



Contents lists available at ScienceDirect

North American Spine Society Journal (NASSJ)

journal homepage: www.elsevier.com/locate/xnsj

Clinical Case Studies

Delayed cerebrospinal fluid (CSF) leak following anterior cervical discectomy and fusion surgery



Sreen Halayqeh, MD^a, Jacob Glueck, BA^{a,b}, Mariah Balmaceno-Criss, BS^{a,b}, Daniel Alsoof, MBBS^{a,b}, Christopher L. McDonald, MD^a, Bassel G. Diebo, MD^a, Alan H. Daniels, MD^{a,*}

^a Department of Orthopedic Surgery, Warren Alpert Medical School, Brown University, 1 Kettle Point Ave East Providence, RI, 02914 United States

^b Warren Alpert Medical School, Brown University, 222 Richmond St., RI, 02903 United States

ARTICLE INFO

Keywords:

ACDF
Complications
Dural tear
CSF leak
Ehlers-Danlos Syndrome

ABSTRACT

Background: An uncommon complication of anterior cervical discectomy and fusion (ACDF) is dura tear, which may be further complicated by cerebral spinal fluid (CSF) leak. Dural tears with CSF leak can lead to catastrophic neurologic outcomes and should be recognized early.

Case Description: This case report describes a 43-year-old female patient with history of Ehlers-Danlos syndrome who presented 1-year post-ACDF with positional headaches and lightheadedness. Imaging revealed ACDF plate subsidence and CSF leak with inferior displacement of the cerebellar tonsils.

Outcome: The patient underwent a revision procedure with removal of index screws and CSF repair using epidural blood patch, fat graft, and Tisseel. The original bicortical screws were replaced with shorter larger diameter unicortical screws. Post-operative imaging at 2 and 6 weeks confirmed resolution of CSF leak.

Conclusions: Healthcare professionals and patients undergoing spinal surgery should be aware of late presentation CSF leaks which can represent gradual decline in neurological function. Surgical candidates at risk to develop CSF leaks should be counseled about possible complications in preoperative planning.

Introduction

Anterior cervical discectomy and fusion (ACDF) is an effective surgery for a multitude of cervical spine pathologies with relatively low rates of morbidity and mortality [1]. Complications following ACDF are commonly mild and can include dysphagia, dysphonia, hematoma, and infection [2]. Dural tears with cerebrospinal fluid (CSF) leak are an uncommon complication which should be recognized early to reduce permanent and potentially catastrophic outcomes [3–5]. Compressive pseudomeningocele and postoperative neurological deficits were both found to be associated with dural tears, indicating the potential for significant CNS deficits following incidental durotomy. Surgical site infection was also found at increased rates with dural tears, and there have been reported cases of intracranial hemor-

rhage in the days following surgery. Increased surveillance and effective treatment of dural tears is required to prevent such outcomes postoperatively [3–5].

Most CSF leaks during ACDF are recognized intra-operatively. However, small dural tears may not be identified, especially if the arachnoid layer remains intact and no obvious CSF leakage occurs. These occult CSF leaks may present in a delayed fashion during the postoperative period. They can occur up to five years post-operatively with fluid leakage from the wound, meningitis, symptoms of mass effect, neck masses, or symptoms of intracranial hypotension including positional headaches, dizziness, and lightheadedness [2,6,7].

This report demonstrates a case of delayed CSF leak more than 1-year after ACDF in a patient who presented with symptoms of intracranial hypotension.

FDA device/drug status: Not applicable.

Author disclosures: **SH:** Nothing to disclose. **JG:** Nothing to disclose. **MBC:** Nothing to disclose. **DA:** Nothing to disclose. **CLM:** Nothing to disclose. **BGD:** Consulting: Spinevision, Clariance (None). **AHD:** Royalties: Stryker (D), Spineart (F), Medicea/Medtronic (A); Consulting: Orthofix (B), Stryker (B), Medtronic (B); Research Support (Investigator Salary, Staff/Materials): Orthofix (D); Fellowship Support: Medtronic (F).

Short sentence summary: This case report describes a delayed cerebrospinal fluid leak 1-year after anterior cervical discectomy and fusion surgery complicated by a past medical history of Ehlers-Danlos syndrome.

