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Review

Pigmented villonodular synovitis of the thoracic spine

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ABSTRACT

Pigmented villonodular synovitis (PVNS) is a proliferative lesion of the synovial membranes. Knees, hips, and other large weight-bearing joints are most commonly affected. PVNS rarely presents in the spine, in particular the thoracic segments. We present a patient with PVNS in the thoracic spine and describe its clinical presentation, radiographic findings, pathologic features, and treatment as well as providing the first comprehensive meta-analysis and review of the literature on this topic, to our knowledge. A total of 28 publications reporting 56 patients were found. The lumbar and cervical spine were most frequently involved (40% and 36% of patients, respectively) with infrequent involvement of the thoracic spine (24% of patients). PVNS affects a wide range of ages, but has a particular predilection for the thoracic spine in younger patients. The mean age in the thoracic group was 22.8 years and was significantly lower than the cervical and lumbar groups (42.4 and 48.6 years, respectively; $p = 0.0001$). PVNS should be included in the differential diagnosis of osteodestructive lesions of the spine, especially because of its potential for local recurrence. The goal of treatment should be complete surgical excision. Although the pathogenesis is not clear, mechanical strain may play an important role, especially in cervical and lumbar PVNS. The association of thoracic lesions and younger age suggests that other factors, such as neoplasia, derangement of lipid metabolism, perturbations of humoral and cellular immunity, and other undefined patient factors, play a role in the development of thoracic PVNS.

1. Introduction

Pigmented villonodular synovitis (PVNS) is a proliferative lesion of the synovial membranes [1–4]. Knees, hips, and other large weight-bearing joints are most commonly affected. Involvement of the axial skeleton is rare with the literature largely comprised of multiple case reports and a handful of small case series. We present a 28-year-old man with PVNS of the thoracic spine and discuss the diagnosis and management of such lesions. In addition, we present to our knowledge the first meta-analysis and comprehensive literature review and, in doing so, offer potential insight into the etiology of these lesions.

2. Case report

An otherwise healthy 28-year-old man presented with 8 months of progressive midline pain in the upper thoracic spine which sometimes radiated to the right scapula. The pain was worse in the morning and in the recumbent position. Physical examination was normal with full strength, normal sensation, and non-pathologic reflexes. Palpation and percussion of the spine did not elicit tenderness. A CT scan revealed a 1.5 × 1.5 cm intrinsic lytic bone lesion involving the right T3 pedicle, hemilamina, and transverse process (Fig. 1A). The lesion extended into the T3–T4 neural foramen and lateral aspect of the spinal canal. A contrast-enhanced MRI demonstrated a uniformly enhancing mass (Fig. 1B, C). A bone scan was negative for any other lesion. Surgery was recommended for gross total resection. A yellowish soft tissue mass was identified emanating from the right T3 pedicle (Fig. 2A). The mass extended partially into the spinal canal and neural foramen. The T3 nerve root was displaced caudally. The right hemilamina, transverse process and pedicle were removed. The entire soft tissue mass was then resected (Fig. 2B). A drill was used to trim the remaining reactive bone surfaces that had been in contact with the mass. His post-operative course was uncomplicated. At 6 month follow-up, he was asymptomatic with complete resolution of his pre-operative pain.

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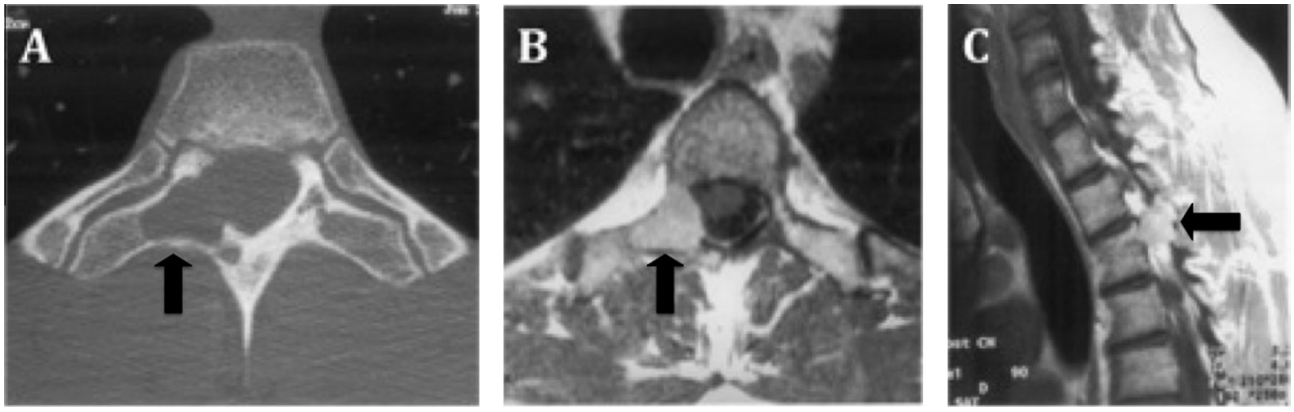

Fig. 1. (A) Axial CT scan demonstrating erosion of the right T3 pedicle, hemilamina (black arrow), and transverse process. (B) Axial contrast enhanced T1-weighted MRI revealing a uniformly enhancing lesion (black arrow) involving the right T3 pedicle, hemilamina, and transverse process. (C) Sagittal contrast enhanced T1-weighted MRI revealing a uniformly enhancing lesion at the T3 level (black arrow).

