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CASE REPORT

Intraosseous pseudomeningocele of the mobile spine: a case report and review of the literature

Emily Rider-Longmaid[1] · Junjian Huang[1] · Ronnie Sebro[1,2,3] · Harvey Smith[3]

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Abstract

Background Pseudomeningoceles most commonly occur due to prior trauma or surgery and are often located in the posterior paraspinous tissues. Here, we report a case of an intraosseous pseudomeningocele that mimicked an intra-osseous T2 hyperintense lesion in the L1 vertebral body.

Case description A 64-year-old male presented with back, left lateral thigh and left knee pain lasting several months. He had no prior history of trauma or surgery. Radiographs of the lumbar spine showed mild levoscoliotic curvature of the lumbar spine, Baastrup's changes between the spinous processes, multilevel degenerative disc disease and facet arthropathy. Magnetic resonance imaging (MRI) of the lumbar spine performed without intravenous contrast showed severe spinal canal stenosis from L1–L2 to L3–L4 and moderate spinal canal stenosis at L4–L5. MRI also showed a 2.5-cm T2 hyperintense lesion involving the posterior aspect of the L1 vertebral body, with questionable contiguity with cerebrospinal fluid. Computed tomography (CT) myelogram was performed instead of biopsy. CT myelogram showed contiguity of the lesion with the intrathecal contrast and a rent in the posterior longitudinal ligament and anterior dura consistent with an intraosseous pseudomeningocele. The patient opted for non-operative management of the pseudomeningocele and his lumbar stenosis due to medical comorbidities.

Conclusions This case illustrates a rare case of an intra-osseous pseudomeningocele and highlights the importance of CT myelogram for diagnosis.

Keywords Radiograph · Magnetic resonance imaging · CT myelogram · Intraosseous pseudomeningocele

Introduction

Spinal pseudomeningoceles were first described by Hyndman and Gerber in 1946, and defined as a dural tear resulting in extravasation of CSF more frequently posterior to the thecal sac [1]. Pseudomeningoceles infrequently present with additional arachnoid tearing and rarely may result in an intraosseous CSF collection [2, 3]. Pseudomeningoceles have been categorized as originating congenitally, iatrogenically, or related to prior trauma [2]. The dura can be compromised by trauma, bony fragments, or punctures in spinal surgeries, particularly laminectomies, resulting in pseudomeningoceles [4]. Pseudomeningoceles arising from accidental dural compromise during surgery was first described by Swanson and Fincher in 1947 [5]. Only two intraosseous pseudomeningoceles of the spine have previously been reported, and these pseudomeningoceles were likely iatrogenic [3, 6]. The majority of pseudomeningoceles are posteriorly located and asymptomatic; however, there is a reported case of an iatrogenic spinal extradural pseudomeningocele that became ossified and symptomatic [7]. We discuss the presentation, and imaging appearance of a case of spinal intraosseous pseudomeningocele in the lumbar spine of a 64-year-old male patient, and review the literature on this entity.

✉ Ronnie Sebro
Ronnie.sebro@uphs.upenn.edu

✉ Harvey Smith
harvey.smith@uphs.upenn.edu

[1] Department of Radiology, University of Pennsylvania, 3400 Spruce Street, Philadelphia, PA 19104, USA

[2] Department of Genetics, University of Pennsylvania, 421 Marie Curie Blvd, Philadelphia, PA 19104, USA

[3] Department of Orthopaedic Surgery, University of Pennsylvania, 3737 Market Street, 6th Floor, Philadelphia, PA 19104, USA

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Case

A 64-year-old male with history of congestive heart failure, type II diabetes, hypertension, coronary artery disease and gout presented with a chief complaint of left leg pain superimposed on a chronic history of back pain in 2015. His severe achy left leg pain that radiated down the left anterior lateral thigh and knee increased over the past several months prior to presentation. His pain was worse with standing from sitting or sleeping. He also complained of back pain, however, his leg pain was subjectively worse. He denied any fever or chills or any bowel or bladder changes. Physical exam showed 5/5 strength bilaterally in the iliopsoas, quadriceps, tibialis anterior and extensor hallucis longus musculature and 4/5 strength bilaterally in the gastrocnemius muscles. Sensation was intact to bilateral lower extremities. He had 1+ symmetric patellar reflexes and trace Achilles reflexes. Babinski test was negative bilaterally. Straight leg raise was positive on the left. Hoffman's sign was negative and there was no clonus noted.

