Catheter-guided multilevel epidural blood patches in an adolescent boy

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ABSTRACT

Background There is increasing evidence for the use of multilevel epidural catheter-guided blood patches to treat spontaneous cerebrospinal fluid leaks in adults. Yet, there are scarce data for their use in children. Furthermore, higher level epidural blood patches are uncommon in both adult and pediatric populations.

Case presentation An adolescent boy with multilevel cerebrospinal fluid leaks associated with status migrainosus failed conservative pain management treatment. As he remained severely symptomatic, epidural blood patches were required to mitigate his symptoms. Given his multilevel leaks, a catheter-guided blood patch approach was used to treat multiple cervicothoracic and thoracolumbar leaks. After three separate uneventful blood patch procedures, the patient was free of symptoms. Other than some rebound intracranial hypertension, the patient maintained full neurological capacity without further complications. Following the resolution of treatment, he has not sought help for pain since his last blood patch over a year ago.

Conclusions Multilevel catheter-guided blood patches have the potential to be administered to higher level spinal regions to the pediatric population suffering from multiple spontaneous cerebrospinal fluid leaks.

INTRODUCTION

Cerebrospinal fluid leaks are a rare pain condition affecting 5 per 100 000 people in the general population.[1] Spontaneous intracranial hypotension occurs secondary to cerebrospinal fluid (CSF) escaping through the dura. The resultant headaches are similar to those following a lumbar puncture. Postural headaches are relieved when supine and can be associated with hearing and visual disturbances.[2] The diagnosis is established by MRI and conservative treatment includes analgesics, bed rest and hydration.[3] If the patient remains symptomatic, epidural blood patches can be used to seal the dura.[4] In adults, these procedures are commonly reported at the lumbar levels; however, thoracic and cervical blood patches are rarer. Furthermore, treating multiple regions of CSF leaks throughout the dura can make this procedure more complicated or unsuccessful.[5] Yet, scarce data have been reported documenting higher and multilevel blood patches in children.

CASE PRESENTATION

An adolescent boy reported a 2-month history of status migrainosus with no identifiable trigger. He described his pain to be orthostatic, characterized as head tightness, usually central and with an average intensity of 6/10 that could worsen to 9/10. He denied aura and photophobia but reported mild phonophobia. His pain had been refractory to medical treatments such as acetaminophen, ibuprofen, metoclopramide, ketorolac and an occipital nerve block.[6] Furthermore, given the patients' physical appearance of an elongated arm span and height for his age, a genetic work up was initiated, and he was later diagnosed with Marfan syndrome having an FRN1 mutation.

An MRI of the head and spine with gadolinium was done, showing findings of extensive cervicothoracic subdural collections in keeping with the presence of a CSF leak with associated intracranial hypotension (figure 1). An MR myelogram demonstrated leaks at T1 with a large communication into the paravertebral soft tissues through the neural foramina, T7–T8 subdural accumulation, L1–L2 extradural redistribution and prominent contrast opacification of the nerve root sleeves to the right of C7/T5/T10/L5, left of T11 and bilateral at T3/L2/L3/L4 (figure 2). The most extensive epidural and subdural contrast opacifications were mainly at the cervicothoracic junction and lumbosacral levels. Though the MR myelogram can contribute to symptomatic headaches, it was felt that this would not greatly change his symptoms in the context of the multiple leakages he already had.

METHODS

Catheter placed blood patches under CT at two locations, cervical–thoracic and thoracic–lumbar, were offered to the patient and his parents. The risk, benefits and novelty of the procedure were discussed with the patient and his family. The decision to use CT imaging was also discussed between the team (radiologist, neurosurgeon and anesthesiologist) and the patients' family. CT imaging was chosen over fluoroscopy, given the significant amount of CSF from different sources seen on MRI. The advantage of using CT was to prevent false-positive accidental punctures of dura mater. CT imaging would allow for more precise identification of the catheter in the epidural space even if there was a CSF leak during the needle placement. After extensive discussions, consent for the procedure was obtained.

