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Pigmented Villonodular Synovitis of the Spine: Two Case Reports and a Literature Review

Kazumasa Nakamura, Hiroshi Takahashi, Katsunori Fukutake, Keiji Hasegawa, Akihito Wada*

Department of Orthopedic Surgery, School of Medicine, Toho University, Tokyo, Japan
Email: *nyagira@med.toho-u.ac.jp

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Abstract

Introduction: Pigmented villonodular synovitis (PVNS) of the spine is a rare condition, with only a limited number of cases documented in the medical literature. In this study, we review imaging findings of two cases of PVNS arising in the spine. **Case 1:** A 52-year-old male presented with right thigh pain of unclear etiology. His condition subsequently deteriorated and he was referred to our hospital. MRI revealed a spinal tumor with high intensity on T1-weighted imaging (T1WI) and iso-intensity on T2WI in the right L2/3 intervertebral foramen. The tumor exhibited heterogeneous enhancement on contrast-enhanced MRI. Computed tomography (CT) myelography showed the presence of an epidural tumor within the right L2/3 intervertebral foramen, situated externally to the spinal canal and compressing the dura mater. Notable scalloping was observed in the posterior margin of the L2 vertebral body on CT images. **Case 2:** A 78-year-old male presented with a complaint of muscle weakness of the left upper limb. His gait gradually deteriorated, which led to suspicion of cervical myelopathy. A tumor was observed on plain MRI and exhibited low to iso-intensity on T1WI and high intensity on T2WI. The tumor was in the left C7/T1 intervertebral segment and the spinal cord was compressed from the left side. CT showed destruction of the medial aspect of the left C7 vertebral arch. **Conclusion:** In spinal PVNS, the signal intensity on MRI is dependent on the timing of hemorrhage within the tumor, and there are no distinctive features. However, if an extradural tumor is suspected and CT shows bone erosion, scalloping or osteolytic changes, PVNS should be considered as a differential diagnosis.

Keywords

Spinal Pigmented Villonodular Synovitis, Imaging Diagnosis, Computed Tomography, Magnetic Resonance Imaging

1. Introduction

Pigmented villonodular synovitis (PVNS) is a rare disease with an estimated incidence of 1.8 per million population. The knee and hip joints are the common sites of involvement. Fewer cases of spinal involvement have been reported and there are numerous uncertainties regarding radiographic evaluation, diagnosis, and treatment [1]-[7]. Spinal PVNS presents in the epidural space and frequently compresses neural elements, resulting in axial pain and neurological dysfunction. Given its locally aggressive nature and relatively high recurrence rate, it is of the utmost importance to differentiate PVNS from other spinal tumors in preoperative diagnosis. Thus, awareness of the possibility of spinal PVNS is crucial for orthopedic surgeons and neurosurgeons. In this report, two cases of PVNS are described that developed at the cervicothoracic junction and lumbar spine and were successfully treated surgically, with medium and long-term postoperative follow-up.

2. Case Presentation

2.1. Case 1

The patient was a 52-year-old male who presented with a chief complaint of right thigh pain. The pain had manifested without an evident etiology 10 months prior to his visit to our hospital. Over the preceding month, the condition had deteriorated, prompting the patient to seek medical advice. A physical examination showed pain, numbness, and hypesthesia on the anterior surface of the right thigh, suggesting L2 and L3 nerve root distributions, with a limited walking distance of only a few meters due to intermittent claudication. No bladder dysfunction or lower limb motor weakness was observed. Preoperative JOA scores for low back pain were 20/29 points.

Plain lumbar radiographs were unremarkable. On plain MRI, a tumor with high intensity on T1-weighted images (T1WI) and iso-intensity on T2-weighted images (T2WI) was present in the right L2/3 foramen. The lesion showed heterogeneous enhancement on contrast-enhanced MRI, and compressed the dural tube while being continuous with the light L2/3 facet joint. A CT myelogram revealed the presence of an epidural tumor within the right L2/3 foramen, situated over the spinal canal. The dura was excluded by the tumor and scalloping was observed at the posterior margin of the L2 vertebral body (**Figure 1(A)-(D)**).

