

Cervical Pseudomeningocele-Induced Hydrocephalus Following Traumatic Brachial Plexus Injury – A Case Report

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Abstract

Introduction: We report a case of a 49-year-old man who sustained a left brachial plexus injury and traumatic brain injury after a motor vehicle accident and subsequently developed a giant left cervical pseudomeningocele. The patient suffered multiple fractures in the cervical and thoracic ribs, transverse processes and the scapula. Physical examination revealed a giant left supraclavicular mass restricting his ability to turn his head ipsilaterally, with head tilted to the right, consistent with complete plexus avulsion. Neurological examination showed progressive muscular atrophy and a positive Tinel's sign and paresthesias of the left hand.

Methods: MRI and CT revealed a giant cervical pseudomeningocele. Left hemilaminectomy and partial medial facetectomy were performed for an extradural repair of the cyst. Three days later, the pseudomeningocele recurred; C6-T2 cervical laminectomy and a combined intra- and extradural repair of CSF leak with tensor fascia lata graft were performed. One day after the second surgery, the patient developed acute communicating hydrocephalus (CH) with progressive neurological decline.

Results: Ventriculoperitoneal shunt placement successfully resolved neurological symptoms associated with CH. The patient continued receiving treatment for neuropathic pain and spasms in the left upper arm at one-year follow up.

Conclusion: We present one of the few documented cases of acute CH after a successful repair of a giant cervical pseudomeningocele. It is important for physicians to be aware of changes in CSF flow dynamics that occur in patients with traumatic brain injury. A repair of a large chronic pseudomeningocele can lead to acute CH in patients and cause rapid neurological decline. It is critical for clinicians to be mindful of the potential complication of acute hydrocephalus in patients who undergo repair of a large chronic pseudomeningocele secondary to trauma.

Keywords: Cervical pseudomeningocele; Brachial plexus; Hydrocephalus; Muscular atrophy

Introduction

Patients present with varying degrees of weakness and muscle atrophy in the upper limbs after sustaining a traumatic brachial plexus injury. One of the complications of brachial plexus injury is the development of pseudomeningoceles, and the reported incidence ranges from 21 to 57% among different case series [1-4]. The mechanism underlying pseudomeningocele is a traction injury to the brachial plexus causing avulsion of the nerve roots from the spinal cord, which can be either partial or complete. This can result in the disruption of the dura-arachnoid layer of the meninges [2,5] leading to continual flow of CSF into the extraspinal space [6,7], ultimately forming a fluid-filled space lined by a fibrous cavity. Most cases of pseudomeningoceles are small and asymptomatic [7], as extraspinal accumulation of CSF only creates marginal pressure on surrounding muscle or bone [2]. However, large lesions can occur in rare instances, causing local pain, and if the size of the dural defect is significant, the spinal cord can herniate into the pseudomeningocele [3,4].

We report a case of a 49-year old man who sustained a traumatic brachial plexus injury from a motorcycle accident and subsequently developed a giant left cervical pseudomeningocele. After his pseudomeningocele was surgically repaired, the patient acutely became obtunded and was found to have an acute communicating hydrocephalus. Ventriculoperitoneal shunt placement was performed emergently and he returned to his baseline neurological status.

Case Report

History and physical examination

Our patient is a 49-year old man who sustained a left brachial plexus

injury 15 months prior from a motorcycle accident. At the time of the accident, he had a Glasgow Coma Scale (GCS) of 8, multiple cervical and thoracic ribs and transverse processes fractures, subarachnoid haemorrhage, and left scapular fracture. He also had a subdural hematoma for which he underwent burr hole evacuation one month after the accident. He was referred from the orthopaedics department to neurosurgery, for evaluation of swelling in the left supraclavicular region (Figure 1). The supraclavicular mass restricted the patient's ability to turn his head ipsilateral to the lesion. He tended to lean his head towards the right while standing.