The patient was placed in the left lateral position. Epidural catheters 20 gage (Brown medical) were placed under sedation with propofol and dexmedetomidine. The first site of injection of the needle

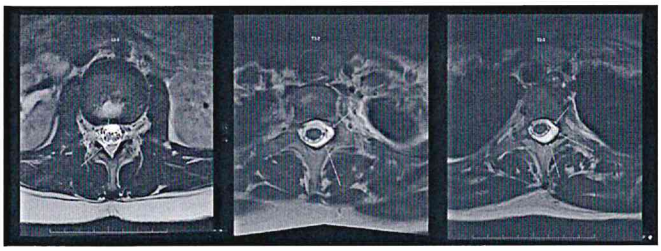

Figure 1 MRI of the brain+C/T/L spine showing extrathecal cerebrospinal fluid in posterolateral cervicothoracic junction and right mid-lumbar region with a potential defect at right posterolateral L2–L3.

was to the mid-thoracic interspace of T5–T6. Within this single-entry point, the catheter was then advanced 12 cm within the epidural space to the estimated level of the cervicothoracic junction. A second injection was into the low lumbar level at the L5–S1 interspace and this catheter was advanced 12 cm in the epidural space to the estimated lower thoracic junction. Under CT guidance, the site of CSF leaks was confirmed (figure 3). The tip of the catheter was located at the level of C6 and was therefore pulled out by 2.5 cm at the T1 vertebral body. The second catheter was found to be coiled; it was therefore reinserted through the L3–L4 space and confirmed on CT to be at the level of L1.

The patient's sedation was then discontinued to allow for close neurological monitoring. Blood for the injection was drawn from the radial artery. He was placed in the prone position and blood was injected through the catheter at incremental doses of 2–4 mL as the catheter was being pulled back by 1 cm through the epidural space. The cervical–thoracic patch was placed first, injecting 15 mL total into the epidural space, followed by the thoracic–lumbar patch injecting 17 mL into the epidural space. At each step, the patient was asked to move his extremities and indicate if he felt any pain to monitor his neurological status. He remained comfortable, responsive and able to move all four limbs at all times. Both catheters were removed once the procedure was complete. At the end of the procedure, his neurological exam remained normal.

After the procedure, an MRI confirmed the circular and lateral spread of the blood at both injection levels without complications (figure 4). He was admitted to the pediatric ward and kept on bed rest while his pain significantly improved. He was discharged from the hospital 2 days later and remained on bed rest for 5 days at home.

Just over 2-week later, the patient's pain symptoms returned. It was decided to repeat blood patches given the suspicion of persistent CSF leaks. This time, fluoroscopy was used instead of CT, given the reduction of CSF in the epidural space.

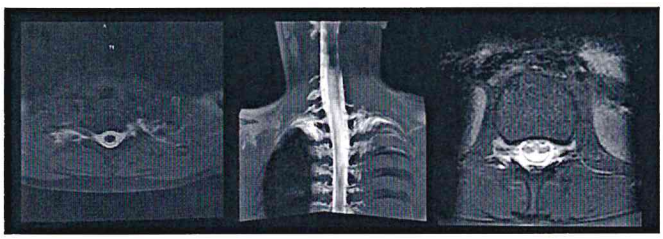

Figure 2 MRI of the spine with intrathecal gadolinium showing massive contrast extension into paravertebral soft tissues at T1 through the neural foramina. Also, multiple other areas of contrast opacifications of nerve root sleeves through the lumbar and thoracic spine.

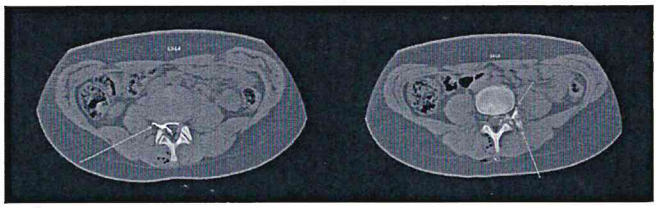

Figure 3 CT showing extravasation of contrast into paraspinal tissues at L3–L4 and L4–L5.

Cervical–thoracic and thoracolumbar epidural catheters were placed under C-arm guidance while the patient was sedated and placed in the left lateral position. The tip of the first catheter was placed into the T5–T6 interspace, and then advanced 12 cm and the second into the L5–S1 interspace and advanced 10 cm with C-arm guidance. One milliliter of epidural contrast was injected

