Spine

SPINE Volume 40, Number 12, pp E735-E739

CASE REPORT

Tenosynovial Giant Cell Tumor, Diffuse Type/ Pigmented Villonodular Synovitis in a Pars Defect

A Case Report

Tetsuya Kimura, MD,* Toshihiko Nishisho, MD, PhD,* Toshinori Sakai, MD, PhD,* Ryo Miyagi, MD,* Shoichiro Takao, MD, PhD,† Seiji Iwamoto, MD,† Kosaku Higashino, MD, PhD,* Yoichiro Takata, MD, PhD,* Yuichiro Goda, MD,* Shunichi Toki, MD,‡ and Koichi Sairyo, MD, PhD*

Study Design. Case report.

Objective. To describe a rare case of tenosynovial giant cell tumor, diffuse type/pigmented villonodular synovitis (PVNS) in a pars defect in a patient with lumbar spondylolysis.

Summary of Background Date. PVNS rarely occurred in lumbar spine, and no studies in the English literature have reported PVNS in a pars defect in lumbar spondylolysis.

Methods. The patient was a 14-year-old female presented with a 5-month history of low back pain. Plain radiography showed spondylolysis at L5 and computed tomography revealed a 1 × 2-cm slightly eroding tumorous mass at the left L5 pars. On magnetic resonance imaging, the mass showed intermediate intensity and gadolinium enhancement on T1-weighted images (WI) and high intensity on T2-WI and T2 STAR-WI. After undergoing computed tomography–guided needle biopsy, a pathological diagnosis of PVNS was made and total gross resection was performed.

Results. The gross appearance and the postoperative pathological diagnosis were consistent with PVNS. The postoperative clinical course was uneventful and postoperative computed tomography and magnetic resonance imaging revealed no residual lesion.

Conclusion. This is the first report of PVNS occurring in spondylolysis.

Key words: tenosynovial giant cell tumor, diffuse type, pigmented villonodular synovitis, spondylolysis, lumbar spine.

Level of Evidence: N/A

Spine 2015;40:E735–E739

From the Departments of *Orthopedics and †Radiology, Institute of Health Biosciences, Tokushima University Graduate School, Tokushima, Japan; and ‡Department of Orthopedics, National Kochi Hospital, Kochi, Japan.

Acknowledgment date: July 28, 2014. Revision date: December 7, 2014. Acceptance date: February 12, 2015.

The manuscript submitted does not contain information about medical device(s)/drug(s).

No funds were received in support of this work.

No relevant financial activities outside the submitted work.

Address correspondence and reprint requests to Toshihiko Nishisho, MD, PhD, Department of Orthopedics, Institute of Health Biosciences, Tokushima University Graduate School, 3-18-15 Kuramoto 770-8503, Tokushima, Japan; E-mail: tnishisho@tokushima-u.ac.jp

DOI: 10.1097/BRS.0000000000000923

Tenosynovial giant cell tumor, diffuse type/pigmented villonodular synovitis (PVNS) is a slowly progressing mass lesion of the synovial membrane with uncertain etiology that occurs predominantly in the knee and hip joints. Spondylolysis is a defect of the pars interarticularis and is a known sequela of stress fracture. Pars defects are considered to communicate with both the cranial and caudal facet joints. Furthermore, they are sometimes accompanied by inflammation and cause low back pain in the form of communicating synovitis, which is spread from the adjacent facet joints.[1]

Although PVNS rarely occurs in the spine, several case studies have reported PVNS in the facet joints.[2] However, no studies in the English literature have reported PVNS in a pars defect in lumbar spondylolysis. Here, we present a case of PVNS in a pars defect in a patient with lumbar spondylolysis.

CASE REPORT

A healthy 14-year-old female was referred to our hospital with a 5-month history of low back pain. She belonged to a basketball team and had not experienced any obvious traumatic episodes. Physical examination revealed tenderness around the L5 spinous process and in the bilateral L4–L5 paravertebral muscles. She had no neurological symptoms such as sensory disturbance or motor weakness in either lower limb. Plain radiographs revealed bilateral spondylolysis at L5 (Figure 1A–D), and computed tomographic scans revealed a 1 × 2-cm tumorous mass located on the left pars defect at L5, slightly eroding the L5 lamina (Figure 2A and Figure 3A).

