Posterior epidural fibrotic mass associated with Baastrup's disease

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Received: 13 December 2009 / Published online: 9 January 2010
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Abstract A few reports have demonstrated rare cases of Baastrup's disease that involve epidural cysts that cause dural compression. However, there have been no reports of a midline epidural fibrotic mass being associated with Baastrup's disease. A 60-year-old man presented with neurogenic claudication that had lasted for 5 years. Radiography showed anterolisthesis at the L4–L5 level, magnetic resonance imaging demonstrated severe stenosis due to a posterior noncystic mass, and the linear fluid signal tracked into the posterior epidural space at the L4–L5 level. A cleft in the ligamentum flavum was identified by probe at surgery, and this enabled the probe to be inserted into the epidural space without excising ligamentum flavum. Histological analysis showed that the fibrotic mass consisted of a collagen matrix that had a cystic component and exhibited a peripheral inflammatory reaction. This report shows that it is possible for an extended epidural cystic mass that occurs in Baastrup's disease to change over time through peripheral inflammation into a cyst-containing fibrotic mass.

Keywords Baastrup's disease · Spinal stenosis · Epidural mass · Fibrosis

Introduction

Baastrup's disease is a well-known disease that is characterized radiologically by the close approximation of

opposing spinous processes, resulting in spinal degeneration such as sclerosis and flattening of the adjacent spinous processes and adventitious bursa formation in the intervening interspinous soft tissues [11]. The causal relationship between this condition and clinical back problems remains unclear [1, 3, 5, 7]. In fact, it has been shown that pain improves after surgical excision of the spinous process [2] or the injection of local anesthetic agents [8]. However, the possibility has been raised that Baastrup's disease may not actually be a disease entity, but is rather just one of a spectrum of degenerative spinal diseases [2, 7, 10], because it is usually associated with various degenerative spinal pathologies [2, 7].

In rare cases [4, 9], Baastrup's disease is associated with the epidural cystic extension of an interspinous bursa that results in dural compression, especially when the lumbar spine is extended. Here, we report a case of Baastrup's disease that is associated with a fibrotic mass in the posterior midline epidural space that shows a cystic component in histological findings and induces dural compression.

Case report

A 60-year-old man presented with neurogenic claudication that had lasted for 5 years and intermittent but not severe back pain. Upon physical examination, tenderness at the L4–L5 level and aggravation of the back pain when placed in the extended position of the lumbar spine were not observed, although the patient did sometimes complain of back pain while lifting a weight in the flexed position of the lumbar spine. Radiographs taken under flexion–extension stress revealed a degenerative lumbar spine with Grade 1 anterolisthesis (Fig. 1a, b). Additionally, magnetic resonance

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Fig. 1 **a, b** Stress radiographs showing the anterolisthesis at the L4–L5 level. **c, d** T1- and T2-weighted sagittal magnetic resonance imaging (MRI) revealed that the linear fluid signal tracked into the posterior epidural space. Also observed was a round mass (*arrows*) in

the epidural space, which was responsible for dural compression and showed low signal intensity on the T1- and T2-weighted images. **e, f** The central portion (*arrowhead*) of the epidural mass showed low signal intensities on the T1- and T2-weighted axial images

imaging (MRI, 3.0-T Achieva Intera, Philips, Netherlands) demonstrated severe spinal stenosis with a bulging annulus and facet joint hypertrophy, and that the linear fluid signal tracked into the posterior epidural space between the spinous processes of L4 and L5 (Fig. 1c–f). A round mass in the posterior epidural space at the L4–L5 level showed low-intensity signals in T1- and T2-weighted sagittal images (Fig. 1c, d). T1- and T2-weighted axial images showed a round mass in the posterior midline epidural space that consisted of a round rim with a high signal intensity and a central portion of low signal intensity (Fig. 1e, f).

The patient was treated with an epidural block at the L4–L5 level and medication and his symptoms improved to some extent. However, 6 months after his first visit to our clinic, he presented with an aggravation of the neurologic claudication in spite of conservative treatment. The claudication distance was <50 m.

Surgery was performed to decompress the spinal canal and to fuse the L4–L5 level. A total en bloc excision that encompassed half of the spinous process of L4 to half of the spinous process of L5 was made following bilateral pedicle screw instrumentation of L4 and L5. An empty, linear cavity in the interspinous ligament at the L4–L5 level was identified; the cavity wall was lined with a tissue resembling the synovial membrane (Fig. 2a) and the cleft on the midline of the ligamentum flavum was determined by probing longitudinally in the operative field. The cleft enabled the insertion of the probe into the posterior epidural space without the excision of any ligamentum

flavum. A round mass in the posterior epidural space causing posterior dural compression was observed after the thickened ligamentum flavum was excised through the midline cleft; the mass showed some adhesion on the dural side (Fig. 2b). The mass had a solid consistency and did not feel like a cyst. Histological analysis of the mass revealed that it largely consisted of collagen matrix that had been produced by fibroblasts as a result of a peripheral inflammatory reaction; the fibrotic mass also contained a cystic lesion that was lined with epithelial cells (Fig. 3).