Surgery was conducted via a posterior approach. There was evidence of black staining in the right L2/3 facet joint capsule and yellow ligament. Following resection of the right L2 inferior articular process, a black discoloration of the right L2/3 articular surface was also apparent. The tumor was adherent to the dura and encompassed the dura and the right L2 and L3 nerve roots. We postulated during surgery that the most probable diagnosis was PVNS. The right L2/3 facet joint was entirely excised, and the tumor was completely resected, followed by L2/3 transforaminal lumbar interbody fusion (TLIF) with pedicle screw instrumentation under fluoroscopic guidance was added to prevent postoperative segmental

instability due to the right facetectomy. The surgery was completed without any complications.

A pathological examination revealed an outgrowth and cicatrization of the villous synovial membrane, as well as hemosiderin deposition. Multinucleated giant cells and foam cells were also observed, leading to the final diagnosis of PVNS (Figure 2(A), Figure 2(B)).

Right thigh pain resolved immediately following surgery and gait disturbance resolved postoperatively. No neurological deficiencies manifested, the JOA score improved to 28/29 points, and there has been no recurrence up to 5 years postoperatively (Figure 2(C)-(E)).

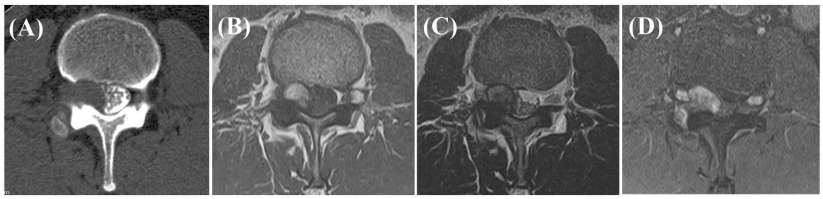

Figure 1. Case 1. (A) Axial computed tomography myelography showed a lesion of 11 × 13 mm located in the right L2/3 foramen, with the dural tube compressed from the right side. Slightly lytic expansive destruction and scalloping of the L2 posterior vertebral wall were observed. (B) The lesion had a high signal intensity on T1-weighted MRI (axial view). (C) The lesion had an intermediate intensity on T2-weighted MRI (axial view). (D) On T1-weighted MRI with fat suppression (axial view), the mass showed heterogeneous enhancement and communication with the joint space of the right facet joint.

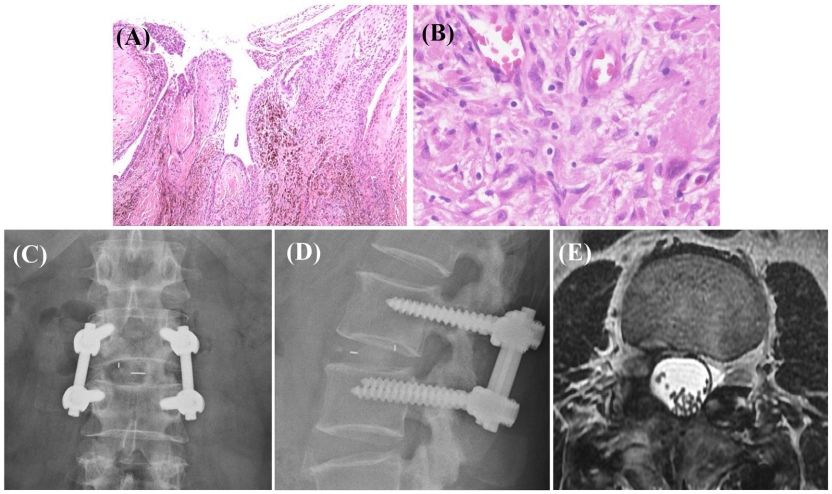

Figure 2. Case 1. (A) Outgrowth of the synovial membrane and hemosiderin deposition. Hematoxylin-eosin staining, × 40. (B) Multinucleated large cells and foam cells were present. Hematoxylin-eosin staining, × 400. (C) Frontal view on a postoperative plain radiograph. (D) Lateral view on a postoperative plain radiograph. (E) Decompression of the dura was favorable on plain T2-weighted MRI at 5 years after surgery, with no evidence of tumor recurrence.

