polygonal stromal cells, a number of haemosiderin-laden macrophages and evenly distributed osteoclast-like multinucleated giant cells (Figure 3).

The location of the lesion, namely, the temporomandibular joint, is of particular interest, as the occurrence of pigmented villonodular synovitis in this region is extremely rare. The first two cases were reported by Lapayowker *et al.* in 1973. Since then, additional cases have been described, mostly in the form of single case reports (Barnard, 1975; Miyamoto *et al.*, 1977; Raibley, 1977; Makek and Drommer, 1978; Takagi and Ishikawa, 1981; Rickert and Shapiro, 1982; Gallia *et al.*, 1982; Curtin *et al.*, 1983; O'Sullivan *et al.*, 1984; Dawiskiba *et al.*, 1989), and reviewed by Eisig *et al.* (1992). A number of additional case reports were also published recently (Syed *et al.*, 1993; Franchi *et al.*, 1994; Ohira, 1994; Shapiro *et al.*, 1996; Youssef *et al.*, 1996; Yu *et al.*, 1997; Tanaka *et al.*, 1997). Including the present case, a total of about 22 cases of pigmented villonodular synovitis of the temporomandibular joint are thus found in the literature. However, it is particularly worth noting that, in the majority of these reported cases, there was no explicit categorization of the lesions into either the localized or the extra-articular type.

Careful analysis of the clinicopathological description of these reported cases of pigmented villonodular synovitis of the temporomandibular joint revealed that most of them belonged to the extra-articular variant. For instance, the lesion described by Franchi *et al.* (1994) consisted of a 1.5 × 1 cm reddish-brown mass adherent to the external surface of the capsule of the left temporomandibular joint and therefore presented as a parotid swelling, and in the case reported by Dawiskiba (1989), the lesion was lateral to the right temporomandibular joint. The extra-articular location of these lesions was also supported by the fact that 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). Moreover, many of these reported lesions showed extensive local infiltration into surrounding tissue. In fact, including our present patient, the lesions extended into the temporal bone and even the middle cranial fossa in seven instances (Dinerman and Myers, 1977; Geiger and Pesch, 1980; Eisig *et al.*, 1992; Ohira, 1994; Shapiro *et al.*, 1996; Tanaka *et al.*, 1997). This local aggressive infiltrative behaviour is also more consistent with that of the extra-articular rather than localized variant of pigmented villonodular synovitis.

Since the vast majority of these lesions are extra-articular in location, the commonest presenting symptom of pigmented villonodular synovitis of the temporomandibular joint is that of a swelling in the pre-auricular region, masquerading as a parotid tumour in more than one third of the cases (Eisig *et al.*, 1992; Syed *et al.*, 1993; Franchi *et al.*, 1994; Yu *et al.*, 1997). In addition, there may be compression of the auditory canal, resulting in hearing loss and/or tinnitis (Lapayowker *et al.*, 1973; Dinerman and Myers, 1977; Eisig *et al.*, 1992). Obviously, the other clinical signs and symptoms relate to the temporomandibular joint, including limitation of mouth opening, pain, trismus and clicking of the joint.

In accordance with the high local recurrence rate of the extra-articular variant of pigmented villonodular synovitis, local recurrence has indeed been observed in lesions of the temporomandibular joints. The case reported by Takagi and Ishikawa (1981) recurred five years after initial excision, and the patient described by O'Sullivan *et al.* (1984) suffered from three recurrences in a seven-year period, diffusely involving the neighbouring tissues and requiring radiation therapy. Our patient remained well with no evidence of local recurrence two years after operation. Similarly, the duration of follow-up in most of the reported cases of pigmented villonodular synovitis of the temporomandibular joint has been short, and the rate of local recurrence would be expected to be higher with longer periods of follow-up. It is therefore entirely logical to advocate, for the effective treatment of this condition, wide local excision, aiming to remove the lesion as completely as possible without producing severe disability for the patient, coupled with long-term follow-up, as recommended by Eisig *et al.* (1992).

References

Barnard, J. D. W. (1975) Pigmented villonodular synovitis in the temporomandibular joint: a case report. *British Journal of Oral Surgery* **13:** 183–187.

Curtin, H. D., Williams, R., Gallia, L., Meyers, E. (1983) Pigmented villonodular synovitis of the temporomandibular joint. *Computerized Radiology* **7:** 257–260.

Dawiskiba, S., Eriksson, L., Elner, A., Johansen, C. C., Hansson, L. G., Westesson, P. L. (1989) Diffuse pigmented villonodular synovitis of the temporomandibular joint diagnosed by fine needle aspiration cytology. *Diagnostic Cytopathology* **5:** 301–304.

Dinerman, W. S., Myers, E. N. (1977) Pigmented villonodular tenosynovitis of the temporomandibular joint: a case report. *Transactions of the American Academy of Ophthalmology and Otolaryngology* **84:** 132–135.