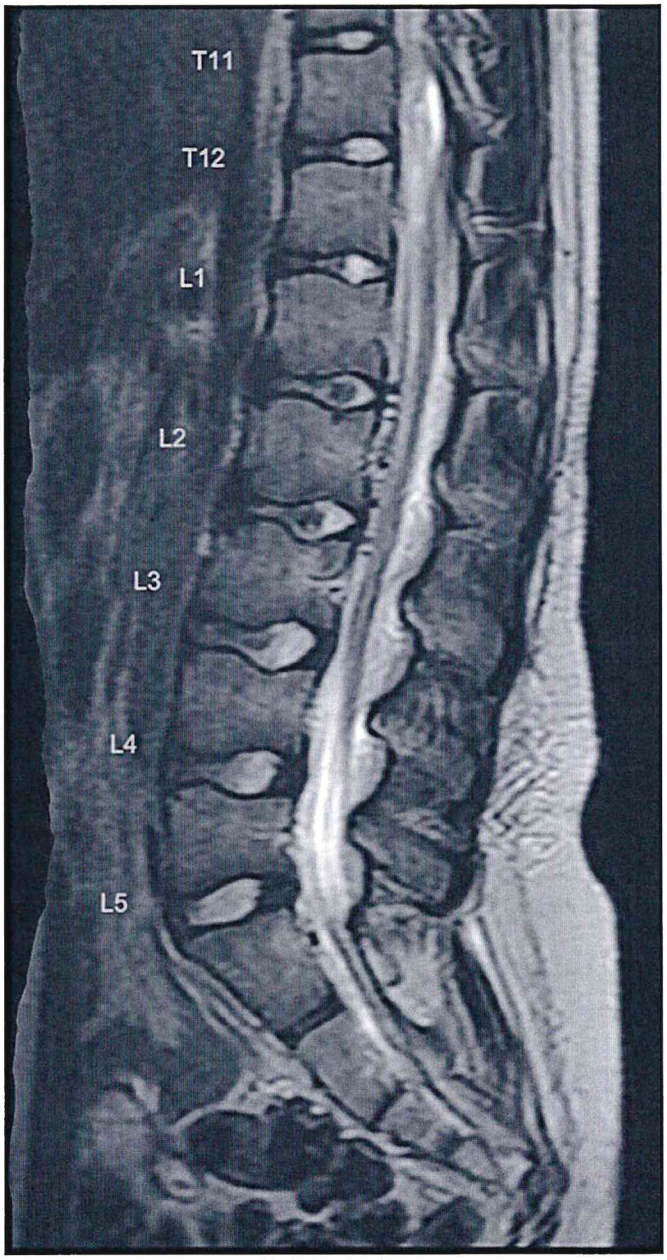

Figure 4 Postprocedural MRI showing mixed gas and blood at anterior thoracolumbar spine without associated complication.

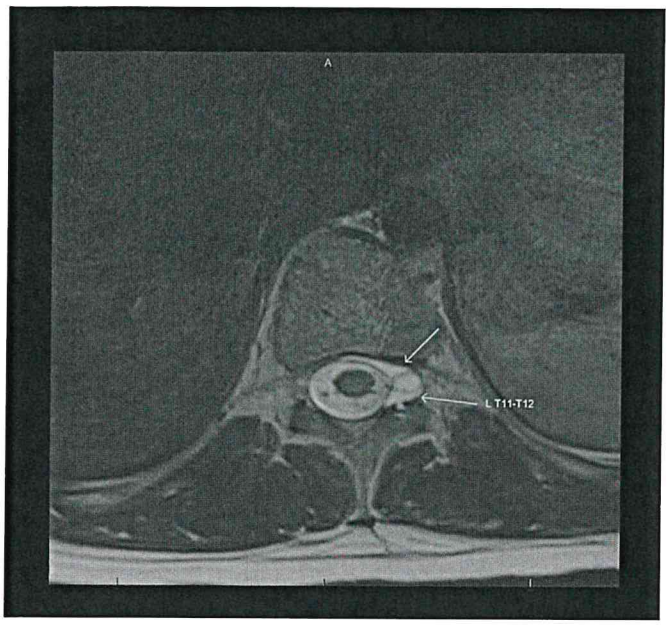

Figure 5 Repeat MRI showing persistent cerebrospinal fluid leak at the meningocele/pseudomeningocele at the T11–T12 neuroforamen.

through each catheter to confirm their position with fluoroscopy. Once confirmed, the sedation was discontinued. Using the same technique as described previously, a total of 8 mL of blood was injected into the cervicothoracic region and 16 mL into the lumbar region, both at incremental doses of 2 mL as the catheters were slowly pulled back by 1 cm at a time. The patient remained comfortable, responsive and with full neurological capacity as with the first procedure.

At follow-up 5 days after the blood patch, the patient noted being able to play and swim. Yet, he did report a mild headache without neck rigidity and that resolved with acetaminophen. At follow-up of 10 days, the patient expressed being even more active, describing 75% improvement and mentioned only a short minimal headache that resolved when supine. However, at day 11, he reported more headaches, dizziness, vomiting and pain at the neck.

This time, the patient's headaches were treated as a migraine. Yet, his symptoms did not resolve. An MRI was repeated, which demonstrated improvement/resolution of findings compatible with intracranial hypotension that were previously seen. However, within the spine, there was suspicion of persistent CSF leaks most conspicuously near the cervicothoracic junction from C7–T2–T3 and a small meningocele/pseudomeningocele at the T11–T12 neural foramen (figure 5).

