Clinical Case Studies

Pediatric Group A streptococcal spinal epidural abscess presenting with recurrent symptoms of viral illness: An operative case report

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A B S T R A C T

Background: Spinal epidural abscess (SEA) in children is a rare condition with dangerous sequelae, and with only 22 other cases reported in the literature, treatment algorithms are poorly understood. Quick identification of the classic triad of sepsis, back pain and neurological deficit is critical. Source identification difficult and often cannot be identified. Reported pathogens include varicella-zoster virus, S. aureus, and S pyogenes.

Case description: We report a case of spontaneous pediatric SEA in a 22-month old female without obvious neurologic deficit, who underwent a T10-11 decompressive laminotomy and evacuation of abscess and subsequent 3-week course of intravenous ceftriaxone for culture positive S. pyogenes

Outcome: The patient showed marked improvement in symptoms after decompression. 5 weeks postoperatively after transitioning from intravenous ceftriaxone to oral cefin, the patient redeveloped a deep space infection and was taken back for a repeat debridement. The cultures from this procedure were negative and the patient was discharged on oral clindamycin.

Conclusions: Pediatric spontaneous SEA is a rare condition and early diagnosis and surgical intervention if indicated can prevent dangerous sequelae. Further studies into the surgical indications for decompression will aid in algorithmic decision making.

Background

Spinal epidural abscess (SEA) in children is an extremely rare, dangerous, and occult condition [1]. A total of 22 case reports of spontaneous pediatric SEA have been documented in the literature [1–20]. Expedient diagnosis, surgical decompression where indicated, and long-term targeted antibiotic therapy have been shown to be most effective [2,15,21].

The classic clinical triad of SEA (sepsis, back pain, and neurologic deficit) is uncommonly recognized in the pediatric population, which Unfortunately leads to misdiagnosis and further neurologic compromise upon presentation [15,22–26]. In the immunocompetent host, specific risk factors lead to either hematogenous or contiguous bacterial invasion of the epidural space: sickle cell disease; Hirschsprung’s disease; chronic intestinal pseudo-obstruction; varicella infection; cat scratch disease; skin, soft tissue, urinary, or respiratory tract infection; osteomyelitis; muscular abscess; or iatrogenic inoculation via invasive procedure (15% of cases) [1,16,27–29]. A bacterial source cannot be identified in up to 30-40% of reported cases, leading to the postulation of a silent bacteremia seeding the epidural space in these instances [16,27].

We report a case of spontaneous pediatric SEA in a 22-month-old female presenting with 1-week history of fevers, dry cough, back pain, and refusal to bear weight, without obvious neurologic deficit. The patient underwent a T10-11 decompressive laminotomy and evacuation of abscess, followed by 3 weeks of IV ceftriaxone for epidural tissue culture positive for Group A Streptococcus (S. Pyogenes).

Case description

History and physical examination

A 22-month-old female represented to our institution with a persistent, 7-day history of febrile malaise (41°C on presentation), painful bowel movements, abdominal pain, lower thoracic back pain, and refusal to bear weight. She had been assessed and discharged from the emergency department 4 days prior with similar symptoms. At that time and in the context of no focal findings, a presumptive diagnosis of non-specific viral illness was made. The parents were instructed to continue supportive care and return to the emergency department if there was no improvement. Of note, patient care transpired in its entirety prior to the COVID-19 pandemic.

Past medical history was significant for an episode of herpetic gingivostomatitis and viral bronchiolitis 2 months prior to presentation, treated successfully with acyclovir. Past surgical history was significant for repair of a facial laceration after sustaining a dog bite (domestic, vaccinated) 1 year prior. Patient was delivered at term via elective c-section, vaccinations were up to date, and she had reached normal developmental milestones for her age. Epidemiological history was significant for multiple domestic cats and dogs at the patient’s home, as well as an aunt with active herpetic cold sores. The patient’s paternal history was normal without any significant medical or surgical history.

Physical Examination revealed a well-nourished female child, with behavior appropriate for her age. She appeared lethargic and uncom-
portable laying on her back. She had tenderness to palpation around the thoracolumbar junction. She had no apparent motor deficits and sensory dermatomes were grossly intact to light and sharp touch. Clonus was absent, she had a negative crossed adductor response, and Babinski’s response was down going. She had no pain with active and passive range of motion of all joints in her lower extremities.

**Vital signs and clinical workup**

Her temperature was 41°C; respiratory rate 40 breaths/min; oxygen saturation 99% on room air, blood pressure 100/60 mmHg via blood pressure cuff; and heart rate was 174 beats/min.

Initial investigations failed to identify a source for a systemic inflammatory response. Chest radiograph was interpreted as normal, she had negative blood cultures initially and throughout the hospital course. A respiratory viral panel yielded negative results, pharyngeal sputum swab was negative and there was a negative urinalysis.

