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Case Report

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Intramedullary spinal abscess associated with intramedullary dermoid and infected dermal sinus:case report and review of the literature

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Abstract

Intramedullary spinal abscesses are extremely rare. Most occur secondary to cardiopulmonary spread;contiguous origin is less frequent. Few intramedullary spinal abscesses have been reported secondary to dermal sinus tracts. A dermoid sinus is a congenital lesion (closed simple dysraphism) with rare incidence.Dermoid sinuses can ease local invasion and spinal infections (meningitis, intradural extramedullary and intramedullary abscesses), usually with thoracolumbar involvement.

Here we describe acase of a 20-month-old boy who presented with fever and refusal to walk 2 days prior to admission. On examination of the lumbar spine, a small sinus with skin stigmata was noted.Neurological evaluation revealed paraparesis more prominent on the left side, reduced tone and reflexes, left leg hypoesthesia,reduced anal tone, and urinary retention.MRI scan demonstrated intramedullary abscess extending from L2 to S1 level.

The patient was urgently admitted for drainage of intramedullary abscess and excision of the dermal sinus tract. Proper antibiotic treatment was completed for 6 weeks with gradual improvement and ultimately full ambulation ability.

Keywords: Dermal Sinus, Intramedullary Spinal Abscess.

Introduction

Congenital dermal sinustract is a rare type of neural tube defect (dysraphism) which results from incomplete separation of epithelial ectoderm and neuroectoderm. It is estimated to occur with an incidence of 1/2500 live births. Dermal sinus tracts have epithelial-lined tracts with 1-2 mm of skin openings, associated with cutaneous stigmata (hair, pigmentation, dimples and hemangioma) [1-4] .

Intramedullary spinal cord abscesses (ISCA) are rare central nervous system infections in children [5]. First described by Hart in 1830,fewer than 150 cases have been reported since then [6,7]. Fewer than 2/3 of cases of ISCA occur among the pediatric age group [7]. Spinal cord abscesses may affect any level of the spinal cord and even may spread to include the total spinal axis(holo-cord). Nevertheless thoraco-lumbar region remains the most frequently involved site,and few reports described holo-cord abscess abscess [8,9]. The most frequently reported risk factors for ISCA include and not restricted to congenital heart disease, immune system disorders, long-term use of vascular access devices, spinal cord tumors, dermal cyst and dermal sinuses, with the latter being the most common cause among spinal dysraphism with an incidence of 25-75% [8,10].

Case

Clinical presentation

A 20 month-old-boy with normal development and no prior admissions, presented to the emergency department due to fluctuating fever for the past two weeks with an absolute refusal to walk 2 days prior to his referral.At the time of arrival, the

patient's temperature was 36.6°C. A primary physical evaluation at the emergency department revealed a poorly compliant and uncooperative child with slight spontaneous movements of the lower limbsand lefthip tenderness. A pelvic and hip joints XR were normal.Blood culture and complete blood count were performed; the latter showed WBC-15K with left shift, PLT-506K and ESR was 46.A more thorough physical examination revealed a small sinus with extending hair and a hemangioma was noted on examination of lumbar spine.No erythematous or cellulitis changes were noted. Neurological examination showed normal upper limbs motor power while lower limbs were para-paretic, with the left limb affected more so than the right limb and distal more than proximal.Additionally,there was reduced tone and reflexes, left leg hypoesthesia, reduced anal tone, and urinary bladder rest catheterization was positive for urinary retention (>400cc).

Radiological investigation

Prompt MRI scan was performed, T1weighted image showed a markedly enlarged conus medullaris filling the spinal canal. T1 with contract showed L2-S1 hypointense loculated lesion with peripheral enhancement strongly suggesting intramedullary abscess, accompanied with an enhanced dermal sinus extending caudally. The spinal cord appeared tethered down to the S1 level (Figure 1).

