

# Delayed Diagnosis of Congenital Dermal Sinus Tract Presenting with Recurrent Meningitis and Paraplegia: Complete Neurological Recovery After Surgical and Targeted Therapy

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## Case Report

**Keywords:** Congenital dermal sinus tract, spinal dysraphism, recurrent meningitis, tethered cord syndrome, meningocele, paraplegia

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# Abstract

## Background

Congenital dermal sinus tract (CDST) is a rare form of spinal dysraphism caused by incomplete disjunction between the cutaneous ectoderm and the neural tube during embryogenesis. Clinically, it typically presents as a small midline cutaneous dimple; however, its potential connection with the spinal canal poses a significant risk for severe complications, including meningitis, intraspinal abscess formation, and progressive neurological deficits.

## Case Presentation:

We report a 6-month-12-day-old male infant who presented with a history of recurrent meningitis, swelling and erythema in the sacrococcygeal region, and loss of movement in the lower extremities. On physical examination, the patient was lethargic, with a bulging anterior fontanelle and paralysis of the lower extremities. Spinal magnetic resonance imaging revealed a cystic lesion measuring approximately 35 × 27 mm at the lumbosacral junction, extending to the subcutaneous tissue and containing neural elements, consistent with meningomyelocele. In addition, tethered cord, expansion of the spinal cord, and contrast enhancement suggestive of meningitis/inflammation were observed. The patient underwent neurosurgical intervention, during which laminoplasty was performed at the L2–L4 levels and the infected abscess tissue was resected. Postoperatively, empirical antibiotic therapy with vancomycin and meropenem was initiated. Following the growth of *Escherichia coli* in abscess cultures and according to antibiotic susceptibility results, vancomycin was discontinued and treatment was continued with meropenem. After a total of 28 days of meropenem therapy, inflammatory markers normalized. Muscle strength in the lower extremities, which was initially consistent with paraplegia, completely recovered, and the patient was discharged in good clinical condition. Follow-up contrast-enhanced spinal MRI at one month was unremarkable.

## Conclusion

This case emphasizes that early recognition of subtle cutaneous findings is critical and demonstrates that complete neurological recovery is achievable even in advanced presentations when prompt surgical and targeted antimicrobial therapy are provided.

## Introduction

Congenital dermal sinus tract (CDST) is a rare form of spinal dysraphism that arises from incomplete separation of the cutaneous ectoderm from the neural tube during embryogenesis (Fig. 4). It is characterized by an epithelium-lined tract extending from the skin surface to the spinal canal. The

prevalence of neural tube defects varies according to ethnicity, with higher rates reported among Hispanic infants compared to other populations. (1,2).

Congenital dermal sinus tract most frequently occurs in the lumbosacral region and typically manifests as a small midline cutaneous dimple. While often regarded as a benign sacral finding, the potential extension of the tract into the spinal canal poses a significant risk for serious complications. Therefore, an underlying dermal sinus tract should be strongly considered in infants presenting with recurrent or unexplained meningitis (3).

Congenital dermal sinus tract (CDST) establishes a direct communication between the skin surface and the subarachnoid space, facilitating bacterial contamination. Consequently, it may lead to severe complications, including spinal and epidural abscesses, intradural infections, infected dermoid or epidermoid cysts, and tethered cord syndrome. These conditions can result in progressive neurological impairment, ranging from lower extremity weakness to complete paralysis (4).

Magnetic resonance imaging (MRI) is the preferred modality for delineating the depth and extent of the sinus tract, as well as identifying associated cystic and infectious complications (Fig. 2a and 2b). Prompt diagnosis followed by surgical excision is essential to prevent irreversible neurological sequelae (5).

## Case Report

A 6-month-12-day-old male infant presented with swelling and erythema in the sacrococcygeal region, accompanied by restricted movement in the lower extremities (Fig. 3). According to the medical history, a progressively enlarging mass had developed in the sacrococcygeal area over the past week, initially the size of a hazelnut and increasing to approximately the size of a walnut, with surrounding hyperemia and tenderness. There was no discharge from the lesion, and no fever was reported during this period. There was no history of trauma.

Two days prior to admission, restricted movement developed in the lower extremities, more pronounced in the right lower limb. No visible erythema or significant swelling was noted in the lower extremities; however, decreased responsiveness to tactile stimuli was reported. According to the parents, a small dimple had previously been noticed in the sacrococcygeal region, but no medical evaluation had been sought.

