

Case report

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# Anterior cervical pseudomeningocele causing syncope after spinal surgery: A case report

Reza Ehsanian<sup>a</sup>, Arshad Ali<sup>b</sup>, Harminder Singh<sup>c,d</sup>, Stephen L. McKenna<sup>d,e</sup>, Mariam N. Mian<sup>e,\*</sup>

<sup>a</sup> Division of Physical Medicine and Rehabilitation, Department of Orthopaedics & Rehabilitation, University of New Mexico School of Medicine, Albuquerque, NM, USA

<sup>b</sup> Rehabilitation Research Center, Santa Clara Valley Medical Center, San Jose, CA, USA

<sup>c</sup> Division of Neurosurgery, Santa Clara Valley Medical Center, San Jose, CA, USA

<sup>d</sup> Department of Neurosurgery, Stanford University School of Medicine, Stanford, CA, USA

<sup>e</sup> Department of Physical Medicine and Rehabilitation, Santa Clara Valley Medical Center, San Jose, CA, USA

ARTICLE INFO	A B S T R A C T
<i>Keywords:</i> Ossification of posterior longitudinal ligament Pseudomeningocele Syncope	Introduction and importance: Pseudomeningocele formation from incidental durotomy is a known risk in spine surgery. We present a case of incidental durotomy leading to anterior neck pseudomeningocele, compressing the carotid body (CB) resulting in syncopal episodes. To our knowledge, this is the first case report implicating syncopal episodes to CB compression via a pseudomeningocele. <i>Case presentation:</i> A mid sixty-year-old patient with history of obesity, hypertension, and diabetes presented with gait impairment and hand weakness. Ossification of posterior longitudinal ligament (OPLL) was diagnosed with computed tomography imaging (CT) and magnetic resonance imaging (MRI). Elective surgery was completed with an anterior and posterior approach for decompression and fusion. Hospital course (San Jose, CA, USA) was complicated by respiratory depression and incomplete tetraplegia. On post-operative day (POD) six, CT revealed anterolateral soft tissue neck swelling; subsequent CT and MRI showed fluid collection expansion, with associated syncopal episodes on POD thirty-nine. Despite interventional radiology drainage, the fluid collection and symptoms returned five days later. The patient ultimately underwent durotomy revision and repair with muscle patch. <i>Clinical discussion:</i> This case highlights the challenges in managing anterior cervical dural tears resulting in pseudomeningocele. Risk factors include anterior cervical corpectomy and decompression, as well as an underlying diagnosis of OPLL. Untreated dural tears may develop into pseudomeningoceles which can contribute to life-threatening outcomes. <i>Conclusion:</i> This case report presents the serious consequences of incidental durotomy, the unique post-surgical complication of syncope due to compression of the CB from a pseudomeningocele, and the challenges of managing a persistent pseudomeningocele.

# 1. Introduction

Incidental durotomy is a known risk and one of the complications of cervical spine surgery [1,2]. The incidence of dural tear in the anterior approach is reported between 0%–8%, being more common in anterior cervical corpectomies than anterior cervical disc fusion surgeries [3,4]. In patients with known ossification of the posterior longitudinal ligament (OPLL), the incidence increases to about 6.7%–31.8% [3]. While there are several different techniques to address dural tears [5–8], their persistence can be problematic [1,9]. Several case reports detail pseudomeningoceles causing airway obstruction, dysphagia, and headaches

[10–13]. Pseudomeningoceles have also been associated with loss of consciousness in Marfan syndrome secondary to intracranial pressure changes [14] as well as syncope and transient anoxic seizures [15]. Both presentations involved the lumbar spine with proposed mechanisms of changes in intracranial pressure or mechanical compression on the spinal cord (SC) leading to syncope. Cervical pseudomeningoceles have been reported to cause bradycardia, oxygen desaturation, and mental status changes [16].