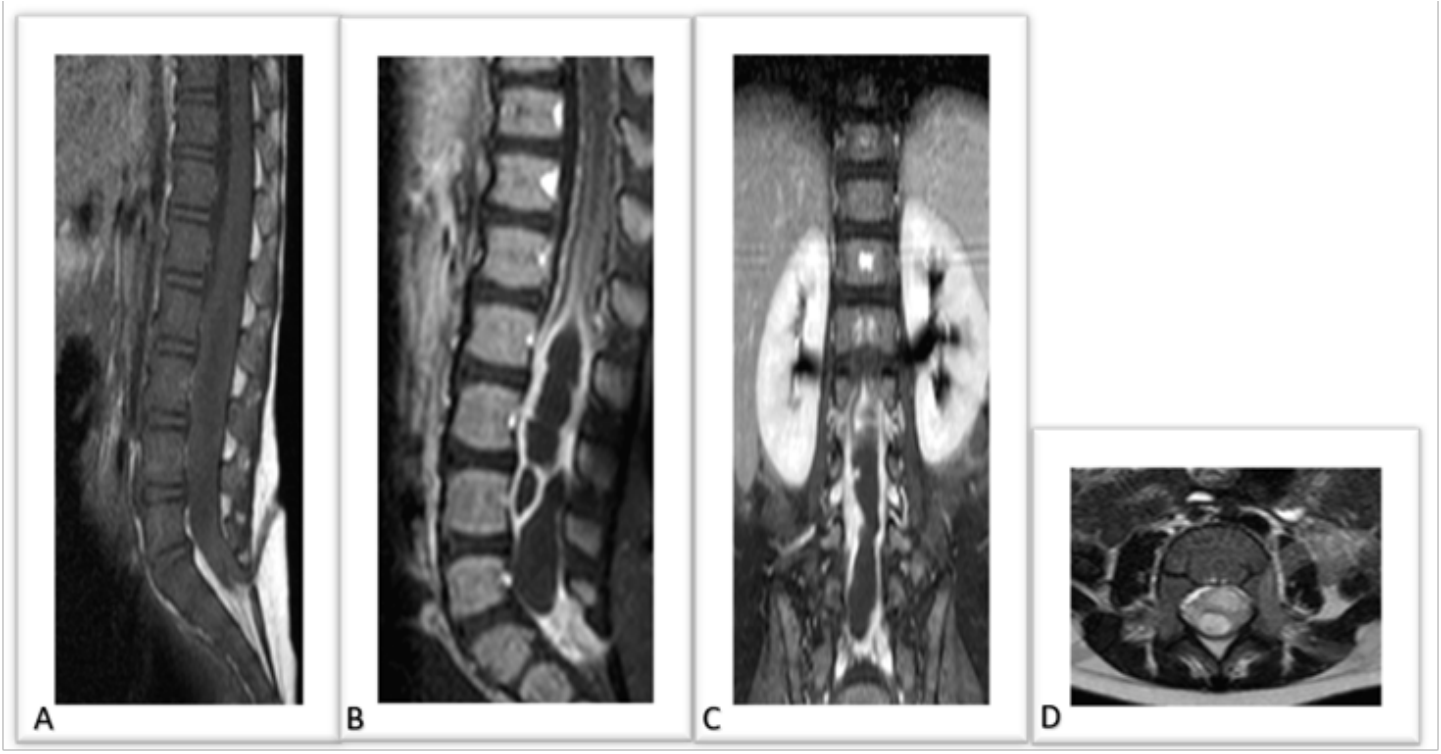

Figure 1: A) sagittal T1 MRI; B) sagittal T1 MRI with contrast; C) coronal T1 MRI with contrast; D) axial T1 MRI with contrast.