* Corresponding author. Department of Orthopedic Surgery, Warren Alpert Medical School of Brown University, 1 Kettle Point Ave East Providence, RI 02914 USA. Tel.: 401-330-1420

E-mail address: alandanielsmd@gmail.com (A.H. Daniels).

<https://doi.org/10.1016/j.xnsj.2023.100271>

Received 13 June 2023; Received in revised form 18 August 2023; Accepted 18 August 2023

Available online 27 August 2023

2666-5484/© 2023 The Author(s). Published by Elsevier Inc. on behalf of North American Spine Society. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>)

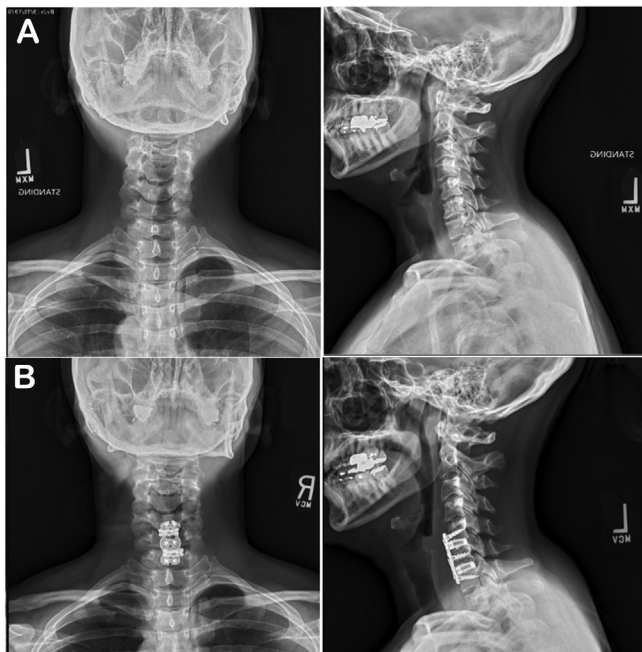


Fig. 1. (A) Preoperative posterior-anterior (PA) (left) and lateral (right) cervical radiographs demonstrating multilevel spondylosis with associated C6 on C7 retroolisthesis. (B) Postoperative PA (left) and lateral (right) radiographs demonstrating interval C5–7 ACDF with adequate alignment and no hardware complications.

Case report

A 43-year-old female patient attended the orthopedic spine clinic with complaints of bilateral upper extremity numbness and weakness over several years, more recently affecting her quality of life and daily activities. Past medical history was notable for Ehlers-Danlos syndrome (EDS). After confirming her numbness and weakness by physical examination, subsequent cervical magnetic resonance imaging (MRI) indicated spinal cord compression at C6–C7 with associated T2 hyperintensity on MRI confirming a diagnosis of cervical myelopathy. Anterior cervical discectomy and fusion were performed with full postoperative symptom resolution and no immediate postoperative complications (Fig. 1). At 6-month follow-up, the patient reported improvement in cervical pain and neurological symptoms, with a normal cervical spine examination.

However, over 1-year postoperatively the patient returned to the office due to severe positional headache and lightheadedness which had been progressively worsening and became constant. The patient did not seek additional medical care during the first postoperative year. A radiograph demonstrated subsidence of the ACDF plate (red arrow in Fig. 2). Due to these concerning symptoms, the patient was admitted and evaluated with a cervical MRI and CT scan. MRI demonstrated a CSF leak and

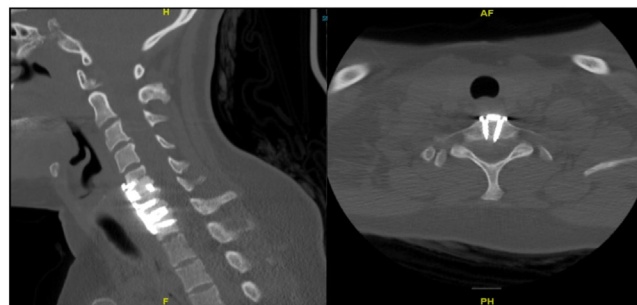


Fig. 3. Sagittal (left) and axial (right) cuts demonstrating evidence of C5–7 ACDF with the left C7 vertebral body screw penetrating the posterior cortex of the vertebral body.

signs of intracranial hypotension with inferior displacement of cerebellar tonsils. Computed tomography scan further demonstrated posterior cortex perforation of the left C7 vertebral body screw as well as interval mild subsidence and kyphosis of the ACDF construct (Figs. 3 and 4).

