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CASE REPORT

Three cases of L4–5 Baastrup's disease due to L5–S1 spondylolytic spondylolisthesis

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Abstract

Purpose Baastrup's disease is characterized by degeneration of spinous processes and interspinous soft tissue, which may cause spinal stenosis. Purpose of this article is to report the possible new cause of Baastrup's disease and result of surgical treatments.

Methods Authors treated three cases of Baastrup's disease on L4–L5 with L5–S1 spondylolytic listhesis. Conservative treatment did not relieve the pain; therefore, surgical treatments were planned according to each specific disease condition.

Results In one case, anterior lumbar interbody fusion of L5–S1 was performed, and after surgery, the size of epidural cyst on L4–L5 was decreased. L4–L5 bilateral laminectomy was performed to directly decompress posterior epidural cyst in a case with stable L5–S1 spondylolytic listhesis. In last case, facet joints and spinous process were removed by L5–S1 posterior lumbar interbody fusion (PLIF) surgery. After the surgery, patients' back and leg pain was improved and postoperative MRI revealed successful decompression of the spinal canal. Improvement in back and leg symptoms was noted at 12-month follow-up.

Conclusions Baastrup's disease at the L4–L5 level may have developed from the instability caused by L5–S1 spondylolytic spondylolisthesis. Viable treatment options include the fusion of L5–S1 or a laminectomy at the L4–L5 level.

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Introduction

Baastrup's disease, first described in 1933 and known as "kissing spine," is a condition characterized pathological changes in the lumbar spine of the appositional surfaces of adjacent spinous processes and the soft tissue between them [1]. The disease, associated with lower back pain, develops owing to the close approximation and impingement of one spinous process on another. Images show contact between adjacent lumbar spinous processes with hypertrophy, sclerosis, and flattening of the appositional surfaces. Bursa, which is filled with synovial fluid, can form in the interspinous region. The propagation of a bursa into the dorsal epidural space can result in posterior epidural cysts that may cause symptomatic spinal stenosis [2–10]. Baastrup's disease manifests clinically as localized midline lumbar tenderness and pain on spine extension, which can be relieved by spinal flexion. Fluoroscopy image-guided interspinous injection or surgical options are potential treatments for Baastrup's disease.

Here, we report three cases of Baastrup's disease along with intraspinal posterior epidural cysts and lower level spondylolytic spondylolisthesis, which resulted in varying degrees of spinal stenosis. The appropriate treatment for each case was decided in accordance with the degree of spinal stenosis.

Case report

Case 1

A 52-year-old man presented with radiating pain in his left calf and Grade 4 weakness in his left ankle plantar flexion experienced 1 month prior. He also complained of lower

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back pain that he experienced for 3 years. The pain was aggravated by activity, and he had developed symptoms of neurogenic intermittent claudication in the past 6 months. Conservative treatment and selective nerve root blocks had been unsuccessful. Local spinal tenderness and paraspinal muscle spasm on flexion were present. The patient stood with a stooped posture and had painful restriction of extension. Radiographs showed the presence of Grade 2 spondylolytic spondylolisthesis at the level of the L5 over S1. T2-weighted magnetic resonance imaging (MRI) revealed the presence of neoarthrosis between the spinous processes of L4 and L5 with associated interspinous bursitis, indicative of Baastrup's disease. Lumbar interspinous bursitis is defined as a fluid-like signal that intervenes between consecutive spinous processes. There was also a cyst in the midline in the posterior epidural space at the same level, which compromised the canal as a result (Fig. 1a, b).

We performed an anterior lumbar interbody fusion (ALIF) and percutaneous pedicle fixation (PPF) of L5 and S1 to restore the disc height, thereby indirectly decompressing the left foramen at the L5–S1 level. Immediate postoperative MRI showed a size reduction in the cyst that was located in the upper level of the fusion segment, even without spinous process excision and direct decompression of the cyst (Fig. 1c, d). The space between the L4 and L5 spinous processes was increased, from 0 to 6 mm after fusion of the L5–S1 segment, owing to the restoration of the L4 to S1 segmental lordosis (from 37° to 30°) and disc height (6–14 mm; Fig. 1e, f). Following surgery, the patient noted a significant improvement in back pain, radiating left leg pain, and neurogenic claudication. At the 12-month postoperative follow-up, his left ankle plantar flexion weakness had improved to Grade 5.

Case 2

An 80-year-old man complained of progressive mechanical back pain and both leg numbness for >5 years. He had developed symptoms of neurogenic claudication in the past 3 months and was treated with medication, with no improvement. Grade 1 spondylolytic spondylolisthesis of L5 over S1 and instability was evident on flexion–extension radiography and computed tomography (CT; Fig. 2a). MRI confirmed severe L4–L5 spinal stenosis from a large central posterior epidural extension of a Baastrup's interspinous bursal cyst and an L5–S1 foraminal stenosis with left subarticular facet synovial cyst (Fig. 2b–d).