Magnetic resonance imaging (MRI) comprising T1-weighted images (WIs), short-tau inversion recovery, T2-WI, T2 STAR-WI, and gadolinium-enhanced fat saturated T1-WI was performed with a field strength of 1.5 T. The mass lesion showed intermediate intensity and gadolinium enhancement on T1-WI and high intensity on T2-WI and T2 STAR-WI (Figure 2B–D and Figure 3B–D). The sagittal view in Figure 3 shows that the mass was definitely arising from the pars defect.

After undergoing computed tomography–guided needle biopsy, a preoperative pathological diagnosis of PVNS was made (Figure 4A, B). Photomicrographs showed proliferative

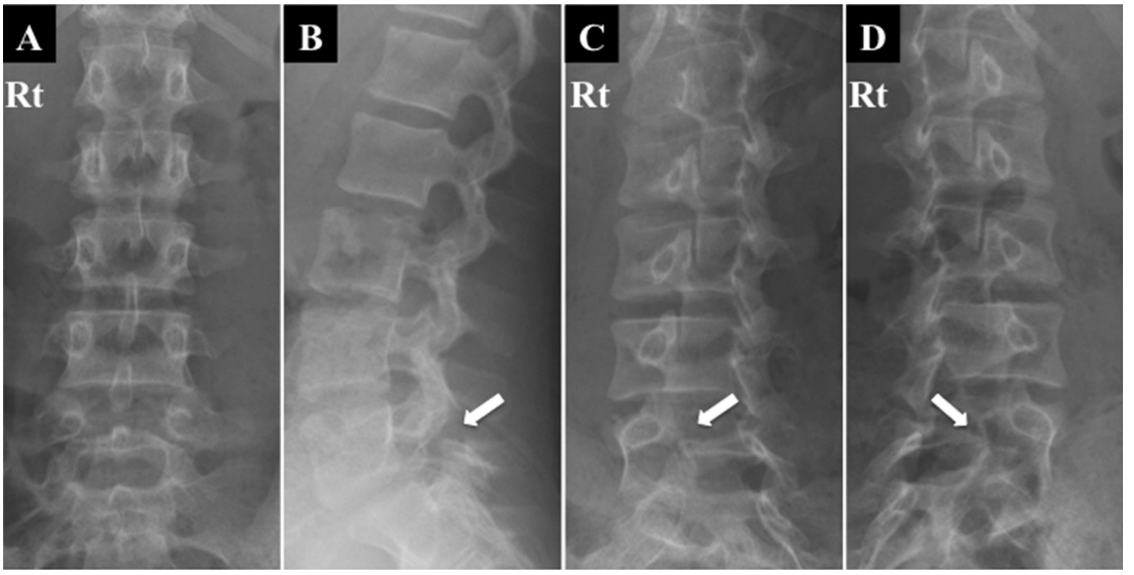

Figure 1. Plain radiographs of the lumbar spine showing spondylolysis at L5 and apparent bone erosion. Anteroposterior view **(A)**, lateromedial view **(B)**, and oblique view **(C** and **D)**. The arrows shows pars defect.

mononuclear histiocyte-like cells and a few multinucleated giant cells. Foamy cells and slight hemosiderin deposition were evident.

Gross total resection and marginal resection of the lamina and flavum were planned (Figure 5A, B). The patient was positioned in the prone position and a midline lumbar incision was made to clear the biopsy tract. Only the left side of the L4–L5 lamina was exposed, leaving the posterior structure of the right side completely preserved. The mass located on the pars defect was considered to communicate with the