2.2. Case 2

The patient was a 78-year-old male who presented with chief complaints of left

upper extremity muscle weakness and gait disturbance. He first developed muscle weakness one year prior to his initial hospital visit. In the three months preceding the visit, he had experienced an increased propensity to fall. Physical findings included motor paralysis of the left C7/C8 innervated region and sensory disturbance of the bilateral lower extremities, as well as promotion of the lower extremity tendon reflex, which indicated radiculomyelopathy. The preoperative JOA scores for cervical myelopathy were 13/17 points.

Plain radiographs were unremarkable, aside from evidence of degenerative spondylosis. Plain MRI showed the presence of a tumor with low iso-intensity on T1WI and high intensity on T2WI at the level of the left C7-T1. The tumor was also in contact with the left C7/T1 facet joint and compressed the spinal cord from the left side (**Figure 3(A)-(C)**). Plain CT revealed encroachment of the medial aspect of the left C7 vertebral arch (**Figure 3(D)**).

Surgery was conducted via a posterior approach. To extract the tumor mass, laminectomy of C7 - T1 and left medial facetectomy were performed. Intraoperative findings revealed the presence of hemosiderin deposition in the yellow ligament and synovial membrane outgrowth at the level of the left C8 nerve root bifurcation. Additionally, a tumor mass arising from the left C7/T1 facet joint was compressing the dural tube and left C8 nerve root. The tumor was carefully dissected and completely excised. Posterior fusion was successfully performed using pedicle screw instrumentation with use of an O-arm/Stealth Station navigation system (Medtronic Sofamor Danek, Memphis, TN, USA) and autologous bone graft at the C7/T1 level to prevent postoperative instability. There were no perioperative complications such as pedicle screw malposition and neurovascular injuries.

The tumor was composed of proliferative mononuclear histiocyte-like cells and a few multinucleated giant cells, as well as CD68-positive interstitial mono-or multinuclear macrophages (**Figure 4(A)**, **Figure 4(B)**). Partial hemosiderin deposition was also observed in select areas, leading to diagnosis of PVNS.

Following surgery, gait disturbance and paralysis of the left upper extremity gradually improved, allowing the patient to ambulate independently. At 3 years postoperatively, the JOA score has improved to 16/17 points, and there has been no tumor recurrence (**Figure 4(C)-(E)**).

3. Discussion

PVNS is a proliferative and locally aggressive tumorous lesion of uncertain etiology involving the synovial membranes of joints, tendon sheaths, and bursae. PVNS is classified as a localized or diffuse type. Both forms are histologically identical to giant cell tumor of the tendon sheath, and are characterized by the presence of giant cells, foamy histiocytes, fibrous tissue, and hemosiderin-laden macrophages. PVNS typically occurs in large joints such as the knee, hip, elbow and ankle. It rarely involves the spine, but in such cases PVNS arises in the articular facet joint [1] [2]. Although the condition is benign, spinal PVNS can cause

various clinical and neurological problems. The narrow bony structure and proximity of nerves to the lesion can result in pain, radiculopathy, and myelopathy, with symptoms depending on the location of the tumor [3]. Minor bleeding within the tumor may be asymptomatic, but can also cause severe pain and neural palsy. Given the high recurrence rate of PVNS, surgical gross total resection is the recommended treatment. Therefore, a meticulous evaluation of preoperative imaging findings is required to facilitate optimal surgical planning. In this regard, we conducted a review of the radiographic characteristics of spinal PVNS documented in the existing literature.

