Subdural fluid collection rather than meningitis contributes to hydrocephalus after cervical laminoplasty: A case report

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Abstract

BACKGROUND
Hydrocephalus following dural tear after spinal surgery is rare. Although a few cases of obstructive hydrocephalus caused by subdural fluid collection and communicating hydrocephalus associated with meningitis have been reported, the mechanism remains uncertain. Herein we describe a patient complicated with hydrocephalus after cervical laminoplasty in whom subdural fluid collection in the cervical spine and posterior cranial fossa rather than chronic meningitis was the main mechanism.

CASE SUMMARY
A 45-year-old man underwent cervical laminoplasty for cervical spondylotic myelopathy at a local hospital. Ten days postoperatively, a high fever occurred and magnetic resonance imaging (MRI) showed cerebrospinal fluid (CSF) leakage. Pseudomeningocele liquid test showed high levels of protein and white blood cell (WBC) count with negative bacterial culture. The patient was treated with short-term intravenous antibiotic and discharged with normal body temperature. The patient was uneventful during the first 8 months follow-up although repeated MRI showed persistent pseudomeningocele. At the 9th month postoperatively, the patient gradually presented with dizziness and headache accompanied by recurrent weakness of his left arm. Imaging examinations demonstrated hydrocephalus and a cystic lesion around the cervical spinal cord. CSF test from lumbar puncture indicated chronic meningitis. MRI on 1 d after pseudomeningocele drainage showed a significant decrease in the cystic volume, suggesting that the cystic lesion would be subdural fluid collection rather than adhesive arachnoiditis. After dural defect repair, the patient’s symptoms completely resolved and hydro-
CONCLUSION
Subdural fluid collection rather than meningitis contributes to the hydrocephalus formation after cervical laminoplasty.

Key Words: Hydrocephalus; Cerebrospinal fluid leakage; Cervical laminoplasty; Subdural fluid collection; Meningitis; Case report

INTRODUCTION
Previous studies have shown the incidences of dural tear after spinal surgery ranging from 1.7% to as high as 17.4% [1,2]. Moreover, destruction of the cerebrospinal fluid (CSF) barrier due to dural tears may result in serious complications, such as pseudomeningocele, meningitis, arachnoiditis, hemorrhage, and extremely rare hydrocephalus. Although a small number of cases have been reported, the mechanism remains uncertain due to the lack of specific evidence. Here, we report a case of CSF leakage complicated with hydrocephalus after cervical laminoplasty, and systematically analyze the possible mechanisms and related risk factors.

CASE PRESENTATION

Chief complaints
The patient is a 45-year-old man who presented with dizziness and headache accompanied by recurrent weakness of his left arm at the 9th mo after cervical laminoplasty.

History of present illness
The patient presented with symptoms of aggravated sensory and motor disturbances in the limbs and unstable walking. He underwent cervical laminoplasty for cervical spondylotic myelopathy at a local hospital (Figure 1A). Although the patient complained of dizziness, the brain magnetic resonance imaging (MRI) before surgery did not reveal any abnormalities (Figure 1B). After the surgery, the patient felt a significant improvement in his condition. However, on the 10th day after operation, a high fever occurred, accompanied by abnormal blood test [white blood cells (WBCs): 13.22 × 10^9/L; neutrophils: 10.42 × 10^9/L; C-reactive protein: 18.55 mg/L; ESR: 37 mm/h]. The skin around the incision was slightly red, but there was no pressure pain or exudation. Cervical MRI was performed due to local doctors' concerns about possible surgical site infection, and unexpectedly revealed occult CSF leakage. Pseudo-
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Figure 1 Previous imaging data at a local hospital. A: Sagittal T2-weighted cervical magnetic resonance (MR) image before laminoplasty surgery revealing cervical spinal stenosis; B: Axial T2-weighted MR image before laminoplasty surgery showing a normal ventricle; C and D: Sagittal T2-weighted MR image showing obvious cerebrospinal fluid leakage without subdural fluid collection at 1 and 8 mo after cervical laminoplasty; E: Cervical MR image at 9 mo after laminoplasty surgery demonstrating a cystic lesion around the cervical spinal cord and medulla oblongata; F: Head computed tomography scan at 9 mo after cervical laminoplasty revealed hydrocephalus with marked enlargement of the ventricular system without any occupying lesion.

meningocele fluid test showed high levels of protein and WBC count (Table 1). Although the patient had no obvious symptoms of neurologic deficits and meningeal irritation, and bacterial culture of pseudomeningocele fluid was negative, mild acute central nervous system infection could not be excluded. Therefore, he was treated with short-term intravenous antibiotic and discharged with normal body temperature.

