Long-Term Outcomes of Conservative Management for Pyogenic Myelitis

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Radiology Case. 2026 January; 20(1):1-9 :: DOI: 10.3941/jrcr.6030

AUTHORS' CONTRIBUTIONS

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DISCLOSURES

All the authors have indicated that they have no financial relationships relevant to this article to disclose.

CONSENT

Yes, oral informed consent was obtained from the patient for the publication of this case report and the accompanying images.

HUMAN AND ANIMAL RIGHTS

The procedures were conducted according to the Declaration of Helsinki and the World Medical Association regarding human experimentation developed for the medical community.

ABSTRACT

Intramedullary spinal cord abscesses are uncommon, with only a few documented cases. They typically present with lower-limb neurological symptoms and signs of sepsis, often leading to misdiagnosis, particularly in children, where symptoms may be atypical. The present case report details the distinctive presentation of a 4-year-old boy with recurrent urinary tract infections who subsequently developed lower-limb weakness. Initially, his symptoms were attributed to recurrent urinary tract infections, delaying neurological evaluation. However, magnetic resonance imaging revealed a diffuse intramedullary spinal cord abscess extending from the cervical to lumbar regions, linked to an S2–S3 congenital dermal sinus tract. These uncommon congenital dermal sinus tracts promote infections, often leading to overlooked abscess formation in children. Magnetic resonance imaging confirmed whole-spine involvement and follow-up imaging revealed significant resolution with residual fibrotic changes.

CASE REPORT

BACKGROUND

This case report highlights an exceptionally rare presentation of holocord pyogenic myelitis secondary to a congenital dermal sinus, a diagnostic and therapeutic challenge in pediatric patients. Almost all cases described in the literature require surgical drainage and antimicrobial therapy; however, our case uniquely demonstrates complete resolution through conservative management alone. Imaging plays a pivotal role in the early diagnosis and differentiation of intramedullary spinal cord pathologies and excluding mimickers. This case also broadens the clinical understanding of the natural course of intramedullary abscesses, emphasizes the variability in presentation.

CASE REPORT

A 4-year-old boy with a history of malodorous urine and recurrent urinary tract infections (UTIs) presented. The mother noted that these episodes commenced at 18 months of age

and were associated with urinary retention. She also observed limping and gradual asymmetry in the lower limbs, which progressed over time. Notably, the patient had no history of fever, weight loss, seizures, or cognitive impairment. The child was fully immunized, and his developmental milestones were within normal limits.

Upon examination, the patient exhibited no dysmorphic features. Nevertheless, lower-limb asymmetry and muscle weakness were noted. The absence of the left ankle reflex was observed, and the child exhibited a hemiplegic gait. Additionally, two dimples were identified on the lower back (Figure 1), along with skin discoloration that the parents had previously missed as they were clinically asymptomatic.

Upon further inquiry, the mother reported that the urinary tract symptoms were treated with antibiotics in an emergency setting. A neurologist was consulted to assess the lower-limb symptoms, and an enhanced magnetic resonance imaging (MRI) was recommended.

Image Findings

The MRI findings (Figures 2,3) revealed widespread swelling of the spinal cord, with high T2 signal intensity extending to the medulla oblongata. Initially, this was interpreted as a holocord astrocytoma.

The lesion exhibited low signal intensity on T1-weighted images, heterogeneous multicystic hyperintensity on T2-weighted images, and thick-rim enhancement after contrast administration. Furthermore, a fistulous dermal sinus tract (Figure 4) extended from the skin dimple and communicated intraspinally at the S2–S3 level. This sinus tract exhibited T1 and T2 hypointensity, along with linear enhancement on postcontrast imaging.

Management

The patient was managed conservatively without the need for antibiotics, surgery, drainage, or invasive procedures. The patient is currently on temporary catheterization, as recommended by his urologist, for behavioral therapy. He uses the catheter mainly during long trips and at school. Furthermore, there has been a notable enhancement in his bladder adaptation and control, resulting in a reduced need for catheterization.