Laboratory evaluation yielded a white blood cell count: 7,500 mm$^3$ (68% neutrophils, 22% lymphocytes, 9% monocytes on differential). Inflammatory markers were significantly elevated with an erythrocyte sedimentation rate (ESR) of 73 mm/hr, a C-reactive protein (CRP) level of 92.5 mg/L and a procalcitonin level of 4.87 ng/ml. Basic metabolic panel showed an elevated alkaline phosphatase of 1710 IU/L.

Abdominal ultrasound was performed to rule out and acute abdominal process and yielded negative results. Because of persistent fevers and ongoing back pain gadolinium contrast-enhanced MRI of the cervical, thoracic, and lumbar spine were performed under anesthesia. Empiric antibiotic treatment with intravenous ceftriaxone was initiated along with subsequent spine surgery consultation.

**Imaging**

T2 STIR and T1 fat-suppression MRI with gadolinium contrast revealed a loculated lower thoracic epidural collection extending from T5-T11 (Fig. 1). The lesion was most prominent at the T9-10 level anteriorly on the left side, causing rightward displacement of the spinal cord. Of note, there was significant dilatation of the central spinal canal extending from T3-11, presumably due to the inflammatory environment.

There was no evidence of discitis or osteomyelitis. Incidentally, there was evidence of a contrast-enhancing fluid collection in the dependent portion of the left pleural cavity.

**Fig. 1.** T2 STIR MRI sequence with gadolinium enhanced contrast depicting a loculated epidural collection extending from T5-T11 with significant dilatation of the central spinal canal extending from T3-T11 without evidence of discitis or osteomyelitis.

**Fig. 2.** T2 STIR MRI sequence with gadolinium enhanced contrast depicting a loculated epidural collection extending from T5-T11 with significant prominence around T9-10 on the ventral and left side of the thecal sac, causing rightward displacement of the spinal elements. Incidentally, there was evidence of a contrast-enhancing fluid collection in the dependent portion of the left pleural cavity.

**Treatment and disease course**

The patient received IV ceftriaxone with marginal improvement in back pain and persistent fevers. A multidisciplinary team including pediatrics, infectious disease, orthopaedic spine surgery, and interventional radiology deliberated over the optimal treatment strategy with the need for a biopsy or aspiration to identify of the offending microbe and deliver targeted antibiotics in the context of negative blood cultures taking precedent.

Several additional factors were also considered. These included the loculated nature of the collection, the known relatively low yield of culture-positive results in the literature warranting optimal fluid and tissue samples, the significant technical challenges for percutaneous biopsy. Furthermore, the associated relatively high radiation exposure of interventional radiology and the opportunity for expedient source control favored surgical management. Treatment with an open surgical procedure was elected by the multidisciplinary team including the family.

Because the nidus of the epidural collection was located on the left side at the T9-T10 level, a left-sided posterior midline approach was utilized to expose the laminae of T9-10. We then proceeded with a left-sided T9 laminotomy, T10 laminectomy, and partial, medial facetectomy exposing the T9 and T10 pedicles. Markedly adherent and abnormal ligamentum flavum was encountered, dissected free from the dura, and sent for culture. The T9 nerve root was medialized, exposing a large loculated collection at the anterolateral edge of the dura, causing obvious displacement posteriorly and compression of the thecal sac. The loculation was incised and purulent material was expressed, which was sent for culture. Solid portions of the loculation were also sent for culture and pathology. After significant irrigation and further debridement of the abscess, the fascia, subcutaneous tissue, and skin were approximated in a layered fashion. The patient was awoken from anesthesia and transferred to the recovery area in stable condition without complication.

**Outcome**

Intraoperative cultures grew Group A beta-hemolytic streptococci (*Streptococcus pyogenes*). The infectious disease team elected to continue...
to treat with intravenous ceftriaxone therapy twice daily for 21 days via peripherally inserted central venous catheter. The patient showed marked improvement in back pain, became afebrile, and began to ambulate freely in the acute perioperative period. Post-operative inflammatory markers trended downward. On post-operative day four, ESR reduced to 65mm/hr (73mm/hr), CRP 8.6mg/L (92.5mg/L). The patient was deemed adequate for discharge with continued intravenous ceftriaxone therapy on post-operative day four.

At outpatient follow-up on post-operative day 24, markers improved significantly to WBC 6,600/mm³, CRP 0.4 mg/L, ESR 6mm/hr. Alkaline phosphatase also normalized to 282 IU/L. Clinically, the patient had no back pain, recurrent viral symptoms, or evidence of surgical site infection.