Neurosurgical Intervention

The patient underwent urgent neurosurgical intervention. A midline skin incision was preformed from L1 to S2, encircling the dermal sinus ostium(figure 2). The sinus tract was followed and traced from L5 to S1, where it split around S1and continued to course caudally.Trapdoor laminotomy was performed at L2 through L5, with clean epidural space encountered (figure 3). Durotomy was performed around the sinus, with no pus or adhesions observed in the subdural space andthe conus medullaris was found to be bulging and tense with the sinus attached to its lower end (figure 4). A median myelotomy incision on the conus released pus within the medullary abscess cavity which also contained pearly, flaky tissue and hair (figure 5). After aspiration, dissection of the cyst and the sinus root was performed (figure 6), followed by untethering of the filum terminale, and tight water closure of the of dura, subcutaneous tissue, and skin-layer by layer. Prompt broad spectrum antibiotics treatment initiated.

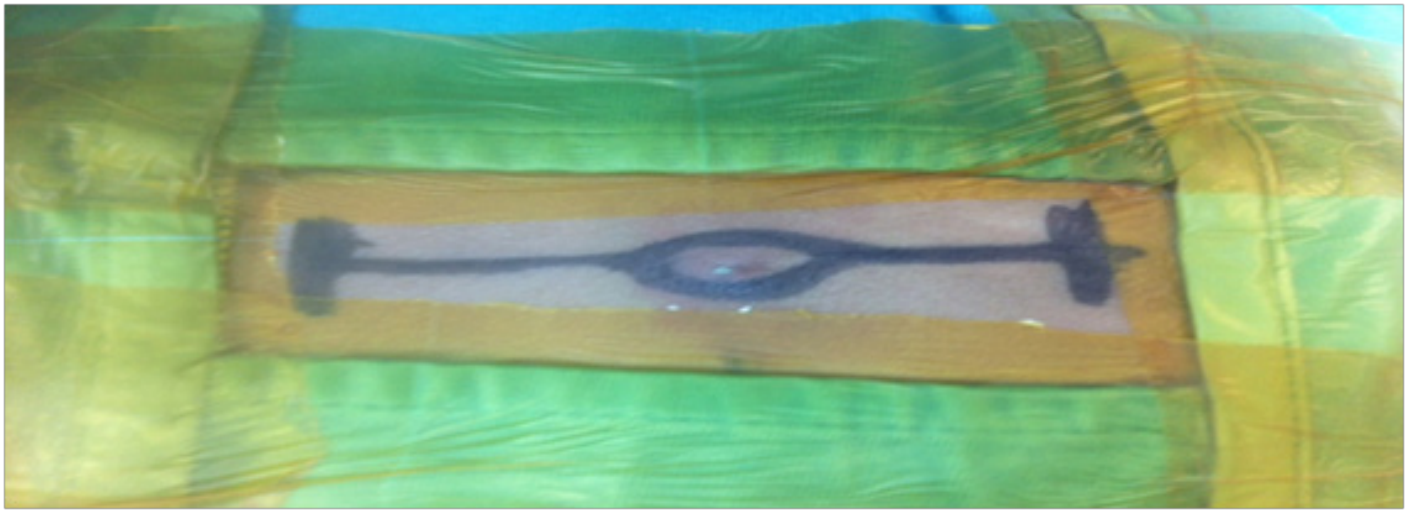

Figure 2: Planed L1-S2 midline skin incision, encircling the dermal sinus ostium.

