Spinal pseudomeningoceles and cerebrospinal fluid fistulas

Daniel Couture, M.D., and Charles L. Branch, Jr., M.D.

Department of Neurosurgery, Wake Forest University School of Medicine, Winston-Salem, North Carolina

Spinal pseudomeningoceles and cerebrospinal fluid (CSF) fistulas are rare extradural collections of CSF that result following a breach in the dural–arachnoid layer. They may occur due to an incidental durotomy, during intradural surgery, or from trauma or congenital abnormality. The majority are iatrogenic and occur in the posterior lumbar region following surgery. Although they are often asymptomatic, they may cause low-back pain, headaches, and even nerve root entrapment. Leakage of CSF from the wound may cause a fistulous tract, which is a conduit for infection and should be repaired immediately.

Diagnosis can be confirmed on clinical examination or imaging studies including magnetic resonance imaging, computerized tomography myelography, and radionuclide myelography. Treatment must be specific to each patient because the timing, size, symptoms, and location of the dural breach all affect the choice of therapy. Nonsurgical methods may be used, but more frequently operative repair is required.

In this article, the authors review the diagnosis and treatment of spinal pseudomeningoceles and CSF fistulas.

Key Words • pseudomeningocele • cerebrospinal fluid fistula • incidental durotomy

Spinal pseudomeningoceles and CSF fistulas are caused by similar mechanisms and can be considered to be on a continuum. A precise definition of a pseudomeningocele is difficult given the various mechanisms that have been used to explain its pathophysiology. A true meningocele is an extradural collection of CSF in an arachnoid-lined capsule. Most investigators would define a pseudomeningocele as an extradural collection of CSF that results from a dural breach. This extradural fluid may be contained in an arachnoid-lined membrane or a fibrous capsule. Other names for a pseudomeningocele include “pseudocyst,” “false cyst,” and “meningocele spurious.”39,45,47 A CSF fistula results if the extradural fluid communicates with the external environment or with some other body cavity.

Incidence of Pseudomeningoceles

By far, the majority of pseudomeningoceles are iatrogenic, resulting from incidental durotomies during spinal or intradural surgery. Incidental durotomies are a frequent complication of lumbar spine surgery, with a reported incidence between 0.3 and 13%.24,31,42,43 Oppel, et al.,43 reviewed data from 3038 lumbar discectomy operations and found the incidence of durotomy during bone removal or retraction to be 5.9%. The true incidence of pseudomeningoceles following incidental durotomy is unknown because many cases are asymptomatic. Schumacher, et al.,54 reported the incidence of pseudomeningoceles to be less than 0.1% in a study of 3000 patients who had undergone lumbar laminectomy. Note that the incidence is much higher in more complicated cases such as spinal dysraphism or dural scarring, which can occur in patients who have undergone previous surgery or radiation treatment.64 Zide, et al.,63 described a 43% incidence of pseudomeningoceles and a 13% incidence of CSF fistulas in patients undergoing release of a tethered spinal cord. There is also a high incidence of postoperative pseudomeningoceles in patients who have undergone certain cervical procedures. Treatment of ossification of the posterior or longitudinal ligament is linked to particular risk, with an incidence ranging from 4.5 to 32%.34,57

PATHOPHYSIOLOGICAL FEATURES

A pseudomeningocele can occur any time there is an opening of the dura mater. It can be occult or apparent and found incidentally or purposely. Its most common cause is

Abbreviation used in this paper: CSF = cerebrospinal fluid.
by far iatrogenic, but traumatic and congenital causes may also be implicated.6,10,12,17,23 During extradural surgery such as a laminectomy, the dura may be breached by direct trauma, excessive dural or nerve root traction, or dural laceration from sharp bone fragments. Lumbar puncture, inadvertent dural puncture after placing an epidural catheter, or myelography needle puncture are other causes of CSF leaks. In cases that require invasion into the dura, such as repair of a tethered cord or resection of intradural tumors, a dural defect may occur if a watertight closure has not been achieved. Leakage may result even if one recognizes improper suturing of dural defects, particularly in anterior defects, which are difficult to repair.

Cerebrospinal fluid fistulas occur in the immediate postoperative period when fluid communicates through the wound. Occasionally, there is very low fluid flow and it is difficult to detect the fistula. Pseudomeningoceles are caused by the egress of CSF into the soft tissue, but not through the skin closure, eventually forming a fibrous capsule. When the dura is breached but the arachnoid remains intact, the arachnoid can become herniated through the dura and an arachnoid-lined sac becomes the pseudomeningocele. The CSF pulsations can push the pseudomeningocele into the soft tissue and affect its size, shape, and location.