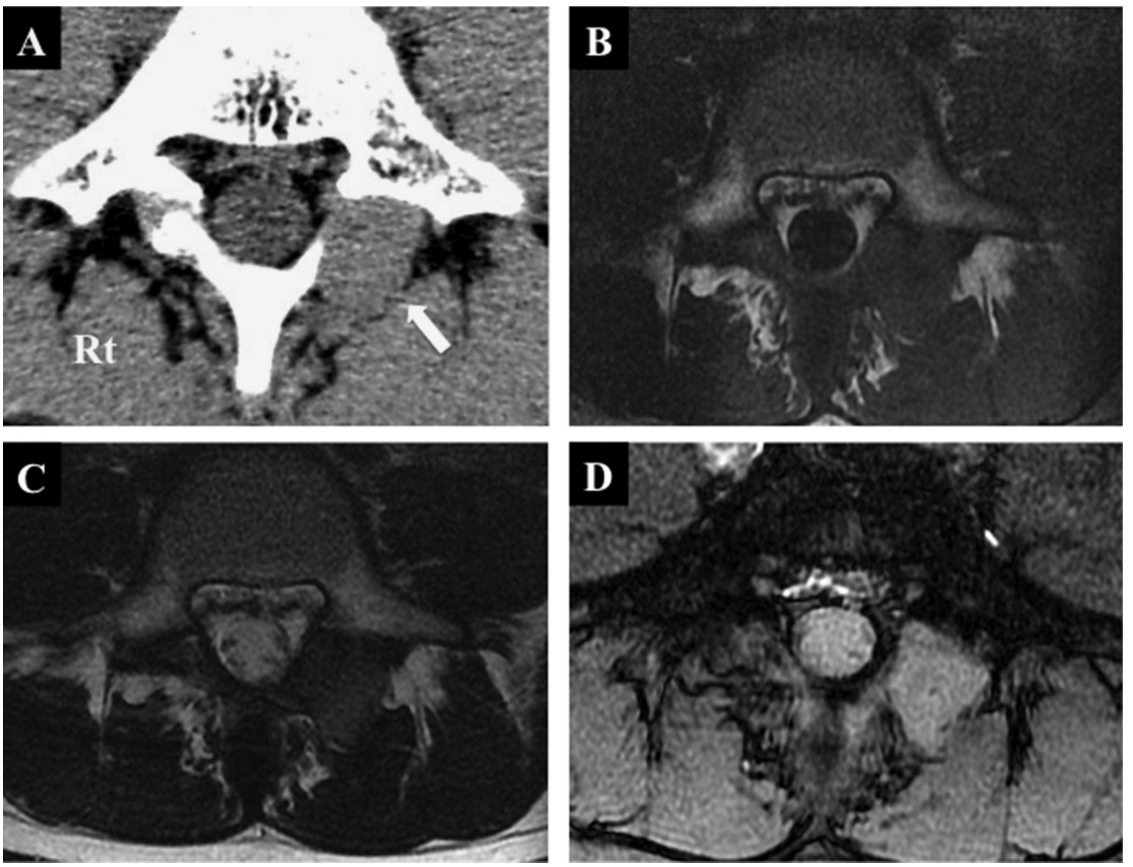

Figure 2. A 1 × 2-cm tumorous mass located on the left pars defect at L5, slightly eroding the L5 lamina. The mass presented with intermediate intensity on T1-weighted images (WIs) and high intensity on T2-WI and T2 STAR-WI. Axial computed tomographic scan **(A)**, T1-WI (axial view) **(B)**, T2-WI (axial view) **(C)**, and T2 STAR-WI (axial view) **(D)**. The arrows shows tumor mass.