Approximately 5 weeks post-operatively, the patient was re-admitted for surgical site dehiscence 1 week after intravenous ceftriaxone was transitioned to oral cefdinir and MRI demonstrated a deep fluid collection without spinal cord involvement indicating either acute bleeding or an infectious process. The patient was afebrile with benign inflammatory markers. The patient was taken to the operating room for a revision irrigation and debridement deep through a laminotomy window at the same T9 level and no active purulent material was found. Culture from this procedure showed no growth, and patient was treated on a short course of IV cefazolin and discharged on oral clindamycin, and the patient recovered well (Fig. 3).

Discussion

Pediatric spinal epidural abscesses have been historically been associated with high rates of morbidity and mortality due to delayed diagnosis or misdiagnosis and non-specific symptoms at disease onset [15]. Streptococci represent 8-17% of the reported infecting organisms in pediatric SEA, with staphylococcus aureus accounting for 50-90% of cases, and gram negative bacteria 10-17% [2,21]. Bacteria gain access to the epidural space occurs via contiguous spread from adjacent foci of osteomyelitis or muscular abscess in 10-30% of cases, or by hematogenous dissemination from a remote source (i.e. skin or soft tissue, urinary, or respiratory tract infections) in 50% of cases.

Unfortunately, a bacterial source cannot be identified in up to 30-40% of cases [27,30,31]. Lack of source identification can lead physicians down dangerous diagnostic pathways, especially in a decompen-sating pediatric patient with pyrexia of unknown origin. Specifically, these patients often undergo lumbar punctures that are both low-yield and high-risk for intradural bacterial inoculation [13,16,32,33]. Laboratory inflammatory markers, blood cultures, abscess cultures, and advanced imaging modalities like gadolinium-enhanced magnetic resonance imaging have helped to guide effective diagnostic and therapeutic treatment practices for pediatric SEA, which can prevent the sinister sequelae of permanent neurological deficit [1,4,13–15,34]. Nevertheless, there is a lack of consensus regarding criteria for surgical intervention versus medical therapy alone [1,7,15,34]. This is ultimately due to the rare nature of pediatric SEA, with the reported incidence as 1.5 cases per 10,000 admissions from 2002-2011 [15]. This case report represents the 23rd published case of spontaneous pediatric SEA [1–20] which will hopefully add more data to aid in establishing diagnostic and therapeutic algorithms.

Our patient presented with a myriad of symptoms including fevers, painful bowel movements, abdominal pain, back pain, and refusal to bear weight. Over a two-month period prior to presentation, she also had multiple emergency department evaluations: sinusitis treated with amoxicillin for which she re-presented 10 days later with herpetic ginvostomatitis treated with acyclovir; viral bronchiolitis treated with supportive care; and constipation treated with fiber-rich diet and supportive care. This presentation correlates with the reported literature stating only 10-15% of cases present with the classic SEA triad of fever, back pain, and neurological deficits [2,34,35].

Darouiche et al. described four clinical stages of SEA in adults: Stage one is characterized by spinal pain, fever and local tenderness; in Stage two, radicular pain, nuchal rigidity and changes in the reflexes appear; in Stage three, motor weakness and bowel and bladder dysfunction arise; paralysis occurs in Stage four [28]. Deciphering localized back pain, nuchal rigidity, and radicular pain in the pediatric population is often difficult or impossible. Thus, it is critical to further investigate a child with back pain and a high fever for spinal pathology in the absence of alternative infectious sources.

Although a bacterial source cannot be identified in up to 30-40% of SEA cases, [27,30,31] specific risk factors have been reported in the literature. Varicella-zoster virus has been suggested to be a predisposing risk factor for SEA [16,26,33,36,37] due to the high prevalence of
associated skin and soft tissue superimposed bacterial infections (typically S. aureus or S. pyogenes). The pathophysiology of bacterial infection is attributed to skin barrier disruption and a transient viral-induced immunosuppression [16,38,39]. In our case, the patient was diagnosed with herpetic gingivostomatitis treated with acyclovir for a short period 2 months prior to admission. There are no reported cases discussing Herpes Simplex Virus (HSV) as a predisposing risk factor for pediatric SEA, and perhaps a similar skin-barrier disruption can occur, leading to silent bacteremia and seeding of the epidural space.

In this case, the child did present with viral bronchiolitis one-month prior which was treated with supportive care and on MRI there was evidence of a contrast-enhancing fluid collection in the dependent portion of the left pleural cavity, around the level of T7-8. Contiguous spread of bacterial pathogens has been described to occur in 10-30% of cases in pediatric SEA, but is typically from diskitis/vertebral osteomyelitis or adjacent psoas muscular abscess [27,30,31].

