Case Report

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Positive-pressure ventilation attenuates subarachnoid-pleural fistula after thoracic spinal surgery: A report of two cases

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Abstract

Background: Dural tear and cerebrospinal fluid (CSF) leakage is known to be a complication of anterior thoracic spine surgery. If dural tear occurs on the ventral side of dura in combination with a pleural injury, it potentially becomes a subarachnoid-pleural fistula. The pressure gradient permits continuous flow of CSF from the subarachnoid space into the cavum thorax, resulting in an intractable subarachnoid-pleural fistula. We report two cases of successfully treated subarachnoid-pleural fistula using noninvasive positive-pressure ventilation (NPPV). **Methods:** Two patients, a 52-year-old man and a 54-year-old woman, underwent anterior thoracic spine surgery to treat thoracic myelopathy caused by spinal tumor and ossification of the posterior longitudinal ligament. During surgery, dural tear and CSF leakage to the cavum thorax due to perforation of the dura was observed. We treated with polyglycolic acid sheet (Neovel®) in combination with fibrin glue; a suction drainage tube was placed at the subfascial level and the wound was drained with negative pressure. However, after removal of the drainage tube, subarachnoid-pleural fistula was proven. We applied NPPV to the patients. **Results:** We used the application of NPPV for 2 weeks in the first patient and 1 week in the second patient. In both of them, subarachnoid-pleural fistula was attenuated without apparent adverse events. **Conclusion:** NPPV is noninvasive and potentially useful therapy to attenuate subarachnoid-pleural fistula after thoracic spinal surgery.

Keywords

cavum thorax, noninvasive positive-pressure ventilation, surgical complication, thoracic spine

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Introduction

Cerebrospinal fluid (CSF) leakage is a postoperative complication of spinal surgery with a reported incidence of 1.4%.[1] Appropriate treatment is important for a persistent CSF leakage from a fistula, which might cause meningeal irritation including headache, nausea and vomiting, cerebral hemorrhage,[2,3] meningitis, and the formation of a pseudocyst. If visible dural damage and CSF leakage occur during spinal surgery, it is clear that a primary suture must be made as the first choice.[4] Where the location of a dural tear is on the ventral side of the dural tube, which cannot be treated with a primary suture, or where postoperative CSF leakage is without a visible dural tear, alternative treatment must be considered. Especially in spinal tumor and ossification of the posterior longitudinal ligament (OPLL) cases, there is

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frequently tight adhesion between ventral dura mater and posterior wall of vertebral body, resulting in higher incidence of ventral dural tear during excision of tumors or ossification foci.

One of the alternative intraoperative dural repair methods is the application of a polyglycolic acid sheet (Neovel®) with fibrin glue, and suction drainage with slight negative pressure.[5,6] Although Neovel patch with fibrin glue cannot be compared with the direct repair, it can be helped to perform such suboptimal procedure in severe circumstances of ventral dural injury during anterior thoracic surgery for spinal tumors and OPLL. Well-known postoperative treatment for CSF leakage is spinal drainage using a subarachnoid catheter. Otani et al. reported favorable outcomes for spinal drainage of CSF leakage.[7] Shapiro and Scully reported a favorable outcome for 94% (101 of 107) of patients treated with spinal drainage for a CSF fistula. By contrast, spinal drainage might cause severe complications including meningitis (2%), overdrainage with neurological decline (3%), and occlusion requiring replacement of the drainage tube (9%) according to its substantial invasiveness.[2] Therefore, effective treatment for CSF fistulas without excessive invasiveness is needed.

Noninvasive positive pressure ventilation (NPPV) is ventilatory support applied without the use of an endotracheal tube, and included continuous positive airway pressure with or without inspiratory pressure support. It has been increasingly used for the treatment of acute and chronic respiratory failure, obesity hypoventilation syndrome, and cardiogenic pulmonary edema.[8] We hypothesized that NPPV is useful for treatment for postoperative subarachnoid-pleural fistula via the increase of hydrostatic pressure in the cavum thorax.

Here we report two cases of successfully treated CSF leakage to the cavum thorax as a result of an anterior surgical procedure for the thoracic spine using NPPV, which is a candidate for an effective treatment modality for CSF fistula to the cavum thorax.

Case presentation

Case 1

A 52-year-old man was referred to our institute with spinal tumor at the T6 vertebral level. He complained of numbness on his bilateral lower extremities and spastic incomplete paraparesis. Computed tomography (CT) revealed destructive change at the T6 vertebra, pedicle, and lamina. Magnetic resonance imaging showed a mass lesion protruding to the spinal canal and compressing the spinal cord (Figure 1).