An essential component of his management involved the discontinuation of diaper use, resulting in a significant improvement in urinary function, including reduced episodes of UTIs and urinary retention.

Follow-up

During follow-up, the patient exhibited significant improvement. The patient's neurological symptoms were completely resolved without any residual deficits. Furthermore, the patient no longer displayed urinary symptoms.

One year ago, his family observed a foot deformity and consulted an orthopedic specialist, after which he underwent tendon-lengthening surgery.

Imaging findings (Figure 5) confirmed nearly complete resolution of the spinal lesion, with residual fibrotic changes observed in the thecal sac. These changes were subsequently confirmed by a Computed Tomography (CT) scan to be a non-ossified structure. The patient remained entirely asymptomatic and resumed his regular daily activities.

DISCUSSION

Etiology and Demographics

Intramedullary spinal cord abscesses are rare [1]. The initial description of intramedullary spinal cord abscesses was provided by Hart in 1830. Subsequently, this infrequent condition has been reported in fewer than 120 cases, highlighting its rarity and the complexities linked to its diagnosis and management [2–6]. Common symptoms include weakness, motor deficits, and signs of infection; however, typical signs of infection, such as fever, may be absent [3,4,7]. In children, the symptoms can be particularly uncharacteristic, further complicating the diagnosis [3,8]. In rare instances, patients may exhibit atypical symptoms that obscure diagnosis. One such unusual presentation is recurrent UTIs, which may divert attention from the underlying spinal pathology [9].

Most intramedullary abscesses are localized, and cases involving the entire spinal cord are rare. Such extensive involvement can pose diagnostic challenges, often resembling a holocord tumor and complicating both clinical management and treatment decisions. In this report, we describe a distinctive case of a whole-spine intramedullary abscess in a patient initially presenting with recurrent UTIs.

The dermal sinus tract is a rare congenital malformation resulting from the incomplete closure of the neural tube during embryogenesis, affecting approximately 1 in every 2500 live births [10,17]. This epithelium-lined channel connects the skin to deeper neural structures and is predominantly located in the lumbosacral region [10,11]. It is a rare formation lined with epithelial cells that can terminate at various depths, ranging from the subcutaneous layers to the thecal sac. These tracts may also be associated with other pathologies such as lipomyelomeningocele, myelomeningocele, split-cord malformation, tethered cord, filum abnormalities, and inclusion tumors [12]. Most patients are in the pediatric age group. These tracts often act as conduits for infections, potentially resulting in complications such as meningitis or abscess formation— including intramedullary spinal cord abscesses [13]. If left untreated, these conditions can lead to progressive neurological deficits [13,14]. Meningitis is the most common septic complication, whereas intramedullary abscess is the least common [14].

Additionally, by reviewing the literature, we identified a case of an initially asymptomatic patient with a dermal sinus who developed an infection that progressed to paraplegia due to secondary myelitis. This infection can affect the spinal cord and nerves, potentially triggering myelitis. This pathway explains how an undetected dermal sinus can lead to secondary myelitis, highlighting the potential for serious infections [15]. Notably, in our case, the patient's symptoms were associated with an infected dermal sinus, leading to inflammation of the spinal cord and subsequent myelitis, thereby elucidating the neurological findings.

Clinical and Imaging Findings

A dimple or sinus opening is usually found at the midline, appearing as a small cutaneous defect in or near this region. Owing to its benign nature, this feature is often overlooked by primary care physicians. Patients may initially be asymptomatic or may present symptoms such as backache, bowel and bladder dysfunction, paraparesis, or paraplegia. In some cases, the opening may become infected, leading to symptoms such as fever, neck stiffness, seizures, and sudden neurological deficits due to secondary complications [1]. Despite the potential severity of this condition, diagnosis is frequently delayed due to the misinterpretation of subtle symptoms [14,16]. Moreover, individuals with dermal sinuses may exhibit a range of neurological symptoms, including motor weakness, urological issues, pain, and bony deformities [17].

