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Intracranial subdural empyema after surgery for lumbar lipomyelomeningocele: A rare complication

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Abstract

Background: Surgery is routinely recommended for lumbar lipomyelomeningocele, especially in the setting of tethered cord syndrome. The most common complications are wound infections and cerebrospinal fluid (CSF) leak, which remain confined to the surgical site. To the best of our knowledge, there have been no prior reports relating an intracranial subdural empyema following detethering surgery. Prompt diagnosis is essential since subdural empyema is a neurosurgical emergency.

Case Description: The patient was an 11-month-old male who underwent detethering surgery for a lumbar lipomyelomeningocele. This was followed by wound drainage consistent with CSF leak, requiring revision. Cultures grew three aerobes (*Escherichia coli*, *Enterococcus*, and *Klebsiella*) and three anaerobes (*Clostridium*, *Veillonella*, and *Bacteroides*). He was started on cefepime, vancomycin, and flagyl. The patient required two more wound revisions and placement of an external ventricular drain (EVD) secondary to persistent wound leakage. A subsequent magnetic resonance imaging (MRI) brain was carried out due to protracted irritability, which revealed extensive left subdural empyema along the parietooccipital region and the inferior and anterior temporal lobe. He underwent evacuation of the subdural empyema where cultures exhibited no growth. Subsequently, he progressed well. His lumbar incision continued to heal. Serial MRI brains and inflammatory markers were reassuring. He weaned off his EVD and went home to complete a 6-week course of antibiotics. Upon completion of his antibiotics, he returned for a clinic visit; he exhibited no interim fevers or wound issues; cranial imaging documented no evidence of a residual or recurrent subdural empyema.

Conclusion: Intracranial subdural empyema may occur after wound complications from detethering surgery despite early initiation of broad-spectrum antibiotics. Possible etiology may be local wound infection that seeds the subdural space and travels to the cranium, leading to meningitis and subdural empyema. Such a scenario should prompt surveillance imaging of the head as undiagnosed subdural empyema may lead to devastating consequences.

Key Words: Lipomyelomeningocele, subdural empyema, tethered cord syndrome

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INTRODUCTION

Lumbar lipomyelomeningocele is a form of occult spinal dysraphism that can cause tethered cord syndrome, leading to neurological, urological, and orthopedic dysfunction.[31] Surgery is typically recommended for tethered cord syndrome. The most common surgical complications are wound infections and cerebrospinal fluid (CSF) leak, which are generally confined to the surgical site.[4,12,13,15,17,19,20,23,25,31] To the best of our knowledge, there have been no prior reports relating an intracranial subdural empyema to detethering surgery. We report a child who received surgery for lipomyelomeningocele, developed wound complications and, unfortunately, sustained an intracranial subdural empyema. Moreover, we postulate the underlying mechanisms for this rare, but potentially devastating complication.

CASE PRESENTATION

The patient was a male who had been followed for an S-shaped gluteal crease since age 2 weeks. At age 3 months, a magnetic resonance imaging (MRI) L spine revealed a lumbar lipomyelomeningocele (2.7 cm rostrocaudal × 1.4 cm transverse × 1 cm anterior-posterior). There was absence of the posterior elements at S3, S4, and S5 vertebrae. The conus medullaris terminated at S2 [Figure 1]. On examination, he had good motor function of his lower extremities. At age 7 months, a voiding cystourethrogram revealed a smooth walled bladder without trabeculation or reflux; urethra appeared normal during voiding. However, formal urodynamic testing demonstrated over-reactivity of the detrusor muscle, with concern for a neurogenic bladder. At age 11 months, he underwent a lumbosacral laminectomy and detethering of his spinal cord with debulking of the lumbar lipomyelomeningocele.