It was noted that approximately one month prior, the patient had presented to another medical center with fever and decreased feeding, was treated for nine days with a preliminary diagnosis of meningitis, and was subsequently discharged. Due to worsening of the current symptoms, the patient was admitted to our clinic for further evaluation and management.

In the prenatal history, the mother was 34 years old and had received only vitamin supplementation during pregnancy, with no history of alcohol or tobacco use. The patient was born at term via

spontaneous vaginal delivery. Apart from a previous episode of meningitis in the postnatal period, there was no known comorbidity or history of surgery. Immunizations were up to date for age, and the patient was being breastfed. There was no family history of genetic, metabolic, or malignant diseases.

On physical examination, the general condition was assessed as moderate to poor, and the patient was lethargic. Body weight was 6.5 kg (3rd percentile), and length was 63 cm (50th percentile). Vital signs were as follows: body temperature 36.3°C, heart rate 131 beats/min, respiratory rate 35 breaths/min, and blood pressure 88/57 mmHg. A bulging anterior fontanelle was noted. Bilateral cervical lymphadenopathy measuring approximately 2 × 2 cm was detected.

Neurological examination revealed paralysis of the lower extremities. Deep tendon reflexes were hypoactive. No pathological reflexes were observed. Nuchal rigidity was absent. The Babinski reflex was bilaterally negative. Edema of the lower extremities was present at a + 2 level.

Laboratory investigations showed a white blood cell count of  $15.77 \times 10^3/\mu\text{L}$ , hemoglobin level of 10.5 g/dL, and platelet count of  $328 \times 10^3/\mu\text{L}$ . C-reactive protein level was 41.2 mg/L. Biochemical parameters were largely within normal limits. Urinalysis revealed no pathological findings.

Spinal magnetic resonance imaging (MRI) was performed. Preoperatively, lumbar spine MRI and non-contrast MRI of the sacrum-coccyx region were obtained. Postoperative follow-up included T2-weighted imaging and contrast-enhanced MRI after the second surgical intervention.

## Imaging Findings

Spinal magnetic resonance imaging (MRI) demonstrated a 35 × 27 mm cystic lesion at the lumbosacral junction, extending into the subcutaneous tissue and containing neural elements, consistent with meningocele. The spinal cord was positioned inferiorly, with imaging features indicative of a tethered cord.

Post-contrast imaging revealed peripheral enhancement of the caudal nerve roots and perimedullary regions, consistent with inflammatory changes suggestive of meningitis. Additionally, an expansile morphology of the lumbar spinal cord was observed.

## Surgical Management

In light of the clinical, laboratory, and radiological findings, the patient underwent neurosurgical intervention. The operation was performed under general anesthesia in the prone position. A midline incision was used to access the lumbar abscess. After dissection of the skin and paraspinal muscles, laminoplasty was carried out at the L2–L4 levels.

Microsurgical dissection was performed to separate the dura mater from the surrounding tissues, followed by midline durotomy and suspension. Intradural exploration demonstrated a thick neural

structure resembling the spinal cord rather than typical cauda equina fibers. The infected abscess tissue was completely excised (Fig. 1).

Following meticulous hemostasis, the dura was closed primarily, reinforced with tissue adhesive, and laminoplasty was completed. The wound was closed in layers, and the patient was transferred intubated to the postoperative intensive care unit.

## Postoperative Course

Postoperatively, the patient was managed in the pediatric department and continued on empirical intravenous vancomycin and meropenem therapy. *Escherichia coli* was isolated from the abscess culture. In accordance with antimicrobial susceptibility results demonstrating sensitivity to meropenem, and following consultation with pediatric infectious diseases specialists, vancomycin was discontinued and meropenem monotherapy was maintained.

Meropenem was administered for a total of 28 days. Acute phase reactants showed a gradual decline to normal levels. The patient, who initially presented with paraplegia, demonstrated marked neurological recovery, with complete restoration of muscle strength in the lower extremities.

Vital signs and hemodynamic parameters remained stable throughout the clinical course. The patient was discharged in good clinical condition upon completion of therapy. Follow-up contrast-enhanced spinal MRI at one month was unremarkable.