The patient underwent total en bloc spondylectomy at the T6 vertebral level. During surgery, a dural tear was observed at the ventral side of the dural tube with bilateral pleural injury. Dural injury and pleural injury were treated with polyglycolic acid sheet (Neovel®) in combination with fibrin glue; a suction drainage tube was placed at the subfascial level and the wound was drained with slight negative pressure. After removal of the wound drainage tube the day after surgery, the patient complained of dyspnea and blood oxygen saturation level showed a marked decrease. An X-ray image and CT showed apparent fluid correction in the cavum thorax (Figure 2). We consulted a thoracic surgeon to puncture the cavum thorax 3 days after surgery, resulting in serous fluid collection. Therefore, CSF leakage to the cavum thorax was proven. Fluid collection in the cavum thorax immediately relapsed, and repeated puncture resulted in 700 mL of fluid collection. After the second puncture, we applied NPPV to the patient. Spontaneous mode, which adds positive pressure in coordination with spontaneous respiration, and spontaneous/timed (S/T) mode, which can assist ventilation for apnea in addition to the S mode, were applied until dyspnea disappeared 2 weeks after surgery. The patient was discharged without any dyspnea 1 month after surgery (Figure 3).

Figure 1. (a) Sagittal CT and (b) T2-weighted MRI of case 1 demonstrating a mass lesion protruding to the spinal canal and compressing the spinal cord at T6. CT: computed tomography; MRI: magnetic resonance imaging.

Case 2

A 54-year-old woman was referred to our institute with thoracic OPLL at the level of T2–T4. She showed spastic paraparesis and she was not walking on admission. A myelogram and CT revealed marked spinal cord compression by the ossification foci at the level of T3–4. Anterior OPLL

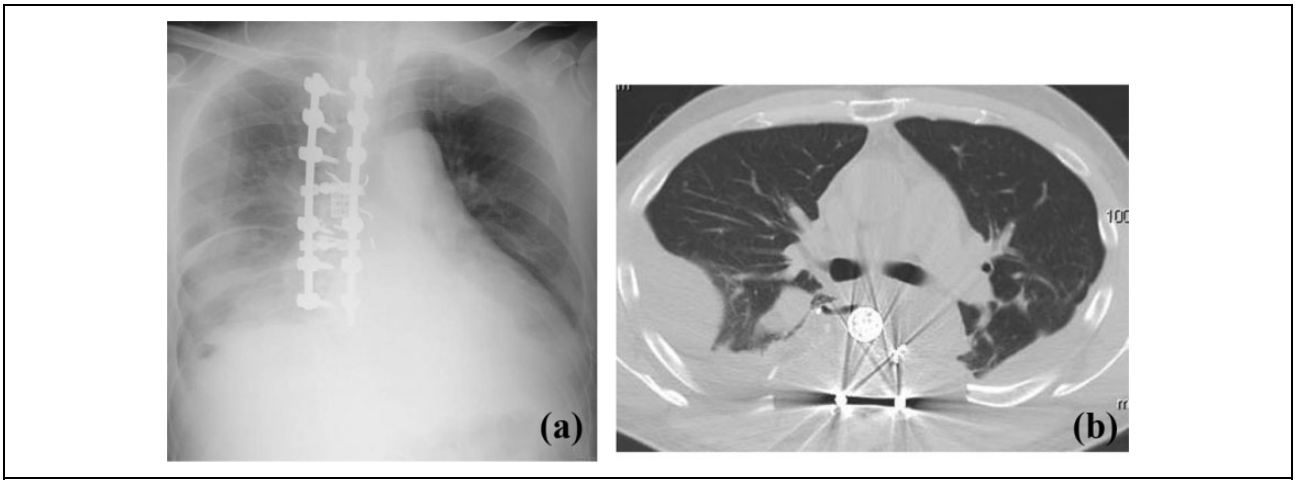

Figure 2. Postoperative plain (a) chest X-ray image and (b) CT of case 1 on day 2 revealed apparent fluid correction in the cavum thorax. CT: computed tomography.