The patient was taken back to the operating room for revision ACDF and repair of the CSF leak which was performed emergently. After reaching the required plane, the wound was thoroughly irrigated, and the previous plate was identified. All previous screws were removed, and bilateral C5, bilateral C6, and left C7 screws were reinserted with shorter, larger diameter screws, avoiding bicortical screws which may have been a risk factor for a leak in this patient with an underlying connective tissue disorder. The right C7 screw was removed, and a large amount of CSF emanated from the hole. After full emanation of the CSF, we first placed autologous blood in the screw hole. Then a fat graft was used, Tisseel was placed, and multiple other small amounts of fat were added before a short-diameter screw was inserted. There was no further emanation of CSF. Following irrigation, thorough wound exploration found no bleeding or injury to visceral structures. The surgery occurred without any complications.

The patient was observed in the orthopedic unit for 48 hours before being discharged home with bedrest instructions. Pain medication and acetazolamide were prescribed, but no physiotherapy was needed. The patient’s headache soon resolved. Repeated imaging at 2 weeks and 6 weeks postoperatively confirmed no evidence of persistent CSF leak, and radiographs showed a revision construct with shorter unicortical ACDF screws (Figs. 5 and 6). Physical examination was normal with no further interventions needed. We used close clinical and MRI monitoring to ensure adequate repair over time since it was not possible to confirm the repair intraoperatively given limited visualization. The patient has consented to the submission of this case report.

Discussion

This report describes a unique case of intracranial hypotension presenting 1-year following ACDF. The patient presented with orthostatic headache and lightheadedness caused by a delayed CSF leak, likely

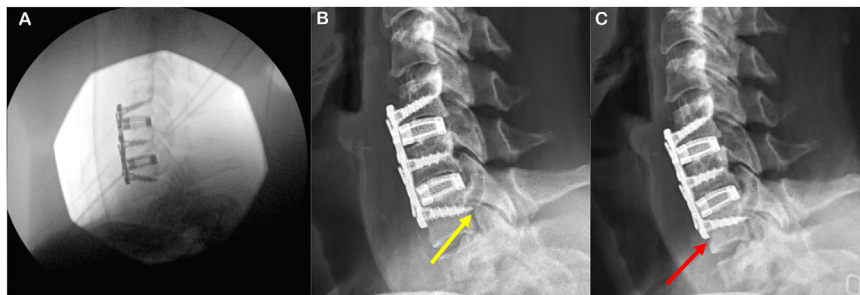


Fig. 2. (A) Intraoperative fluoroscopy. (B) Immediate radiograph of C5–7 ACDF. (C) Compared to 1 year postoperatively which demonstrates subsidence (red arrow).

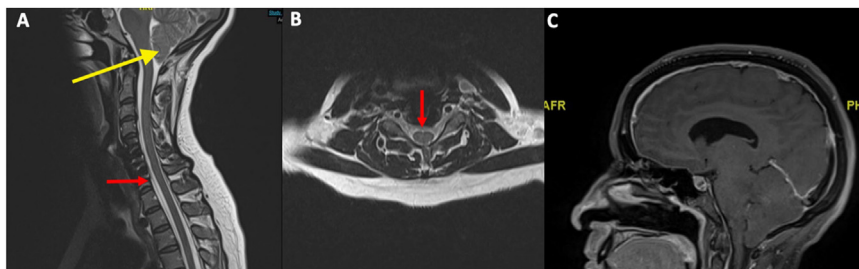


Fig. 4. Preoperative T2 weighted sagittal (A) and axial (B) MRI demonstrating the presence of a ventral hyperintense fluid collection consistent with CSF leak (red arrow). The yellow arrow in (A) demonstrates tonsillar herniation. Preoperative midsagittal T1 brain MRI (C) demonstrating inferior displacement of the cerebellar tonsils. Postoperative T2 sagittal MRI.