In the adult literature, there have been few reports of pleural empyema extending to the epidural space leading to an epidural abscess [40–45]. It may be possible that in our case, an indolent undiagnosed bacterial pneumonia lead to the development of a pleural empyema that through invasion of the fascial planes separating the parietal pleura from the epidural space lead to the presenting features of epidural abscess, but it may also be possible that the epidural abscess developed via silent bacteremia and subsequent abscess rupture lead to the development of the contrast-enhanced fluid collection in the adjacent pleural cavity. Outside of the history of viral bronchiolitis, the patient did not exhibit any clinical features suggesting empyema or bacterial pneumonia other than an elevated procalcitonin, which has shown specificity in community-acquired pneumonia but is non-specific in the setting of empyema [46,47]. These findings did not change the treatment algorithm for the patient but could possibly point to potential source of infection. Blood cultures were negative in up to 40% of reported cases of SEA, and were negative in this case as well [34]. Inflammatory markers were elevated however (ESR 73 mm/hr, CRP 92.5 mg/L), but differentiated complete blood panel showed a normal white blood cell count (7,500/mm³) albeit with a high percentage of neutrophils (68%). ESR has shown to be highly sensitive for SEA and can thus be used as a screening tool. However, as is in this case WBC has been shown to be normal in many pediatric SEA cases [24,36,48,49]. When the differential includes other infectious processes like myelitis, meningitis, or alternative parainfectious processes, previous authors have routinely performed lumbar puncture prior to imaging with poor diagnostic outcomes and a high risk of intradural bacterial inoculation [13,16,32,33]. In a cohort of patients with SEA, Rubin et al. performed lumbar punctures and found clear infectious signs in only 52% of cases, non-specific results in 38%, and normal CSF in 10% [13]. It can be concluded that the danger of intradural inoculation with lumbar puncture is high, and imaging should be obtained prior to invasive diagnostic procedures, especially in children. Gadolinium-enhanced magnetic resonance imaging, which carries no risk of radiation, has been shown to have a >90% sensitivity and specificity for SEA. This technology has allowed for earlier diagnosis and focused, appropriate interventions with antimicrobial therapy plus or minus or surgical intervention [1,4,13–15,34].

In our case, Gadolinium-enhanced MRI clearly identified the nidus of the epidural collection. The presence of a neurologic deficit in this setting would warrant emergent surgical decompression, but in the absence of neurological symptoms obtaining an abscess culture via any means remains paramount. CT-guided needle aspiration and culture by interventional radiology has been reported in the pediatric SEA literature as potentially offering both diagnostic and therapeutic benefit [1,15]. Hawkins et al reported on 9 pediatric SEA cases from 2002-2011 in which 2 patients underwent surgical decompression and 4 undergoing CT-guided needle drainage with successful outcomes in all patients without post-operative neurological deficits [15]. Auletta et al reviewed 8 pediatric SEA cases from 1984-1999: 6 received surgical drainage, 1 received CT-guided needle drainage, and 1 received no surgical inter-

vention. There were no deaths in the series, and only 2 patients had minor neurologic sequelae [1].

Because SEA is more likely to develop in a larger epidural spaces rich in fat, abscesses are more common dorsal to the thecal sac, especially in children, [1,16] In these cases, CT-guided needle drainage is a viable option, but ventral, lateral, and intra-foraminal abscesses render percutaneous interventions technically challenging and potentially dangerous. [1,29,35] Moreover, the adult literature has shown that there is a risk of neurological deterioration with conservative treatment with antibiotics alone, even in those without neurologic deficits [1,50,51]. Despite targeted antibiotic therapy, 19–23% of adult patients have developed worsening neurological symptoms in the absence of surgical intervention [35,52]. Therefore, in the absence of a bacterial source, surgical decompression and debridement followed by targeted antibacterial therapy of a localized epidural abscess, especially one that is more ventrally located, is warranted regardless of the neurological status of the pediatric patient. Certain contraindications for surgical intervention must be ruled out, which have been described in the adult literature: panspinal epidural abscess, >48-72 hours of neurological deficit or paraplegia, and extensive comorbid conditions that would render surgical intervention fatal [15,24,25,53,54]. Furthermore, there is a lack of consensus regarding antibiotic duration. The reported literature suggests a mean duration of 4-6 weeks especially in cases without surgical intervention, but there is significant variability amongst infection disease providers.

In conclusion, pediatric SEA is difficult to identify and easily misdiagnosed. Once an epidural abscess is discovered, surgical intervention should be considered highly, with the goals of decompressing the collection, isolating the infectious organism, and ensuring eradication. The paucity of literature in pediatric SEA has led to controversial conclusions regarding antibiotic therapy alone versus both surgical decompression and targeted antibiotic therapy. Future studies should aim to establish an algorithm based upon the given literature in order to prevent delays in diagnosis and permanent neurological sequelae in pediatric patients with spinal epidural abscesses.

Declaration of Competing Interest

None.

Patient Informed Consent Statement

The authors declare that informed patient consent was taken from all the patients.

Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:10.1016/j.jxlsj.2021.100067.

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