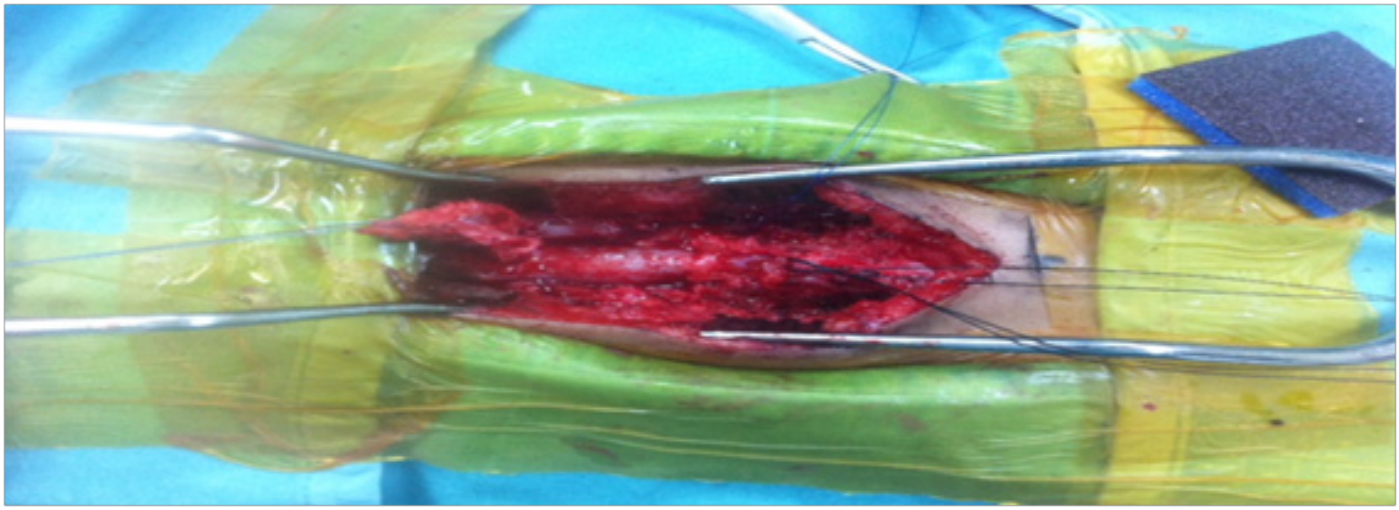

Figure 3: L2-L5 Trapdoor laminotomy with clean epidural space.

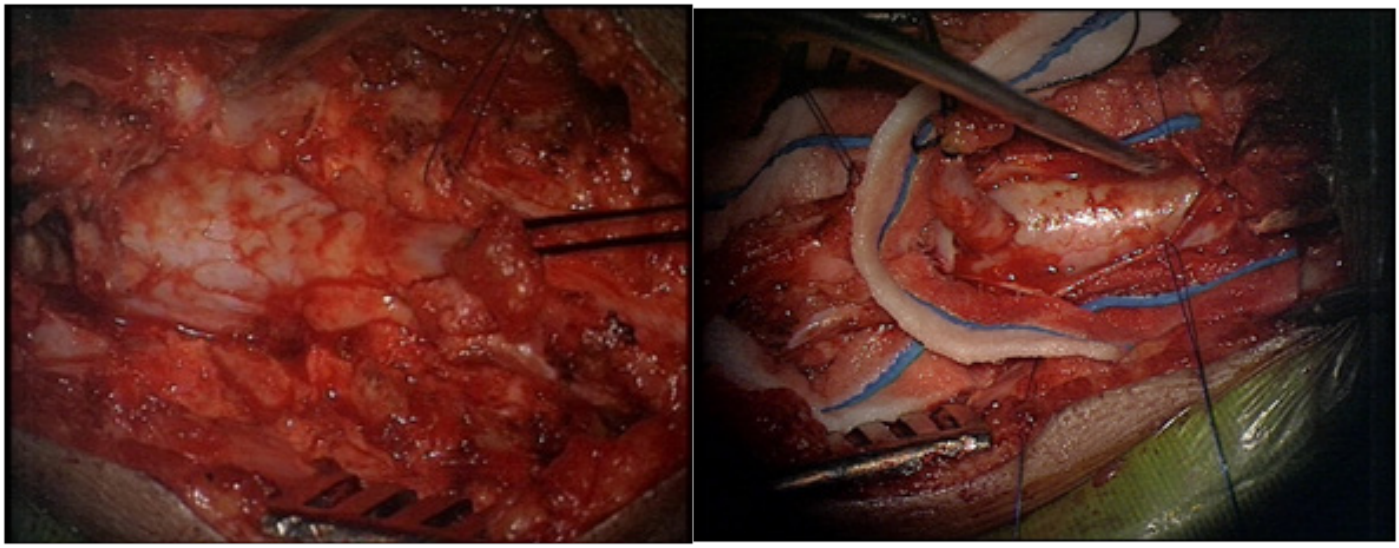

Figure 4: Peri-sinus durotomy was performed, neither subdural pus or adhesions were observed , conus medullaris was found to be bulging and tense with the sinus attached to its lower end.

