Repair of a traumatic subarachnoid-pleural fistula with the percutaneous injection of fibrin glue in a 2-year-old

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Subarachnoid-pleural fistulas (SPFs) are rare clinical entities that occur after severe thoracic trauma or iatrogenic injury during anterolateral approaches to the spine. Treatment of these fistulas often entails open repair of the dural defect. The authors present the case of an SPF in a 2-year-old female after a penetrating injury to the chest. The diagnosis of an SPF was suspected given the high chest tube output and was confirmed with a positive β 2-transferrin test of the chest tube fluid, as well as visualization of dural defects on MRI. The dural defects were successfully repaired with CT-guided percutaneous epidural injection of fibrin glue alone. This case represents the youngest pediatric patient with a traumatic SPF to be treated percutaneously. This technique can be safely used in pediatric patients, offers several advantages over open surgical repair, and could be considered as an alternative first-line therapy for the obliteration of SPFs.

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THE first case of a subarachnoid-pleural fistula (SPF) after a gunshot wound to the chest was reported by Milloy et al. in 1959.¹⁵ Over the last 60 years, the literature on SPF has been sparse, although its 2 most commonly reported causes are thoracic trauma and iatrogenic injury during anterolateral approaches to the spine. Documented cases of pediatric SPF are even rarer. Nonetheless, there is consensus that an anomalous connection between the subarachnoid and pleural spaces is not likely to resolve spontaneously and requires surgical intervention to repair the dural defect. Here, we present the youngest pediatric case of SPF secondary to penetrating trauma, as well as the first report of successful fistula obliteration with CT-guided percutaneous epidural injection of fibrin glue over the site of the CSF leak.

Case Report

History and Examination

A previously healthy 2-year-old female was an unfortunate bystander to a domestic dispute and suffered a through-and-through gunshot wound to her left upper chest. An entry wound was identified in her left midclavicular line approximately 3 cm medial and superior to the nipple, and an exit wound was seen in the midline of her upper thoracic region. She was intubated and a left-sided chest tube was placed for the treatment of a left hemopneumothorax and returned serosanguineous fluid. Neurological exam was significant for bilateral lower extremity plegia, a T-3 sensory level, and absent rectal tone, consistent with an American Spinal Injury Association (ASIA) Grade

ABBREVIATIONS ASIA = American Spinal Injury Association; CISS = constructive interference in steady state; EBP = epidural blood patch; NIPPV = noninvasive positive pressure ventilation; SPF = subarachnoid-pleural fistula.

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A spinal cord injury at T-3. Computed tomography scanning revealed extensive left-sided pulmonary contusions without mediastinal injury, as well as a left-sided comminuted pedicle and laminar fractures at T-3 with several osseous fragments within the lateral aspect of the spinal canal. There was no significant spinal canal stenosis (Fig. 1). Magnetic resonance imaging of the spine demonstrated intramedullary T2 hyperintensity at the level of T-2 and T-3, indicative of spinal cord contusion. Additionally, a subdural hygroma was seen at the lower thoracic and upper lumbar levels with anterior displacement of her lower thoracic cord, conus medullaris, and cauda equina (Fig. 2).

Treatment

She received a blood transfusion for anemia, was started on norepinephrine to maintain perfusion pressure, and was transferred to the pediatric intensive care unit. Her chest tube was set to suction for 24 hours.

Spinal stabilization was not deemed immediately necessary. The patient was weaned from pressors, was successfully extubated, and had her chest tube set to water seal on hospital Day 2. On hospital Day 3, she was noted to have increased serous output from her chest tube (274 ml over 24 hours), and a sample tested positive for $\beta 2$ -transferrin. She was transferred to the floor in stable condition on hospital Day 4. Between hospital Days 4 and 7, she was kept on bed rest but continued to have high serous output from her chest tube (range 50–230 ml over 24 hours). On hospital Day 8, her chest tube became dislodged and was removed. A subsequent thoracic ultrasound on Day 9 revealed a large pleural effusion. She remained asymptomatic but was noted to have enlargement of her pleural effusion on serial chest radiographs. On hospital Day 10, thoracic MRI with constructive interference in steady state (CISS) sequence was performed, demonstrating a right posterolateral dural defect at the level of T-2 and left lateral dural defect at the level of T-3 (Fig. 3). A 12-Fr chest tube was reinserted on the same day, with the return of a large amount of clear fluid. Over the next 24 hours, the chest tube put out 272 ml of serous fluid. She underwent CT-guided percutaneous epidural injection of 2 ml