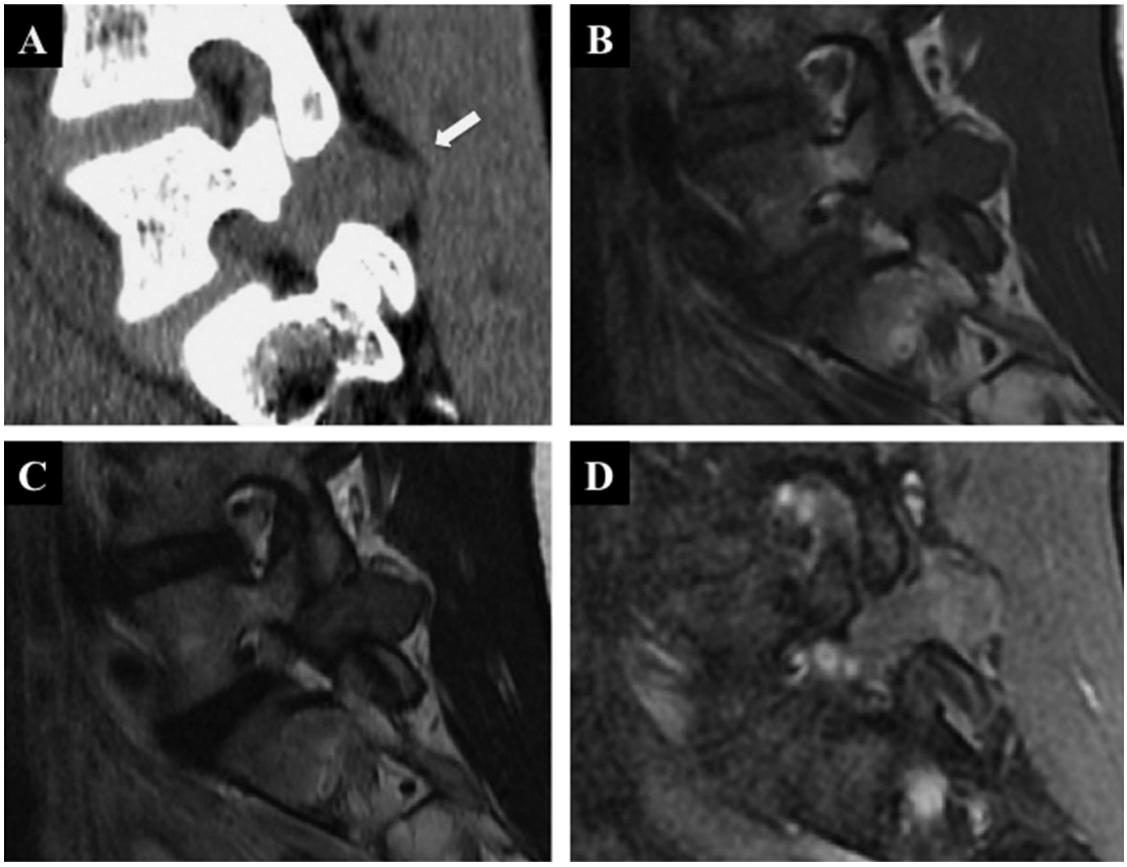

Figure 3. The mass located on the left pars defect at L5. Sagittal computed tomographic scan **(A)**, T1-weighted image (WI; sagittal view) **(B)**, T2-WI (sagittal view) **(C)**, and gadolinium-enhanced, fat saturated T1-WI (sagittal view) **(D)**. The arrows shows tumor mass.

adjacent facet joints (L4–L5 and L5–S); it adhered to the ligamentum flavum at L4–L5 and was adjacent to the infralamina at L4 and the supralamina at L5. Therefore, total gross resection with partial laminectomy at left L4 and L5 and partial facetectomy at left L4–L5 and L5–S were required, while exercising care to protect the L5 root that passes underneath the pars defect. The resected mass was a softly elastic and light yellow-reddish oval lesion measuring 2.5 × 1.5 × 1.5 cm (Figure 6A, B). Both the gross appearance and the postoperative pathological diagnosis were consistent with PVNS. The postoperative clinical course was uneventful. Postoperative computed tomographic scans (Figure 7A, B) and magnetic resonance images (Figure 7C, D) revealed no residual lesion and no apparent instability.