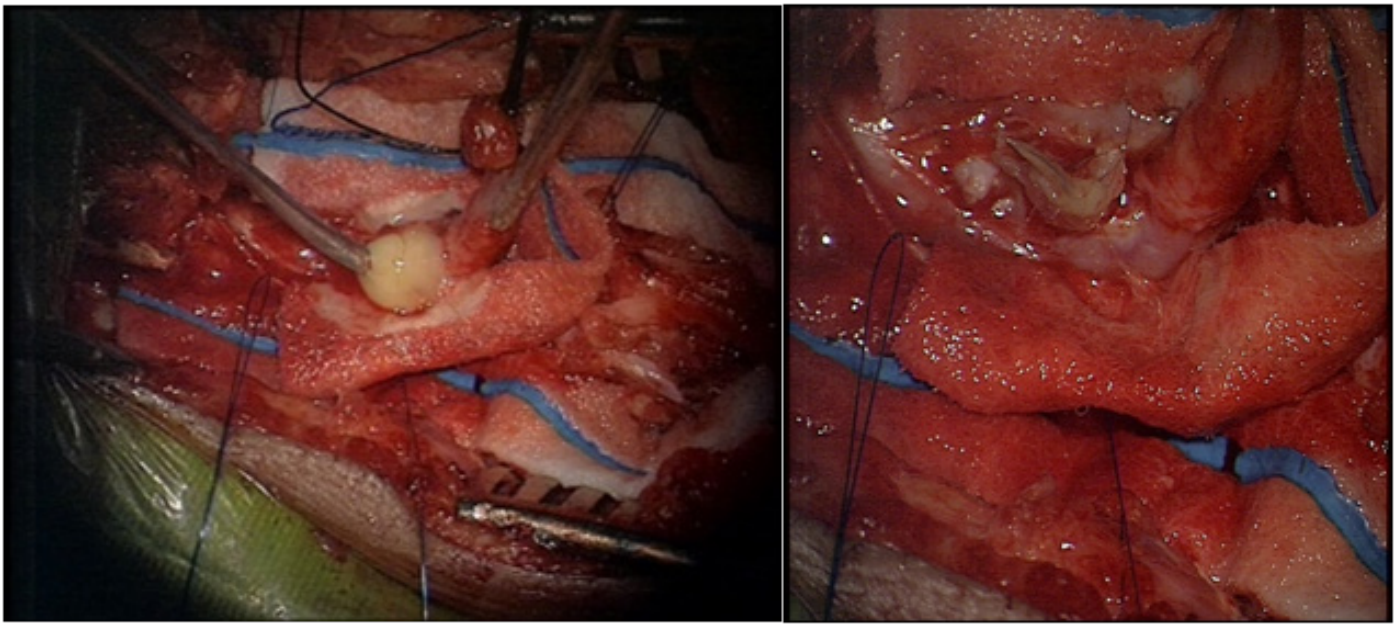

Figure 5: A median myelotomy incision on the conus released pus within the medullary abscess cavity (also contained pearly, flaky tissue and hair).