FIG. 1. Axial CT scan of the thorax demonstrating a penetrating ballistic injury to the left chest with associated extensive left upper lobe contusion and chest tube placement. There are comminuted ballistic fractures of the left pedicle and lamina of T-3 with several small osseous fragments protruding into the left aspect of the spinal canal.

FIG. 2. **Upper:** Axial MRI T2 STIR sequence obtained at T-3, demonstrating intramedullary T2 hyperintensity suggestive of a spinal cord contusion (green arrow). **Lower:** Sagittal T2 STIR sequence of the lower thoracic and lumbar spine depicting a large subdural fluid collection with anterior displacement of the lower thoracic spinal cord and conus. Imaging characteristics on T1- (not shown) and T2-weighted imaging were consistent with CSF. The clinical significance of this hygroma was unknown. Lumbar puncture was offered for drainage, but the family declined this intervention. Figure is available in color online only.

of fibrin glue (Tisseel, Baxter International) under the T-2 lamina via a 20-gauge, 9-cm spinal needle, and this provided sufficient coverage of both dural defects at T-2 and T-3 (Fig. 4).

Posttreatment

Postprocedure, her chest tube output was 0 and 8 ml on hospital Days 11 and 12, respectively. She remained asymptomatic, and serial radiographs were stable without

reaccumulation of pleural fluid. Her thoracostomy tube was removed on Day 12, and she was transferred to inpatient rehabilitation on hospital Day 13. She completed a 6-week inpatient rehabilitation course without any pulmonary or neurological complications. At the time of discharge from rehabilitation, her neurological status remained an ASIA Grade A. She was seen as an outpatient at 9 weeks postprocedure with an unchanged neurological exam. Chest radiograph did not show recurrence of the pleural effusion and suggested closure of the SPF. Additionally, there was no evidence of thoracic kyphosis as a result of the left T-3 pedicle and laminar fractures, and she will continue to be managed conservatively.

Discussion

Subarachnoid-pleural fistulas are unique clinical entities that can develop with disruption of both the parietal pleura and the thoracic dura.⁸ The 2 most common scenarios in which they are encountered are severe thoracic trauma or iatrogenic injury during anterolateral approaches to the thoracic spine. Given the rarity of these fistulas, their diagnosis requires a high index of suspicion from clinicians. Symptoms of SPF stem from hydrothorax (dyspnea, hypoxia, respiratory distress) or intracranial hypotension (postural headache, altered mental status, nausea and vomiting, diplopia, auditory abnormalities, and even chronic cerebellar hemorrhage²²). Additional clinical signs can include recurrent pleural effusion, transudative effusion, abnormally high thoracostomy tube output, and imaging findings consistent with intracranial hypotension. Pleural fluid positive for $\beta 2$ -transferrin increases diagnostic confidence as this test reaches 100% sensitivity and 95% specificity for the presence of CSF.²³ While CT myelography is the imaging modality of choice to visualize SPFs, MRI with thin-section T2-weighted sequences (such as CISS) and contrast-enhanced cisternography have become reasonable alternatives as they have been reported to have 76% and 100% sensitivity, respectively, in detecting CSF leaks.²