Radiographs of the lumbar spine showed mild levoscoliotic curvature of the lumbar spine, Baastrup's degenerative changes between the spinous processes, multilevel degenerative disc disease and facet arthropathy (Fig. 1a, b). Magnetic resonance imaging (MRI) of the lumbar spine was performed, which showed severe spinal canal stenosis from L1–L2 to L3–L4 and moderate spinal canal stenosis at L4–L5. MRI also showed a T2 hyperintense lesion involving the posterior aspect of the L1 vertebral body with suggestion of contiguity with the cerebrospinal fluid (Fig. 2). There was no herniation of the neural elements into the lesion.

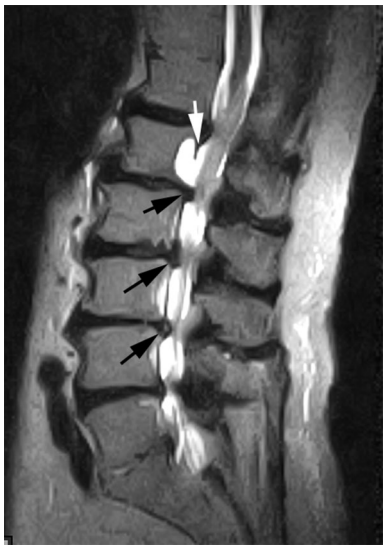

Fig. 2 Sagittal T2-weighted MR image of the lumbar spine (repetition time (TR) = 4710 ms, echo time (TE) = 125 ms, slice thickness 5 mm, interslice gap of 1 mm and acquisition matrix of 256 × 192) shows severe spinal canal stenosis from L1–L2 to L3–L4 and moderate spinal canal stenosis of L4–L5 (black arrows). A 2.5 cm craniocaudal × 1.2 cm anterior–posterior × 2.2 cm T2 hyperintense lesion (white arrow) is seen involving the posterior aspect of the L1 vertebral body

Computed tomography (CT) myelogram showed clear continuity of the lesion with the intrathecal contrast and a rent in the posterior longitudinal ligament and anterior dura consistent with an intra-osseous pseudomeningocele (Fig. 3a, b). CT also showed several calcified disc bulges, including

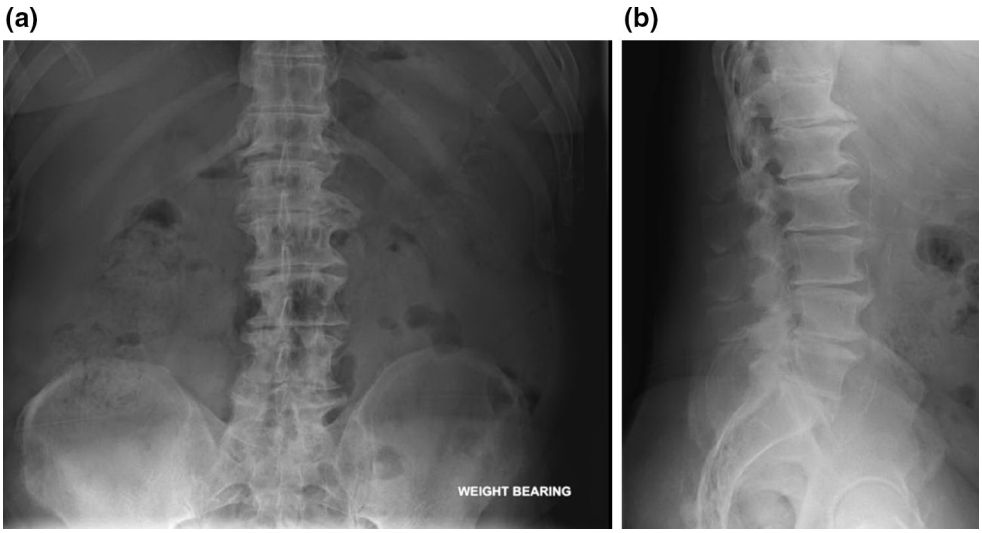

Fig. 1 **a** Anterior–posterior radiograph of the lumbar spine demonstrating levoscoliotic curvature of the lumbar spine, multilevel degenerative disc disease and facet arthropathy. **b** Lateral radiograph of the lumbar spine showing Baastrup's degenerative changes of the spine, multilevel degenerative disc disease and facet arthropathy