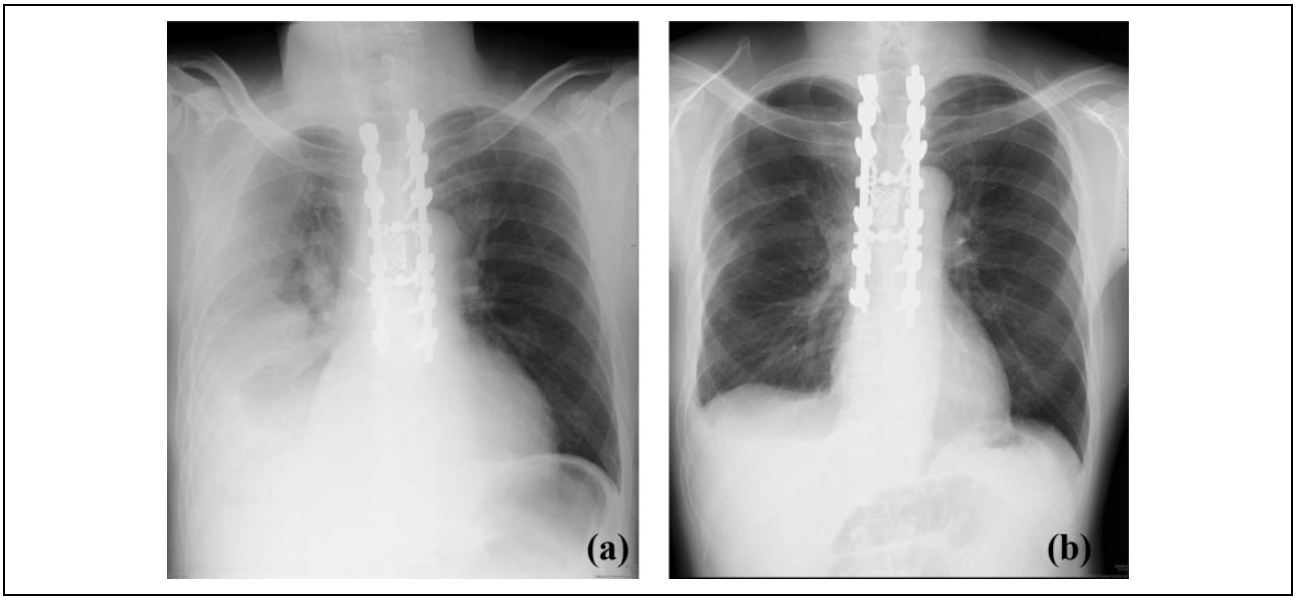

Figure 3. Plain chest X-ray image of case 1 (a) pre-NPPV treatment and (b) post-NPPV treatment showed fluid correction in the cavum thorax attenuated by NPPV treatment. NPPV: noninvasive positive-pressure ventilation.

excision followed by a bone graft via an extrapleural approach was performed. Ossification foci were successfully excised and autologous fibula was placed as a strut graft. During separation of the ossification foci from the dura mater, CSF leakage from dural defect was observed. Pleural injury at the approach site was also visible. Dural injury and pleural injury were treated with polyglycolic acid sheet (Neovel®) in combination with fibrin glue; a suction drainage tube was placed at the subfascial level and the wound was drained with negative pressure. After removal of the wound drainage tube the day after surgery, apparent fluid correction in cavum thorax was observed with X-ray and CT scan. Ten days after surgery, we punctured the thorax resulted in 700 mL of fluid collection, and applied NPPV to the patient for 1 week. After the application of NPPV, dyspnea was gradually attenuated.

Discussion

Dural tear and CSF leakage during anterior surgical procedures in thoracic spine is often difficult to treat because of its specific location and surrounding anatomical structures. Especially pleura, which is adjacent to the thoracic spine, can be easily damaged during anterior surgical procedures for the thoracic spine. If a dural tear occurs on the ventral side of dura in combination with a pleural injury, it potentially becomes a CSF fistula to cavum thorax. The cavum thorax has negative hydrostatic pressure, while the pressure

