Infected Spinal Dermal Sinus Tract with Meningitis: A Case Report

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Abstract-

Purpose: Congenital dermal sinus tract (DST), an uncommon entity of cranial or spinal dysraphism, occurs along the midline neuraxis that may arise from nasion and occiput down to the lumbar and sacral region. It is often diagnosed in infants and children for skin signs, neurological deficits, local infection, meningitis, or abscess. For spinal DST, there is a paucity of case or series report in Taiwan.

Case Report: In this paper, we report a case in a 6-year-old girl. The girl presented with midline lumbar skin dimple, hypertrichosis, and history of bacterial meningitis. She was successful treated by surgical excision of the DST with local infection that ended within the subarachnoid space between L2-3 vertebrae.

Conclusion: This case highlights the importance of a thorough examination of the midline craniospinal axis in children with meningitis or history of meningitis.

Key Words: dermal sinus tract, spinal dysraphism, meningitis

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INTRODUCTION

Spinal dermal sinus tract (DST) is a rare entity of spinal dysraphism in childhood. It is usually located at the lumbar or lumbosacral region. DSTs are lined by stratified squamous epithelium and extend from the surface through the deeper tissue into the cranial or spinal cavity [(1)]. Cutaneous abnormalities, neurological deficits, local infection, and meningitis are the main manifestations with infection causes major morbidity. Early diagnosis of DSTs can be achieved by thorough clinical examination and neuroimaging studies including plain x-ray, sonography, and magnetic resonance imaging (MRI). The authors report an unusual, but successfully treated case of an infected dermal sinus tract. This case highlights the importance of examination of the midline craniospinal axis in children with recurrent meningitis or a history of meningitis.

CASE REPORT

A 6-year-old girl was born full-term with a birth

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mark on the midline lumbar region. A pinhole opening associated with focal hypertrichosis were also noted (Fig. 1). She grew up uneventfully until 5 years old. She was diagnosed to have bacterial meningitis and was treated successfully with antibiotics. There was no residue neurological sequela. However, at 6 years old, she was admitted to our service because of fever of unknown origin. She had dull headache with vomiting. There was no seizure. Glasgow coma scale was evaluated as E4M6V5 and she had intact cranial nerve functions. Neck stiffness and urinary retention were the major findings. Laboratory data showed leukocytosis with elevated C-reactive protein level. Chest x-ray did not reveal any abnormal patches and routine urinary examination was within normal ranges. Empiric antibiotics with ceftazidime and vancomycin were prescribed after admission. She had undergone lumbar magnetic resonance imaging (MRI) studies for lumbar skin stigmata and highly suspicion of central nervous system infection. The study showed a dermal sinus tract that extended deeply into the subcutaneous tissue and connected to the dura at level between L2-L3 vertebrae (Fig. 2A). It was isodense on T1 weighted image with significant regional subcutaneous tissue enhancement after contrast medium injection (Fig. 2B). Bony elements were normal without spinal bifida occulta. Under the impression of infected spinal dermal sinus, she received surgical intervention. The lesion was explored with a longitudinal incision extending from the skin opening. We traced the sinus tract layer by layer down to L2 lamina. Necrotic tissue and pus were sealed-off in the subcutaneous layer. The pus was drained and the necrotic tissue was completely debrided with curette after L2 laminectomy. The remaining infected tract connected to and incorporated into the dura (Fig. 3). After dura opening, we found that the tract ended and adhered to the

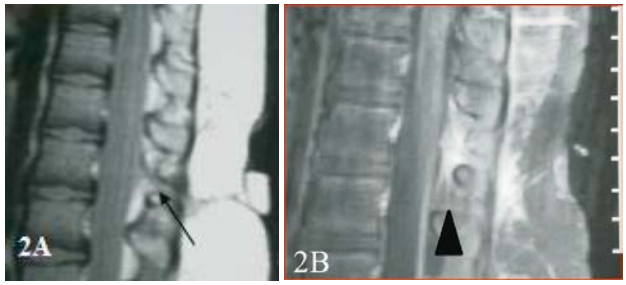

Figure 2. (2A): T1 weighted sagittal lumbar MRI of patient showing a dermal sinus tract extended into the spinal canal (arrow). (2B): Contrast-enhanced T1 weighted sagittal lumbar MRI showing subcutaneous enhancement (arrowhead). Conus medullaris is at L2 level.