Neurological exam showed diffuse left upper extremity weakness without activation of bicep, rotator cuff, or deltoid. Significant muscular atrophy was noted on the left upper limb, with mild winging of the left scapula. He was unable to extend the wrist or the digits. He had no digital flexion. There was no interosseous function in the left hand and a positive Tinel's sign was noted over the supraclavicular fossa with paresthesias in the left hand. Muscle strength was normal on the right side. He denied any headache, photophobia, nausea, vomiting, or difficulty with balance or gait. Review of systems and laboratory studies were non-contributory otherwise.

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Imaging

Magnetic resonance imaging of the cervical and thoracic spine



Figure 1: A marked left supraclavicular bulge is noticeable upon physical examination. The patient leans his head to the right.

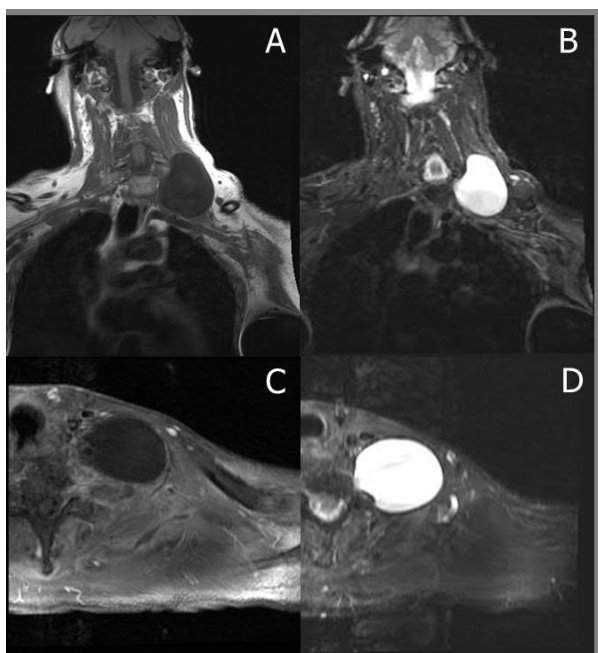


Figure 2: (A and B) T1 and T2 coronal MRI of the cervicothoracic region, showing a giant left cervical pseudo-meningocele abutting the brachial plexus and depressing the clavicle. (C and D) T1 and T2 axial MRI with gadolinium, showing a large paraspinous mass.

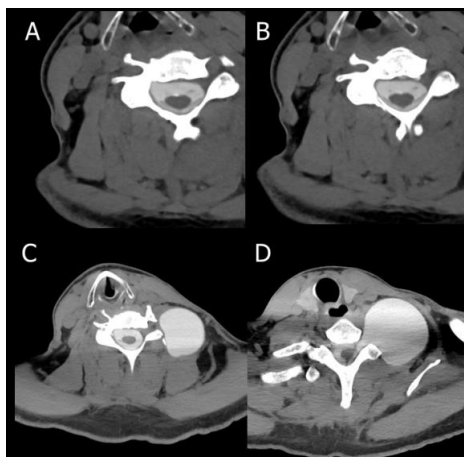


Figure 3: (A-D) Axial cervical myelogram from levels C5 through C8, respectively, showing leakage of cerebrospinal fluid through the left C8 foramina.

showed paraspinous fluid accumulation abutting nearby soft tissues (Figure 2). CT myelogram studies revealed a giant pseudomeningocele over the left C8 nerve root (Figure 3).

First pseudomeningocele repair procedure

The patient underwent left C7-T1 hemilaminectomy and partial medial facetectomy to obtain proper exposure. The extradural repair of the pseudomeningocele was performed. A small piece of muscle was harvested locally and was sutured over the C8 nerve root sleeve with a 4-0 Nurodon (Ethicon, New Jersey), which was supplemented with muscle and fat. Gelfoam (Pfizer, New York) and DuraSeal (Integra, New Jersey) were used to provide additional reinforcement. A lumbar drain was placed at the end of the case, which drained at 10cc per hour, to promote another pathway for CSF to escape while the repair site was healing. The patient tolerated the procedure well with no acute events occurring overnight.