Different theories exist as to the likelihood of a connection remaining patent. One theory holds that when intact arachnoid herniates through the dura, it is more likely for the communication to remain open and form a pseudomeningocele; if an arachnoid tear occurs as well, it is more likely that the communication will close.59 Others assert that the volume of leaked fluid is most important; for example, with a small leak the fluid is more easily absorbed and the dura heals more readily.60 Another theory holds that a ball-valve mechanism can lead to a one-way flow of CSF and thus a higher probability of developing a pseudomeningocele.8

The majority of dural tears heal spontaneously, but certain conditions can prevent healing. Large dural defects, poor overlying soft tissue coverage (as in dysraphism), scar tissue, radiation, infection, nutritional deficits, steroid agents, and elevated CSF pressure can all contribute to poor healing of dural tears. Occasionally, nerve roots can herniate out of the sac and cause pain and radicular symptoms in addition to preventing healing of the breach.

Traumatic causes of CSF fistulas and pseudomeningoceles are rarer and their true incidence is unknown. The majority of these entities occur after an injury to the brachial or lumbar plexus because the nerve roots are anchored between two mobile parts.13 Stretching of the nerve root causes avulsions and ultimately the development of a nerve root meningocele. In such cases, the arachnoid and dura that invest the nerve root are torn and CSF leaks into the soft tissue surrounding the nerve, thus framing a cavity. Wallerian degeneration and CSF pulsations may lead to enlargement of the cavity. Sporadic cases of pseudomeningoceles following blunt trauma, which are not related to nerve roots, have been described but are much rarer.3,7,25,31,41,44,65 Although there is a high rate of dural tears following thoracolumbar burst fractures, these tears rarely progress to pseudomeningoceles or CSF fistulas.7 In contrast, penetrating injuries to the spine have a high incidence of dural tears and CSF leaks.59

Congenital disorders such as neurofibromatosis and Marfan syndrome have also been associated with pseudomeningoceles.69 Note that patients with such disorders often have true meningoceles rather than pseudomeningoceles. It is believed that pseudomeningoceles form due to CSF pulsations on a more elastic dura. Dural ectasia is frequently present in Marfan syndrome, although often it causes no symptoms. Congenital pseudomeningoceles are frequently located anteriorly and are associated with defects in the vertebral bodies.

**Clinical Features**

Patients with pseudomeningoceles and CSF fistulas present with a wide variety of signs and symptoms. The diagnosis of a cutaneous CSF fistula can often be made by inspection of the wound. A watery discharge from the wound, especially if the discharge is augmented during the Valsalva maneuver or associated with headache, are common signs of a fistula. Headaches are believed to be the result of a reduction in CSF volume and lowered intracranial pressure, which in turn causes traction on the meninges and blood vessels and pain. Frequently, a halo sign—a light brown halo that surrounds a central stain on bedding or another absorbent surface—is present. A sample of the fluid may be sent for laboratory examination, a very sensitive and specific test, to determine if β-transferrin is present. Note that β-transferrin is a protein isoform arising by the action of cerebral neuraminidase and is found only in the central nervous system. A very small sample of fluid is required (< 1 ml) and no special handling is needed.69 Measuring the glucose level is an unreliable method, as is comparing the serum and fluid chloride levels. Fever or meningismus indicates bacterial or aseptic meningitis.

As stated previously, pseudomeningoceles are often asymptomatic; however, they can be diagnosed based on a subcutaneous or subfascial fluid collection that increases with Valsalva maneuvers such as sneezing and coughing. Cervical and thoracic pseudomeningoceles are more easily palpable than lumbar ones, but occasionally lumbar collections can track into the subcutaneous tissues. Serum, liquefied hematoma, wound infection, and abscess should be ruled out to reach a definite diagnosis. Patients with pseudomeningoceles can present with localized back pain and postural headaches. Localized nerve root entrapment, herniation, or adhesions can produce pain or radicular symptoms even several weeks or months after surgery.1,6,16,21,31,51,62 Authors of several case reports have described patients with progressive myelopathy, including one patient who was found to have spinal cord herniation into the pseudomeningocele together with cardiac and respiratory effects that caused bulging.4,8,15,19,20,22

**Imaging Studies**

A patient history and physical examination can be augmented by imaging studies to localize the CSF fistula tract or pseudomeningocele. Magnetic resonance imaging is the diagnostic study of choice because of its ability to visualize soft tissue. Note that magnetic resonance imag-
ing typically reveals low signal intensity on T₁-weighted images and high signal intensity on T₂-weighted images, which is consistent with CSF. These images may demonstrate the level of communication with the thecal sac as well as spinal cord compression or nerve root entrapment.²²,²⁹

Computerized tomography myelography is sometimes better for visualizing the location of the pseudomeningocele or fistulous communication relative to the surgical site because of its ability to demonstrate bone. Occasionally, calcification or erosion into the bone is apparent.²⁹,⁵³,⁵⁴,⁶⁰ Delayed computerized tomography myelography is useful in detecting a slow-filling pseudomeningocele.²⁹ Nerve root pseudomeningoceles can also be revealed on myelography. Retrograde radionuclide myelography can be used to detect pleural fistulas or slow, intermittent leaks after lumbar puncture or traumatic injury.⁸,¹⁸,²¹,²⁶

Avoidance and Conservative Treatment

The avoidance of iatrogenic pseudomeningoceles and CSF fistulas can be achieved with a careful surgical technique and the use of an intraoperative microscope. If a breach in the dura mater does occur, it should be closed with a watertight primary closure.

