Percutaneous treatment of subarachnoid-pleural fistula with Onyx

Case report

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Subarachnoid-pleural fistula is a well-described complication after anterior surgery for thoracic disc herniation, but is difficult to treat by means of traditional chest and lumbar drains due to interference by positive ventilation pressures that may keep the fistula open and prevent proper closure. Current treatment strategies include surgical repair, which is technically challenging, and noninvasive positive pressure ventilation, which can take several weeks to be effective. In this report, the authors describe a novel treatment for subarachnoid-pleural fistula using percutaneous obliteration with Onyx.

Surgery for removal of a T7–8 disc herniation associated with ossification of the posterior longitudinal ligament was performed in a 56-year-old woman via an anterior transthoracic transpleural approach. Ten days after surgery, she presented with diplopia due to a subarachnoid-pleural fistula that was confirmed by CT myelography. Percutaneous injection of Onyx was performed under local anesthesia. Postprocedure CT showed complete obliteration of the fistula with no adverse events. A CT scan obtained 1 month later showed complete resolution of the pleural effusion. Neurological examination at 3 months postsurgery was normal. Clinical and radiological follow-up at 1 year showed complete recovery and no sign of fistula recurrence. Percutaneous treatment for subarachnoid-pleural fistula is an easy, safe, and effective strategy and can therefore be proposed as a first-line option for this challenging complication.

Key Words • subarachnoid-pleural fistula • embolization • thoracic disc herniation • Onyx • percutaneous • trauma

S ubarachnoid-pleural fistula is a rare type of CSF fistula that has been reported following traumatic injury,11,18 thoracotomy for tumor resection,9,16 and thoracic disc herniation.14,15 Unlike other types of CSF fistula that can resolve spontaneously or with lumbar drainage, treatment of subarachnoid-pleural fistula is usually difficult because of the negative pressure in the pleural cavity.18 We report a case of subarachnoid-pleural fistula occurring after anterolateral thoracotomy for T7–8 disc herniation, treated by percutaneous obliteration. To the best of our knowledge, this is the first report of this type of management for this challenging complication.

Case Report

History and Presentation. A 56-year-old woman, with no significant medical history, presented with lower-limb pain that progressively increased for 1 year. Physical examination revealed pyramidal syndrome with right Babinski sign, and the patient reported tenderness on palpation of the back. No motor or sensory deficit was detected. Magnetic resonance imaging and a CT scan showed a calcified T7–8 disc herniation with considerable spinal cord compression and ossification of the posterior longitudinal ligament (Fig. 1). Somatosensory evoked potentials confirmed thoracic myelopathy.

Operation and Postoperative Course. Surgical decompression was performed via a right transthoracic transpleural approach, using a high-speed drill for discectomy and thinning of the ossified portion of the ligament. A dural tear with CSF leak was noted during drilling of the strongly adherent ligament. The tear was repaired using autologous muscle graft packed with fibrin glue, and dural substitute (TachoSil; Takeda Pharmaceuticals International) was applied onto the dura. No CSF leak was observed at the end of the procedure and no chest tube was placed.

No immediate postoperative neurological or respiratory complications were observed; a mild pleural effusion was noted on chest radiography on postoperative Day 1, but remained stable over the following days. However, on Day 10, the patient complained of headache and diplopia. Neurological examination revealed a palsy of cranial nerve VI, which was suggestive of intracranial hypoten-
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Fig. 1. Preoperative imaging features. Sagittal T2-weighted MR image (A) and sagittal and axial (B and C) CT reconstructions showing calcified T7–8 disc herniation with spinal cord compression.

sion. Computed tomography examination of the brain showed no intracranial complication. A chest CT scan followed by CT myelography confirmed the presence of a subarachnoid-pleural fistula (Fig. 2A and B).

Fistula Treatment and Follow-Up. We decided to perform percutaneous obliteration of the fistula under local anesthesia. The fistula was located without stereotactic guidance under biplane fluoroscopy after initial identification at the right posteroinferior part of T-7 on CT myelography. A 22-gauge needle was positioned just above the T-8 pedicle via a right costotransverse approach, and 0.5 ml of Onyx 34 (ev3) was progressively injected until a shape similar to the one seen on the CT myelogram was obtained, suggesting complete filling of the fistula (Fig. 2C). Immediate postprocedure CT scanning confirmed complete occlusion of the subarachnoid-pleural fistula (Fig. 2D and E). The foramen was not obliterated (Fig. 2C), and the patient did not complain of any radicular pain.

No neurological complication due to the procedure was observed after treatment. Symptoms of intracranial hypotension resolved progressively, and the patient was discharged 8 days later. Pleural effusion had almost completely resolved by 1 month postprocedure (Fig. 3B), and CT scanning performed at 1 year showed the usual fibrous pleural scar (Fig. 3C) and no sign of fistula recurrence. A lateral radiograph obtained 1 year after the procedure (Fig. 2F) showed no change in the Onyx cast, thereby demonstrating that Onyx holds up well even to pulsatile CSF. Neurological examination 3 months after surgery was normal. At the 1-year follow-up, the patient showed no sign of oculomotor dysfunction, in particular no diplopia, and no headache. She resumed working as a housekeeper 6 months after surgery, although she still complained of moderate thoracic pain when exercising.