Second pseudomeningocele repair procedure

On post-operative day 3, the patient's pseudomeningocele recurred as a new, fluid-filled supraclavicular mass accompanied by intermittent headaches. Re-exploration of the wound was performed and a wider exposure was obtained with C6-T2 cervical laminectomy. The previous repair was removed and a continued CSF leak through our previous repair was noted. The dura was opened at midline, and the fistula point was identified intradurally at the left C8 root sleeve. Fascia lata was harvested from the right thigh to use as a graft to supplement the dura both intra and extradurally. DuraSeal (Integra, New Jersey) was used for closure. The procedure resulted in obliteration of the supraclavicular bulge, and the patient was transferred to the SICU post-operatively. He was extubated on the same day without acute events overnight.

Ventriculo-peritoneal shunt placement

On the post-operative day 4 from the second surgery, the patient's mental status acutely declined after the lumbar drain was removed. The patient became obtunded, his speech was incomprehensible, and he was unable to follow commands. At the time, the patient had just finished walking in the halls with physical therapy. He had wide fluctuations in his blood pressure indicative of changes in intracranial pressure; initially, his systolic blood pressure was in the 60s; repeat measuring revealed a systolic blood pressure in the 130s, then subsequent readings showed a drop down to the 70s. Examination of the pupil revealed the right side to be 5-6 mm, and the left to be 3 mm. His heart rate varied widely between 50s and 100s switching between sinus rhythm and junctional rhythm. In addition, the patient had an episode of bowel incontinence when transported back to his bed.

Due to an acute decline in his mental status, CT of the head was done immediately, and was compared to the scan taken two weeks prior (Figure 4A). The new scan revealed acute communicating hydrocephalus, with moderate enlargement of the lateral and third ventricles without evidence of intracranial haemorrhage or mass (Figure 4B). This finding prompted the patient to undergo an emergent right ventriculoperitoneal shunt placement (Figure 4C).

The patient returned to his baseline neurological examination after the shunt placement. The patient made a good recovery and was discharged on post-operative day 14 to an inpatient rehab. At the time of discharge, he no longer had fullness over his left supraclavicular region.

Left brachial plexus exploration and intercostal nerve transfer to biceps branch of musculocutaneous nerve

On post-operative day 29, the patient made a good recovery

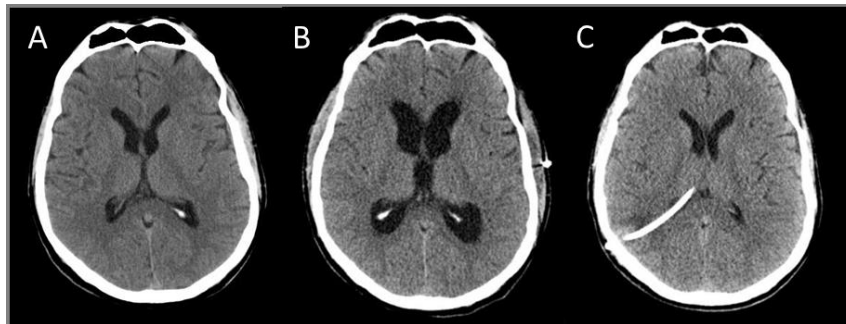


Figure 4: (A) Head CT scan taken two weeks prior to the second pseudo-meningocele repair surgery shows normal size of the ventricles. (B) CT scan taken after the second pseudo-meningocele repair surgery shows marked dilatation of the lateral and third ventricles including the temporal horns, consistent with communicating hydrocephalus. (C) CT scan taken after placement of a right ventriculo-peritoneal shunt demonstrates normal sized ventricles.