## Discussion

Congenital dermal sinus tract (CDST) is a rare spinal dysraphism arising from failure of embryological disjunction between the surface ectoderm and neural ectoderm. It is defined by an epithelium-lined tract that may extend from the skin surface to deeper neural structures, including the dura mater, conus medullaris, filum terminale, or spinal cord. Clinically, CDST most frequently involves the lumbosacral region and may initially manifest as subtle cutaneous markers, such as a small midline dimple, skin tag, hyperpigmentation, hemangiomatous lesion, or hypertrichosis. Despite their innocuous appearance, these findings may be deceptive, as CDST is associated with a substantial risk of infection and progressive neurological impairment (6).

In the present case, the most striking contributor to delayed diagnosis was the initial underestimation of the sacrococcygeal dimple present since birth. This finding is in line with the literature, where cutaneous markers often constitute the earliest clinical sign; however, diagnosis is frequently delayed until the onset of complications such as meningitis, local infection, tethered cord syndrome, or lower extremity neurological deficits. Ackerman and Menezes, in their large case series, reported that patients most commonly presented with cutaneous findings, neurological impairment, and infection. Likewise,

Radmanesh et al. highlighted that CDST typically becomes clinically apparent through cutaneous anomalies, infection, or neurological dysfunction (1, 4).

A particularly noteworthy aspect of this case is the concomitant presence of recurrent meningitis and paraplegia. Recurrent bacterial meningitis should prompt thorough investigation for an underlying structural defect. Contemporary radiological literature identifies spinal dermal sinus tract as a well-recognized congenital cause of recurrent bacterial meningitis in the pediatric population. Furthermore, these reviews emphasize the critical role of spinal ultrasonography and especially MRI in delineating the sinus tract and detecting associated intraspinal abnormalities in suspected cases (6).

The identification of *Escherichia coli* in the abscess culture represents a clinically significant finding. In CDST cases complicated by recurrent meningitis, particularly those involving the lumbosacral region, *E. coli* is frequently implicated due to fecal contamination. Masuoka et al. emphasized that *E. coli* is among the most commonly reported pathogens in CDST-associated recurrent meningitis. Accordingly, in infants with sacral midline cutaneous lesions and meningitis, the isolation of enteric organisms should strongly suggest the presence of an underlying structural spinal communication (6).

MRI findings were pivotal for both diagnostic confirmation and surgical planning in the present case. The literature consistently identifies MRI as the gold standard for evaluating the extent of the dermal sinus tract and detecting associated abnormalities, including low-lying conus/tethered cord, dermoid or epidermoid lesions, infected cystic formations, and intradural or intramedullary abscesses. Lane et al. demonstrated the critical utility of MRI in delineating tract depth and associated intraspinal anomalies in pediatric lumbosacral CDST cases. Furthermore, Masuoka et al. reported that gadolinium-enhanced spinal MRI effectively demonstrates inflammatory changes, including peripheral enhancement and abscess formation around the spinal cord. The MRI findings in our case—namely peripheral enhancement, tethered cord, and expansile cord morphology—are in agreement with these reports (6, 7).

In our case, the presence of a cystic lesion consistent with meningocele in association with a tethered cord suggests a complex form of spinal dysraphism rather than a simple dermal sinus tract. It is well established that congenital dermal sinus tracts (CDST) may coexist with additional anomalies, including dermoid and epidermoid tumors, a low-lying conus medullaris, and tethered cord. Masuoka et al. reported that a low-lying conus medullaris is frequently observed in patients with CDST and that dermoid/epidermoid lesions may be present at any level along the tract. Therefore, in patients presenting with superficial cutaneous findings, focusing solely on the sinus opening is insufficient; a comprehensive evaluation of the entire neuraxis, particularly the lumbosacral region, is essential (6).

Surgical management represents the cornerstone of treatment in these patients, not only for infection control but also for the prevention of recurrent infections and progressive neurological deterioration. Both classical series and more recent reports consistently describe the standard approach as complete excision of the sinus tract, with intradural exploration when indicated, combined with drainage or resection of associated abscesses and release of tethering. In a series of 21 patients reported by Singh et al., surgical intervention was the mainstay of treatment in individuals presenting with infection and

neurological deficits; notably, this cohort included cases presenting with acute meningitis and sudden paraplegia (5).

The complete restoration of muscle strength in the lower extremities—initially paraplegic—following postoperative, culture-directed antibiotic therapy with meropenem for 28 days represents a remarkable clinical outcome. This finding highlights the critical role of early surgical intervention combined with prolonged, targeted antimicrobial therapy in optimizing neurological recovery in infected CDST cases. Consistent with this observation, contemporary reports have documented favorable neurological outcomes, particularly in patients with intradural or intramedullary infectious complications who receive timely surgical and intravenous antibiotic treatment (3).