The patient was uneventful during the first 8 mo follow-up although repeated MRI showed persistent pseudomeningocele (Figure 1C and D). However, at the 9th mo, the patient gradually presented with dizziness and headache accompanied by recurrent weakness of his left arm. MRI at this time revealed pseudomeningocele, as well as cystic lesion around the cervical spinal cord and medulla oblongata (Figure 1E). And cranial computed tomography (CT) scans showed marked enlargement of the ventricular system (Figure 1F). Then, he was admitted to our hospital for further treatment.

History of past illness
The patient had no previous history of any illnesses.

Personal and family history
The patient had no relevant personal or family history.

Physical examination
Physical examination showed no obvious abnormality except decreased muscle strength of the left upper limb.

Laboratory examinations
CSF analysis at our hospital indicated chronic meningitis (Table 1). Repeated bacterial culture of CSF was negative. The hematology test was normal.

Imaging examinations
On the second day after admission, computed tomography myelography showed that the dural-arachnoid defect was located at the level of C5, near the lower edge of the fixed plate (Figure 2).

FINAL DIAGNOSIS
The patient was diagnosed with hydrocephalus, chronic meningitis, and CSF leakage. We concluded that subdural fluid collection was the main cause of patient’s discomfort.

TREATMENT
Dural repair was used to eliminate the source of subdural fluid collection (Figure 2E and F). No antibiotic treatment was given because the patient had no obvious fever or meningeal irritation.
Table 1 Results of cerebrospinal fluid culture during hospitalization and follow-up

| Date | Sample | WBCs | RBCs | Glucose | Protein | Chloride | CSF culture | Implication |
|------|--------|------|------|---------|---------|----------|-------------|-------------|
| 1    | Cyst puncture | WBCs: 450 × 10⁶/L | 0 | 5.92 mmol/L | 1.478 g/L | Normal | Negative | Acute meningitis |
| 2    | Lumbar puncture | WBCs: 303 × 10⁶/L; monocytes, 14% | 0 | Normal | 4.24 g/L | 117 mmol/L | Negative | Chronic meningitis |
| 3    | Lumbar puncture | WBCs: 10 × 10⁶/L | 0 | 5.09 mmol/L | 2.02 g/L | 130 mmol/L | Negative | Chronic meningitis |

1: 13 d after cervical operation at a local hospital; 2: Readmission due to hydrocephalus at our hospital; 3: 21 mo after dural repair. WBCs: White blood cells; RBCs: Red blood cells; CSF: Cerebrospinal fluid.

Figure 2 Imaging examination and treatment at our hospital. A: Computed tomography myelography revealing that the defect of dural-arachnoid was located at the C5 level and close to the lower edge of the fixed plate; B: Sagittal and axial view of cervical magnetic resonance imaging before pseudomeningocele drainage; C: Sagittal and axial view after pseudomeningocele drainage revealing a significant decrease in the cystic volume; D: The patient undergoing pseudomeningocele drainage; E: Intraoperative photograph demonstrating the dural-arachnoid defect; F: Dural-arachnoid defect was repaired with autologous fascia.

OUTCOME AND FOLLOW-UP

The patient showed an immediate improvement in his disease after the surgery. Postoperative cervical MRI showed significantly decreased pseudomeningocele and subdural fluid collection (Figure 3A). During the follow-up period, repeated imaging examination revealed that subdural fluid collection disappeared completely (Figure 3B), and ventricular size gradually returned to normal (Figure 3C and D). CSF analysis at the 21-mo follow-up showed that the levels of protein and WBC count decreased significantly without long term antibiotic treatment (Table 1).