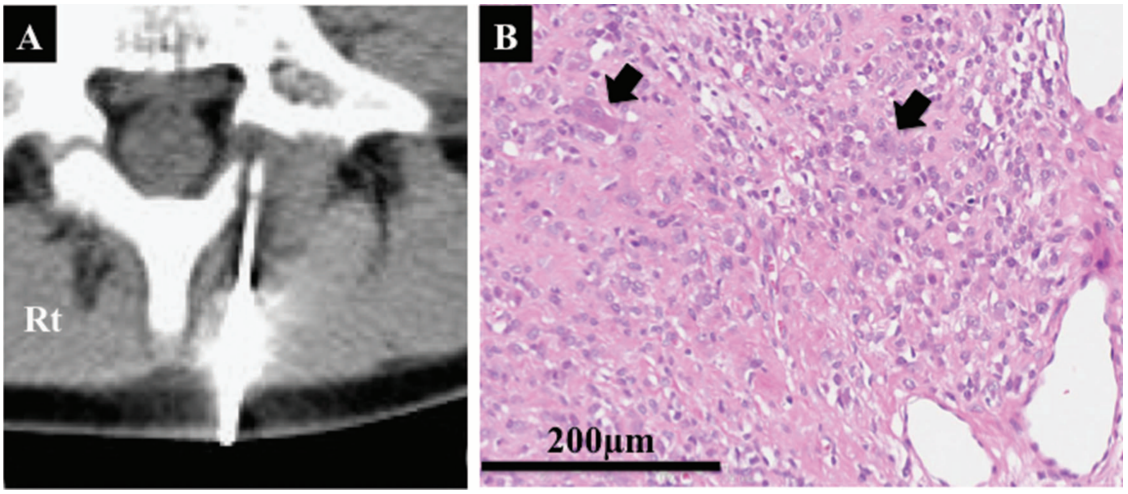

Figure 4. Biopsy findings. Biopsy tract after computed tomography–guided needle biopsy **(A)**. Photomicrograph showing proliferative mononuclear histiocyte-like cells and a few multinucleated giant cells (arrows) **(B)**. Hematoxylin-eosin staining, ×200. Scale bar, 200 μm.

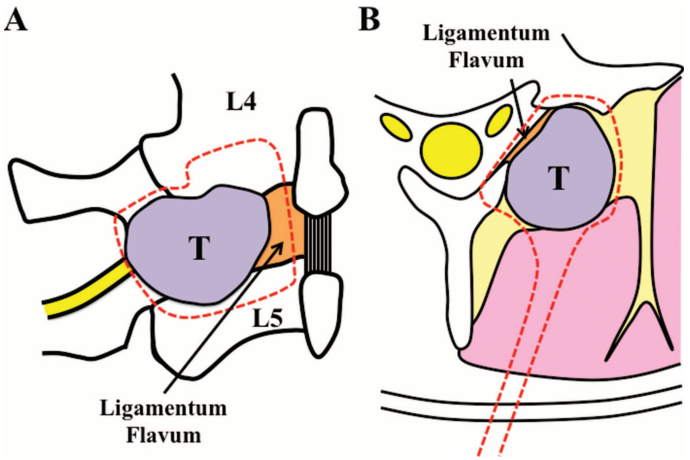

Figure 5. Surgical margin. The tumor was resected by partial laminectomy at left L4 and L5 and partial facetectomy at left L4–L5 and L5–S with the ligamentum flavum. Coronal view **(A)**; axial view **(B)**. T indicates tumor.

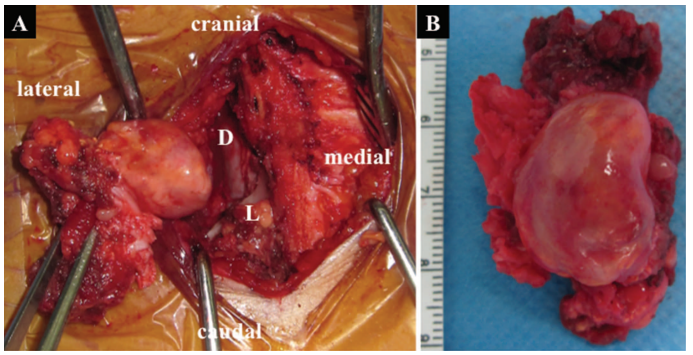

Figure 6. Intraoperative and postoperative findings. The mass adhering to the ligamentum flavum during surgery **(A)**. The 2.5 × 1.0-cm mass was softly elastic and light yellow-reddish **(B)**. L indicates lamina; D, dural tube.

DISCUSSION

PVNS has an occurrence of 80% in the knee joint and 15% in the hip joint but it rarely occurs in the spine.[3,4] Kleinman *et al*[5] reported the first case of PVNS in the cervical spine in 1980. Because of the rarity of spinal PVNS, precise examination is needed to obtain a correct diagnosis. Various symptoms of PVNS in the spine have been reported and include low back pain, radiculopathy, and myelopathy, whereas one-third of cases present with no symptoms and the tumors are found incidentally by imaging. Our patient presented with only low back pain, which is so common that it cannot be used alone to distinguish between PVNS and spondylolysis.

