Anterior sacral pseudomeningocele following minimal trauma

Case report

MICHAEL J. COOLS, M.D., WAJD N. AL-HOLOU, M.D., WILLIAM R. STETLER JR., M.D., FRANK LA MARCA, M.D., AND JUAN M. VALDIVIA-VALDIVIA, M.D.

Department of Neurosurgery, University of Michigan, Ann Arbor, Michigan

Sacral fractures are rare and seldom result in formation of a sacral pseudomeningocele. Treatment of these pseudomeningoceles usually consists of conservative management with flat bedrest or open operative management. The authors describe the case of a 55-year-old woman with an anterior sacral pseudomeningocele that was successfully treated using a lumbar drain for temporary continuous CSF drainage. The patient first presented to an outside institution several days after sacral trauma from an ice skating fall. Initial symptoms included throbbing headaches relieved by lying flat. Head and cervical spine CT demonstrated no abnormality. As symptoms worsened, she presented to another institution where MRI of the lumbar spine indicated sacral fracture with pseudomeningocele. The patient subsequently transferred to the authors’ facility, where symptoms included headaches and occasional mild sacral pain. Given her headaches and the authors’ concern for CSF leak, another head CT scan was performed. This revealed no subdural hematoma or other abnormality. A subsequent CT myelogram revealed an anterior sacral pseudomeningocele at S3–4 with an anterior irregular linear filling defect, likely representing torn dura. Treatment included placement of a lumbar drain (10 ml/hr) and flat bedrest. Resolution of the CSF leak occurred on postprocedure Day 9. At the 4-week follow-up visit, the patient had no clinical symptoms of CSF leak and no neurological complaints. To our knowledge, this is the first description of temporary continuous CSF drainage used to treat a posttraumatic sacral pseudomeningocele. This technique may reduce the need for potentially complicated surgical repair of sacral fractures associated with CSF leak in select patients.

(http://thejns.org/doi/abs/10.3171/2013.6.SPINE12956)

Key Words • cerebrospinal fluid • conservative management • lumbar drain • pseudomeningocele • sacrum • trauma • technique

Sacral fractures are uncommon injuries usually caused by a significant trauma.17 Most often these fractures occur below S-2, as both S-1 and S-2 are rigidly fixed to the pelvis.12 Neurological injury occurs in 15%–25% of cases, with the majority of neurological injuries being radiculopathies.8,16,21 Cauda equina syndrome also occurs, but is much rarer.2,7,8,16,21 Overall, traumatic fractures involving the lumbosacral spine can result in pseudomeningoceles; however, this is most commonly seen at L-4 and L-5 as a result of nerve root avulsion.19 In the case of sacral fractures, pseudomeningocele is a rare complication and is considered to be secondary to nerve sheath avulsion19 or the movement of bony elements resulting in a dural tear.12 Furthermore, as the dural sac commonly ends at S-2, fractures below S-2 should not commonly result in a pseudomeningocele.

Most clinicians initially advocate conservative management for sacral pseudomeningoceles.2,18 However, if the CSF leak or symptoms are persistent, then intervention may be necessary. Interventions that have been described include a blood patch,19 continuous CSF drainage using a lumbarperitoneal shunt,2 and operative dural closure.3 In this report, we describe the case of a woman who presented with a complex sacral fracture at S-3 as-
Sacral pseudomeningocele following minimal trauma

associated with an anterior pseudomeningocele following minimal trauma. We also report on the use of temporary continuous lumbar drainage for management of a sacral pseudomeningocele, a technique that has not been previously described.

**Case Report**

**History and Physical Examination.** A previously healthy 55-year-old woman presented with worsening headaches. She reported that 6 days prior she had been ice skating and fell on her back. Initially she noted severe pain in her sacral region, but she was able to continue ice skating. She denied hitting her head. Over the next few days she began having throbbing headaches when she sat up quickly; the headaches resolved when she lay flat. She initially presented at an outside institution, where head and cervical spine CT demonstrated no abnormalities. As her symptoms continued to worsen, she presented to another outside institution, where MRI of her lumbar spine revealed concern for a sacral fracture and pseudomeningocele.

The patient subsequently transferred to our institution. Other than the headaches and occasional mild sacral pain, the patient noted no other symptoms. Of note, she denied any numbness, paresthesias, or lower-extremity weakness. She also reported no bowel or bladder symptoms. Physical examination was notable for mild tenderness to palpation over the distal pelvis, but no hip tenderness or pelvic instability was observed. Neurological examination was notable for full strength in her lower extremities with normal sensation in her legs and perineum.

Review of her MR images demonstrated a fracture of the S-3 vertebral body, as well as a T2-hyperintense collection extending from the sacral canal to the presacral region (Fig. 1). This collection measured 1.9 cm in the anterior-posterior dimension, 2.5 cm transversely, and 3.4 cm craniocaudally and was contiguous with the dural sac. The conus medullaris terminated at T12–L1, and there were no other abnormalities noted. The sacral fracture noted on MRI could not be appreciated on radiographs, and a pelvic radiograph obtained at that time demonstrated no other fracture. Given the patient’s headaches and our concern for a CSF leak, a noncontrast head CT was obtained; this study revealed no evidence of a subdural hematoma or other abnormality concerning for severe intracranial hypotension.