DISCUSSION

Previous cases have found that hydrocephalus can develop following cervical laminoplasty and fusion[3-6], thoracic and lumbar decompression[7-12], intraspinal tumors resection[13-17], and even cervical myelogram[18] (Table 2). There are two types of hydrocephalus. One is obstructive hydrocephalus. Different etiologies, including compressive effect of subdural fluid collection or cerebellar enlargement due to cerebellar hemorrhage[7,13], obstruction of clot formation after brisk bleed enters the subarachnoid space and ventricle system[18], had been reported.
Table 2: Summary of previously reported cases on hydrocephalus after spinal surgery

| Ref.                      | Age (yr), sex | Spine procedure                      | Dural tear | Fever | CSF test                              | Risk factors of hydrocephalus                                                                 | Intervention                                      | Outcome |
|---------------------------|---------------|--------------------------------------|------------|-------|---------------------------------------|------------------------------------------------------------------------------------------------|-------------------------------------------------|---------|
| Bland and McDonald[14], 1992 | 58, M         | Cervical tumor resection             | Yes        | None  | Elevated protein and red blood cell count | Elevated CSF protein, subarachnoid hemorrhage                                                   | VP shunt                                        | Full recovery |
| Maezawa et al[4], 1996    | 69, F         | Cervical laminoplasty                | Yes        | None  | Elevated protein (64 mg/L)             | Systemic hypertension, elevated CSF protein level, subdural hydroma, suboccipital arachnoiditis | VP shunt                                        | Full recovery |
| Aghi et al[18], 2004      | 52, F         | Cervical myelogram                   | None       | None  | Elevated leukocytes and erythrocytes   | Hemorrhage in cervical subdural space                                                           | EVD, suboccipital craniectomy, C1–C2 laminectomies | Full recovery |
| Koerts et al[10], 2008    | 45, M         | Lumbar surgery                       | Yes        | None  | Moderate increase of WBCs, elevated protein level (69 mg/L) and lactate | Multiple lumbar surgery, CSF infection, and spinal adhesive arachnoiditis                        | EVD                                             | Full recovery |
| Morofuji et al[11], 2009  | 51, M         | Thoracic decompression               | Yes        | None  | None                                  | Remote cerebellar hemorrhage                                                                    | Suboccipital decompression                       | Full recovery |
| Lindley et al[3], 2011    | 14, M         | Oc–C2 fusion + rhBMP                 | None       | None  | None                                  | Intense inflammatory response to rhBMP, wound serumoma formation, Epidural fluid extending from the surgical site into the epidural space | EVD, wound exploration, and drain               | Full recovery |
| Stovell et al[8], 2013    | 63, F         | C1–C2 fixation                       | Yes        | None  | None                                  | Potential subarachnoid blood, injury of vessel                                                  | VP shunt                                        | Full recovery |
| Cavanilles et al[7], 2013 | 65, F         | Lumbar fusion and decompression      | Yes        | None  | None                                  | Caudal sagging of cerebellum, mass effect with compression in the posterior fossa               | EVD                                             | Mild motor deficits |
| Kaloostian et al[9], 2013 | 77, M         | T11–S1 posterior decompression and instrumented fusion | Yes | None | None                                  | Subarachnoid blood in the cerebellar folia                                                      | VP shunt                                        | Cognitive defects |
| 81, M                     | L4–5 decompression |                                    | Yes        | None  | None                                  | Cerebellar hemorrhage                                                                           | Venticulostomy                                   | Died |
| 64, F                     | L1–S1 posterior decompression and instrumented fusion |                       | Yes | None | None                                  | Large cerebellar hemorrhage, brainstem compression, and hydrocephalus                           | -                                              | Died soon |
| Matsushima et al[5], 2016 | 65, M         | Cervical laminoplasty                | Yes        | None  | Elevated protein (75 mg/L)             | Increased CSF protein levels, spinal cord subarachnoidal hemorrhage                              | Dural repair and VP shunt                       | Full recovery |
| Endriga et al[8], 2016    | 62, F         | Lumbar decompression                 | Yes        | None  | None                                  | Subarachnoid hemorrhage, extensive subdural fluid collection, pseudomeningocoele                  | VP shunt                                        | Full recovery |
| Benedetto et al[13], 2016 | 31, M         | Cervical tumor resection             | Yes        | None  | None                                  | Subdural fluid collections                                                                      | Dural repair                                    | Full recovery |
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| Authors          | Year | Gender | Operation                          | Postoperative Symptoms | Diagnosis                                      | Treatment                        | Outcome       |
|------------------|------|--------|------------------------------------|------------------------|-----------------------------------------------|----------------------------------|---------------|
| Esfahani et al   | 2017 | M      | Cervical neurinetric cyst resection| Yes, High fever        | Contamination of high cytokeratin content or other debris in the CSF, chemical meningitis | VP shunt                         | Full recovery |
| Kobayashi et al  | 2018 | M      | Cervical tumor resection           | Yes, None              | Aseptic meningitis, microhemorrhage, and fibrinogenic components | VP shunt and dural repair       | Full recovery |
| Prior et al      | 2018 | F      | Lumbar tumor resection            | Yes, None              | Possible dissemination of fat droplets in the subarachnoid spaces, aseptic | VP shunt and dural repair       | Full recovery |
| Tan et al        | 2018 | F      | L3-S1 laminectomies and fusion     | Yes, None              | Intraventricular hemorrhage                    | EVD                              | Full recovery |