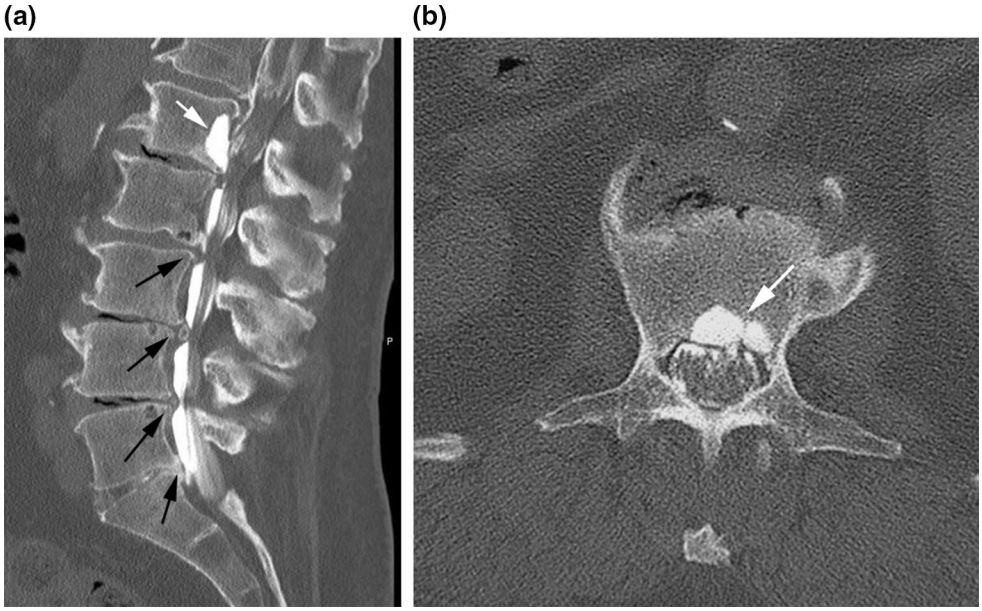

Fig. 3 **a** Sagittal CT image of the lumbar spine from CT myelogram demonstrates iodinated contrast filling the thecal sac and extending into the lesion at L1 (white arrow). Note multilevel degenerative disc disease (black arrows) as well as Baastrup's degenerative changes of the lumbar spine. **b** Axial CT image of the L1 vertebral body from CT myelogram shows iodinated contrast extending through a defect in the posterior longitudinal ligament through the posterior cortex of the L1 vertebral body inferiorly at the level of the L1–L2 disc space (white arrow)

a calcified disc bulge at the L1–L2 level. The patient opted for a trial of non-operative management of his symptoms.

Discussion

Spinal pseudomeningoceles were first described by Hyndman and Gerber in 1946, and defined by a dural tear resulting in extravasation of CSF, frequently posterior to the spinal cord [1]. Intraosseous pseudomeningoceles are secondary to tears of the dura causing CSF leakage into intraosseous cavities [6]. Iatrogenic pseudomeningoceles are most frequently caused by lumbar laminectomies. In 1947, Swanson et al. reported an incidence of pseudomeningocele after lumbar laminectomy to be 0.07% in 1700 cases [5]. In 1983, Teplick et al. reported the incidence of pseudomeningocele after lumbar laminectomy to be 2% in 400 cases [8]. In 1988, Schumacher et al. reported the incidence of pseudomeningocele after lumbar laminectomy to be less than 1% in 3000 cases of lumbar laminectomies [9]. However, intraosseous pseudomeningoceles with no history of trauma or surgery, as seen in this case, are so rare that their true incidence is unknown. These lesions are correctly termed pseudomeningoceles because they lack the arachnoid covering that meningoceles possess [1].

The two previously reported intraosseous pseudomeningoceles of the spine occurred in an 18-year-old male patient, and a 41-year-old male patient [3, 6]. The 18-year-old male patient presented with an intraosseous cystic lesion involving the C2 vertebra 14 years after a Chiari decompression [3]. The 41-year-old patient presented with an intraosseous pseudomeningocele involving the spinous process of the L5 vertebra 10 years after surgical discectomy at that same level [6]. Our case is similar to the first case, where the intraosseous pseudomeningocele involves the vertebral body itself rather than the posterior elements, but differs from both cases as there was no history of prior surgery.