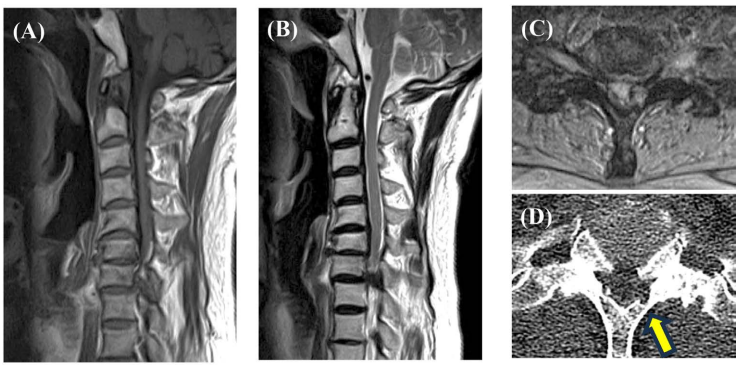

Figure 3. Case 2. A lesion of 8 × 9 mm was located in the left C7/Th1 intervertebral segment, slightly scalloping the C7 lamina. On MRI, the mass had iso-intensity on T1-weighted imaging (T1WI) and high intensity on T2WI. (A) T1WI (sagittal view); (B) T2WI (sagittal view); (C) T2WI (axial view); (D) Axial CT. The yellow arrow indicates encroachment of the left C7 lamina.

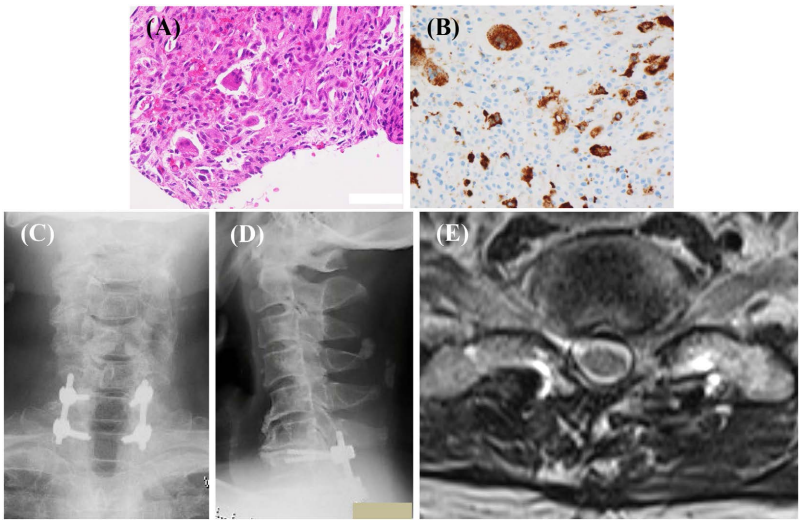

Figure 4. Case 2. (A) Photomicrograph showing proliferative mononuclear histiocyte-like cells and a few multinucleated giant cells. Hematoxylin-eosin staining, × 400. (B) CD68-positive interstitial mono-or multinuclear macrophages on immunohistochemical staining with anti-CD68 antibody as the primary antibody, × 400. (C) Frontal view on a postoperative plain radiograph. (D) Lateral view on a postoperative plain radiograph. (E) Decompression of the dura was favorable on plain T2-weighted MRI at 3 years after surgery, with no evidence of tumor recurrence.

To the best of our knowledge, there have been 73 English-language case reports of spinal PVNS. Imaging findings have been reported on X-ray radiographs and CT images in 57 cases [1] [3] [5]-[32], on MRI T1WI in 20 cases [3] [5] [7] [12] [14] [16] [22], on MRI T2WI in 27 cases [1] [3] [5]-[7] [11] [12] [14] [16] [19] [23], and on contrast-enhanced MRI in 13 cases [3] [7] [14] [16] [22] [27] [29]-[31] [33] [34].