Owing to severe stenosis, a posterior lumbar interbody fusion (PLIF) and PPF of L5 and S1 were performed for the direct removal of the posterior epidural cyst of Baastrup's disease and left facet synovial cyst. Postoperatively, the patient's back and leg pain resolved. Immediate postoperative MRI revealed decompression of the spinal canal by removal of abutting L5 spinous process and lamina (Fig. 2e, f). At 12-month follow-up, the patient was found to have experienced significant clinical improvement.

Fig. 1 **a**, **b** T2-weighted magnetic resonance imaging (MRI) revealed an interspinous bursitis (*empty arrow*) and posterior epidural cyst (*white arrow*), with the resultant canal compromise. **c**, **d** Postoperative MRI showed a reduction in size of the cyst. **e** Preoperative lateral lumbar X-ray shows the apposition of the L4 and L5 spinous process (*arrow*) with sclerosis. **f** The space between the L4 and L5 spinous processes (*arrow*) was increased after the surgery

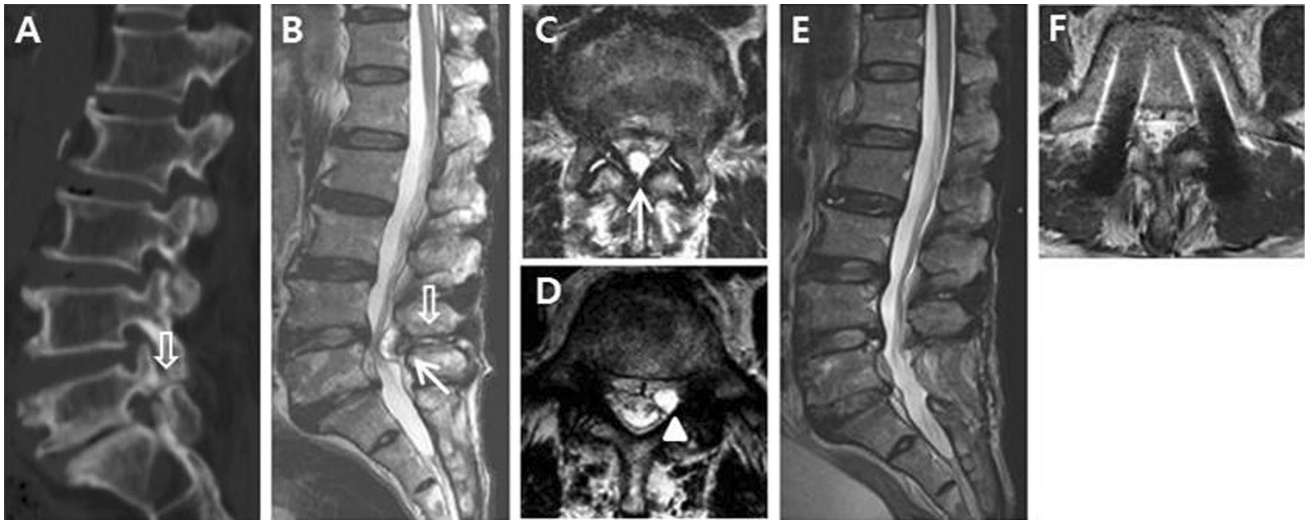

Fig. 2 **a** Computed tomography (CT) image showing spondylolytic spondylolisthesis (*empty arrow*). MRI confirmed **b**, **c** an interspinous bursitis (*empty arrow*) and a large central posterior epidural cyst causing spinal stenosis (*arrow*). **d** A subarticular facet synovial cyst (*arrow head*), which was responsible for foraminal stenosis, was also observed. **e**, **f** Postoperative MRI revealed a well-decompressed spinal canal

Case 3

A 70-year-old man presented with lower back pain and radiating pain in both legs. His pain was aggravated by extension and neurogenic claudication distances <200 m. Radiography revealed Grade 1 spondylolytic spondylolisthesis of L5 over S1, with no evidence of spinal instability in flexion–extension X-ray and CT (Fig. 3a–c). MRI demonstrated a large posterior epidural cyst and central disc protrusion resulting in severe spinal stenosis at the L4–L5 level and lumbar interspinous bursitis between the L4 and L5 spinous process (Fig. 3d, e).

The patient underwent an L4–L5 laminectomy and excision of spinous process for the resection of the epidural cyst. After surgery, the patient's back pain and neurogenic claudication were relieved. Postoperative MRI revealed successful removal of the large posterior epidural cyst (Fig. 3f, g). Improvement in back and leg symptoms was noted at 12-month follow-up.