In the absence of specific symptoms, radiological findings are vital for making a correct diagnosis in cases of PVNS. In particular, PVNS often exhibits bone erosion on plain radiograph and computed tomographic scan. Magnetic resonance imaging of PVNS typically shows low intensity on both T1- and T2-WIs, with the low intensity on T2 STAR-WI indicating deposition of hemosiderin.[2] Furthermore, if gadolinium-enhanced fat saturated T1-WI shows contrast enhancement, the lesion may be a soft tissue tumor, including PVNS.[6] In the present case, the tumor showed intermediate intensity and gadolinium enhancement on T1-WI and high intensity on T2-WI and T2 STAR-WI, indicating a soft tissue tumor, but not specifically PVNS. Therefore, computed tomography–guided needle biopsy was needed to make a definitive diagnosis. Although hemosiderin depositions are characteristic feature in PVNS, it was reported that PVNS of the spine often was less pigmented.[7,8] Therefore, this lumbar case also showed poor hemosiderin depositions.

Because PVNS is a benign but locally aggressive tumor, complete excision is required.[9,10] If complete resection is not achieved, the probability of recurrence is high.[11,12] In the present case, gross total resection and marginal resection of the lamina and flavum were attempted because a histological

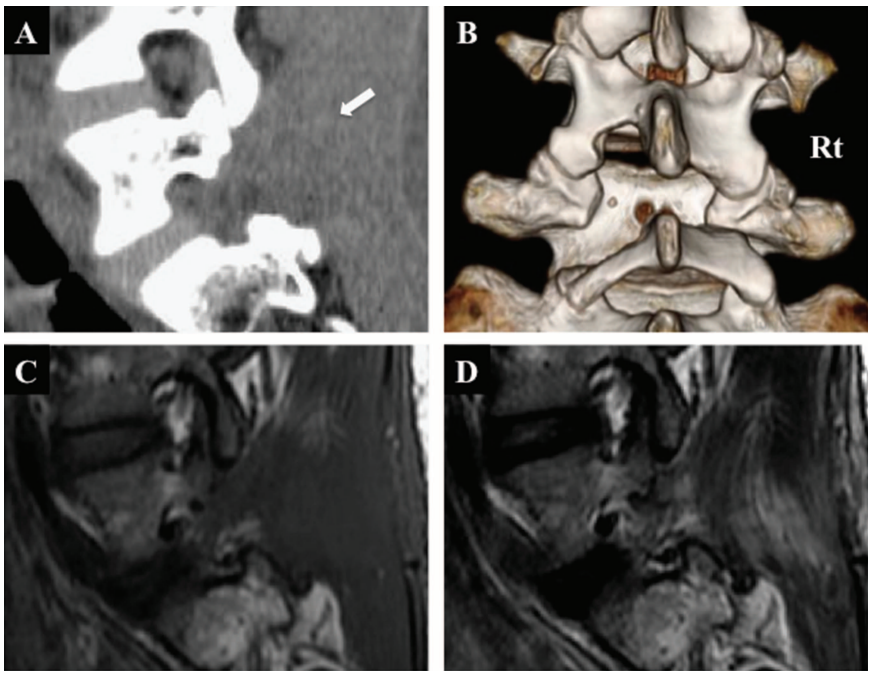

Figure 7. Computed tomographic (CT) scans obtained 5 days and magnetic resonance images obtained 3 months postoperatively, showing no residual lesion and no apparent instability. Sagittal CT scan **(A)**, 3-dimensional CT scan **(B)**, sagittal T1-weighted image (WI) **(C)**, and sagittal T2-WI **(D)**.

diagnosis was obtained before surgery. However, careful follow-up is needed because PVNS has a high recurrence rate.

The etiology of PVNS has not been elucidated. In 1941, Jaffe *et al*[13] used the term PVNS for the first time to describe a proliferative, tumefacient villonodular lesion of the synovium. However, whether or not PVNS is an inflammatory lesion or a neoplasm is controversial, and the cells of origin have not been elucidated.[14,15] We could not clarify whether the present tumor originated from the L4–L5 facet or the L5–S facet because it adhered to the pseudoarthritic pars defects. As Sairyo *et al*[1] and McCormick *et al*[16] showed that pseudoarthritic pars defects in lumbar spondylolysis communicate with both the cranial and caudal facet joints, we speculated that PVNS in the present case arose from the adjacent facet joint of the pars defect in spondylolysis.