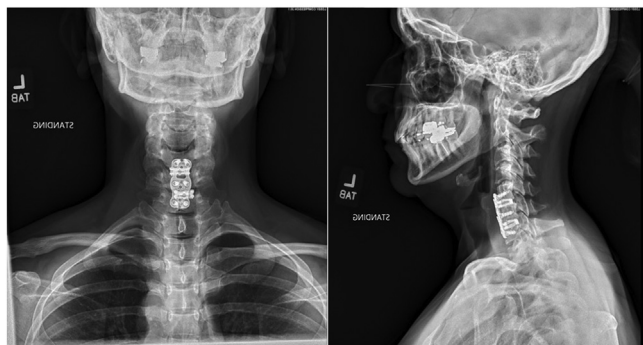


Fig. 5. Cervical spine X-ray 2-weeks after the revision surgery. PA (left) and Lateral (right) views demonstrate the replacement of the C7 screws with shorter screws of increased diameter and maintained lordosis without any immediate postoperative hardware complications.

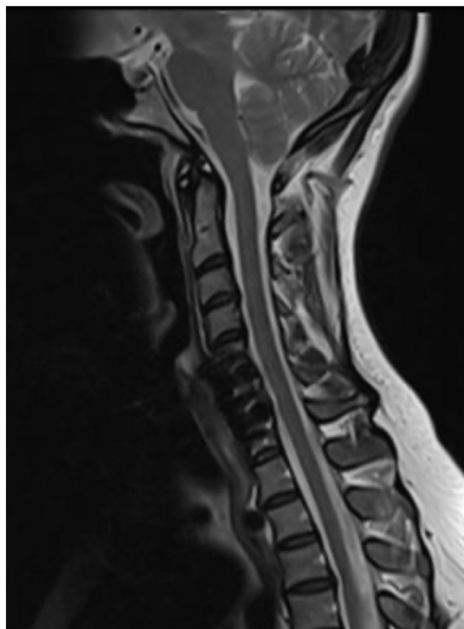


Fig. 6. A 6-week postoperative T2 weighted sagittal MRI demonstrating resolution of CSF leak.

due to a combination of mild cage subsidence and subsequent cervical kyphosis, which led to screw tip penetration through the posterior vertebral body into the spinal canal. Preoperatively, the presence of a CSF leak with resultant signs of intracranial hypotension and brainstem herniation was confirmed with both MRI and CT scans. Intraoperatively this was confirmed with a large rush of CSF from that screw hole after screw removal.

After a comprehensive literature review, we identified one case of an oropharyngeal CSF fistula 5-years after ACDF [7] and 1 case dis-

cussing the use of a lateral ventriculo-peritoneal shunt for delayed CSF leak postcervical laminoplasty. The leak was likely caused by posterior spinal cord drift after decompression, leading to friction with the posterior bony structure and resulting in thinning of the dural membrane, gradually appearing small cracks, and finally leading to large amounts of CSF exposure [8], indicating the rarity of this complication.

Cage subsidence after ACDF is not rare and occurs in 13.2% to 62.5% of cases [9]. Cervical kyphosis is a subsequent deformity which may occur secondary to subsidence as reported in the literature [9]. However, the correlation between the occurrence of cage subsidence and long-term clinical outcomes like screw penetration of the dura leading to dural tear and CSF leak remains obscure [9,10].

Cerebrospinal fluid leaks are more common in anterior cervical corpectomy (ACCF) with a 3-times increased risk compared to ACDF. Moreover, the risk of CSF leak is also elevated in patients with an ossified posterior longitudinal ligament (OPLL) [2,11–13]. Other conditions that increase the risk of CSF leak include dural ectasia in neurofibromatosis (NF) type 1 or Marfan disease, intradural disc herniation, revision operations, compressive osteophytes, arthrodesis, non-White patients, and male gender [6,12,14,15].