Additionally, this case emphasizes that the evaluation of recurrent meningitis in children should extend beyond immunodeficiency to include a systematic search for underlying structural abnormalities. Spinal dysraphism must be considered in infants presenting with midline lumbosacral cutaneous lesions, enteric pathogen-associated meningitis, or lower extremity neurological deficits. Early diagnosis and timely surgical management are crucial in preventing irreversible neurological sequelae, whereas delayed recognition may lead to severe complications such as paraplegia, sphincter dysfunction, and recurrent infections (6).

To the best of our knowledge, the coexistence of recurrent meningitis, paraplegia, an *E. coli*-associated abscess, and complete neurological recovery following delayed diagnosis of CDST has been exceedingly rare. This case underscores that even in advanced clinical presentations, timely surgical intervention combined with targeted antimicrobial therapy can result in favorable neurological outcomes.

Despite increasing awareness, delayed diagnosis remains a significant clinical challenge, particularly when subtle cutaneous findings are overlooked during early infancy. This case reinforces the importance of careful examination of sacrococcygeal cutaneous findings during routine pediatric evaluations, as early detection may prevent severe neurological complications.

## Conclusion

In conclusion, this case underscores that a sacrococcygeal dimple present since birth should not be dismissed as a benign cutaneous finding. In infants with recurrent meningitis, local infectious signs, or lower extremity neurological deficits, congenital dermal sinus tract and associated complex spinal dysraphism must be carefully considered. Prompt evaluation with contrast-enhanced spinal MRI, followed by timely surgical intervention and culture-directed antimicrobial therapy, is critical for preventing irreversible neurological damage. As demonstrated in this case, complete neurological recovery is achievable even in advanced presentations.

## Declarations

# Consent for Publication

Written informed consent for publication was obtained from the patient's parents.

# Ethics Statement

Written informed consent for publication was obtained from the patient's parents. This case report was prepared in compliance with patient confidentiality, and all personal identifiers were removed to ensure anonymity.

# Conflict of Interest

The authors declare that they have no conflict of interest.

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This study received no specific grant from any funding agency in the public, commercial, or not-for-profit sectors.

# Author Contribution

Author Contributions Adnan Erseçkin: Conceptualization, patient management, data collection, literature review, manuscript drafting. Abdulmutalip Karaaslanlı: Neurosurgical management, surgical intervention, critical revision of the manuscript. All authors approved the final manuscript.

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# Availability of Data and Material

Data sharing is not applicable to this article as no datasets were generated or analyzed.

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## Figures

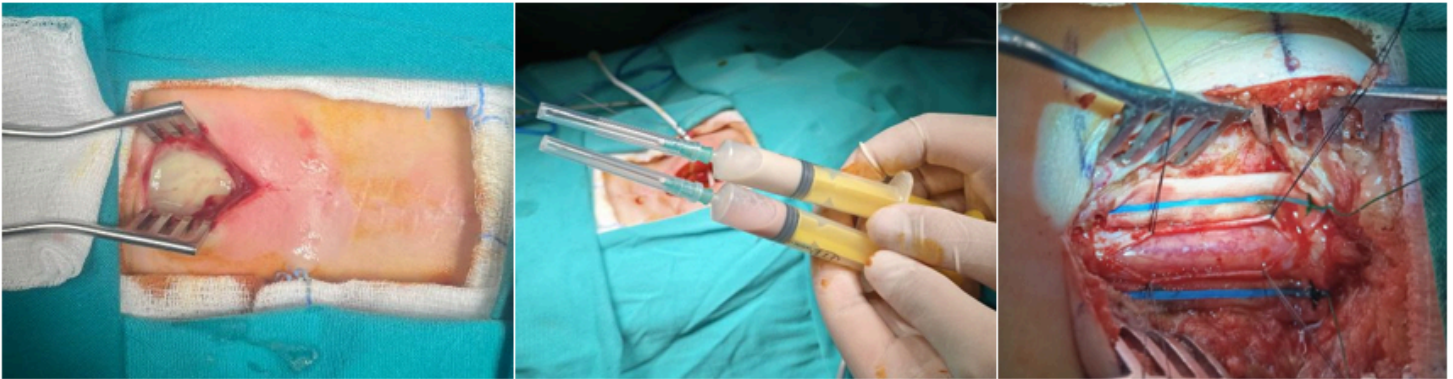


**Figure 1**

Intraoperative image showing the lumbar midline incision exposed with retractors, revealing an infected cystic lesion consistent with an abscess cavity.