Pseudomeningoceles are commonly asymptomatic, but patients sometimes present with headaches, back pain, and radicular pain due to involvement of the neural elements or stenosis [4]. Sensory and/or motor loss may also accompany radicular pain [2]. Pseudomeningoceles may be palpable when CSF projects into the posterior subcutaneous tissues, and can be noted as an increase in size or firmness with Valsalva maneuvers [4]. Lumbar pseudomeningoceles are less commonly palpable, though watery discharge can occur at operative sites if the pseudomeningocele is iatrogenic in nature [2]. Rarely, patients may present with myelopathy due to spinal cord herniation or focal cord compression, secondary to the pseudomeningocele [2].

MRI and CT myelogram are the best imaging studies for evaluation and diagnosis of an intraosseous pseudomeningocele [10–12]. MRI findings typically show T2 signal hyperintensity similar to that of CSF, indicative of CSF as the content of the cyst [12]. MRI can confirm CSF communication with subarachnoid space and surrounding tissue, and indicate intraosseous CSF collections through the appearance of honeycomb lattice [12]. Although MRI is suggestive, it is often not diagnostic. Other T2 hyperintense lesions, including hemangiomas, Schmorl's nodes, chordomas, and metastases are included in the differential diagnosis in a patient with no known history of surgery. If the patient has a history of surgery, then the differential diagnosis includes an epidural hematoma, and a sterile versus infected post-operative fluid collection [13]. CT myelogram is often required for definitive diagnosis. CT myelogram

shows intra-osseous extravasation of intra-thecal contrast confirming communication between the intra-thecal space and the intra-osseous lesion [11, 14]. Intra-osseous pseudomeningoceles have been described to have a honey-comb appearance on radiographs [15], however, the imaging appearance is sufficiently nonspecific, and further evaluation with MRI and CT myelogram is required for diagnosis. Biopsy is potentially problematic as this may result in the development of a fistula, increasing the risk of arachnoiditis and meningitis.

The definitive treatment for intraosseous pseudomeningoceles is surgical intervention due to the natural history of progression [12]. Surgical intervention involves debridement of the pseudomeningocele cavity, and then primary repair of the dural defect. Primary repairs are frequently bolstered with fascia, or muscle. Synthetic dural patches or allografts are used to augment the dural repair, and bone is remodeled or removed for osseous decompression [2, 11, 12]. Fibrin or hydrogel sealants are frequently used to bolster the dural repair, and patients are placed on flat bed rest for at least 2 days [2]. Ventral defects can be particularly difficult to repair primarily. If a water-tight primary repair is not achieved, a lumbar drain may need to be placed to divert CSF from the repair site. Although the dural tear at the L1 level was asymptomatic, it is possible that one of the neural elements may in future become entrapped and subsequently become symptomatic. The patient's primary presenting symptom was neurogenic claudication, and the surgical goal is decompression of the neural elements. Although the pseudomeningocele presented as chronic and stable, with posterior decompression and subsequent expansion and possible posterior drift of the dura, there is concern that the anterior communication with the pseudomeningocele cavity may be disrupted. Disruption of the anterior pseudomeningocele would result in a cerebrospinal fluid leak. Consequently, it was determined that if at the time of decompression there was extravasation of the cerebrospinal fluid, the dural defect would be repaired. In the event a watertight closure was not possible, the dural defect would have been managed with either a fat graft or managed with onlay ventral collagen graft with a hydrogel sealant. In the setting of a significant ventral dural defect consideration is given to potential use of a cephalad lumbar drain to divert cerebrospinal fluid in the initial postoperative period. In our case, the optimal treatment for the patient would have involved decompression of his lumbar stenosis at L4–L5 and, if needed, repair of the dural defect at L1. However, the patient opted for non-operative management.

The etiopathogenesis of intraossesous pseudomeningoceles is unknown, but patients with this condition all have a dural defect. We postulate that degenerative changes of the disc (herniation, calcification) may cause local mass effect, thin and erode the posterior longitudinal ligament and rupture the dura. We noted that our patient had multiple calcified disc herniations, which may have predisposed the patient to this condition, however this is speculative. If calcified disc herniations are in fact contributory, then this could explain the location of the pseudomeningocele.

Conclusion

We report a rare case of an intraosseous pseudomeningocele involving the L1 vertebral body in a patient with no known history of surgery or trauma. Intra-osseous pseudomeningoceles are best diagnosed using CT myelogram, which would demonstrate communication between the intrathecal subarachnoid CSF and the intra-osseous lesion. Surgery with debridement of the pseudomeningocele cavity and repair of the dural defect is the treatment of choice.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflicts of interest.

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