Overexpression of an inflammatory mediator (*e.g.*, colony-stimulating factor 1) in neoplastic cells has been suggested to lead to an abnormal accumulation of non-neoplastic cells that form a tumorous mass in PVNS.[15,17] It is, therefore, possible that PVNS in the present case was the result of an inflammatory reaction in response to spondylolysis, although this is the only case reported to occur in a pars defect. Consequently, PVNS in pars defects in lumbar spondylolysis is considered to be both reactive and neoplastic.

To the best of our knowledge, this is the first report of PVNS in a pars defect. Although we think that complete resection was successful, we are carefully following up the patient.

➢ Key Points

- ❑ This is the first report of PVNS in a pars defect in a patient with lumbar spondylolysis.
- ❑ Because PVNS is a benign but locally aggressive tumor, complete excision is required. Preoperative computed tomography–guided needle biopsy was very useful to make a definitive diagnosis.
- ❑ Because several case studies reported that pseudoarthritic pars defects in lumbar spondylolysis communicate with both the cranial and caudal facet joints, we speculated that PVNS in a pars defect arose from the adjacent facet joint.

References

1. Sairyo K, Sakai T, Mase Y, et al. Painful lumbar spondylolysis among pediatric sports players: a pilot MRI study. *Arch Orthop Trauma Surg* 2011;131:1485–9.
2. Motamedi K, Murphey MD, Fetsch JF, et al. Villonodular synovitis (PVNS) of the spine. *Skeletal Radiol* 2005;34:185–95.
3. Giannini C, Scheithauer BW, Wenger DE, et al. Pigmented villonodular synovitis of the spine: a clinical, radiological, and morphological study of 12 cases. *J Neurosurg* 1996; 84:592–7.
4. Mimura T, Kawasaki T, Yagi K, et al. Pigmented villonodular synovitis causing osteonecrosis of the femoral head: a case report. *Case Rep Orthop* 2013;2013:756954.
5. Kleinman GM, Dagi TF, Poletti CE. Villonodular synovitis in the spinal canal: case report. *J Neurosurg* 1980;52: 846–8.
6. Bessette PR, Cooley PA, Johnson RP, et al. Gadolinium-enhanced MRI of pigmented villonodular synovitis of the knee. *J Comput Assist Tomogr* 1992;16:992–4.
7. Oh SW, Lee MH, Eoh W. Pigmented villonodular synovitis on lumbar spine: a case report and literature review. *J Korean Neurosurg Soc* 2014;56:272–7.
8. Parmar HA, Sitoh YY, Tan KK, et al. MR imaging features of pigmented villonodular synovitis of the cervical spine. *AJNR Am J Neuroradiol* 2004;25:146–9.
9. Yener U, Konya D, Bozkurt S, et al. Pigmented villonodular synovitis of the spine: report of a lumbar case. *Turk Neurosurg* 2010;20:251–6.
10. Rovner J, Yaghoobian A, Gott M, et al. Pigmented villonodular synovitis of the zygoapophyseal joint: a case report. *Spine* 2008;33:E656–8.
11. Musluman AM, Cavusoglu H, Yilmaz A, et al. Pigmented villonodular synovitis of a lumbar intervertebral facet joint. *Spine J* 2009;9:e6–9.
12. Oe K, Sasai K, Yoshida Y, et al. Pigmented villonodular synovitis originating from the lumbar facet joint: a case report. *Eur Spine J* 2007;16(suppl 3):301–5.
13. Jaffe HL, Lichtenstein L, Sutro CJ. Pigmented villonodular synovitis, bursitis and tenosynovitis. *Arch Pathol Lab Med* 1941; 31:731–65.
14. Nishio J. Updates on the cytogenetics and molecular cytogenetics of benign and intermediate soft tissue tumors. *Oncol Lett* 2013;5: 12–8.
15. West RB, Rubin BP, Miller MA, et al. A landscape effect in tenosynovial giant-cell tumor from activation of CSF1 expression by a translocation in a minority of tumor cells. *Proc Natl Acad Sci U S A* 2006;103:690–5.
16. McCormick CC, Taylor JR, Twomey LT. Facet joint arthrography in lumbar spondylolysis: anatomic basis for spread of contrast medium. *Radiology* 1989;171:193–6.
17. Stacchiotti S, Crippa F, Messina A, et al. Response to imatinib in villonodular pigmented synovitis (PVNS) resistant to nilotinib. *Clin Sarcoma Res* 2013;3:8.