We present a case of a pseudomeningocele formed after an anterior and posterior approach for cervical decompression in a patient with OPLL, resulting in recurrent syncope. Our report is the first to associate

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<sup>\*</sup> Corresponding author at: 751 South Bascom Ave, San Jose, CA 95128-2604, USA. *E-mail address:* mariam.mian@hhs.sccgov.org (M.N. Mian).

pressure from a pseudomeningocele on the carotid body with syncope, consistent with syncope associated with right carotid massage [17–19]. In this case, drainage of the fluid collection provided temporary improvement in symptoms. However, the pseudomeningocele rapidly reaccumulated, secondary to dissection of soft tissue plains within the cervical spine from the initial fluid collection, leading to recurrent syncope. Our report suggests that definitive surgical repair may be needed in cases of CSF accumulation in Zone II of the neck [20] in order to prevent syncopal episodes secondary to compression of the carotid body. In our case, the definitive surgical intervention consisted of a surgical re-exploration with removal of hardware, revision of the anterior cervical fusion at C3–4 and repair of the durotomy with a muscle patch graft. This work has been reported in line with the SCARE 2020 criteria; ethics-committee approval #22-001CR [21].

#### 2. Case presentation

A mid sixty-year-old male with a history of morbid obesity, sleep apnea, hypertension, hyperlipidemia, and cervical myelopathy presented to an outside hospital (San Jose, CA, USA) with progressive gait imbalance and bilateral hand weakness. Magnetic resonance imaging revealed cervical spondylotic myelopathy with OPLL at C3-C5 (Images not available). The patient underwent a posterior C2–5 decompressive laminectomy and lateral mass instrumented fusion, as well as an anterior C3-C4 corpectomy and fusion. No issues were documented intraoperatively. Post-operatively, he was not able to move his legs and had decreased strength in his arms. Immediate post-operative CT cervical spine imaging was unremarkable. Clinically, he was classified as incomplete tetraplegia, C5 American Spinal Injury Association (ASIA) impairment scale B (AIS B) SC injury [22].

Prior to his admission to acute rehabilitation for his SC injury, his hospital course was complicated by persistent respiratory failure requiring prolonged non-invasive ventilation. On post-operative day (POD) 6 CT with pulmonary angiography obtained to rule out a pulmonary embolism, showed an incidental finding of a "soft tissue swelling and edema in the right anterolateral visualized lower neck probably residual of recent surgery". The patient was monitored and managed with non-invasive ventilation for his respiratory support. Deemed hemodynamically stable, the patient was admitted to our acute rehabilitation hospital on POD 22. On POD 27, he showed respiratory distress with the use of accessory muscles and a vital capacity of 700 mL; the patient was subsequently transferred to the acute side of our hospital for further respiratory support. Contrast enhanced CT imaging of the soft tissue mass of the neck demonstrated a 6.6  $\times$  6.9  $\times$  7.6 cm fluid collection with a thin rim extending from the prevertebral space at C5-C6 to the subcutaneous soft tissues between the lateral aspect of the right thyroid gland and the medial aspect of the right sternocleidomastoid muscle. On POD 39, the patient developed syncopal episodes with breathing treatments and repositioning. On POD 49 MRI of the cervical spine without contrast revealed re-visualization of the anterior cervical soft tissue fluid collection similar to the prior CT imaging from POD 34 and POD 0. Cardiology consult note on POD 56 noted 6-7 bradycardic episodes lasting 1-2 min with a nadir of 29 beaths per minute on telemetry. They attributed the bradycardia to sick sinus syndrome and recommended pacemaker placement.

Interventional radiology (IR) was consulted due to the continued reproducible syncope with supine positioning, and concern that the anterior cervical fluid collection was compressing the carotid bulb, which is typically located around C3-C4, leading to increased vagal tone. IR aspirated 180 cc of clear fluid with resolution of syncopal symptoms on POD 51 (Fig. 1). On POD 56, additional events of syncope ensued prompting IR re-consultation due to presumed reaccumulation of the pseudomeningocele. The IR noted reproducible bradycardia with ultrasound probe compression of the anterior cervical fluid collection. The original neurosurgeon was notified, and the patient was repatriated to the referring hospital for pseudomeningocele repair. On POD 57 a lumbar drain was placed for decompression of the pseudomeningocele. This was unsuccessful and on POD 69 the patient underwent a re-exploration with removal of the original hardware and revision of the anterior cervical fluid neurosurging anterior cervical fluid collection.