Finally, a third blood patch procedure was done to target the final small leaks, taking place 3 weeks after the second procedure. This final procedure was done with similar methods, injecting 9 mL of blood through the T3–T4 interspace under C-arm guidance. The patient remained responsive, with normal sensation, strength and reflexes within all extremities. After the procedure, he remained in hospital for 4 days and was discharged without pain or other symptoms.

RESULTS

Two weeks after the last patch, the patient described a completely normal life. He went back to enjoying swimming, diving, biking and playing without any restrictions. He took vitamin B2 to prevent migraines. He described his global impression of change as resultant of treatment to be 100% improved. A month later, he sustained normal activities and had gone back to regular school. He continued to be followed regularly by ophthalmology and neurosurgery, as there was suspicion of some rebound intracranial hypertension in the context of a brief self-resolving headache. He was treated with medications to decrease intraocular pressure and continued to do well. As he expressed complete physical and social activity recovery, he was discharged from the Center for Complex pain. At this point in time, he has not sought help for pain since his last blood patch over a year ago.

DISCUSSION

This case report describes the use of lumbosacral and cervicothoracic epidural blood patches in the treatment of multiple level CSF leaks in an adolescent with a new diagnosis of Marfan syndrome. This case report suggests that catheter-guided high level blood patches have the potential to be administered to the pediatric population with spontaneous CSF leaks.

The most common level of dural CSF leaks occurs at the thoracic spine.[7] While cervical CSF leaks exist, epidural blood patches at this level are rarely performed, as these procedures have been thought to be higher risk.[4] Serious complications of blood patches include epidural abscess, paraparesis, cauda equina syndrome and radicular pain.[8] Refractory intracranial hypertension, as demonstrated by our patient's papilledema, is another potential complication that should be closely monitored for.[9] Yet, at the cervical level, spinal cord compression is the most feared complication.[10]

A literature review of 19 adults who received cervical epidural blood patches suggested the possibility that they could be performed without significant risk of serious complications in adults.[10] Three studies documented adverse events including back pain and neck pressure, though no cases of neurological or vascular complications were described.[10]

Furthermore, the use of epidural blood patches is rarely reported in children, as spontaneous intracranial hypotension is rare in the pediatric population. Schievink *et al*[11] describe the use of epidural blood patches in 23 pediatric patients. In this study, patches were performed mainly in the lumbar region with few thoracic but no cervical level patches described.[11]

Our procedures involved epidural catheters that entered at single points to administer blood patches to multiple sites of leakage. Typically, extra points of entry would have been necessary to treat our patient's many CSF leaks. This approach avoided further disruption to the epidural space and allowed for safer treatment at many levels.[5 12] This novel treatment was originally described by Ohtonari *et al*[5] in five patients in 2012, and has since been incorporated into practice.

In contrast to Ohtonari, we used two catheters to treat multiple levels. During our procedure, it was noted to be difficult to navigate the catheter when it traveled more than 4–5 levels above the insertion point. The catheters we used were thin and did not have the ability to direct their tips. The patient also had multiple dural defects, which made navigation even more challenging. Therefore, using two insertion points allowed for easier manipulation of the catheters.

While the blood patches showed benefit for our patient, he required repeated procedures to achieve continued full functionality and pain relief. In the literature, success rates have been found to vary from 30% to 90%.[3] When clinical benefit is not perceived, repeated patches can be performed at least 7 days apart.[3] Other options may include the use of percutaneous injections of fibrin or surgical CSF leak repair.[7]

While the exact mechanism of these leaks remains unknown, 20% of CSF leaks in adults have been associated with connective tissue disorders, whereas in children, over half of patients have been found to have connective tissue disorders.[11]

This case report should be extrapolated with caution. Although our patient demonstrated a successful outcome, the procedure remains high risk and there is likely a bias in publication as poor outcomes may not be reported. It is difficult to truly know how safe multilevel catheter-guided blood patches are with the limited amount of data available on the topic. In our case, we took necessary precautions in keeping our patient awake to monitor his neurological status thoroughly throughout the procedure.

Multilevel catheter-guided blood patches have the potential to be an effective treatment option for spontaneous cervicothoracic and thoracolumbar CSF leaks. However, they should be administered by professionals with extensive training in pediatric regional anesthesia and within a pediatric institution with resources available to treat the potential complications.

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Contributors LW contributed to the conception, drafting and revision of the manuscript. SA contributed to the clinical care of the patient and revision of the manuscript. ZL contributed to the selection of imaging for the manuscript, drafting and revision of the manuscript. CS-M contributed to the clinical care of the patient and the selection of imaging for the manuscript. MdCN contributed to the clinical care of the patient and revision of the manuscript. PMI contributed to the clinical care of the patient, revision of manuscript and is guarantor.

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