In the present case, the child initially experienced recurrent UTIs with urinary retention and was solely treated with antibiotics by a local physician. However, suspicion of neurological involvement arose only after the child developed lower-limb weakness and asymmetry, prompting the recommendation for an MRI. In a previous study, leg deformity and thinning and shortening of the leg were observed during clinical examination [17]. This feature was also observed in our patient, who later developed leg shortening. These findings reinforce the importance of early recognition and evaluation of clinical signs to prevent complications.

MRI is the preferred neurodiagnostic procedure for evaluating the spinal column and cord [18–20]. MRI allows for the assessment of vertebral column involvement; detection of epidural and subdural infections; identification of dermal sinus, epidermoid, or intramedullary tumors; and evaluation of spinal cord abnormalities [20–22]. Typical MRI findings include spinal cord expansion with hyperintense signals on T2-weighted images, while contrast-enhanced T1-weighted sequences demonstrate peripheral rim enhancement, outlining the extent of the intramedullary cavity.

Treatment and Prognosis

The management of congenital dermal sinuses, particularly in complicated cases, necessitates surgical excision of the sinus tract to prevent or treat infections and neurological deterioration. For individuals with ongoing infections, such as meningitis or intramedullary abscesses, combining antibiotic treatment with surgery can yield excellent outcomes [23]. However, in the present case, spontaneous resolution without surgical or antibiotic intervention was observed, representing an uncommon clinical course. Supportive measures, such as urinary catheterization and strict hygiene practices, helped manage the symptoms of our study patient and prevent further complications.

Differential Diagnoses

The differential diagnosis for a holocord intramedullary abscess is wide and includes both infectious and non-infectious etiologies. Given the rarity of this entity, considering other conditions that may present with similar clinical features and overlapping MRI findings, particularly those causing diffuse or longitudinally extensive spinal cord involvement, is crucial. Among the most important mimickers are syringohydromyelia, holocord astrocytoma, and acute disseminated encephalomyelitis (ADEM). Syringohydromyelia typically presents with progressive motor weakness and sensory dissociation, particularly in children with associated congenital anomalies such as Chiari malformation. MRI demonstrates a non-enhancing cerebrospinal fluid-intensity cavity without surrounding edema or diffusion restriction, which helps to differentiate it from an abscess.

In contrast, holocord astrocytoma is more common in the pediatric population and presents with slowly progressive neurological deficits. On MRI, the lesion is usually eccentric and poorly demarcated, with fusiform expansion of the spinal cord. Approximately 40% have a cystic component, particularly in pilocytic subtypes, and the solid tumor component typically enhances after contrast administration. **ADEM** is characterized by an acute or subacute onset, usually after a recent viral infection or vaccination. Its clinical features include multifocal neurological deficits and altered mental status. MRI findings show multifocal, T2 hyperintense lesions in the brain and spinal cord, typically with patchy and asymmetric distribution. These lesions are non-expansile and lack ring enhancement or diffusion restriction, which helps differentiate them from intramedullary abscesses.

Conclusion

Failure to identify the congenital dermal sinuses could lead to serious infections of the central nervous system. A delayed diagnosis, as demonstrated in this case, may lead to severe consequences such as intramedullary spinal cord abscesses. MRI is crucial for diagnosing and assessing congenital dermal sinuses and their associated complications. It aids in determining the severity of spinal abnormalities, the presence of sinus tracts, and any related congenital malformations while also distinguishing them from conditions such as malignancies.

TEACHING POINT

Pyogenic myelitis is a rare but serious condition that requires early recognition and appropriate management to prevent

permanent neurological deficits. In pediatrics, congenital dermal sinus is the most frequent underlying cause. MRI typically reveals spinal cord swelling, intramedullary hyperintensity on T2-weighted images, and sinus tracts, aiding in differentiation from other myelopathies and guiding treatment decisions.

