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CASE REPORT

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

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Author contributions: Huang HH, Cheng ZH, Ding BZ, Zhao J, and Zhao CQ designed the study; Huang HH collected the data; Huang HH, Cheng ZH, Ding BZ, Zhao J, and Zhao CQ analyzed the data and developed the methodology; Huang HH was a major contributor in writing the manuscript; Huang HH, Cheng ZH, and Zhao CQ analyzed and interpreted the patient's data; Zhao CQ oversaw the study; Huang HH and Cheng ZH contributed equally to this work; all authors read and approved the final manuscript.

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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 mo follow-up although repeated MRI showed persistent pseudomeningocele. At the 9th mo 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-

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cephalus gradually disappeared. CSF analysis at the 21-mo follow-up revealed significantly decreased protein level and WBC count.

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

Core Tip: Hydrocephalus following dural tear after spinal surgery is rare, and the mechanism remains uncertain. Although this case is not the first case of subdural fluid collection or chronic meningitis accompanied with hydrocephalus after spinal surgery, it is the first case of hydrocephalus accompanied with both subdural fluid collection and chronic meningitis. It confirmed that subdural fluid collection rather than meningitis mainly contributes to hydrocephalus after cervical laminoplasty for the first time. Combined with this case and literature review, it provided a reliable explanation for the mechanism of hydrocephalus after spinal surgery.

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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-

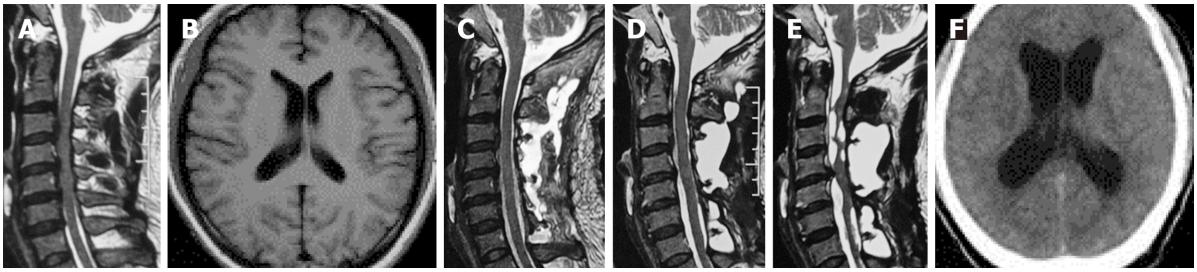

Figure 1 Previous imaging data at a local hospital. A: Sagittal T2-weighted cervical magnetic resonance (MR) image before laminoplasty surgery revealing cervical spinal stenosis; B: Axial T2-weighted MR image before laminoplasty surgery showing a normal ventricle; C and D: Sagittal T2-weighted MR image 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 × 10^6/L	0	5.92 mmol/L	1.478 g/L	Normal	Negative	Acute meningitis
2	Lumbar puncture	WBCs: 303 × 10^6/L; monocytes, 14%	0	Normal	4.24 g/L	117 mmol/L	Negative	Chronic meningitis
3	Lumbar puncture	WBCs: 10 × 10^6/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.