Other possible contributing factors to delayed CSF leak have been hypothesized, including advanced age, history of prior surgery in the same region, length of surgery, and direct trauma [16]. A recent review on the surgical repair of syndromic scoliosis found that EDS patients have a high rate (>10%) of dural tears. The dura mater is primarily composed of type 1 collagen, so EDS mutations affecting expression of type 1 collagen might contribute to the high rate of dural tears. However, there is little data demonstrating a causative factor regarding this relationship, and thus further research investigating dural tear incidence in different EDS subtypes would help to investigate this relationship [17,18].

The anterior approach associated with ACDF provides limited access for repair due to limited access to the dura [11]. Therefore, there is limited information on the management of CSF leaks after ACDF, and management is largely surgeon dependent. The first step in management is identification of the dural puncture intra-operatively, for anterior cervical dural tears access and visualization are often a challenge. If direct visualization and suture repair are to be attempted, a corpectomy is necessary. The downside to corpectomy is the increased dead space created by corpectomy which may encourage ongoing CSF leak or pseudomeningocele if the repair is not water-tight.

After recognition of a leak, it is important to attempt a repair intraoperatively to prevent any further complications and the need for subsequent procedures [2]. Repair methods include primary dural closure with suture, repair with fibrin glue, gelatin foam, DuraSeal, muscle graft, fat and fascia graft, blood patch, or any combination of these techniques [1,11]. Postoperatively, CSF drains and wound-peritoneal shunts have been used as a preventive measure to reduce CSF leaks which resulted in excellent success rates of 83% to 100% [1,11,14]. In this case, the use of neither shunt nor drain was advisable since drainage of additional CSF could be catastrophic in a setting of intracranial hypotension.

In this ACDF revision case, we accessed the dural tear through the pre-existing screw hole given our proposed mechanism of injury and its

favorable risk-benefit ratio compared with corpectomy. The dura was sealed with an autologous epidural blood patch (EBP) due to the treatment's efficacy in treating incidental durotomy in a cost-effective manner [19]. Epidural blood patch for CSF leaks following dural tears has been shown to have an 85% success rate that climbs to as high as 90% when EBP is repeated [20]. A fat graft was then utilized for its properties as an effective water sealant, revascularization agent, and its nonadherence to neural elements [21]. Tisseel was used as a hemostatic agent due to the inaccessibility and sensitivity of spine revision surgery in a patient with EDS.

Conclusions

Healthcare professionals and patients undergoing spinal surgery should be aware of late presentation CSF leaks. Our case report demonstrates a CSF leak due to cage subsidence with progressive kyphosis causing screw penetration; this required urgent work-up and surgical intervention. Surgical candidates at risk to develop CSF leaks should be counseled about possible complications in preoperative planning. Further research is needed to investigate the role of EDS in developing late CSF leak after spine surgeries, and the ideal management strategies for the late CSF leaks.

Patient Informed Consent Statement

Complete written informed consent was obtained from the patient for the publication of this study and accompanying images.

Declaration of Competing Interest

One or more authors declare potential competing financial interests or personal relationships as specified on required ICMJE-NASSJ disclosure forms.