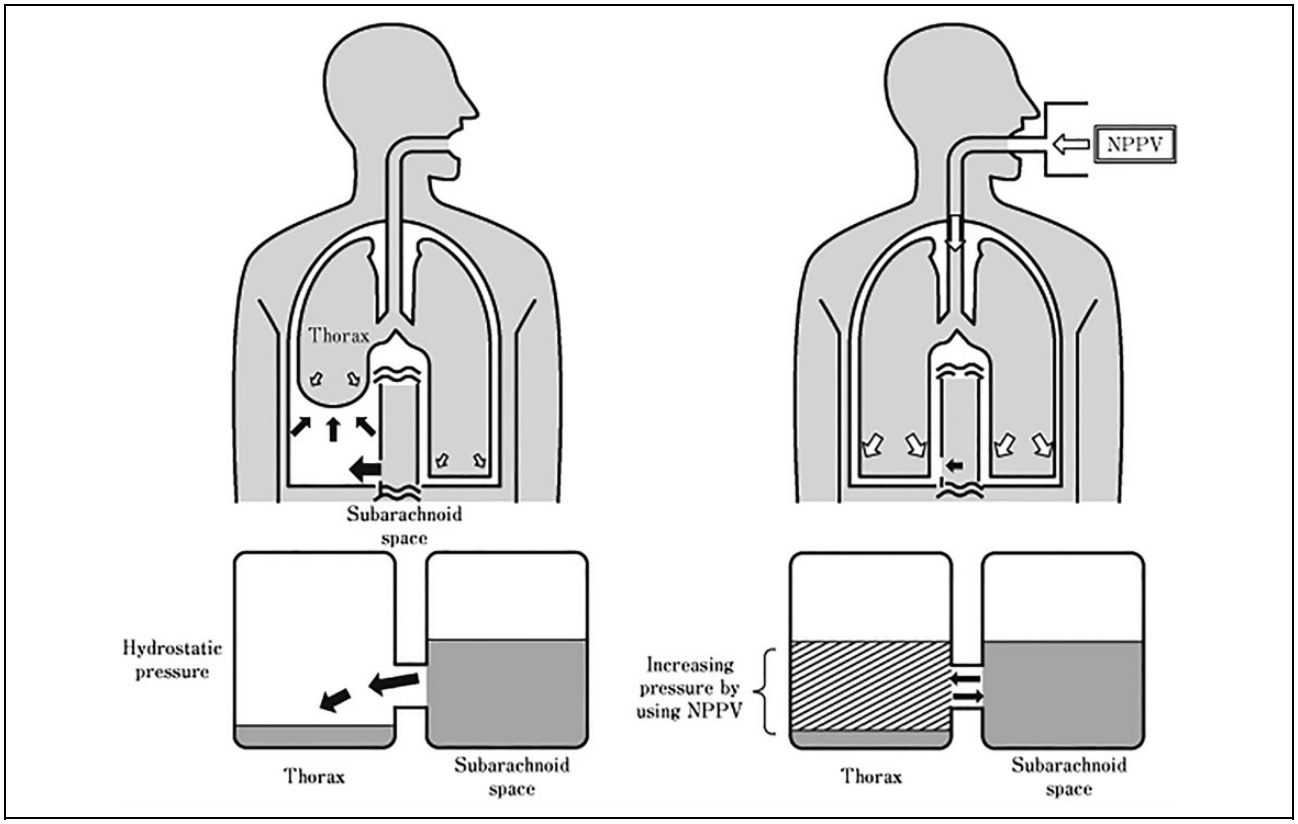

Figure 4. Mechanism of CSF fistula to the cavum thorax. The hydrostatic pressure gradient permits continuous flow of CSF from the subarachnoid space into the cavum thorax, resulting in an intractable CSF fistula to cavum thorax (left). NPPV can increase the hydrostatic pressure in the cavum thorax resulting in attenuation of the hydrostatic pressure gradient between the subarachnoid space and the cavum thorax (right). CSF: cerebrospinal fluid; NPPV: noninvasive positive-pressure ventilation.

in the subarachnoid space is positive.[7,9] This hydrostatic pressure gradient permits continuous flow of CSF from the subarachnoid space into the cavum thorax, resulting in an intractable CSF fistula to cavum thorax. Even in the present two cases, a continuous CSF fistula to the cavum thorax occurred, although dural repair with a polyglycolic acid sheet (Neovel®) in combination with fibrin glue was performed in both cases. Because of the mechanism of CSF fistula to the cavum thorax, which is mainly caused by a hydrostatic pressure gradient between the subarachnoid space and cavum thorax, we applied NPPV (Figure 4). NPPV can increase the hydrostatic pressure in the cavum thorax by positive end-expiratory pressure,[10] possibly resulting in attenuation of the hydrostatic pressure gradient between the subarachnoid space and the cavum thorax. In the present two cases, leakage of CSF from the fistula to the cavum thorax was promptly ameliorated after the application of NPPV without apparent adverse events. The true effectiveness of NPPV for CSF leakage to the thorax has not been established because there are only three case reports of NPPV for CSF leakage to the thorax.[9,11,12] As cons of this treatment, NPPV has risks in patients with disturbed consciousness, and those with an uncontrolled pneumothorax.[9,13] Therefore, careful investigation before application of NPPV is mandatory and close observation of the patient is essential.

Limitations

To date, we have no standard protocol (standard pressure and duration, etc.) on NPPV for CSF leakage to the thorax. Therefore, in case 2, we tried to stop the positive pressure 1 week after the initiation of NPPV. We check the SpO_2 and chest X-ray for several days after stopping the positive pressure. Fortunately, CSF leakage did not deteriorate. Even after the experience of the present cases, we have to grope our way because we have no standard protocol of NPPV for CSF leakage to the thorax.

Conclusions

NPPV is a potentially useful therapy to attenuate CSF leakage into the cavum thorax after thoracic spinal surgery. Therefore, NPPV therapy is a treatment option for postoperative CSF leakage into the cavum thorax.

Declaration of conflicting interests

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