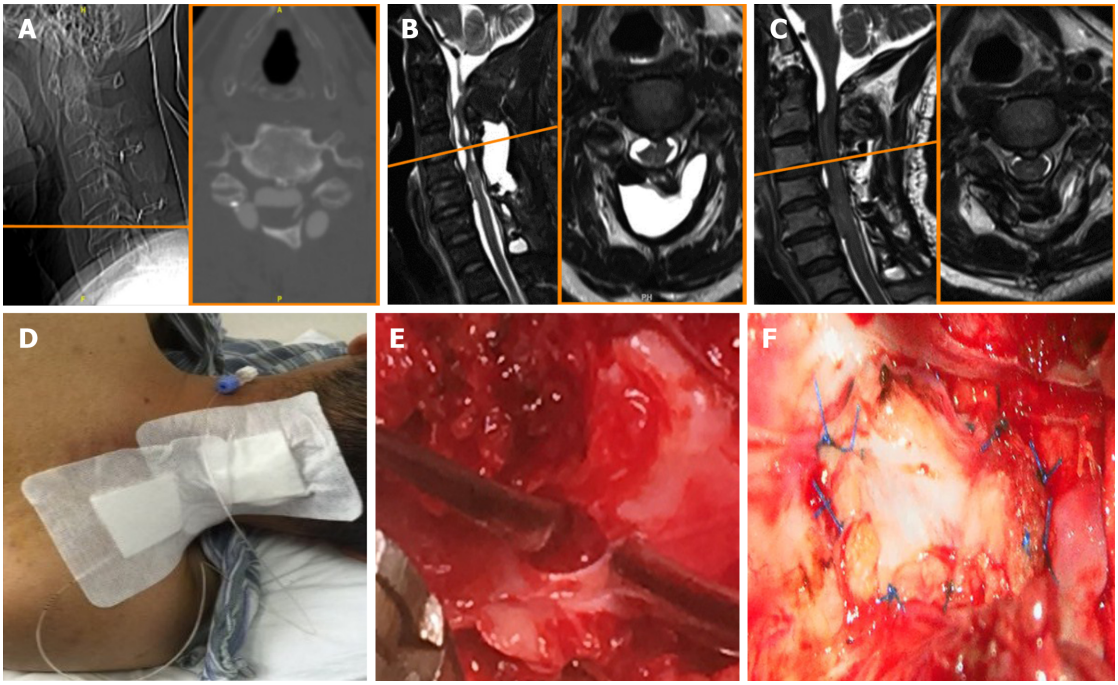

Figure 2 Imaging examination and treatment at our hospital. A: Computed tomography myelography revealing that the defect of dural-arachnoid was located at the C5 level and close to the lower edge of the fixed plate; B: Sagittal and axial view of cervical magnetic resonance imaging before pseudomeningocele 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 oppressive 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 CSF 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, and 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 seroma formation, Epidural fluid extending from the surgical site into the epidural space	EVD, wound exploration, and drain	Full recovery
	8,M	Oc–C1–C2 fusion + rhBMP	None	None	None			Full recovery
Stovell *et al*[6], 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	Luambar 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	Ventriculostomy	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, pseudomeningoceole	VP shunt	Full recovery
Benedetto *et al*[13], 2016	31, M	Cervical tumor resection	Yes	None	None	Subdurall fluid collections	Dural repair	Full recovery

Esfahani *et al*[15], 2017	7, M	Cervical neurenteric cyst resection	Yes	High fever	None	Contamination of high cytokeratin content or other debris in the CSF, chemical meningitis	VP shunt	Full recovery
Kobayashi *et al* [16], 2018	39, M	Cervical tumor resection	Yes	None	None	Aseptic meningitis, microhemorrhage, and fibrinogenic components	VP shunt and dural repair	Full recovery
Prior *et al*[17], 2018	6, F	Lumbar tumor resection	Yes	None	None	Possible dissemination of fat droplets in the subarachnoid spaces, aseptic	VP shunt and dural repair	Full recovery
Tan *et al*[12], 2018	76, F	L3-S1 laminectomies and fusion	Yes	None	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 conformed 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

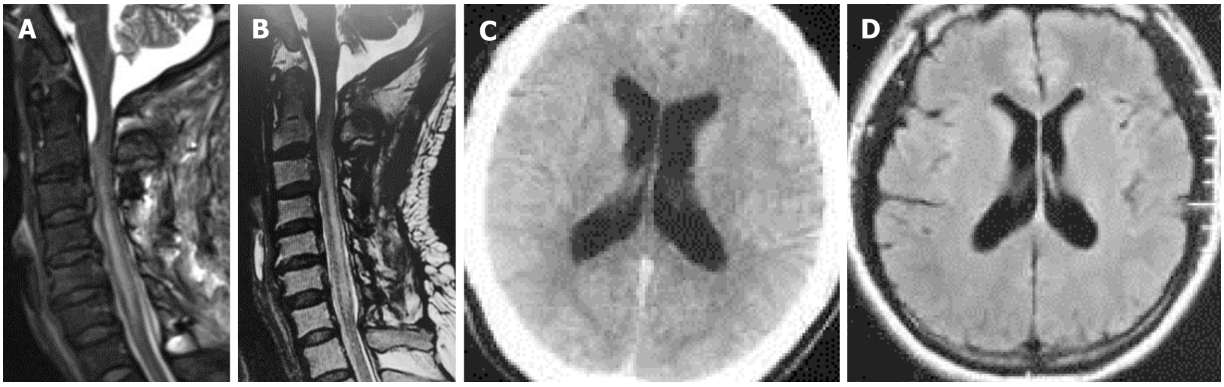

Figure 3 Cervical magnetic resonance imaging and brain computed tomography in follow-up period. A: Cervical magnetic resonance imaging at 2 d after dural repair showing significantly deceased pseudomeningocele and subdural fluid collection; B: Subdural fluid collection disappeared completely 15 mo after dural repair; C: Computed tomography-scan at 3 mo after dural repair demonstrated reduced hydrocephalus compared with pre-operation; D: Brain magnetic resonance at 1 year after dural repair showed that the ventricular system almost returned to normal shape.

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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