QUESTIONS

Question 1: What imaging characteristics are seen in intramedullary spinal cord abscesses on MRI scans?

A) Hyperintense lesion on T1-weighted images without contrast
B) Low signal intensity on T1-weighted images (applies)
C) High signal intensity on T2-weighted images (applies)
D) Ring enhancement after contrast administration (applies)
E) Normal MRI findings in all cases

Explanation:

Typical MRI features include low and high signal intensities on T1- and T2-weighted images, respectively. Contrast administration also typically reveals ring enhancement.

Question 2: Which clinical symptoms are commonly associated with infected myelitis?

A) Progressive limb weakness (applies)
B) Fever (applies)
C) Hyperreflexia in early stages
D) Sensory deficits (applies)
E) Bowel and bladder dysfunction (applies)

Explanation:

Infected myelitis commonly presents with progressive limb weakness, sensory deficits, and autonomic dysfunction, including bladder disturbances. Fever is usually present due to the infectious nature of the disease. However, hyperreflexia is not an early finding since acute spinal cord involvement typically causes hyporeflexia or areflexia.

Question 3: Which of the following statements about congenital dermal sinus are accurate?

A) It is a rare form of spinal dysraphism with an incidence between 1 in 2000 and 3000 live births (applies)
B) It occurs due to abnormal separation between neuroectoderm and surface ectoderm during fetal development (applies)
C) Most commonly occur in the cervical spine region
D) It can serve as an entry point for pathogens causing central nervous system infections (applies)
E) All patients show bacterial growth in cultures obtained from abscesses

Explanation:

Congenital dermal sinus is a rare condition that develops due to abnormal adhesion or incomplete separation between neuroectoderm and surface ectoderm. It can serve as a portal for bacterial entry, causing central nervous system infections and most commonly occurs in lumbar and sacral areas rather than the cervical area, and approximately 30% of cultures may be sterile.

Question 4: What are the possible complications of a congenital dermal sinus if left untreated?

A. Meningitis (applies)
B. Spinal intramedullary abscess (applies)
C. Hydrocephalus
D. Permanent neurological deficits (applies)
E. Neurogenic bladder (applies)

Explanation:

Untreated congenital dermal sinus can lead to life-threatening central nervous system infections, such as meningitis and spinal cord abscesses, and typically result in permanent neurological damage.

Question 5: Which treatment approach is typically recommended for congenital dermal sinus cases with infectious myelitis?

A) Conservative management without intervention
B) Antibiotic treatment alone
C) Surgical excision of the sinus tract combined with antibiotics (applies)
D) Radiation therapy
E) Corticosteroid therapy only

Explanation:

The recommended treatment generally includes surgical excision of the dermal sinus tract along with antibiotic therapy to manage or prevent further infections and neurological damage.

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FIGURES

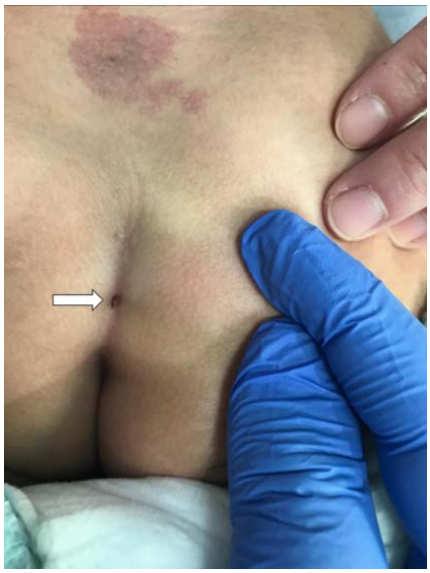

Figure 1: A 4-year-old boy with recurrent urinary tract infections who subsequently experienced lower-limb weakness due to pyogenic myelitis secondary to congenital dermal sinus.
FINDINGS: Sacral skin dimple (arrow), which is located above the gluteal folds.
TECHNIQUE: Photograph