Given that the patient’s headache was postural and not severe, a CT myelogram was deemed to be safe. The CT myelogram demonstrated a sacral spinal canal pseudomeningocele at S3–4, with an anterior irregular linear filling defect, likely representing frayed or torn dura (Fig. 2). Distal to this, there was an area of extravasation of contrast medium anteriorly into the presacral retroperitoneal space.

**Treatment.** Given the findings of a symptomatic CSF leak, a lumbar drain was placed, with drainage set at 10 ml per hour. The patient was maintained in a flat supine position initially. Activity was gradually increased, and on postprocedure Day 3, the patient was ambulating with the lumbar drain clamped. Her headache returned and she was treated with flat bedrest with continuous drainage. On postprocedure Day 9, a repeat myelogram was performed, revealing resolution of the CSF leak (Fig. 3). The drain was clamped and subsequently removed. The patient noted some urinary retention after removal of the lumbar drain. The consulting urologist attributed the urinary retention to sacral nerve irritation from the lumbar drain and narcotics and did not consider urodynamic studies to be indicated. The patient had no other symptoms concerning for cauda equina syndrome, and the results of her neurological examination were otherwise normal. At her 4-week follow-up visit to the neurosurgery and urology clinics, she had no clinical symptoms of CSF leak and no neurological complaints. Her urinary retention had resolved and the results of her neurological

**Fig. 1.** Sagittal (left) short tau inversion recovery (STIR) and axial (right) pretreatment T2-weighted MR images demonstrating sacral pseudomeningocele. Hyperintense T2-signal fluid collection can be seen in the presacral region contiguous with the spinal canal, measuring 1.9 cm in the anterior-posterior, 2.5 cm in the transverse, and 3.4 cm in the craniocaudal directions.

**Fig. 2.** Sagittal (left) and axial (right) pretreatment CT myelograms revealing an anterior sacral pseudomeningocele. Intrathecal contrast medium was administered using fluoroscopic guidance to the L4–5 level, with immediate extravasation of contrast material outside the dural sac into an irregular collection in the sacral spinal canal around the S3–4 level. The contrast material then became distributed anteriorly and bilaterally into the retroperitoneal space. A fracture was also identified at S-3 and S-4, with retropulsion of a bony fragment into the sacral spinal canal at S-3.
examination were normal. She remained asymptomatic at the 1-year follow-up visit.

Discussion

Here, we present the case of a 55-year-old woman with an S-3 fracture who presented with an anterior sacral pseudomeningocele after minimal trauma. This patient was successfully treated with continuous lumbar drainage, which previously has not been reported as a treatment modality for such injuries.

Sacral fractures are rare injuries that usually result from a significant trauma or occur in the setting of sacral insufficiency secondary to osteoporosis. However, on rare occasions they can result from minimal trauma. Sacral fracture from minimal trauma results in neurological deficit in only 5% of cases, and the deficits usually are minor and resolve with observation.

Pseudomeningoceles are rare complications of sacral fractures. Most of the lumbosacral injuries resulting in pseudomeningoceles described in the literature occurred in the lumbar spine, although in some cases, both the lumbar and sacral spine were involved. Pseudomeningoceles isolated to the sacral region are rare and often involve a preexisting sacral defect such as spina bifida occulta. A pseudomeningocele at the S-3 level would not be typically expected, given that the dural sac usually ends at the S-2 vertebral level. Hadley et al. described a similar finding in a case of an S-4 fracture that also resulted in a pseudomeningocele, but that was a result of significant trauma. Padberg and Coene also described an isolated sacral pseudomeningocele in the sacral spine at S-1, but in that case, there was no associated fracture.

In patients who do present with traumatic lumbosacral pseudomeningocele, the most common presenting symptom is headache, although focal neurological symptoms secondary to direct nerve injury have been described. Cases have also been reported in which patients have had lower-extremity weakness and numbness and low-back pain. Patients with posterior pseudomeningoceles have even presented with a fluctuant mass over the sacrum. An anterior posttraumatic pseudomeningocele could present with neurological deficits as a result of compression of the lumbosacral plexus.

There are several theories that propose a mechanism for the formation of traumatic lumbosacral pseudomeningoceles. While in most cases these injuries are secondary to significant trauma such as a motor vehicle accident, there are several descriptions of such injuries occurring after seemingly trivial trauma. In the initial description of this phenomenon, Nosik felt the pseudomeningocele was caused by a propagated pressure wave directly putting pressure on the cone-shaped nerve sheaths in the lumbar spine, as well as by an increase in intraabdominal pressure. Nosik and others have also noted that there is likely a preexisting defect that predisposes patients to pseudomeningocele formation, given the frequency of falls without pseudomeningocele. Support for this hypothesis comes from reports of sacral pseudomeningoceles in patients with spina bifida occulta identified after a trauma. In each case, the pseudomeningocele occurred at the level of the defect and extended posteriorly.