VP: Ventriculoperitoneal; EVD: External ventricular drainage; CSF: Cerebrospinal fluid.

The other is communicating hydrocephalus. There are possible relationships between postoperative communicating hydrocephalus and subarachnoid hemorrhage, infection, contamination of the CSF with blood, multiple surgeries, increased CSF protein levels, high blood pressure, and meningitis[4,5,8,10,19]. Researchers speculated that these factors may lead to the obstruction of arachnoid granulation and arachnoid villi, the reduction of CSF compartment compliance, and the rise of CSF circulation resistance, which in turn causes the disorder of CSF absorption and circulation[16,20,21]. However, most of them are speculative conclusions without direct evidence.

This is the first case of hydrocephalus accompanied with both subdural fluid collection and chronic meningitis after spinal surgery. In this case, it is significant to differentiate subdural fluid collection from spinal adhesive arachnoiditis. Spinal adhesive arachnoiditis is a disease characterized by inflammation and scarring of the arachnoid membrane of the spinal cord, and only surgical intervention may provide temporary relief[22]. Reduced volume of cystic lesion and improved condition after pseudomeningocele drainage successfully confirmed its subdural fluid collection nature. Dural repair eliminated the patient’s discomfort, and hydrocephalus gradually disappeared, suggesting that subdural fluid collection rather than meningitis contributes to hydrocephalus. At the 21-mo follow-up, the patient still had asymptomatic meningitis with decreased CSF protein and WBC compared with before, which further confirmed the dominant role of subdural fluid collection in the formation of hydrocephalus. Subdural fluid collection in this case bought about the compression and backward displacement of the medulla oblongata, and caused the stenosis of the fourth ventricle outlet and following hydrocephalus. In addition, it also hindered the flow of CSF below the cervical spine and reduced the compliance of the CSF circulation system, which further promoted the formation of hydrocephalus[23].

Notably, the occurrence of chronic meningitis is a complex process, and one third of patients are still unable to determine the specific pathogenic factors[24]. In this case, further etiological analysis could not be accessed because of the lack of pathogen detection methods. Considering the long-term asymptomatic state of the patient and the gradual decrease of CSF protein level and WBC, it is reasonable to keep...
observation and follow-up of the patient, which can avoid excessive examination and overtreatment.

**CONCLUSION**

The development of hydrocephalus after cervical laminoplasty in this case was mainly caused by subdural fluid collections rather than meningitis, which provides original insight into the pathogenesis of hydrocephalus after spinal surgery. Priority could be given to the relief of obstruction in similar cases permitted under patients’ condition.

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