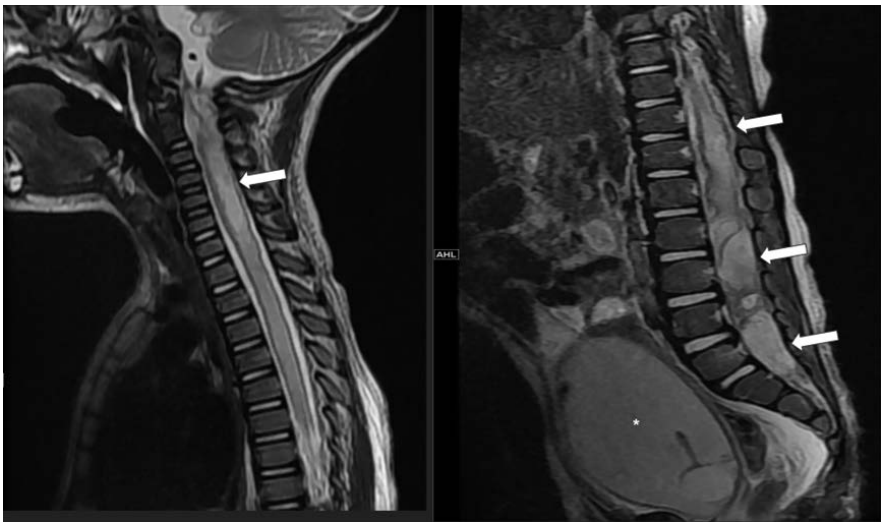

Figure 2: A 4-year-old boy with recurrent urinary tract infections who subsequently experienced lower-limb weakness due to pyogenic myelitis secondary to congenital dermal sinus.
FINDINGS: A heterogeneous hyperintense lesion expanding the cord and reaching the craniocervical junction. Notice the distended elongated urinary bladder (asterisk), a feature of the neurogenic bladder. TECHNIQUE: Sagittal STIR MRI of the spine. STIR, Short Tau Inversion Recovery; MRI, magnetic resonance imaging

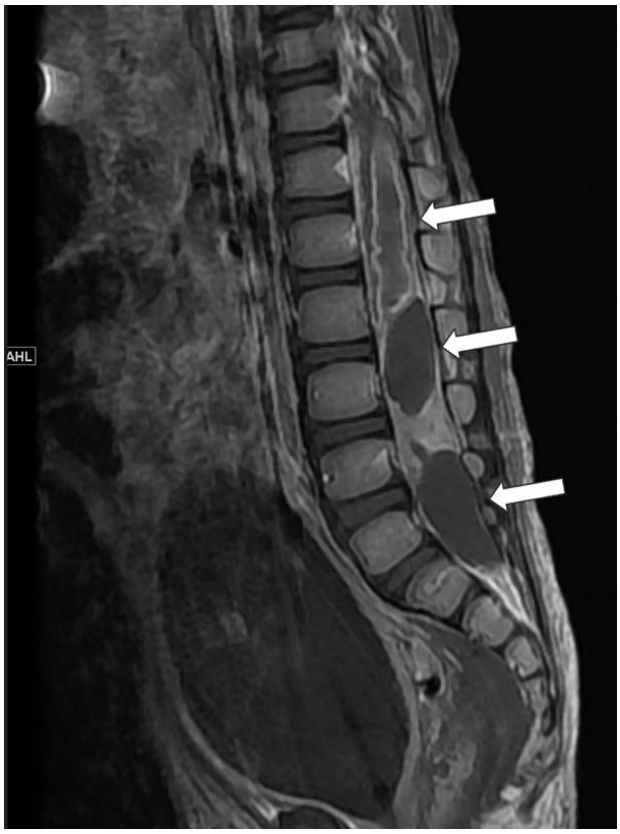

Figure 3: A 4-year-old boy with recurrent urinary tract infections who subsequently experienced lower-limb weakness due to pyogenic myelitis secondary to congenital dermal sinus.
FINDINGS: Expansion of the distal spinal cord at the level of the conus medullaris, along with a loculated collection at the cauda equina.
TECHNIQUE: Sagittal T1 fat-sat MRI with contrast. MRI, magnetic resonance imaging

