Pigmented villonodular synovitis originating from the lumbar facet joint: a case report

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Abstract The authors successfully treated a rare case of pigmented villonodular synovitis (PVNS) that originated from the lumbar facet joint (L4-5). A 43-year-old man presented with a complaint of left severe sciatica causing difficulty in walking. Magnetic resonance imaging (MRI) demonstrated an extradural mass on the left side at L4 and the mass compressed the dural tube and was continuous with the left L4-5 facet joint. A computed tomography myelogram revealed an extradural defect of contrast medium at the L4 level and an erosion of the L4 lamina. A total synovectomy with unilateral osteoplastic laminectomy was performed. The histological findings were a diagnosis of PVNS. The patient's symptoms resolved completely and the MRI at postoperative 3 years demonstrated no recurrence of PVNS. It is important to totally remove the synovium, which is the origin of PVNS in order to prevent the recurrence. We think that our procedure is reasonable and adequate for lumbar PVNS.

Keywords Pigmented villonodular synovitis · Lumbar spine · Synovectomy · Juxtafacet cyst · Laminoplasty

Pigmented villonodular synovitis (PVNS) is a slowly progressive mass lesion that arises in association with villous or nodular overgrowth in the synovial membranes

of tendon sheaths, joints and bursae [12, 14, 31]. The etiology of PVNS remains controversial but degenerative change and trauma have been implicated [1, 2, 6, 9, 11–14, 20, 31]. It is well known that PVNS occurs in young individuals and affects the appendicular skeleton, particularly the knee and hip joints [2, 9, 16, 18, 19, 22, 23, 28]. The occurrence of PVNS in the axial skeleton is quite rare and there have been few reports of PVNS of the lumbar spine; specifically, only 18 cases are found in the English literature [4, 7, 8, 10, 16, 21, 23, 24, 26, 27, 30, 31]. There have been no reports focusing on surgical treatment for PVNS although it is important that the synovium is totally removed to prevent the recurrence [5]. We successfully treated a case of PVNS that originated from the lumbar facet joint (L4-5) using total synovectomy with unilateral osteoplastic laminectomy.

Case report

A 43-year-old man presented with a 2-year history of low back pain and he had undergone conservative medical treatment with a diagnosis of L5 spondylolysis. He consulted us with a complaint of left severe sciatica causing difficulty in walking for 3 weeks. He had no obvious history of trauma. Neurological examination revealed normal deep tendon reflexes in the lower legs bilaterally (patellar tendon reflex and Achilles tendon reflex) and a negative result upon performing the straight leg raising test. There were no sensory disturbances, motor weakness or urinary incontinence. Plain lumbar spine radiographs showed spondylolysis and anterolisthesis of L5. Magnetic resonance imaging (MRI) demonstrated an extradural mass on the left side at L4. This mass lesion showed mixed high and iso-intensity on T1-weighted imaging, and mixed high and

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Fig. 1 Sagittal MRI showed the compression of the dural tube due to an extradural mass at the L4 level. *Left* The mass lesion showed mixed high and iso-intensity on T1-weighted imaging. *Right* The mass lesion showed mixed high and low intensity on T2-weighted imaging

low intensity on T2-weighted imaging (Fig. 1). The mass also exhibited clear rim enhancement upon T1-weighted imaging with Gadolinium (Gd) (Fig. 2). The mass compressed the dural tube and was continuous with the left L4-5 facet joint. A myelogram of the lumbar spine showed a large defect of contrast medium extending from the level of the L4 pedicle to the L4-5 facet joint on the left side; in addition, the left L4 and L5 roots were not described. A computed tomography (CT) myelogram revealed an extradural defect of contrast medium at the L4 level and an erosion of the L4 lamina (Fig. 2).

The patient was taken to the operating room for spinal decompression, resection of the mass and histological

diagnosis. He was placed in the prone position and a 7-cm-long midline skin incision was made, so as to expose the L4 lamina on only the left side. The paraspinal muscle on the right side was not divided. A unilateral osteoplastic laminectomy was performed for the left L4 lamina. The left lamina was cut at a slanting angle at the level of the pedicle, while exercising care to protect the L4 root. The spinous process was split in half using a micro-bone saw and chisel. The left lamina was temporarily removed en bloc after the articular capsule of the left L4-5 facet joint was incised. The softly elastic and dark reddish-brown mass lesion originating from the left L4-5 facet joint was observed at the dorsum of the ligamentum flavum. The mass adhered to the L4 and L5 roots