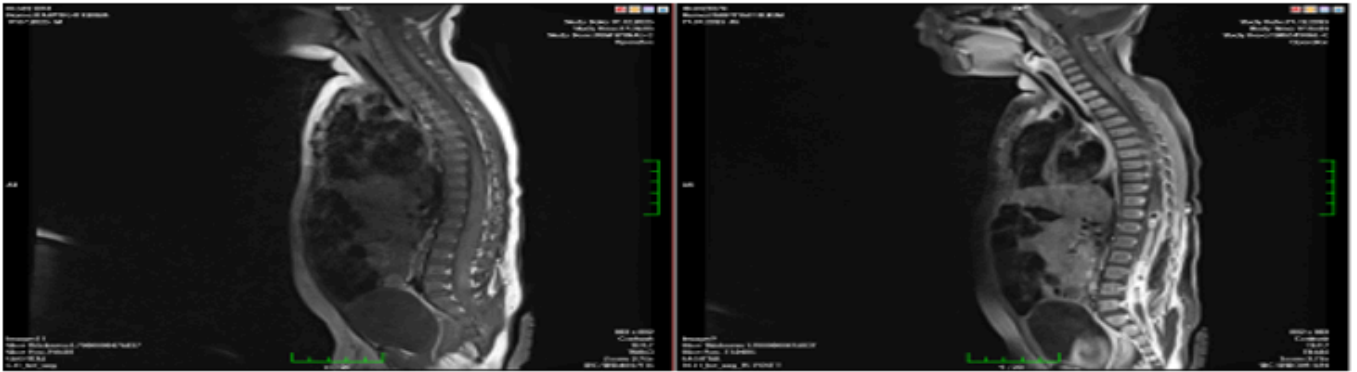


**Figure 2a**

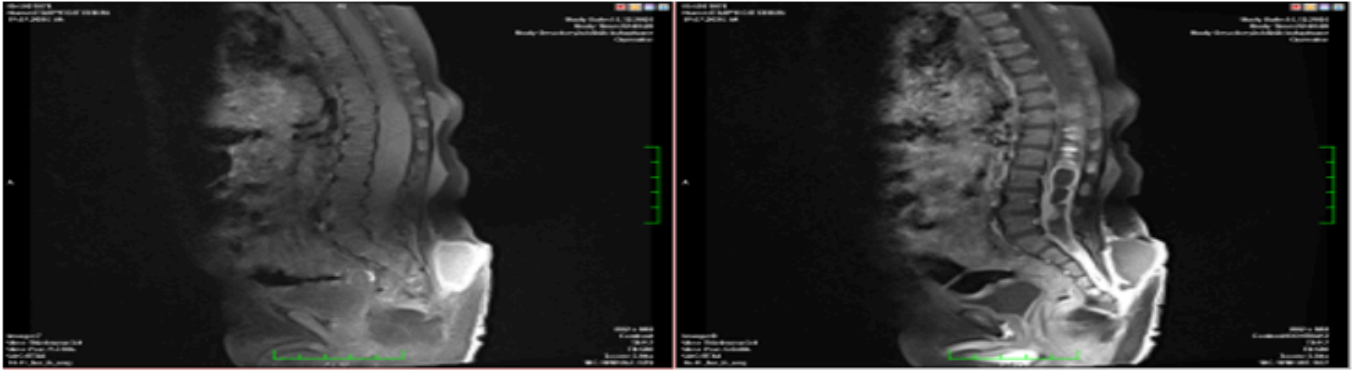


**Figure 2b**

**Figure 2**

**2a.** Preoperative spinal magnetic resonance imaging (MRI).

**2b.** Postoperative magnetic resonance imaging (MRI) demonstrating follow-up findings.

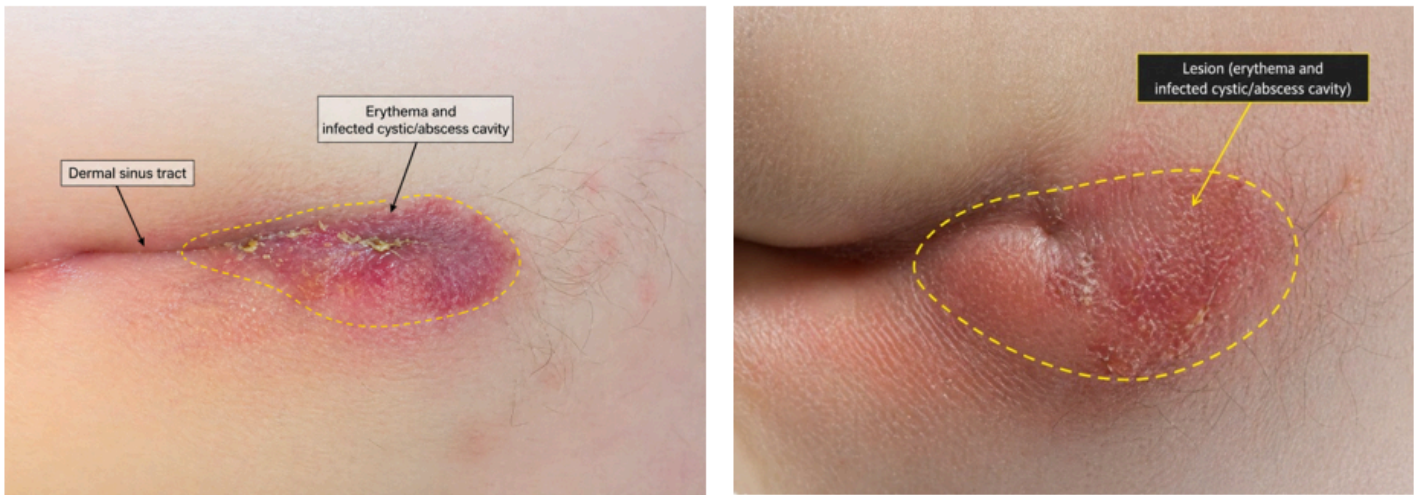