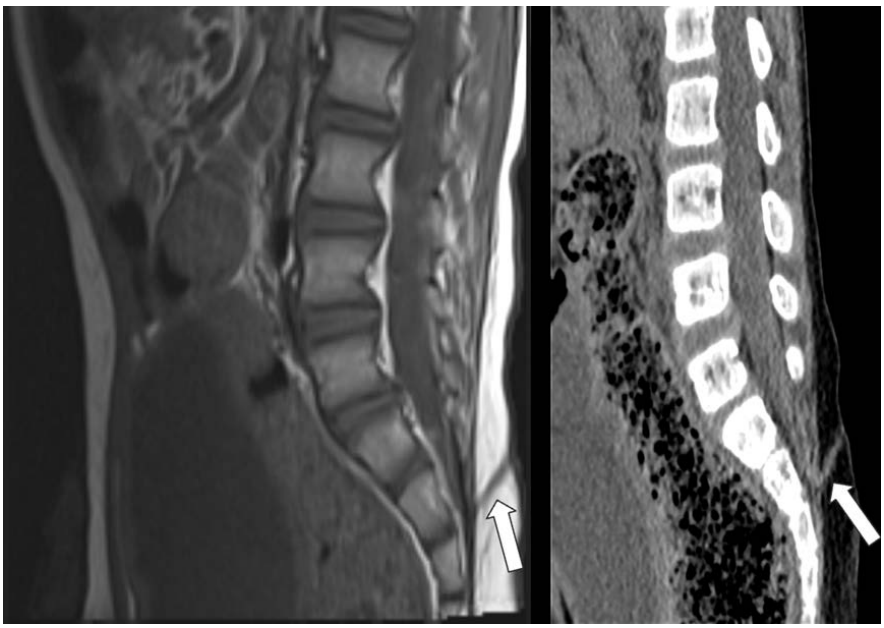

Figure 4: A 4-year-old boy with recurrent urinary tract infections who subsequently experienced lower-limb weakness due to pyogenic myelitis secondary to congenital dermal sinus.
FINDINGS: Dermal sinus tract (arrows) extending from the skin dimple with intraspinal communication at the level of S2-S3.
TECHNIQUE: Sagittal T1 MRI (A) and sagittal CT (B) through the lumbar spine. MRI, magnetic resonance imaging; CT, computed tomography