Postoperatively, he exhibited his baseline neurological function. He was kept flat for 2 days, sedated with a dexmedetomidine drip in the Intensive Care Unit. Initial course was complicated by profuse diarrhea, emesis, and intermittent fevers up to 40.3°C, attributed to viral gastroenteritis. On 5 days later, the wound was draining CSF. Wound exploration revealed a CSF fistula at the lower third of the incision with no gross signs of infection. The child was started on vancomycin, cefepime, and flagyl. Cultures grew three aerobes (*Escherichia coli*, *Enterococcus*, and *Klebsiella*) and three anaerobes (*Clostridium*, *Veillonella*, and *Bacteroides*). Because of persistent fevers and CSF drainage 2 days later, a right frontal external ventricular drain (EVD) was placed, and the lumbar wound was reexplored. The leak emanated from the superior 1 cm part of the incision where devitalized tissue was evident; this was debrided to bleeding tissue and closed. EVD was left at 5 cm above his external auditory canal. He was kept flat bed rest, intubated and sedated. Unfortunately, the wound exhibited further wound drainage 4 days later and was again re-explored. Frank purulent discharge was immediately encountered as the wound fell open superiorly. The suprafascial space was cultured, which eventually grew *E. coli*. There was no pus extending from the epidural/subfascial space. He was kept sedated and paralyzed postoperatively for 3 days to allow for wound healing.

MRI brain performed because of persistent irritability showed extensive left-sided subdural empyema, greatest at the parietooccipital region and the inferior and anterior temporal lobe [Figure 2]. MRI spine was negative for obvious signs of infection. Via two separate linear incisions, two different craniotomies were performed along the left temporal and left parietooccipital region to evacuate the subdural empyema collections. No significant membranes were encountered, but fairly extensive firm and fibrous debris was encountered at both sites and were removed with copious irrigation/gentle pituitary work/gentle aspiration; cultures remained negative for aerobes and anaerobes. Subsequently, patient progressed well with reduced irritability. His lumbar incision continued to heal. Serial MRI brain and inflammatory markers were reassuring. He weaned off his EVD; CSF was tested daily since its insertion and remained negative throughout the

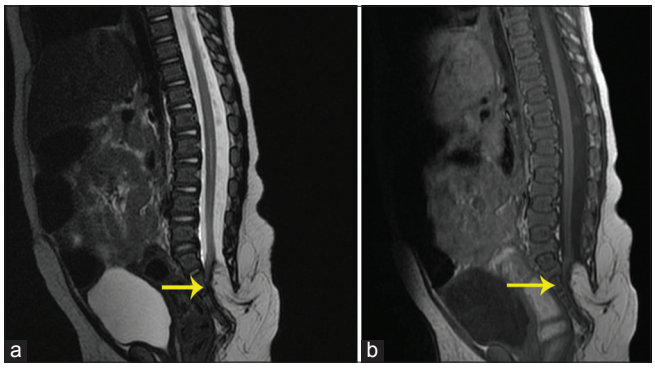

Figure 1: (a) Magnetic resonance imaging L spine T2 (arrow) and (b) T1 demonstrates lumbar lipomyelomeningocele (arrow)

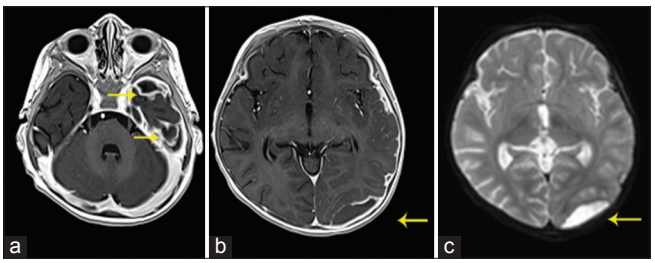

Figure 2: (a) Magnetic resonance imaging brain T1 with contrast demonstrates extra-axial rim enhancement along left anterior temporal (arrow) and (b) left occipital (arrow). (c) Left occipital collection demonstrates diffusion-weighted restriction (arrow)

hospitalization. He continued ampicillin, meropenem, vancomycin, and flagyl; once operating room (OR) cultures from the craniotomy were negative, vancomycin was discontinued. Ampicillin remained for enteroccocal coverage, meropenem for Gram-negative coverage, and flagyl for anaerobic coverage. At discharge, he remained on intravenous meropenem and enteral metronidazole to complete a 6-week course. Upon completion of his antibiotics, patient returned for a follow-up clinic visit; he exhibited no interim fevers and his cranial imaging [Figure 3] documented no evidence of a residual or recurrent subdural empyema. His lumbosacral wound was well healed.