Fig. 2 The mass compressed the dural tube and was continuous with the left L4-5 facet joint. *Left* The mass displayed rim enhancement with Gd on T1-weighted imaging. *Right* The mass lesion invaded the lamina

Fig. 3 The intraoperative photos. *Upper left* Removing the lamina en bloc temporally, the hemorrhagic tumor mass (*black arrows*) originated from the left L4-5 facet joint and was observed in the dorsum of the ligamentum flavum. The mass was softly elastic and dark reddish-brown in color; it adhered to the L4 and L5 roots on the left side. *Upper right* After removing the mass and completing the

total synovectomy, the dural tube, L4 root and L5 root were completely decompressed. *Lower left* The mass (*black arrows*) adhered to the L4 lamina and was accompanied by bone erosion. *Lower right* We replaced the laminae to an original position after having removed the tumor. (SP spinous process, D dural tube, P pedicle, SF superior facet, IF inferior facet, and asterisk root)

on the left side, and was approximately 25 mm in diameter. The mass was totally resected along with the capsule of facet joint (Fig. 3). Finally, the previously resected lamina was replaced en bloc and was firmly fixed using a 30 mm cancellous screw, 4.5 mm in diameter (SYNTHES, Davos, Switzerland) as a lag screw.

Pathologically, the mass was composed of mononuclear cells and showed varying degrees of cellularity. There was marked synovial invasion by masses of polygonal and round cells with round nuclei. Scattered osteoclast-like giant cells and hemosiderin-laden macrophages were also present (Fig. 4). CD68 staining was positive for macrophages [8]. The histological findings were consistent with a diagnosis of PVNS.

Postoperatively, the patient recovered with a complete relief of his symptoms. Three years after surgery he still had no pain and his neurological examination was normal. Plain lumbar spine radiographs and a CT scan showed the union of L4 laminae in the original position (Fig. 5). An MRI at that time demonstrated no recurrence of PVNS (Fig. 6).

Discussion

In 1980, Kleinman et al. [17] reported the first case of spinal PVNS and thereafter 48 cases were reported in the English literature. However, there have only been 18

Fig. 4 Photomicrograph from a histologic section of the resected mass showing mononuclear round cells, foamy cells and hemosiderin-laden macrophages. (H&E, original magnification $\times 20$ and $\times 4$)

reported cases of PVNS of the lumbar spine. The pathologic findings of spinal PVNS are similar to those seen in lesions of the appendicular skeleton [16]. In addition, imaged-based diagnoses can be readily made when spinal PVNS exists. CT findings in spinal PVNS usually demonstrate bone involvement (including erosion, scalloping and destruction) and increased attenuation, presumably related to iron within hemosiderin [25, 29]. Calcification is

Fig. 5 Radiographs obtained 3 years after surgery showed the union of L4 laminae

Fig. 6 T1-weighted imaging with Gd of MRI at 3 years after surgery demonstrated no recurrence of PVNS

not a feature of PVNS. Involvement of the facet joint and discontinuity from the vertebral space should suggest the diagnosis and warrant an evaluation of PVNS. In general, on MRI, the intensity of the fluid in the mass is variably

altered due to the process of hemorrhage. Moreover, synovial tissue, scar and hemorrhage can be confused within the mass of PVNS. On MRI, the intensity of PVNS is not definite, but the demonstration of continuity between the mass and the facet joint helps support the diagnosis of PVNS.

The management options for spinal PVNS include surgery, radiation therapy, and radioisotope infusion [3]. The role of radiation therapy has not been clearly defined. Surgical resection is the primary treatment for this lesion. However, the rate of local recurrence for spinal PVNS has been reported from 17 to 46%, such that it is relatively common occurrence [10, 22, 31]. In recent years, research has suggested that deoxyribonucleic acid ploidy status and proliferative index might be related to the potential for recurrence [6]. There have been no reports focused strictly on the value of various surgical procedures (some authors recommend wide synovectomy and/or laminectomy), although it is important to totally remove the synovium in order to prevent the recurrence [5]. In our case, a total synovectomy with a unilateral osteoplastic laminectomy was performed. In 1980, Kawai et al. [15] introduced osteoplastic laminectomy as a decompression procedure for lumbar spinal canal stenosis. We applied this procedure in a unilateral way to this patient. Using unilateral osteoplastic laminectomy, we were able to not only excise the mass under direct observation but also complete the total synovectomy. Consequently, we experienced no recurrence of PVNS as of 3 years following surgery. We think that this procedure is reasonable and adequate for PVNS of the lumbar spine.

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