**Fig. 1.** POD 51 Computed Tomography Angiogram (CTA). Imaging demonstrates an anterior cervical soft tissue fluid collection (a-c, arrows) compressing the carotid bulb. Note this image was obtained after the removal of 180 cm<sup>3</sup> of fluid by Interventional Radiology. Sagittal image (a) demonstrated the extent of the fluid collection at the largest cross-sectional area. Axial images (b and c) are provided to demonstrate the fluid collection relative to the structures in the neck.

right inferior corner of the original dural patch with an active CSF leak was identified and repaired with a muscle patch graft. This surgical repair was noted to be durable, and the patient's symptoms resolved.

## 3. Discussion

This case report presents a patient who underwent a cervical corpectomy and fusion complicated by a pseudomeningocele. The patient had OPLL causing obliteration of the tissue planes between the dura and bone leading to a dural defect during the anterior corpectomy, complicated by the development of a pseudomeningocele. Subsequent attempts to drain the fluid percutaneously and via lumbar drain placement only temporarily resolved the patient's symptoms. Exploration and definitive repair of the defect resulted in the definitive resolution of the pseudomeningocele and symptoms. The fact that an initial post-surgical fluid collection occurred likely dissected the neck tissues and created a new potential space with low pressure supporting reaccumulation of the fluid if a durotomy persists.

The differential diagnosis for syncope is broad, including cardiac syncope, neurally mediated syncope, and orthostatic hypotension syncope [23]. To our knowledge, this is the first report of syncope secondary to carotid sinus compression by pseudomeningocele. The closest case is by Frenkel et al., where a surgical fixation screw at C5 irritated the vagus nerve with movement, resulting in bradycardic and asystole episodes [24].

In patients with known OPLL, the disease causes the dura to become markedly thin or nearly absent as it ossifies with the posterior ligament [25]. This not only increases the risk of a dural tear during decompression, but the size of the defect is likely to be larger [25,26]. Different surgical techniques have been employed to help reduce the risk of a dural tear, including thinning the OPLL and isolating it from other vertebrae instead of its removal ("floating method") and using a posterior surgical approach for the decompression [3]. Despite these efforts, an anterior approach with corpectomy is indicated at times, especially if a kyphotic deformity is present, and CSF leaks are a real risk [4,27,28]. Although primary closure of these tears can occur intraoperatively, there is a risk that suturing itself, may cause new smaller dural defects [27]. Larger defects will likely not resolve with bed rest alone and may require a diverting lumbar drain or re-exploration and muscle graft for closure [26]. There is literature supporting conservative management of cervical pseudomeningoceles. Rahimizadeh et al. presented nonoperative management of cervical pseudomeningocele after anterior cervical corpectomy and fusion [4]. Unlike our case, the patient in Rahimizadeh's case report was asymptomatic [4].

Our case demonstrates a pseudomeningocele causing syncopal episodes directly affecting the health of a patient undergoing inpatient rehabilitation. Conservative measures failed to address symptoms, and ultimately a surgical re-exploration, revealed a 2 mm defect in the dura, requiring a muscle patch graft for definitive repair. A key lesson of this case is that once CSF has dissected tissue planes in the soft tissue of the neck, conservative approaches such as blood patch and lumbar drainage may not be effective.

#### 4. Conclusion

This case highlights the complicated nature and challenges in managing anterior cervical dural tears resulting in pseudomeningocele. In general, we recommend against an anterior approach in patients with OPLL because of the high risk for durotomy and CSF leak. In patients who have good cervical lordosis and OPLL, a posterior cervical decompression and fusion is preferred over an anterior approach. In patients with cervical spine kyphotic deformity, a posterior approach may not adequately decompress the spinal cord and an anterior approach may be unavoidable. In these cases, it is very important to counsel the patient of the high likelihood of durotomy. Intraoperatively to avoid leak, a thin island of bone can be left attached to the dura after the corpectomy. If a leak is encountered, the dura can be patched with a muscle patch or dural replacement graft and fibrin sealant. Postoperatively, a lumbar drain may be used to divert CSF and allow the repair to heal.

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# **Ethical approval**

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#### Consent

N/a.

#### Author contribution

All authors contributed to chart review and writing.

Research registration (for case reports detailing a new surgical technique or new equipment/technology)

N/a.

# Guarantor

Mariam N. Mian.

#### Declaration of competing interest

There are no conflicts of interest to be disclosed by any of the authors.

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