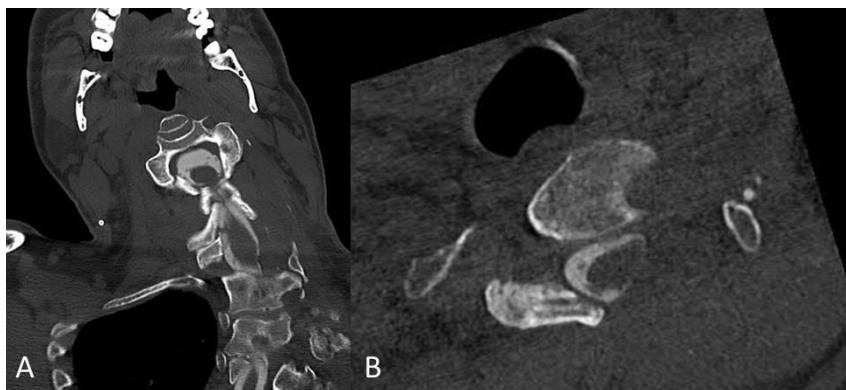


Figure 5: CT myelogram performed at 23-month post-op from the repair. (A) There is an evidence of laminectomy defect and left C7/T1 facetectomy from surgery. There is a complete resolution of pseudo-meningocele over the left C8 nerve root. (B) There was a progression of levoscoliosis in his cervical spine when compared to pre-op from 25 to 40 degrees.

from his surgeries. He underwent a left brachial plexus exploration and intercostal nerve transfer to biceps branch of musculocutaneous nerve. Identifying normal anatomy was extremely challenging due to a considerable amount of scar in the region. C5 and C6 nerve roots appeared to be avulsed from the cervical cord. The spinal accessory nerve was heavily encased in scars and stimulation of these nerves did not result in contractions of identifiable trapezius muscle. Therefore, the decision was made to perform nerve transfer of intercostal nerve 4 and 5 to biceps branch of musculocutaneous nerve. The patient tolerated the procedure well and he was discharged to an inpatient rehab.

At 20-month follow-up from intercostal nerve transfer surgery, the patient still had no movement in his wrist, elbow or shoulder. He had patchy sensation throughout the left hand. He no longer had supraclavicular bulge. A CT myelogram demonstrated a complete resolution of pseudomeningocele at 23 month from the repair. There was a progression of his levoscoliosis in his cervical spine from 25 degree to 40 degree after 3-level laminectomy and a unilateral facetectomy (Figure 5). In the absence of any significant neck pain or other symptoms related to his neck deformity, we decided to continue following the patient clinically.

Discussion

Cervical pseudomeningoceles resulting from traumatic brachial plexus injury are common. In general, they are usually small in size and remain asymptomatic, thus rarely requiring surgical intervention. However, in rare instances, giant cervical pseudomeningoceles can result from traumatic brachial plexus injury.

In 2013, Tedesco-Marchese et al. reported two cases of patients with pseudomeningoceles, who presented with a giant supraclavicular mass with accompanying weakness of the ipsilateral upper limb and Horner syndrome [7]. The first case was a 32-year old male who was involved in a motor vehicle accident with a complete left brachial plexus injury [7]. One month after trauma, he underwent a repair of his pseudomeningocele. Three months later, the patient developed hydrocephalus and underwent placement of a ventriculoperitoneal shunt [7]. When the progressive bulging in the left supraclavicular region returned, with headache, nausea, and dizziness elicited by manual compression of the bulge, cervical MRI and cisternography was performed and revealed an abnormal collection of CSF lateral to the cervical spine, consistent with pseudomeningocele [7]. A lumbar puncture decreased the size of supraclavicular mass only transiently. The patient's ventriculoperitoneal shunt system was exchanged for a new device, which led to the resolution of his bulge and neurological symptoms.

In a second case, a 25-year old patient injured himself in a textile machine. Immediately after the injury, he developed complete paralysis and numbness of the left upper limb [7]. He was surgically explored for hematoma in the supraclavicular region. Four months later, the patient developed a large bulge in the left supraclavicular region, accompanied by mild headache with nausea and dizziness upon compression of the mass, with limited ability to rotate his head to the left. Physical exam showed a large, painless prominence in the left supraclavicular region, without pulsation or murmur, but with significant hypotrophy and complete paralysis and numbness of the

upper limb, and Horner syndrome [7]. The patient underwent lumbar puncture, which transiently relieved the bulging. A lumboperitoneal shunt was placed for a definitive treatment, which resulted in resolution of his pseudomeningocele.