References

- [1] Gazzeri R, Galarza M, Callovi G. Use of tissue sealant patch (TachoSil) in the management of cerebrospinal fluid leaks after anterior cervical spine discectomy and fusion. *Br J Neurosurg* 2021;1–8. doi:10.1080/02688697.2021.1881444.
- [2] Syre P, Bohman L-E, Baltuch G, Le Roux P, Welch WC. Cerebrospinal fluid leaks and their management after anterior cervical discectomy and fusion: a report of 13 cases and a review of the literature. *Spine (Phila Pa 1976)* 2014;39:E936–43. doi:10.1097/BRS.0000000000000404.
- [3] Takenaka S, Makino T, Sakai Y, et al. Dural tear is associated with an increased rate of other perioperative complications in primary lumbar spine surgery for degenerative diseases. *Medicine (Baltimore)* 2019;98:e13970. doi:10.1097/MD.00000000000013970.
- [4] Raudenbush BL, Molinari A, Molinari RW. Large Compressive pseudomeningocele causing early major neurologic deficit after spinal surgery. *Glob Spine J* 2017;7:206–12. doi:10.1177/2192568217694145.
- [5] Zimmerman RM, Kebaish KM. Intracranial hemorrhage following incidental durotomy during spinal surgery. *J Bone Jt Surg* 2007;89:2275–9. doi:10.2106/JBJS.F.01550.
- [6] Hu P, Yu M, Liu X, et al. Cerebrospinal fluid leakage after surgeries on the thoracic spine: a review of 362 cases. *Asian Spine J* 2016;10:472–9. doi:10.4184/asj.2016.10.3.472.
- [7] Weinheimer WJ, Rowley JG, Otto R, Floyd J, Chen PG. Oropharyngeal CSF leak secondary to anterior cervical discectomy and fusion. *Case Rep Otolaryngol* 2018;2018:1–4. doi:10.1155/2018/7629184.
- [8] Gao W, Ren B, Wang X, Zhou X, Han Y, An J. Lateral ventriculo-peritoneal shunt treating delayed cerebrospinal fluid leak after cervical laminoplasty: case report. *Med (United States)* 2019;98:e16917. doi:10.1097/MD.00000000000016917.
- [9] Lee YS, Kim YB, Park SW. Risk factors for postoperative subsidence of single-level anterior cervical discectomy and fusion: the significance of the preoperative cervical alignment. *Spine (Phila Pa 1976)* 2014;39:1280–7. doi:10.1097/BRS.0000000000000400.
- [10] Obermueller T, Wagner A, Meyer B, et al. Radiographic measurements of cervical alignment, fusion and subsidence after ACDF surgery and their impact on clinical outcome. *Acta Neurochir* 2020;162:89–99.
- [11] Elder BD, Theodoros D, Sankey EW, et al. Management of cerebrospinal fluid leakage during anterior cervical discectomy and fusion and its effect on spinal fusion. *World Neurosurg* 2016;89:636–40. doi:10.1016/j.wneu.2015.11.033.
- [12] Yee TJ, Swong K, Park P. Complications of anterior cervical spine surgery: a systematic review of the literature. *J Spine Surg* 2020;6:302–22. doi:10.21037/jss.2020.01.14.
- [13] Rahimizadeh A, Soufiani H, Rahimizadeh S. Remote cervical pseudomeningocele following anterior cervical corpectomy and fusion: report of a case and review of the literature. *Int J Spine Surg* 2016;10:36. doi:10.14444/3036.
- [14] Mitchell BD, Verla T, Reddy D, Winnegan L, Omeis I. Reliable intraoperative repair nuances of cerebrospinal fluid leak in anterior cervical spine surgery and review of the literature. *World Neurosurg* 2016;88:252–9. doi:10.1016/j.wneu.2016.01.014.
- [15] Barber SM, Fridley JS, Konakondla S, et al. Cerebrospinal fluid leaks after spine tumor resection: avoidance, recognition, and management. *Ann Transl Med* 2019;7:217. doi:10.21037/atm.2019.01.04.
- [16] Dannawi Z, Lennon SE, Zaidan A, Khazim R. Orgasmic dural tear: an unusual delayed presentation of postural headache following lumbar discectomy. *BMJ Case Rep* 2014;bcr2014208071 2014. doi:10.1136/bcr-2014-208071.
- [17] Levy BJ, Schulz JF, Fornari ED, Wollowick AL. Complications associated with surgical repair of syndromic scoliosis. *Scoliosis* 2015;10:1–16. doi:10.1186/s13013-015-0035-x.
- [18] Rabenhorst BM, Garg S, Herring JA. Posterior spinal fusion in patients with Ehlers-Danlos syndrome: a report of six cases. *J Child Orthop* 2012;6:131–6. doi:10.1007/s11832-012-0393-3.
- [19] Xiong GX, Tobert D, Fogel H, et al. Open epidural blood patch to augment durotomy repair in lumbar spine surgery: surgical technique and cohort study. *Spine J* 2021;21:2010–18. doi:10.1016/j.spinee.2021.06.011.
- [20] Murphy RETSJPB. Epidural blood patch. NIH NIEHS 2022.
- [21] Fang Z, Tian R, Jia YT, Xu TT, Liu Y. Treatment of cerebrospinal fluid leak after spine surgery. *Chinese J Traumatol - English Ed* 2017;20:81–3. doi:10.1016/j.cjtee.2016.12.002.