Banno et al. note that the area around the defect is fragile and that the underlying dura may tear easily if subjected to direct impact.

Our patient, however, did not have any identifiable spinal defects that would have predisposed her to pseudomeningocele formation. Furthermore, the conus medullaris ended at T12–L1, indicating no evidence of a low-lying cord. A more likely mechanism was suggested by Hadley et al., who noted that a delayed dural tear could be secondary to movement of fractured bony elements.

It is possible that the pathophysiology of the CSF leak was an anterior meningocele that ruptured with the trauma. There have been several case reports of anterior sacral meningoceles presenting with urinary retention, pelvic cysts, or even constipation. However, to date, there have

Fig. 3. Sagittal (A and B) and axial (C) posttreatment CT myelograms demonstrating resolution of the CSF leak. Intrathecal contrast was administered with fluoroscopic guidance to the L2–3 level, which was 2 levels above the lumbar drain. At the S-3 level, a collection of contrast material is confined to the spinal canal, with no leakage of contrast material outside the sacral spinal canal.
only been a few case reports of ruptured meningoceles, and those patients presented with meningitis.6,11,18 Also, given that no meningocele was evident after treatment with lumbar drainage, it is unlikely that a meningocele was present before the injury, although we cannot entirely exclude that possibility. Nevertheless, if in fact our patient had a ruptured sacral cyst, conservative management with lumbar drainage treated the associated symptoms, thereby eliminating the need for surgical repair.

In the present case, the diagnosis was made using CT myelography, which clearly identified the CSF leak. In the setting of concern for intracranial hypotension due to CSF leak, one must be judicious in the performance of lumbar punctures as continued CSF drainage could exacerbate the symptoms. At our institution, when evaluating CSF leaks, we consistently perform head CT to rule out subdural hematomas or other findings of severe intracranial hypotension, which we would consider contraindications to lumbar puncture. Furthermore, our neuroradiology department strictly limits CSF drainage for the myelogram in cases of suspected CSF leak.

Treatment of these pseudomeningoceles has traditionally involved either conservative management with observation or operative management. Conservative management consists of prolonged flat bedrest, which has proven effective in a number of cases.3,9,10,13,18,28 If this fails and symptoms persist, intervention may be necessary. The first treatment described was a blood patch,19 a treatment that is still in use for spinal CSF leaks today. Several authors have described operative management. Kitchen et al.4 used a lumboperitoneal shunt to successfully treat a chronic lumbar pseudomeningocele. Where open operative management is undertaken, the operation usually consists of a laminectomy, identification of the dural defect, and direct repair of the defect.3,4,12,24 These operations have been successful in improving neurological function. The only complications reported have been a postoperative CSF leak, which was managed with a spinal drain,12 and communicating hydrocephalus in the case of a patient with an intrathecal hemorrhage.24

Our patient had symptoms that were significant and severely limiting. We initially attempted conservative therapy with flat bedrest and intravenous hydration. However, symptoms persisted, and it was determined that intervention was necessary. We treated her with continuous lumbar drainage for over 1 week, resulting in resolution of her symptoms and radiographic resolution of the CSF leak. Continuous CSF drainage is a well-known treatment modality used for treatment of both intracranial and spinal CSF leaks and pseudomeningoceles.24 However, in the case of traumatic sacral pseudomeningoceles, temporary continuous lumbar drainage has not been previously described as a treatment modality.

Conclusions

We conclude that lumbar drainage is a viable nonoperative option for patients in whom initial conservative management has failed to resolve a CSF leak associated with a sacral fracture, potentially reducing the need for a complicated surgical repair.

Disclosure

Dr. La Marca reports being a consultant for Globus, Lanx, and Biomet and receiving royalties from Stryker.

Author contributions to the study and manuscript preparation include the following. Conception and design: Valdivia-Valdivia, La Marca. Acquisition of data: Cools, Al-Holou, Stetler. Analysis and interpretation of data: Al-Holou, Stetler. Drafting the article: Cools, Al-Holou. Critically revising the article: all authors. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Valdivia-Valdivia. Study supervision: Valdivia-Valdivia, La Marca.

Acknowledgment

The authors would like to thank Holly Wagner for providing editorial assistance.

References


Manuscript submitted October 5, 2012.
Accepted June 3, 2013.
Please include this information when citing this paper: published online July 5, 2013; DOI: 10.3171/2013.6.SPINE12956.
Address correspondence to: Juan M. Valdivia-Valdivia, M.D., Department of Neurosurgery, University of Michigan, 1500 E. Medical Center Dr., Room 3552 Taubman Center, Ann Arbor, MI 48109-5338. email: juanvald@med.umich.edu.