**Figure 3**

Preoperative image demonstrating cutaneous erythema and a dermal sinus tract in the sacrococcygeal region.

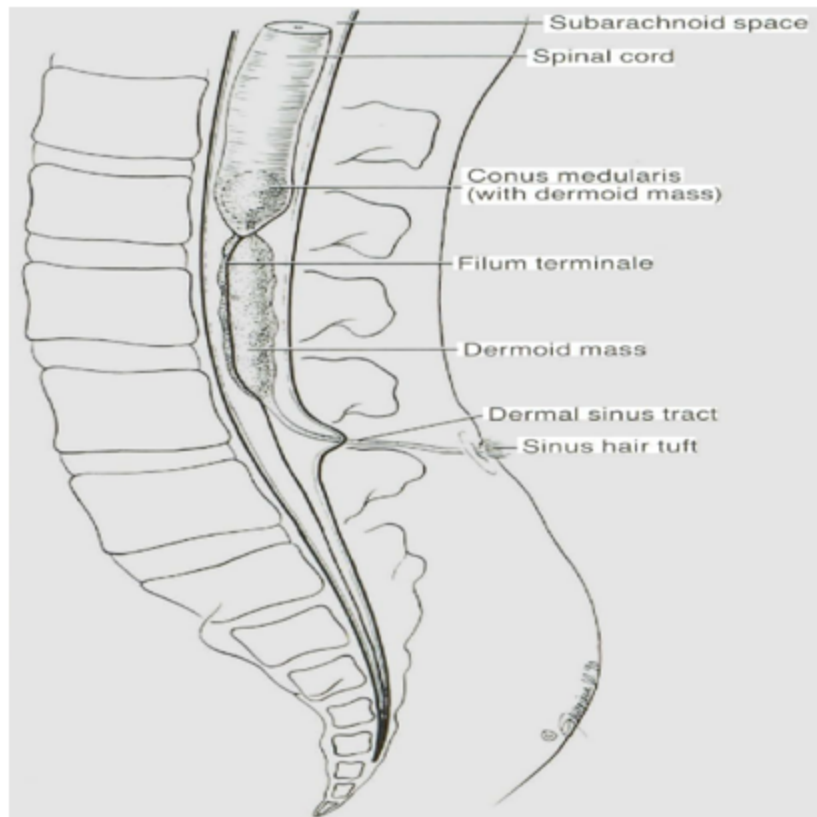


**Figure 4**

Clinical image demonstrating a dermal sinus tract in the sacrococcygeal region (arrow).