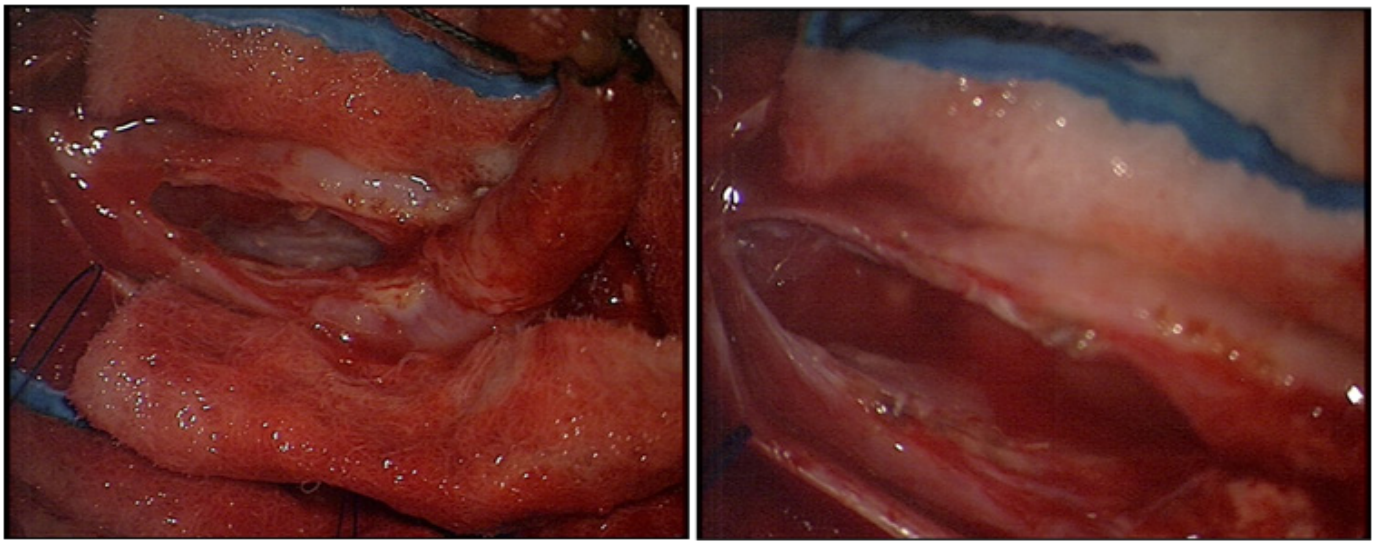

Figure 6: After cyst cavity aspiration, dissection of the cyst and the sinus root was performed.

Histopathological-Microbiological Findings

Post-operative microbiology laboratory culture was positive for sensitive Klebsiella species growth and narrow spectrum specified antibiotics treatment established.

Histopathological results showed hyperkeratotic congested skin tissue with skin appendages and residual dermal sinus lined with squamous epithelium, granulation tissue and residual epithelial glands associated with acute on top of chronic inflammatory process, luminal occlusion and epithelial necrosis.

Post-Operatively

Post-operative radiological follow up confirmed significant resolution following 6 weeks of intravenous antibiotics treatment (figures). Additionally, on neurological examination the patient was able to ambulate with left sided foot drop, hypoesthesia, and had gradual resolution of neurogenic bladder.