Discussion

There were reports about Baastrup's disease with adjacent spodylolisthesis [2, 8]. We propose that Baastrup's disease may have developed from lower level spondylolytic spondylolisthesis. Authors thought of two possible mechanisms. In the case of the L5–S1 spondylolytic spondylolisthesis, the L5 vertebral body is shifted anteriorly, and lumbar segmental hyperlordosis occurs to restore sagittal

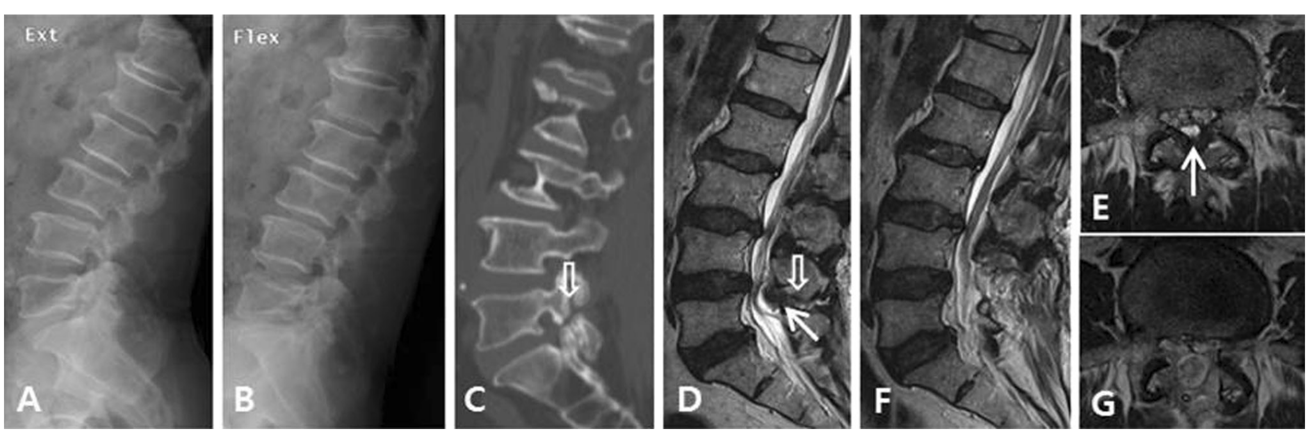

Fig. 3 **a–c** Stress radiographs showed no evident spinal instability but spondylolysis (*empty arrow*) in CT. MRI demonstrated **d** an interspinous bursitis (*empty arrow*) and **e** a large posterior epidural cyst (*arrow*) that caused spinal stenosis. **f**, **g** Successful removal of the large epidural cyst was confirmed on postoperative MRI

Table 1 Review of previously reported cases of posterior epidural cyst associated with Baastrup's disease

Authors (year)	Sex	Age	Symptom	Level	Combined disease	Treatment
Chen et al. [2] (2003)	M/F = 9:1	43–77 years (average 67)	Low back pain: 9 Radiculopathy: 8		Facet joint arthropathy: 4 Spondylolisthesis: 6 Disc protrusion: 2	Conservative treatment: 4 Decompression with spinal fusion: 6
Rajasekaran et al. [3] (2003)	F	50	Low back pain Neurogenic claudication	L3–4	Grade 1 anterolisthesis L3–4 Grade 2 anterolisthesis L4–5 Facet joint arthropathy	Spinous process excision Decompression with spinal fusion L3–L4–L5
Hui et al. [5] (2007)	F	72	Back pain, leg weakness Neurogenic claudication	L4–5	Disc bulging Facet joint arthropathy	Surgical decompression
	M	75	Tender lump in lower back	L3–4		Conservative treatment
Jang et al. [4] (2010)	M	60	Low back pain Neurogenic claudication	L4–5	Grade 1 anterolisthesis L4–5 Facet joint arthropathy	Spinous process excision Decompression with spinal fusion L4–L5
Joseph DeMattia [6] (2016)	M	57	Low back pain radiating both legs	L4–5		Laminectomy L4–L5
Breed et al. [7] (2016)	M	62	Low back pain Neurogenic claudication	L3–4		Minimally invasive image-guided percutaneous procedure
Present cases	M	52	Low back pain, leg pain Neurogenic claudication	L4–5	Grade 2 anterolisthesis L5–S1 Facet joint arthropathy	ALIF and PPF L5–S1 without direct decompression of cyst
	M	80	Low back pain, leg pain Neurogenic claudication	L4–5	Grade 1 anterolisthesis L5–S1	PLIF and PPF L5–S1 with direct decompression of cyst
	M	70	Low back pain, leg pain Neurogenic claudication	L4–5	Grade 1 anterolisthesis L5–S1	Laminectomy L4–L5

M male, *F* female, *L* lumbar, *ALIF* anterior lumbar interbody fusion, *PPF* percutaneous pedicle fixation, *PLIF* posterior lumbar interbody fusion

imbalance [11]. Owing to segmental hyperlordosis, the "kissing" of the L4 and L5 spinous process may occur. Another mechanism is instability. In spondylolysis of L5, L5 lamina and spinous process is separated from pedicle and vertebral body. This unstable part of L5 lamina and spinous process abuts L4 spinous process and posterior epidural cyst is formed on L45 level. Segmental instability leads to ligamentum flavum hypertrophy and spinal stenosis [12, 13]. Synovial facet cyst is also likely to occur in unstable lumbar spine [14]. In Baastrups disease of L45 level with L5S1 spondylolytic spondylolisthesis, L45 posterior epidural cyst may be formed as a result of L5 instability from lysis.