In general, PVNS has low intensity on T1WI and T2WI MRI [35] [36], and the mass often exhibits a mixed signal intensity on T2WI due to the presence of hemorrhage-induced hemosiderin deposition [37]. However, the signal intensity of these scans has varied among reported cases of spinal PVNS (**Table 1**). As MRI is usually performed at the time of symptom onset, the intensity may vary depending on the timing of hemorrhage within the tumor. An enhancement effect on contrast-enhanced MRI was reported in 12 of 13 cases of spinal PVNS (92.3%), with patterns of enhancement similar to those in PVNS arising from large joints of the extremities [37]-[42]. The results of our literature review indicate that MRI signals of spinal PVNS are typically unremarkable and have no distinctive features. However, demonstration of a connection between the tumor and facet joint may provide supporting evidence for diagnosis of spinal PVNS.

Table 1. Summary of reported MRI findings.

Signal intensity	Number of cases (%)
T1 WI	
Low	6 (30.0)
Intermediate	9 (45.0)
High	5 (25.0)
Total	20
T2 WI	
Low	7 (25.9)
Intermediate	5 (18.5)
High	11 (40.7)
Heterogeneous	4 (14.8)
Total	27

Bone erosion, destruction, scalloping, and osteolytic lesions were observed on CT images in 50 of 57 cases (87.7%), which is consistent with findings for PVNS in the knee and hip joints [35]. In our patients, case 1 exhibited vertebral body scalloping, while case 2 showed vertebral arch destruction on CT images. The differential diagnosis of a tumorous lesion originating from the facet joint includes facet cyst and ligamentum flavum hematoma, but these conditions rarely present with bone erosion, destruction, and scalloping on CT. Thus, if preoperative MRI suggests a relationship with the facet joint and CT shows bone erosion, spinal PVNS should be included in the preoperative differential diagnosis, despite the

rarity of this condition.

Several papers have concluded that the basic principle of treatment of PVNS of the spine is surgical gross total resection [7] [28] [32] [43]. However, there is a paucity of optimal surgical methods to achieve such resection. Modern surgical techniques including spinal instrumentation and intraoperative imaging guidance of spine surgery may improve surgical outcomes of spinal PVNS for a longer postoperative duration without recurrence. Previous case reports indicate that laminectomy without spinal fusion is the predominant surgical approach for PVNS extraction, with fusion added in select cases. This practice can be attributed to the global trend of neurosurgeons primarily performing laminectomy, a critical step in tumor removal. Conversely, orthopedic surgeons frequently opt for spinal fusion in combination with laminectomy. Thus, the surgical approach may be influenced by surgeon preference. Furthermore, prior to the advent of spinal navigation systems and rigid spinal fusion implants in the 21st century, orthopedic surgeons lacked the expertise to perform spinal fusions in a safe and effective manner, which may have further contributed to the preference for laminectomy alone. Given that PVNS originates from the synovium of the unilateral facet joint, it frequently occupies the intervertebral foramen. To dissect the tumor from the nerve tissue safely and accomplish complete removal, it is essential to obtain a wide surgical field, which can be achieved through facetectomy. In both of our cases, we were able to perform safe gross total resection of spinal PVNS using a facetectomy. In addition, we were able to avoid segmental instability after surgery by adding instrumented spinal fusion. To date, there has been no recurrence of PVNS in either case and no complications associated with the spinal fusion. However, there is a risk of adjacent segment disease in the postoperative period in patients who have undergone fusion surgery, and longer-term postoperative follow-up is necessary.

4. Conclusion

Imaging findings for spinal PVNS on plain radiographs, CT and MRI are essentially similar to those observed for PVNS in the knee and hip joints. In cases with these findings in the spine, PVNS should be considered in the preoperative diagnosis. Spinal PVNS is a benign condition, but surgical treatment involving gross total resection with the originating facet joint and spinal fusion is indicated due to the high recurrence rate.

Consent to Patient

Patients were informed that data from these cases would be submitted for publication and gave their consent.

Conflicts of Interest

The authors declare that there is nothing to disclose regarding the publication of this manuscript.

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