Case Report

Refractory Cervical Spinal Pseudomeningocele Successfully Treated with a Polycarbonate Face Mask: Technical Note

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Abstract

Introduction: Spinal pseudomeningocele involving the cervical spine is rare and may be symptomatic or asymptomatic. It can occur spontaneously, after trauma or after a surgical procedure. Typically, treatment involves primary closure of the fistula with or without CSF diversion. However, when pseudomeningocele is refractory to surgical revision additional options are limited.

Methods: We describe a case of a 50-year-old female presenting with post-operative pseudomeningocele after 16 days from a C1 schwannoma resection. She failed a revision surgery with primary closure reinforced with Duragen® and Duraseal™ application and a lumbar drain placement. Hence, a novel method was attempted by applying extrinsic compression to the upper cervical spine utilizing a polycarbonate elastic face mask (Jobst Incorporated, Prior Lake, MN, USA).

Results: The elastic face mask with a posterior cervical strap to provide continuous external compression to the surgical wound was worn for 9 weeks. Complete resolution of pseudomeningocele was noted on follow up MRI and the patient remained asymptomatic at 7-month follow up evaluation.

Conclusions: Extrinsic compression with polycarbonate face mask may be a safe nonsurgical option or may be a useful adjunct to surgical repair for post-operative cervical spinal pseudomeningocele.

INTRODUCTION

Spinal pseudomeningocele can occur spontaneously, after trauma, or following surgical procedures involving the spine or neural elements. Spinal pseudomeningoceles can be asymptomatic or symptomatic, with local pain being the most common presenting symptom. Though rare, neurologic complications can occur, including radiculopathy and myelopathy from nerve root and spinal cord herniation, respectively. Postural headache from intracranial hypotension can also occur. Although most cases of spinal pseudomeningocele occur in the lumbar spine, cervical pseudomeningocele can follow iatrogenic dural laceration from cervical or posterior fossa surgery [1-13], dural disruption in atlanto-occipital dislocation [14], and trauma to the upper cervical region [15].

Treatment for pseudomeningocele typically involves CSF diversion, followed by primary operative dural repair [16]. Pseudomeningocele following cranial procedures may be managed conservatively with a local pressure dressing and lumbar drainage (“tap and wrap”), but reliably applying extrinsic compression to the spine, especially the highly mobile cervical segment, presents unique challenges. We report a case of refractory cervical pseudomeningocele after failed primary fistula closure that completely resolved after prolonged external compression. We utilized a commercially available elastic face mask (Jobst Incorporated, Prior Lake, MN, USA) with a posterior cervical strap to provide continuous external compression to the surgical wound for 9 weeks, resulting in complete resolution of the pseudomeningocele. To our knowledge, this is the first report of a spinal pseudomeningocele, and in particular a posterior cervical pseudomeningocele, managed successfully with a pressure dressing. The face mask provided a useful and surprisingly comfortable pressure dressing solution for the posterior cervical spine.

CASE PRESENTATION

A 50-year-old woman underwent suboccipital craniectomy, C1 laminectomy, midline and lateral durotomy, and gross total resection of a right C1 schwannoma. The dural repair was primary and supplemented by an onlay collagen matrix (Duragen®, Integra LifeSciences, Plainsboro, New Jersey, USA) and hydrogel sealant (Duraseal, Covidien, Dublin, Ireland). A valsala maneuver was performed, and no cerebrospinal fluid (CSF) egress was seen, so lumbar drainage was not used. Patient’s head was kept elevated
greater than thirty degrees for the first two days and discharged home on post-operative day four without any symptoms or signs of CSF leak. Wound was clean, dry and intact.

On postoperative day 16, she presented to the emergency room complaining of increasing neck pain and postural headache. To the right of her well-healed scar, she had a palpable fluctuance suggestive of pseudomeningocele. This was confirmed by a computed tomography (CT) scan of her cervical spine (Figure 1). She was taken to the operating room for primary dural closure. A CSF fistula was identified at the right lateral portion of the dural repair. This was closed primarily and watertight closure was again verified by a Valsalva maneuver. Additional Duragen® and Duraseal were applied. A lumbar drain was not used. Four days later, she continued to complain of neck pain and postural headache. A CT scan of the cervical spine was obtained demonstrating reaccumulation of the fluid collection (Figure 2). Lumbar drainage for 5 days was instituted, but the fluid collection remained with persistent fluctuance and postural headache. An elastic face mask was applied with a foam pad inserted between its strap and the patient’s neck posteriorly as an added pressure dressing over the site of the pseudomeningocele (Figure 3). The lumbar drain was then removed, and she was discharged home with the face mask 2 days later.

After 2 weeks of using the pressure dressing around the clock except when showering, she reported complete resolution of her postural headaches. The posterior cervical region exhibited decreased fluctuance. Magnetic resonance imaging (MRI) of the cervical spine after three weeks of pressure dressing use showed a stable pseudomeningocele (Figure 4). At 9 weeks of pressure dressing use, she remained asymptomatic the fluctuance in the neck had completely resolved, and the incision remained well healed (Figure 5). The pressure dressing was discontinued. MRI of the cervical spine 8 months after revision surgery (and approximately 6 months after discontinuing the use of the brace) demonstrated complete resolution of the pseudomeningocele (Figure 6). She remains asymptomatic at 7 months follow up.