DISCUSSION

Intracranial subdural empyema may cause headaches, fevers, altered mental status, motor deficits, and seizures.[9] Devastating consequences include epilepsy, hemiparesis, and death. The infection may result from cranial procedures, meningitis, sinusitis, otogenic infection, or trauma.[5,7,9,14,18,27,28] Mortality rates can be as high as 4.4–24%.[7,21,24,30] Failure to initiate treatment within 1–2 days may lead to a rapid decline toward coma and death.[3] Pathogenesis is contingent on the etiology. A sinus infection may spread along valveless veins existing between extracranial and intracranial structures; direct extension can also occur from an infected sinus or mastoid.[3] Furthermore, a cranial neurosurgical procedure (ventricular shunting or subdural drain placement) may seed bacteria in the subdural space or prompt a secondary infection of a distant subdural effusion.[6,16]

The spread of infection from a spinal location to a cranial location is rare. Lumbar punctures, spinal anesthesia, and infected intrathecal baclofen pumps have been associated with meningitis.[2,11] To the best of our knowledge, however, the spread of infection from a spinal location to a cranial location has not been reported to develop into subdural empyema. Perhaps the most suitable analogy is congenital dermal sinus tracts, which connect the surface of the skin to the central nervous system.[1]

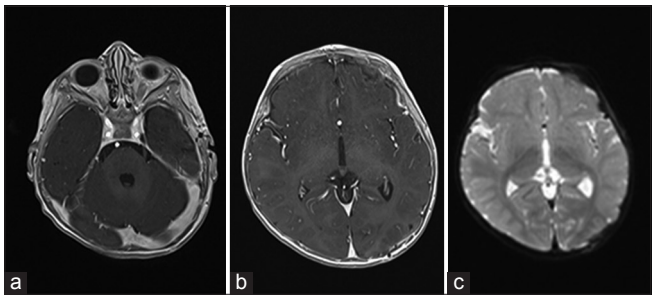

Figure 3: Magnetic resonance imaging brain T1 with contrast demonstrates resolution of subdural along left anterior temporal (a) and left occipital (b). Prior diffusion-weighted imaging signal has also resolved (c)

The pathology has been associated with meningitis[10,22,26] and intraspinal abscess (epidural, subdural, and intramedullary).[22,26] Moreover, Mount[22] reported two instances of intracranial infection (a cerebellar abscess and an infected dermoid cyst) secondary to dermal sinus tracts tracking to the occipital protuberance. Radmanesh *et al.*[26] and Emami-Naeini *et al.*[8] each reported a case with intracranial abscess from a spinal dermal sinus tract.

Our patient had several neurosurgical procedures: A right frontal EVD placement and the initial detethering surgery followed by three wound revisions. It seems reasonable that the etiology of the subdural empyema was from a wound infection after the initial surgery (secondary to wound drainage coupled with profuse diarrhea/wound contamination); the wound infection may seed the CSF space and travel to the cranium to form an empyema from the beginning. On the other hand, the persistent CSF leakage from the wound, compounded by EVD drainage, may induce formation of a left subdural hygroma/effusion, which was subsequently seeded with infection from the lumbar wound. The patient was kept flat for a prolonged period, which may facilitate the spread of infection to the cranium. Nevertheless, his MRI spine rostral to the surgical site did not exhibit any signs of infection. The polymicrobial bacteria from the cultures were consistent with a gastrointestinal source. Another possible source was EVD though its location was contralateral to the subdural empyema; moreover, the CSF was tested daily and remained negative throughout the hospitalization course. In addition, there were no signs of infection along the ventricular catheter on the MRI brain. On the other hand, only about 9% of CSF cultures are positive in patients with subdural empyema.[29]

The patient was on prolonged sedation/paralysis to encourage wound healing, but such a scenario limits the ability to assess for intracranial complications based on a physical examination. Given this rare complication, we recommend routine surveillance cranial imaging to rule out subdural empyema, as prompt diagnosis is associated with better outcomes.

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Conflicts of interest
There are no conflicts of interest.

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