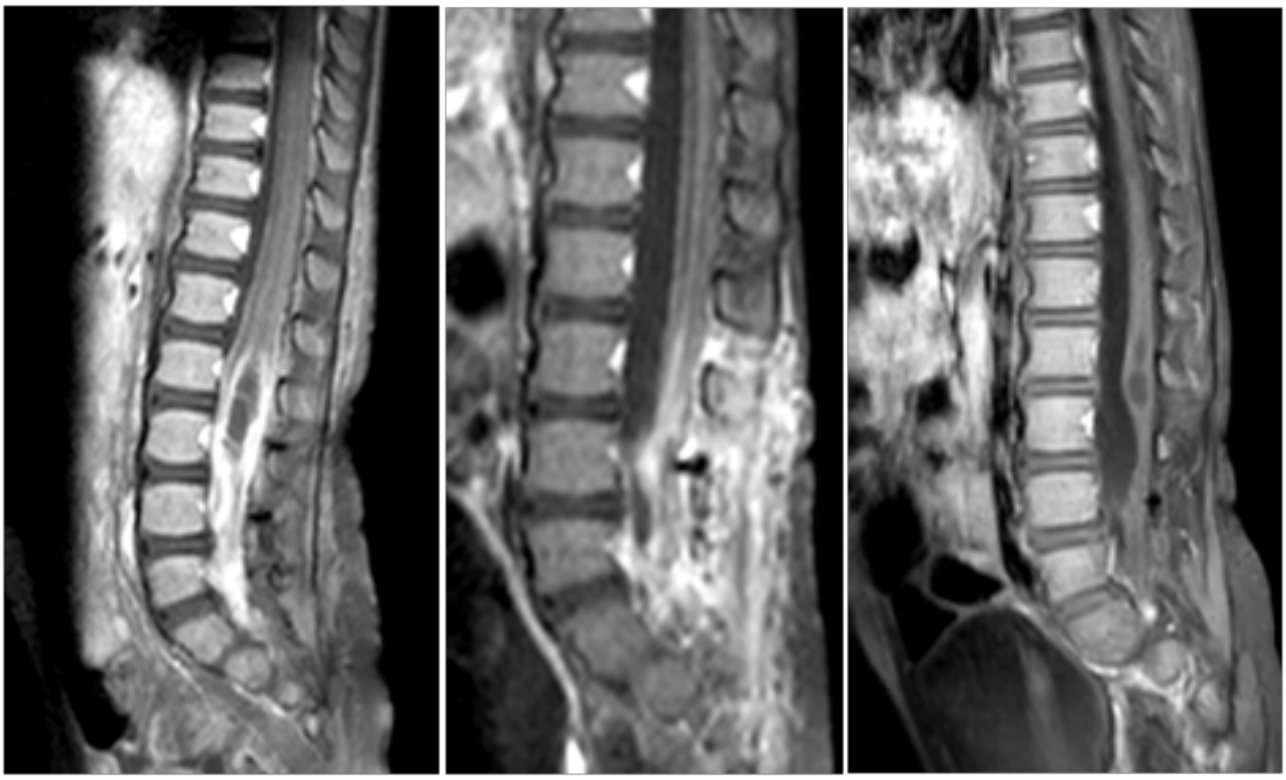

Figure 7: Sagittal T1 MRI with contrast day 1 (A), 19(B), 45(C).

Discussion and Conclusion

Intramedullary spinal cord abscess (ISCA) is a rare pyogenic spinal cord infection of the central nervous system, especially in children [10]. The most well-known etiology is due to hematogenic spread, however, contiguous spread due to dermal sinus can be high as half the cases [11-13].Congenital dermal sinus (CDS) is an uncommon form of spinal dysraphism, that can be significantly morbid if left untreated, leading to CNS infections ranging from recurrent meningitis to holo-cord abscesses [8,9,10,14].No reported survivors among patients who did not undergo surgery. Since the introduction of antibiotics mortality has significantly reduced,however the majority of survivors complain of significant subsequent neurological compromise [10].While almost a third of cases of ISCA reveal sterile cultures, various microorganisms have been isolated, including and not restricted to: Staphylococcus, Streptococcus pneumonia, Hemophilus, Proteus, Listeria,Actinomyces, Pseudomonascepacia and Mycobacterium tuberculosis [15].High clinical suspicion is crucial for diagnosis;the absence of the ISCA triad off ever, pain, and neurological compromise,does not exclude the diagnosis [16]. In two case series the median age reported was 15 and 26 months, with 10 and 4 cases reported respectively. A female predominance and acute presentation within 1-3 days was noted in the 26 month old case series [1,7,8].Prompt proper radiological and surgical intervention remans the gold standard and the only way to achieve good neurological outcomes among surviving patients [7,10,13]. Recurrence of ISCA was associated with about 25% of the cases [18]. In our case a subacute febrile presentation with noticeable neurological deficit led to unsatisfactory musculoskeletal diagnosis. Further evaluations and investigations led to the correct diagnosis. Prompt neurosurgical intervention and antimicrobial treatment resulted in a satisfactory neurological outcome.

For conclusion, an acute febrile disease associated with acute or rapid onset neurological compromise,regardless of pain, and that is associated with spinal defects should be investigated with proper radiological imaging to rule out ISCA and treated accordingly and immediately if proved to be.

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