Dorwart, R. H., Genant, H. K., Johnston, W. H., Morris, J. M. (1984) Pigmented villonodular synovitis of synovial joints: Clinical, pathologic and radiologic features. *American Journal of Roentgenology* **143:** 877–885.

Eisig, S., Dorfman, H. D., Cusamano, R. J., Kantrowitz, A. B. (1992) Pigmented villodular synovitis of the temporomandibular joint: case report and review of the literature. *Oral Surgery, Oral Medicine, Oral Pathology* **73:** 328–333.

Enzinger, F. M., Weiss, S. W. (1995) Benign tumors and tumorlike lesions of synovial tissue. In *Soft Tissue Tumors.* 3rd Edition. C. V. Mosby: St. Louis, pp 735–755.

Franchi, A., Frosini, P., Santoro, R. (1994) Pigmented villonodular synovitis of the temporomandibular joint: report of a case. *Journal of Laryngology and Otology* **108:** 166–167.

Gallia, L. J., Johnson, J. T., Myers, E. N. (1982) Pigmented villonodular synovitis of the temporomandibular joint: a case report. *Otolaryngology – Head and Neck Surgery* **90:** 691–695.

Geiger, S., Pesch, H. J. (1980) Synovitis pigmentosa villonodularis, eine saltene Kiefergelenkerkrankung. *Fortschritte der Kieferund Gesichts-Chirurgie* **25:** 129–132,

Jaffe, H. L., Lichtenstein, L., Sutro, C. J. (1941) Pigmented villonodular synovitis, bursitis, and tenosynovitis: a discussion of the synovial and bursal equivalents of the tenosynovial lesions commonly denoted as xanthoma, xanthogranuloma, giant cell tumor, or myeloplaxoma of the tendon sheath, with some consideration of the tendon sheath lesion itself. *Archives of Pathology and Laboratory Medicine* **31:** 731–765.

Lapayowker, M. S., Miller, W. T., Levy, W. M., Harwick, R. D. (1973) Pigmented villonodular synovitis of the temporomandibular joint. *Radiology* **108:** 313–316.

Makek, M., Drommer, M. (1978) Localized nodular synovitis of the temporomandibular joint: a case report. *Journal of Maxillofacial Surgery* **6:** 302–305.

Miyamoto, Y., Tani, T., Hamaya, K. (1977) Pigmented villonodular synovitis of the temporomandibular joint: case report. *Plastic Reconstructive Surgery* **59:** 283–286.

Ohira, Y. (1994) Pigmented villonodular synovitis (PVS) of the temporomandibular joint (TMJ): A case report. *Otolaryngology - Head and Neck Surgery – Tokyo* **66:** 348–352.

O'Sullivan, T. J., Alport, E. C., Whiston, H. G. (1984) Pigmented villonodular synovitis of the temporomandibular joint. *Journal of Otolaryngology* **13: 123–126.**

Raibley, S. O. (1977) Villonodular synovitis with synovial chondromatosis. *Oral Surgery, Oral Medicine, Oral Pathology* **44:** 279–284.

Rickerts, R. R., Shapiro, M. J. (1982) Pigmented villonodular synovitis of the temporomandibular joint. *Otolaryngology – Head and Neck Surgery* **90:** 668–670.

Schwartz, H. S., Unni, K. K., Pritchard, D. J. (1989) Pigmented villonodular synovitis: a retrospective review of affected large joints. *Clinical Orthopaedics and Related Research* **247:** *243–255.*

Shapiro, S., Kessler, S., McMenomey, S. O. (1996) Pathologic quiz case 1. *Archives of Otolaryngology - Head and Neck Surgery* **122:** 194, 196.

Syed, A., Van Hasselt, C. A., To, K. F. (1993) Pigmented villonodular synovitis of the temporomandibular joint. *Journal of Laryngology and Otology* **107:** 853–854.

Takagi, M., Ishikawa, G. (1981) Simultaneous villonodular synovitis and synovial chondromatosis of the temporomandibular joint: report of a case. *Journal of Oral Surgery* **39:** 699–701.

Tanaka, K., Suzuki, M., Nameki, H., Sugiyama, H. (1997) Pigmented villonodular synovitis of the temporomandibular joint. *Archives of Otolaryngology – Head and Neck Surgery* **123:** 536–539.

Youssef, R. E., Roszkowski, M. J., Richter, K. J. (1996) Pigmented villonodular synovitis of the temporomandibular joint. *Journal of Oral and Maxillofacial Surgery* **54:** 224–227.

Yu, G. H., Staerkel, G. A., Kershisnik, M. M., Varma, D. G. K. (1997) Fine-needle aspiration of pigmented villonodular synovitis of the temporomandibular joint masquerading as a primary parotid gland lesion. *Diagnostic Cytopathology* **16:** 47–50.

Address for correpondence:
Dr Louis T. C. Chow,
Department of Anatomical and Cellular Pathology,
Prince of Wales Hospital,
30–32 Ngan Shing Street,
Shatin,
Hong Kong.

Fax: (852)-2637-6274