Optimal treatment for SPF remains controversial; however, there is consensus that such fistulas are unlikely to resolve spontaneously.⁷ The intrathoracic pressure ranges from -5 to -7.5 cm H₂O, whereas the intradural pressure is between $+10$ to $+15$ cm H₂O; thus, this pressure gradient plays a critical role in maintaining fistula patency.²⁰ Surgical intervention is often required to eliminate this pressure differential and entails direct repair of pleural and dural defects, either with open surgery or in a minimally invasive fashion.^{10,20} This surgical intervention most commonly requires the use of a dural substitute or sealant (Tisseel, DuraSeal) and can be augmented with a vascularized graft.^{7,8,19} Other authors have reported success with secondary occlusion of fistulas by using an epidural blood patch (EBP)¹ or synthetic material such as DuraSeal, Tisseel, and even Onyx;^{4,6,11} however, all of these successes occurred in adult patients. Kurata et al. reported that 5–14 days of continuous noninvasive positive pressure ventilation (NIPPV) can be used as an adjunct to chest tube drainage, with the goal of increasing intrathoracic pressure and reducing the pressure gradient between the

FIG. 3. Reformatted axial thoracic CISS MRI sequences showing areas of CSF leakage (arrows) from a right posterolateral dural defect at the level of T-2 (upper) and a left lateral dural defect at T-3 (lower). Figure is available in color online only.

FIG. 4. Computed tomography–guided percutaneous epidural injection of 2 ml of fibrin glue (Tisseel) under the lamina of T-2 (A). The fibrin glue can be visualized as a hyperdensity in the dorsal epidural space spanning the dural defects at T-2 (B) and T-3 (C). This technique was successful in occluding the SPF.

pleural and subarachnoid spaces.¹² In the pediatric population, continuous NIPPV may be a therapeutic challenge as patient compliance can be difficult and places the patient at risk for side effects of continuous NIPPV (skin breakdown, gastric distention, dry mouth). Furthermore, there is only 1 documented pediatric case of PPV as therapy for SPF; however, the 9-month-old patient in that case required intubation and invasive PPV by endotracheal tube for nearly 2 weeks.²⁴

There are few reports of SPF in the pediatric neurosurgical literature, with sporadic single cases that mainly occur after thoracic tumor resection and trauma.^{9,10,18,24} Common to these cases were requirements for open surgery and repair of the dural defect. Postoperatively, patients in these cases remained in the hospital for 7–14 days for observation and chest tube management. Liang et al. reviewed 40 cases of traumatic SPF occurring between 1959 to 2005 and reported 18 cases of SPF in pediatric patients.¹³ Of these, 7 cases (age range 3–18 years) were secondary to penetrating trauma, and all patients in these cases required intervention in the form of a chest tube or open repair. We were unable to find any cases of spontaneous resolution of SPF in the pediatric population. Reports discussed above are summarized in Table 1.

Our patient represents the youngest reported case of penetrating traumatic SPF and the first report of percutaneous epidural injection of fibrin glue alone for the successful treatment of SPF in a pediatric patient. The abrupt reduction in chest tube output and the serial images that showed no reaccumulation of her pleural effusion allowed for the removal of her chest tube on postoperative Day 2. In this case, we believe the percutaneous epidural technique was advantageous as it obviated the need for open surgery and reduced the chest tube duration and postoperative hospital stay, enabling timely transfer to inpatient rehabilitation.

We do not believe the bullet traversed the spinal cord and venture that dural lacerations were probably attributable to impact from osseous fragments against the thecal sac or secondary shockwave from the blast injury. Interestingly, the dural defect at T-2 was located on the right posterolateral aspect of the cord, at a distance from the bullet trajectory and left pedicle fractures. Given that our patient had multiple, isolated dural defects, we speculate that her dura was probably very friable, even possibly

“shredded,” with smaller defects impossible to visualize by the CISS MRI sequence. Therefore, we do not believe that shredded dura with multiple small defects would be a contraindication for CT-guided percutaneous treatment for SPF—in fact, it may be an ideal first-line treatment for multiple dural defects that would be difficult to access via a single surgical incision.

Our patient presented with a complete spinal cord injury and did not display evidence of spinal canal stenosis; therefore, mass effect from percutaneous fibrin glue injection was less concerning, especially when a small volume (2 ml) of fibrin glue was injected to cover 2 spinal levels. We can only speculate that large volumes may increase the risk. In cases of incomplete spinal cord injury or in those with critical canal stenosis, the percutaneous addition of material into the epidural space may place the patient at risk for neurological decline. Close postprocedure monitoring would be required, and surgical decompression may be necessary if decompensation occurs. Certainly, if a patient had an unstable middle column injury with retropulsed fragments resulting in canal stenosis and an incomplete injury, surgical intervention rather than the use of percutaneous techniques would be more appropriate treatment.