Discussion

Although rare, subarachnoid-pleural fistula has been well described following surgery for either tumor resection,\textsuperscript{2,4,9,16} discectomy,\textsuperscript{14,15} or vertebral fracture.\textsuperscript{10,18} Clinical presentation typically includes respiratory signs due to pleural effusion, signs of intracranial hypotension such as orthostatic headache and oculomotor paralysis, and signs of pneumocephalus.\textsuperscript{2} The fistula can be demonstrated on CT myelography,\textsuperscript{6} MRI, or myeloscintigraphy.

Management of subarachnoid-pleural fistula is challenging, as the negative intrapleural pressure occurring at each inspiration constantly draws CSF into the pleural cavity, thereby promoting persistence of the fistula. A chest tube, although often necessary to drain the pleural collection, increases the negative pleural pressure and can therefore worsen the situation.\textsuperscript{18} Successful treatment by lumbar CSF drainage alone has been reported\textsuperscript{9} but is ineffective in most cases,\textsuperscript{18} particularly when a chest drain has been placed.

Recently, 2 reports\textsuperscript{10,18} of subarachnoid-pleural fistula treated by noninvasive positive-pressure ventilation in combination with chest drainage for 5 to 14 days demonstrated that this type of alternative conservative management is also feasible. However, the consequences of subarachnoid-pleural fistula, such as intracranial bleeding\textsuperscript{15} or massive pneumocephalus,\textsuperscript{2} can be life threatening and may require immediate treatment of the fistula. Successful surgical obliteration, either via video-assisted thoracoscopic repair,\textsuperscript{9} cadaveric dural allograft,\textsuperscript{15} or pericardial fat pad,\textsuperscript{16} has also been reported, but these methods are more invasive.

Onyx is an ethylene vinyl alcohol copolymer that was approved by the European Union in 1999 and by the US FDA in July 2005 as an embolic agent for brain arteriovenous malformations. It has been widely used for endovascular treatment of these lesions for more than 20 years, and has also been used in various other conditions such as brain aneurysm\textsuperscript{17} and dural arteriovenous fistula (both cranial\textsuperscript{3} and spinal\textsuperscript{13}), and also for preoperative embolization of cranial and spinal tumors.\textsuperscript{3} Unlike other liquid embolic agents, such as N-butyl 2-cyanoacrylate or polyvinyl alcohol foam, Onyx only precipitates after diffusion of its solvent carrier to form a spongy material.\textsuperscript{12} In the case presented here, the particular conformation of Onyx allowed complete filling of the fistula. The main safety concerns are the possibility of diffusion of Onyx into the spinal canal, potentially causing cord compression, and direct toxicity to nervous tissue by dimethyl sulfoxide.\textsuperscript{1} Slow injection of Onyx over several minutes facilitates
control of its distribution into the fistula, and the injection can also be briefly suspended to prevent unwanted diffusion of the material. In this case we used Onyx 34, rather than the less concentrated Onyx 18, based on the assumption that a higher concentration would decrease the risk of leakage into the spinal canal. If difficulties in positioning the injection needle are encountered, such as might be the case in smaller fistulas, stereotactic guidance can be used and contrast injection through the needle can be performed to ensure proper needle placement.

Fig. 2. Axial CT myelogram (A) and sagittal CT reconstruction (B) obtained on postsurgical Day 10 showing contrast leaking into the pleural cavity (open arrows). Enhancement of pleural fluid confirms the subarachnoid-pleural fistula. Lateral radiograph (C) obtained during the procedure demonstrating the needle positioned in the area of leakage (black arrow) during Onyx injection (double arrows indicate cast of Onyx). Axial (D) and sagittal (E) postoperative CT reconstructions demonstrating complete filling of the fistula. Lateral control radiograph (F) obtained at 1-year follow-up showing no significant change in the Onyx cast.

Fig. 3. A: Postoperative CT myelogram performed on Day 10 showing contrast extravasation (open arrow) into the pleural cavity with enhancement of pleural fluid, confirming the subarachnoid-pleural fistula. B: Computed tomography scan performed 1 week after Onyx injection showing reduction of pleural effusion, which almost completely disappeared at 1 month. C and D: Computed tomography scans obtained at 1-year follow-up showing complete resolution of pleural effusion (C) and the expected pleural thickening (D). There was no significant change in the appearance of the Onyx plug between the 1-month and 1-year scans.
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Conclusions

Subarachnoid-pleural fistula is a well-described complication of thoracic spine surgery and can be difficult to manage. We describe an original and easy procedure using an embolic agent to percutaneously obliterate the fistula, leading to complete clinical and radiological resolution. This report shows that this type of percutaneous treatment is an effective and minimally invasive technique for solving an old problem.

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 to the study and manuscript preparation include the following. Conception and design: Saliou, Knafo. Acquisition of data: Saliou, Parker. Analysis and interpretation of data: Knafo. Drafting the article: Knafo. 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: Saliou. Study supervision: Saliou.

References


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