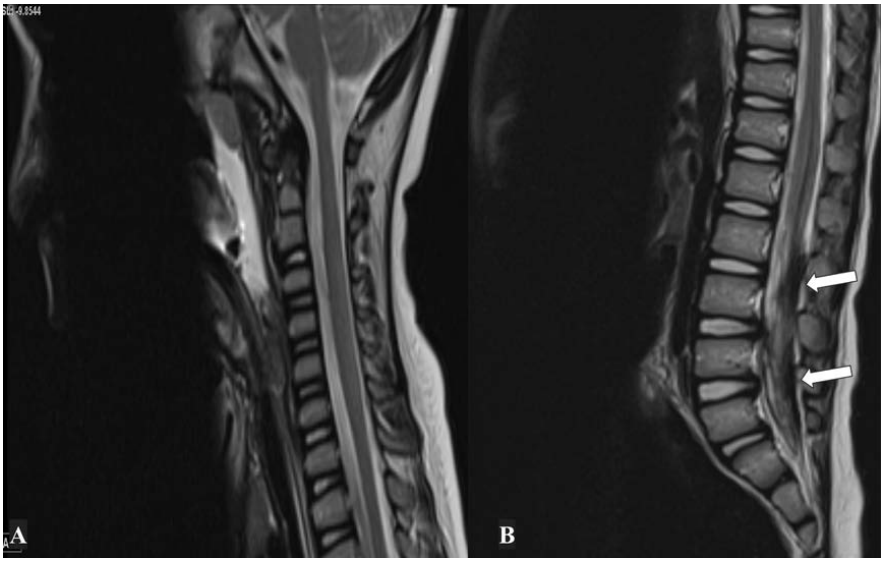

Figure 5: A 4-year-old boy with recurrent urinary tract infections who subsequently experienced lower-limb weakness due to pyogenic myelitis secondary to congenital dermal sinus.
FINDINGS: At follow-up, complete resolution of the abscesses after conservative management, with residual low signal fibrosis (arrows).
TECHNIQUE: Sagittal T2 sequences of the cervical (A) and lumbar (B) spines.

SUMMARY TABLE

Incidence	Intramedullary spinal cord abscess rare with few hunderet cases reported in the literature The majority of pediatric cases involve children below the age of six.
Etiology	In adults, it typically results from hematogenous spread of infections, such as those originating from the urinary or respiratory tracts, whereas in children, congenital dermal sinus is the most frequently identified underlying cause.
Common locations	Spinal cord abscess may develop at any level of the spinal cord, however thoracolumbar is the most common location. With underlying congenital dermal sinus it usually originates in lumbosacral area and may extends cranially. In very rare ocasions, holocord involvement may occur.
Gender variations	Males appear to be slightly more affected by Intramedullary spinal cord abscesses in reported cases.
Clinical features	Clinical features of intramedullary spinal cord abscess include fever, elevated inflammatory markers and neurological symptoms including motor deficit, sensory impairment, hyperreflexia, irritability and urinary or bowel dysfunction.
Imaging findings	MRI is the preferred method for diagnosis, usually showing expansion of the spinal cord with high signal intensity on T2. T1 post contrast reveals capsule-like structures, outlining the extent of the abscess cavity.
Prognosis	In pediatric patients, intramedullary spinal cord abscesses secondary to congenital dermal sinus are associated with more favorable outcomes, with excellent recovery observed in the majority of cases. Mortality is rare in this group and is most often related to delayed diagnosis or insufficient treatment.
Treatment	Treatment typically involves antimicrobial therapy (antibiotic or antifungal) to control the underlying infection, abscess drainage, and Surgical intervention such as a decompressive laminectomy and a myelotomy.

DIFFERENTIAL TABLE

Condition	Clinical features	Imaging features (MRI)
Syringohydromyelia	-Slowly progressive onset (months to years) -Typically no fever or acute illness -Central cord syndrome: dissociated sensory loss (loss of pain and temperature)	-Follows CSF signal on T1 and T2. -No enhancement. -Usually associated with Chiari-1 malformation.
Holocord astrocytoms	-Gradual onset (weeks to months) -Systemic symptoms (fever, infection) usually absent -Progressive motor deficits, gait disturbances, sensory abnormalities -Pain, scoliosis, bowel/bladder dysfunction	-Eccentric, poorly demarcated. -Fusiform cord expansion. 40% have a cystic component (pilocytic subtype) -Solid component enhances usually.
ADEM	-Acute/subacute onset (days to weeks) -Often preceded by recent viral infection or vaccination -Multifocal neurological deficits, altered consciousness	-Multifocal, hyperintense lesions on T2-weighted images -Typically patchy and asymmetric distribution in brain and spinal cord -No ring-enhancement

KEYWORDS

Myelitis; Congenital Dermal Sinus; Intramedullary Abscess; Sacral Dimple; Magnetic Resonance Imaging

ABBREVIATIONS

UTIs = Urinary Tract Infections
MRI = Magnetic Resonance Imaging
ADEM = Acute Disseminated Encephalomyelitis
CT = Computed Tomography