Our decision to use fibrin glue over an alternative material, such as an EBP, partially stems from surgeon preference, and we do not have extensive experience with EBP. To our knowledge, no studies have compared targeted EBP and fibrin glue to suggest the superiority of one over the other. Despite representing the gold standard for post-lumbar puncture headaches, EBP has a widely variable success rate and a failure rate up to 44% and often requires multiple treatments when used for spontaneous intracranial hypotension.^{4,21} The largest case series involving percutaneous epidural fibrin glue injection involved 23 patients and reported a 65% success rate.¹⁶

In pediatrics, EBP requires weight-based dosing, and typically between 0.3 and 0.4 ml/kg is used and larger doses (up to 0.8 ml/kg) have been reported for repeat procedures.^{3,5} Our patient’s weight was approximately 15 kg, and thus between 4.5 and 6 ml of blood would have been used. This is roughly 2–3 times the volume of fibrin glue that was injected. As discussed above, whether a larger volume of material injected into the epidural space would be problematic is subject to speculation.

TABLE 1. Summary of reported cases of pediatric SPFs

Authors & Year	Patient Age	Sex	Mechanism of SPF	Treatment
Patel et al., 2005	18 yrs	M	Blunt trauma (snowboarding accident)	Thoracotomy, DuraGen collagen matrix (Integra), fibrin glue, extra-abdominal omental patch
Valla, 2007	9 mos	M	Rhabdomyosarcoma of the chest wall resection	Intubation, PPV, chest tube
Liang et al., 2008*	All patients: 1–18 yrs; blunt trauma patients: 1–15 yrs; penetrating trauma patients: 3–18 yrs	All patients: 11 M, 7 F; blunt trauma patients: 7 M, 4 F; penetrating trauma patients: 4 M, 3 F	Total no. of SPFs: 18; SPFs due to blunt trauma (MVC, autoped): 11; SPFs due to penetrating trauma (GSW): 7	All patients: chest tube 6, surgery 12; blunt trauma patients: chest tube 2, surgery 9; penetrating trauma patients: chest tube 4, surgery 3
Kitagawa et al., 2010	4 yrs	F	Intrathoracic ganglioneuroma resection	Video-assisted thoracoscopic repair
Karabayir et al., 2012	2 yrs	M	Ganglioneuroblastoma resection	Hemilaminectomy, autologous fascia graft, fibrin glue

autoped = automobile hit pedestrian; GSW = gunshot wound; MVC = motor vehicle collision; PPV = positive pressure ventilation.

* Retrospective review of 40 cases of traumatic SPFs between 1959 and 2005. Of the 40 cases, 18 were reported in pediatric patients.

Given the increasing popularity of CT-guided epidural patching for CSF leaks, we agree with previous authors that it represents a viable alternative to open surgery.^{1,6,17} While this technique may not be applicable for all patients, future studies may identify factors that better stratify patients toward this option. For example, Mihlon et al. demonstrated that CT-guided percutaneous EBP is more likely to be successful with a durotomy size less than 5 mm.¹⁴

Conclusions

Our case represents the youngest pediatric patient with a penetrating traumatic SPF successfully treated with CT-guided percutaneous epidural injection of fibrin glue alone. Our case demonstrates that the CT-guided percutaneous epidural technique can be safely used in pediatric patients and in patients with multiple dural defects and can offer patients a shorter hospital stay if fistula closure is achieved. Factors determining the success rate of the percutaneous obliteration of SPFs require investigation.

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Disclosure

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

Conception and design: Chern. Acquisition of data: Chu. Analysis and interpretation of data: Chu. Drafting the article: Chu. Critically revising the article: Chu, Miller, Chern. Reviewed submitted version of manuscript: all authors. Study supervision: Chern.

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