The underlying pathophysiology of the development of acute communicating hydrocephalus after a repair of pseudomeningocele was an abrupt alternation in CSF dynamics within the patient's ventricular system. We hypothesize that the patient's large cervical pseudomeningocele acted as a "fifth ventricle", which over time had gradually accommodated a substantial volume of CSF outside the central nervous system. Because of chronicity of the supraclavicular mass (the patient was involved in a motor vehicle accident 15 months prior), we speculate that the choroid plexus had "adapted" to the abnormal presence of this extra space that can accumulate CSF; thus, produced more CSF than in an otherwise anatomically and physiologically normal individual. Thus, the definitive repair of the pseudomeningocele forced the "extra" CSF into the ventricular system. After the lumbar drain was removed, there was no route of escape, which resulted in the development of acute communicating hydrocephalus.

Another contributing factor to the development of acute hydrocephalus in our patient is impaired CSF circulation from scarring and fibrosis of arachnoid granulation as a sequela of the head injury from the motor vehicle accident. CSF physiology including production, circulation and absorption is a complex and dynamic process. CSF circulation is an intricate system that allows elimination of compounds from the brain parenchyma. It has been described to play an essential role for normal brain functioning [8]. Often cited as "the third circulation [9], CSF is a heterogeneous fluid composed of plasma filtration and cell segregation products [10]. The choroid plexus and surrounding structures [3] produces CSF, which flows into the ventricles and eventually drains into arachnoid granulations and the main venous system of the brain [11,12]. The described processes maintain CSF homeostasis between production and absorption. When CSF absorption is compromised, as in our patient, inflammatory changes secondary to trauma act as a barrier in the draining system, causing CSF accumulation, resulting in dilatation of ventricular system [13].

The pathophysiology of hydrocephalus from traumatic brain injury is thought to be secondary to an increase in the arachnoid villi outflow resistance. After a traumatic event, inflammation and haemorrhage promote proliferation and fibrosis formation from arachnoid cells, which leads to an obstruction of CSF flow through arachnoid villi into the venous system, ultimately leading to an abnormal accumulation of CSF [13-16]. Treatment options include CSF diversion through shunts [17], endoscopic third ventriculostomy with or without septostomy [18,19], and choroid plexus cauterization. In extreme cases, decompressive craniotomy has been performed as a salvage operation [20]. In addition, a lumbar drain represents another viable option, as demonstrated by Manet et al. in 2016 [21].

Current interventions for hydrocephalus are not without flaws and complications of shunts include infections, hematoma, seizures, CSF leaks, tissue necrosis, and shunt malfunctions secondary to obstruction, disconnection or displacement. Shunt failure rate requiring revisions varies from 22% to 49% [22-25]. Further research is needed to provide additional links between inflammation, scar tissue formation and CSF dysfunction, which may lead to more reliable and lasting treatment options for posttraumatic hydrocephalus.

Although brachial plexus injuries resulting in a large, symptomatic pseudomeningoceles are not frequently encountered, the characteristic signs and symptoms of pseudomeningocele itself, and the subsequent

complications from surgical repair should be carefully reviewed. This is critical as dismissal of early signs of intracranial hypertension such as dizziness, headaches, and nausea can have fatal consequences. Furthermore, it is important to not confound an abnormal supraclavicular mass in post-traumatic patients with other etiologies such as injuries to the bone or to the vasculature (subclavian artery pseudoaneurysm) [7,26,27]. A detailed physical examination evaluating for the presence of murmur, pulse, presence of Horner's syndrome, dizziness and/or headache that can be induced by manual compression of the bulge, is critical in making the correct diagnosis. Imaging studies such as MRI and myelography can be diagnostic in differentiating these conditions.

Conclusion

There is paucity in the literature describing complications of surgery in patients who present with a large, symptomatic cervical pseudomeningoceles from trauma. Long-term follow up in this group of patients is limited. We present a case in which surgical repair of a large pseudomeningocele was complicated by acute communicating hydrocephalus. Thus, the potential for abruptly disrupting the patient's aberrant cerebrospinal fluid dynamics with surgery should be considered during perioperative period. We found that the placement of a ventriculoperitoneal shunt is safe and effective in resolving hydrocephalus. The surgeons should be mindful of this potential complication of acute hydrocephalus in patients who undergo a repair of large chronic pseudomeningocele secondary to trauma.

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