**DISCUSSION**

Dural defects, irrespective of the means by which they occur, permit CSF egress to the surrounding tissues through fistulization. When the CSF pulse pressure exceeds the interstitial pressure of the surrounding tissue, a pseudomeningocele can
form. The fistula may contain a flap of tissue that encourages unidirectional flow (ball valve) and very gradual expansion of the pseudomeningocele with each cardiac cycle. The fluid collects in a cavity lined by a fibrous membrane instead of an arachnoid-lined (leptomeningeal) membrane, hence the term “pseudomeningocele” as opposed to “meningocele,” [16] such a true meningeal or arachnoid diverticulae. Though sometimes asymptomatic, pseudomeningoceles often present with postural headache or local pain [16]. The literature contains rare reports of more serious complications of pseudomeningocele, including herniation of nerve roots [17,18] or the spinal cord causing neurologic decline [12,13,19]. Cervical pseudomeningoceles therefore warrant treatment to avoid potentially catastrophic complications.

Physicians must consider the physical properties of CSF leak involved in sustaining the fistula and preventing its closure prior to relying on the biologic phenomenon of dural fibrosis to eliminate the leak. These mechanisms include a pressure differential across the defect and/or a ball valve that prevents the egress of CSF from the pseudomeningocele during diastole. In the presence of a dural defect, CSF is able to assume a path of flow into the extradural space through the defect when the interstitial pressure in the tissue is less than the CSF pulse pressure.

A variety of methods for cervical pseudomeningocele treatment exists. Primary fistula repair is the most frequently reported means, and this is most frequently reported as being performed with application of some sort of patch graft. Moskowitz et al. compared complication rates between various popular graft materials in a large series of dural substitute applications in closure after posterior fossa neurosurgical procedures and found no clear differences between them [20]. As an adjunct to primary fistula repair, extrinsic compression is effective in raising interstitial tissue pressure and reversing the pressure gradient between the intradural and extradural space. Otologists routinely employ local pressure dressings prophylactically after postauricular approaches to the skull base, and therapeutic local pressure dressings have also been reported as a therapeutic option after development of clinically significant pseudomeningocele in this region [21]. Because of the proximity of the high cervical region to the posterior fossa, the intradural hydrostatic pressure can reasonably be assumed to be similar in the two regions. As such, we hypothesized that application of continuous external compression to the area could similarly collapse the subfascial potential space and promote dural scarring by increasing the pressure in the interstitial space. However, maintaining continuous extrinsic compression to this area using standard materials such as gauze wrap or a cervical collar are technically challenging and unlikely to be tolerated or properly implemented by any but the most motivated of patients. Numerous commercially available masks exist for the treatment of soft tissue swelling from venous or lymphatic insufficiency, and we employed one such mask for this patient.

There have been no randomized trials comparing any single therapeutic intervention in the treatment of cervical pseudomeningocele with any other. The current body of evidence does not support any one therapeutic modality as being definitely superior to any other. On initial presentation of pseudomeningocele, decision was made to perform primary revision due to late presentation of the pseudomeningocele. Lumbar drain is commonly utilized for CSF diversion but was not placed intra-operatively because water-tight closure was felt to be adequate. [22-25] Weng et al. described open excision of the pseudomeningocele to promote CSF absorption into soft-tissue with primary dural repair and CSF diversion as a safe and effective treatment. [1] In this patient, we felt that a repeated invasive attempt at defect closure would not likely result in successful pseudomeningocele resolution, as the initial attempt to do so had failed. In the absence of any definitely superior therapy, we successfully employed a novel and very well tolerated means of resolving the pseudomeningocele.

Cervical pseudomeningocele presents a unique therapeutic challenge, as the complications from mass effect or neural element herniation can be catastrophic. We feel that the operative repair of a dural defect still should be considered as a first line of treatment. However, there are significant risks and costs associated with reoperation in the setting of a dural defect, particularly in the cervical spinal region. If surgical repair is unsuccessful or refused by a patient, long-term external compression should be considered.

The patient had complete resolution of this pseudomeningocele and she tolerated the therapy well. The benefits of this therapy are numerous, increased patient comfort, increased patient convenience, technical ease of implementation for the patient, dramatically reduced costs compared with repeated operative repair, and decreased risk of infection or mechanical injury compared with repeated operative repair. Hence, we pose external cervical compression with a face mask as a therapeutic option for patients with surgically refractory posterior cervical pseudomeningoceles.

**CONCLUSION**

In patients who present with a cervical pseudomeningocele without evidence of myelopathy or radiculopathy, application of continuous external cervical compression with serial outpatient examination and imaging is a safe nonsurgical therapeutic option or may be a useful adjunct to surgical repair.

**REFERENCES**

1. Weng YJ, Cheng CC, Li YY, Huang TJ, Hsu RW. Management of giant