Fig. 2 **a** Intraoperative photograph showing the round mass (*white arrows*) in the posterior epidural space after partial excision of the ligamentum flavum and location (*black arrow*) of the cleft in the ligamentum flavum at the midline. **b** The wall of the empty cavity in the interspinous ligament at the L4–L5 level was lined with a tissue resembling synovial membrane (*arrowhead*)

Fig. 3 **a** Low-power view of the bisected epidural mass (hematoxylin and eosin staining). This revealed that the mass consisted of a solid fibrotic matrix. A cystic component was found within the mass, and the outer portion of the mass bore edematous changes and vascular proliferation (*double-headed arrows*). **b** The solid portion of the mass appeared to be composed of a collagen matrix that may have been produced by proliferating fibroblasts (*thin arrows*) and inflammatory cells (*triangle pointers*). **c** Epithelial cells (*thick arrows*) lined the surface of the cyst

Discussion

Baastrup's disease (kissing spine) is usually characterized by mechanical back pain due to neoarthrosis between apposing spinous processes that is exacerbated by extension of the lumbar spine and is relieved by bending forward [3]. However, whether the characteristic pathologies of Baastrup's disease are actually the cause of the back pain remains unclear, considering the report that surgical excision of the apposing spinous processes does not always eliminate the patients' symptoms [2]. Indeed, in our case, while MRI revealed a large interspinous bursa at L4–L5, the patient did not complain of mechanical back pain that worsened with extension or tenderness at L4–L5. Instead, the patient only complained of intermittent back pain when lifting in the flexed position due to anterolisthesis at L4–L5. Our observations are consistent with the notion that the mechanical back pain that is usually associated with Baastrup's disease is actually secondary to the degenerative spine disease that often accompanies Baastrup's disease, such as disc degeneration, spinal stenosis, and spondylolisthesis, etc. [2, 7]. Indeed, Maes suggested [7] in 2008 that we should "consider lumbar interspinous bursitis as Baastrup's sign instead of Baastrup's disease."

The other, rarer, presentation of Baastrup's disease besides mechanical back pain is neurogenic claudication resulting from posterior dural compression due to the development of an epidural cyst as an extension of an interspinous bursa [4, 6, 9]. The extension into the epidural space may occur as a result of severe degeneration in the

soft tissue of interspinous processes, and it is possible that this extension could be more severe and associated with a large collection of fluid in interspinous bursa, such as popliteal bursa [4].

Three reports [4, 6, 9] have also demonstrated cysts in the epidural space associated with Baastrup's disease that cause dural compression. In contrast, histological analysis of the mass causing posterior dural compression in our case revealed that it was not a cyst; rather, it was a fibrotic mass that contained a cystic component. Hui and Cox [6] have also reported two cases of unusual cystic extensions in Baastrup's disease on the basis of only MRI findings. However, the MRI findings for the first case were insufficient to define the mass as a cyst, because its central portion showed low signal intensity and its peripheral portion showed high signal intensity, which was probably due to peripheral inflammatory reactions seen in the T2-weighted axial image and T2-weighted sagittal image, which also showed low signal intensity throughout the mass with a sparse high signal intensity. These MRI observations are similar to those described in this report.

This leads us to the question: what caused the epidural fibrotic mass in our case? While it is within the range of possibilities that the intraspinal synovial cyst originated at the facet joint, this is unlikely for the following reasons: the posterior midline mass had no connecting structures as observed in any operative field or radiological finding, and there was a midline cleft in the ligamentum flavum identified by longitudinal probing. In particular, the midline cleft in the ligamentum flavum suggests that the posterior

epidural mass was originally connected with the interspinous bursa. Our histological findings, which revealed that it is a fibrotic mass that contains a cyst and bears peripheral inflammatory changes (Fig. 3), suggest that the mass initially had a cystic lesion. It may have then been enlarged by the fibrotic matrix that was laid down by peripheral inflammatory reactions. Alternatively, the initial cyst may have been ruptured or damaged by unknown causes early in development, and this was followed by replacement with the fibrotic matrix. It is likely that either of these possibilities took place over a considerable period of time.

To the best of our knowledge, this is the first report of the association of Baastrup's disease with an epidural fibrotic mass that is connected to the interspinous bursa through the cleft of the ligamentum flavum and that causes severe neurogenic claudication. Our intraoperative and histological observations and previous reports [4, 6, 9] of cystic extensions of interspinous bursa in Baastrup's disease suggest that the mass in our case may have originated from a cystic lesion that occurred as an epidural extension of the interspinous bursa and that changed over time by peripheral inflammation into a cyst-containing fibrotic mass.

Conflict of interest statement This Research was supported by the Chung-Ang University Research Grants in 2008.

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