Surgical treatment of spondylolytic spondylolisthesis is fusion surgery including TLIF, PLIF and ALIF [15–19]. The methods of treatment used in the current series were variable as they were specific to each patient's individual pathology. In the first case, the authors performed an ALIF and PPF without posterior decompression because the posterior epidural cyst was small and most of the symptoms were due to the L5–S1 foraminal stenosis. However, indirect decompression through the restoration of disc height, spondylolisthesis, and lumbar lordosis could have also reduced the size of an intraspinal posterior epidural cyst that was not at the same level. The correction of spondylolisthesis and stabilization of the segment also seem to play a role in relieving stenosis caused by the cyst. In the second case, PLIF was performed on the main lesions, i.e., the foraminal stenosis and facet cyst. The resection of the L5 spinous process subsequently removed the posterior epidural cyst on the L4–L5 level. In the third case, the L5–S1 segment was not unstable, even though spondylolysis was present on L5, and the posterior epidural cyst was large enough to cause spinal stenosis. Decompression of cyst was performed with an L4–L5 laminectomy. Although partial surgery and total surgical excision of the spinous process did not produce clinical improvement in 64 patients, according to Beks et al. [20], these methods have proven particularly useful for patients with posterior epidural cysts [2–6]. However, more follow up is needed for concern of progressive instability since intervertebral disc space was well preserved. Subsequent fusion of L5S1 may be needed if anterolisthesis and the symptoms occur. As in summary, L45 laminectomy only is considered in large sized posterior epidural cyst with stable L5S1. L5S1 PLIF is considered if L5S1 is unstable and posterior epidural cyst is large. L5S1 ALIF is considered if L5S1 is unstable and posterior epidural cyst is small. Size of posterior epidural cyst is important but symptoms of patients should also be considered. Symptomatic posterior epidural cyst will result in both leg claudication rather than one leg radiculopathy.

The chance of developing Baastrup's disease, which occurs owing to the close approximation of adjacent spinous process, increases with age owing to the elevation of degenerative changes that occur with normal ageing [21]. Maes et al. [22] reported an 8.2% prevalence of Baastrup's disease in 539 symptomatic patients undergoing lumbar spinal MRI, while Chen et al. [2] found that the prevalence of intraspinal posterior epidural cyst associated with Baastrup's disease was <1%. Symptomatic spinal stenosis caused by the posterior epidural extension of the interspinous bursal cyst has only been reported in a few cases [2–7]. In the cases presented herewith, the patients with spondylolytic spondylolisthesis had posterior epidural cyst in the upper level. Jang et al. [4] suggested that a cystic lesion can occur as an epidural extension of the interspinous bursa that had changed into a cyst-containing fibrotic mass over time owing to peripheral inflammation.

The role of Baastrup's disease and lumbar interspinous bursitis as a substantial nociceptor is controversial [23, 24]. The majority of patients with Baastrup's disease have substantial degenerative disc disease and facet osteoarthrosis, which are the most likely sources of pain and potential therapeutic targets. From the current cases, we can infer that the correction of excessive lordosis and instability may alleviate lower back pain and decrease the size of the posterior epidural cyst.

The proposed treatments were a conservative course of analgesics and non-steroid anti-inflammatory drugs, CT-guided multiple needle fenestration and injection, or surgical decompression [2–7, 10]. A summary of previously reported cases of symptomatic posterior epidural cyst associated with Baastrup's disease is shown in Table 1. Although the duration of follow-up or treatment response was not reported in previous cases, we report a relatively long-term follow-up with good treatment response.

Further cases are needed to confirm whether L5–S1 spondylolytic spondylolisthesis may cause L4–L5 Baastrup's disease. The outcomes of different surgical methods should be evaluated to determine the best treatment method. This study emphasizes that following confirmation of L4–L5 Baastrup's disease, and careful analysis should be performed at the L5–S1 level to note a spondylolytic lesion to avoid misdiagnosis.

Conclusion

L4–L5 Baastrup's disease may develop owing to the instability caused by an L5–S1 spondylolytic spondylolisthesis. Viable treatment options include the fusion of L5–S1 or a laminectomy at the L4–L5 level.

Compliance with ethical standards

Conflict of interest None.

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