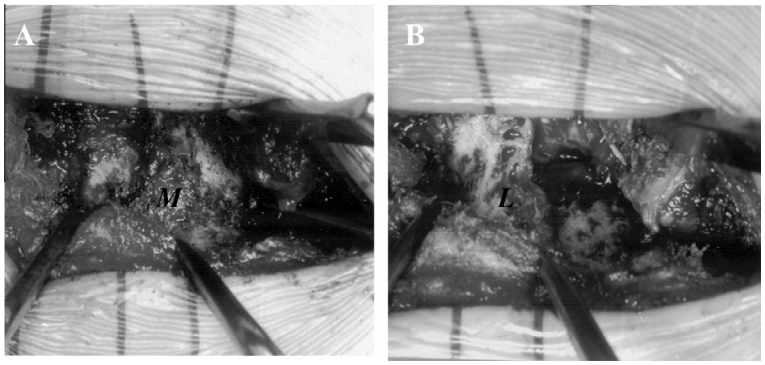

Fig. 2. (A) Intra-operative picture prior to resection. (B) Intra-operative picture after removal of the mass. L = lamina of T3, M = mass.

Follow-up CT scan showed a T3 hemilaminectomy defect with resection of the T3 pedicle and transverse process with no residual mass (Fig. 3A). Post-operative MRI showed similar findings, with preservation of the T3 nerve root (Fig. 3B). Pathologic examination of the surgical specimen showed fibrous tissue with numerous synoviocytes and osteoclast-like giant cells with scattered histiocytes, rare lymphocytes, and scattered areas of hemosiderin deposition consistent with PVNS (Fig. 4).

3. Methods

3.1. Literature review

PubMed was searched using the keywords "PVNS spine," "villonodular tenosynovitis," and "synovitis spine". All case reports and case series that reported on PVNS in the spine were reviewed for diagnosis, age, symptoms at presentation, and location along the

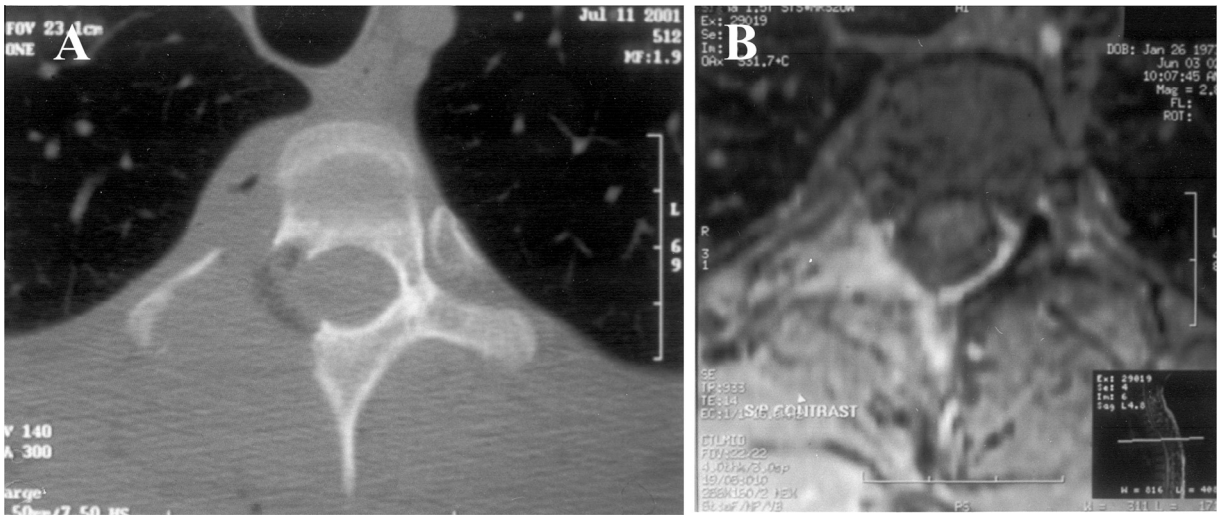

Fig. 3. (A) Follow-up axial contrast enhanced CT scan revealing absent right T3 pedicle, hemilamina and transverse process with no evidence of recurrence. (B) Follow up axial contrast enhanced T1-weighted MRI revealing absent right T3 pedicle, hemilamina and transverse process, with no evidence of recurrence.