Bedrest is frequently the first step in the conservative management of pseudomeningoceles and CSF fistulas. Oversewing of the wound is required in the case of a CSF fistula. Data from studies have shown the resolution of a CSF fistula with bedrest and a watertight skin closure.³⁶,⁶¹ The routine or prophylactic use of antibiotic agents has not been shown to decrease the incidence of meningitis and, in fact, contributes to multidrug-resistant organisms.¹³ Abdominal binders and focal compression have been shown to be helpful in the treatment of pseudomeningocele.³² A suboccipital pseudomeningocele has been treated successfully with a compression dressing and drainage of the collected fluid.³⁸

An epidural blood patch is a well-known modality of treating intracranial hypotension due to lumbar puncture or epidural injection. It has also been used successfully to treat postoperative CSF fistulas and pseudomeningoceles.³⁰,³⁵ The injection of blood promotes clot formation over the dural tear. Note that clot formation has been found to be increased in the presence of CSF.¹¹,⁵² The clot helps to prevent CSF efflux and allows the dura to heal.³⁷ Data from several reports have demonstrated that epidural patching with the aid of fibrin glue can also be used for treatment.²⁷,⁶⁶

Closed subarachnoid drainage has been successfully used in the treatment of CSF fistulas and pseudomeningoceles. The diversion of CSF decreases the CSF pressure differentially between the intradural and extradural space, allowing the dural breach to heal. Drainage of 120 to 360 cc/day for 3 to 5 days has a 90 to 92% success rate in the treatment of a CSF fistula.³⁸,⁶¹ Lumbar drains are not without complications and involve a risk of spinal headaches (60%), infection such as meningitis (2.5%), discitis (5%), and wound infection (2.5%), transient nerve root irritation, and recurrence.⁵⁶ Diversion of CSF alone may not be sufficient, and frequently the combination of CSF diversion and epidural blood patch is required.³⁷

Surgical Treatment

Surgical dural repair is the definitive treatment for CSF fistulas and pseudomeningoceles. It should be the primary treatment undertaken in patients with profuse leaking of CSF or symptomatic pseudomeningoceles. It is also indicated after the failure of conservative treatment. Careful imaging studies should be obtained to determine the location of the dural breach. If a pseudomeningocele is present, it should be delineated and the wall should be entered. Any nerve roots present should be freed and reduced into the dura. The durotomy site should be protected with a cottonoid, and additional exposure and bone removal should be performed if necessary. Historically, some surgeons have treated a small defect by merely opening the dura mater widely to prevent a ball-valve effect; however, most surgeons recommend primary closure of the dura.

The dura should be closed primarily with a nonabsorbable suture on a taper or reverse cutting needle. If a large defect is present, a patch of fascia or cadaveric dura mater should be used to allow direct closure without compression of the neural elements. Several adjuvant agents have been used in addition to suturing to assist in dural healing. DuraGen, autologous fibrin glue, TachoComb, Tisseal, BioGlue, and other agents have all been successfully used in treatment. Either Gelfoam or muscle alone placed over the dural breach is ineffective in stopping the leak.³⁹,⁴⁰,⁵¹,⁵⁵ Fibrin sealant alone can withstand high hydrostatic pressures, but is effective for only 6 to 7 days and thus must be supplemented with a dural, muscle, or fat graft.³,¹⁴,⁴⁸ A pedicle flap is rarely required in closing the defect. Placement of a drain is controversial because it may lead to a persistent communication between the extradural and intradural space. Postoperative bedrest is generally recommended for several days in patients with lumbar defects.

Some durotomies, such as anterior or far-lateral ones, occur in surgically inaccessible areas. In these cases a midline durotomy can be made to allow for the placement of a muscle or fat graft that may be pulled into the defect. Shunt insertion (ventriculoperitoneal or lumboperitoneal) should be reserved for patients with impaired CSF absorption or those with a pleural fistula or a fistula at the occipital–cervical junction, which require a complicated repair.

CONCLUSIONS

Pseudomeningoceles and CSF fistulas are rare complications that are usually caused iatrogenically following lumbar surgery. Although these entities are frequently asymptomatic, they can be associated with a range of signs and symptoms including headaches, infection, back pain, radiculopathy, and myelopathy. Magnetic resonance imaging is the neurodiagnostic modality of choice, but computerized tomography myelography and radionuclide myelography may be helpful in difficult cases. A meticulous technique during the original surgery and a watertight closure in the event of durotomy are key. If one encounters either of these entities, bedrest, epidural patch with blood or fibrin glue, and CSF diversion may be attempted, although direct surgical repair is the definitive treatment.
References

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Address reprint requests to: Charles L. Branch, Jr., M.D., Department of Neurosurgery Wake Forest University School of Medicine, Winston-Salem, North Carolina 27157-1029. email: cbranch@wfubmc.edu.