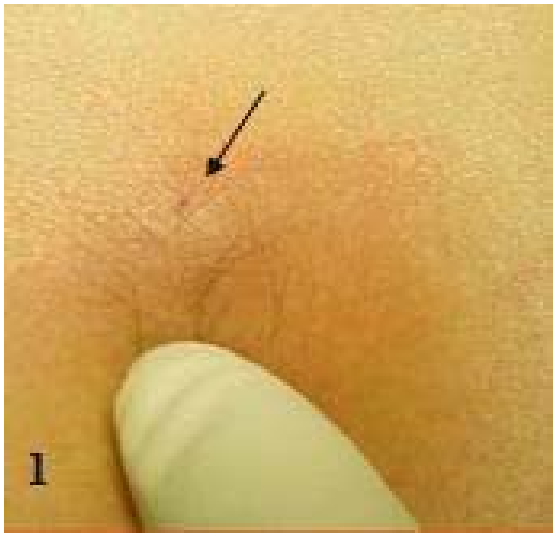

Figure 1. Lumbar region skin stigma: pinhole opening and focal hypertrichosis (arrow).

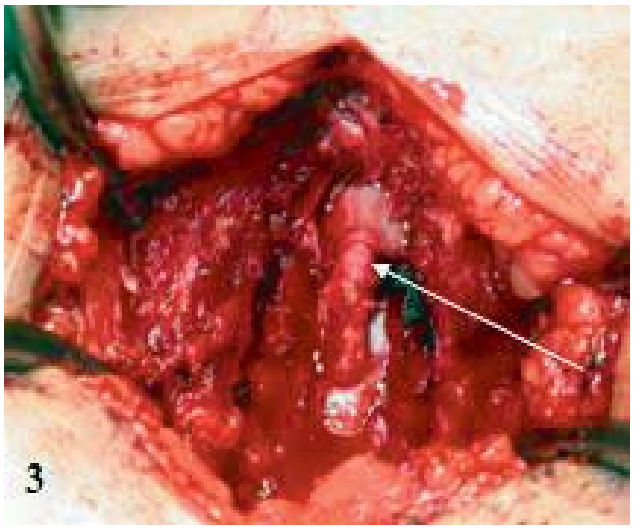

Figure 3. Operative findings after L2 laminectomy and partial excision of the sinus: the deep end of the tract connected and incorporated into the dura (arrow).

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arachnoid membrane. The tract was completely excised. The dura was closed with water-tight suture after the whole operative field was irrigated with saline. Postoperatively, the patient recovered without any neurological deficit. Histologically, the infected DST consisted of stratified squamous epithelium surrounding by granulation tissue with acute and chronic inflammatory cell infiltration.

DISCUSSION

Spinal dermal sinus presented with meningitis was reported by Moise in 1926 [2]. The embryogenesis of DST is theorized to occur from defective separation of the epithelial ectoderm at a point from the neural ectoderm during the 3rd to 5th week of fetal development along the neuraxis. The incomplete disjunction creates a tract lined by epithelia and surrounded by dermal elements that extends from the skin surface to the deep tissue and frequently intradurally and attached on neural tissue [3,4]. In 1990, a review of all published cases of congenital spinal dermal sinuses reported that 1% occurred at the cervical level, 10% at the thoracic, 41% at the lumbar, and 35% at the lumbosacral region [5]. More than 90% of DSTs terminate into subdural space; nearly 60% enter the subarachnoid space and 27% are attached to the neural elements [6]. Associated anomalies of DST include bifid lamina, tethered cord, inclusion tumors (epidermoid and dermoid cyst), split cord malformation, tight filum terminalis, and other forms of spinal dysraphism such as lipomyelomeningocele and myelomeningocele [7]. Lesions involving the nervous elements could cause various neurologic problems. In cases of tethering spinal cord, gait difficulty with lower extremities and bladder dysfunction are common neurologic presentations. Unlike adult, pain is not common with tethered cord in childhood. DSTs are associated with an overall of 37.1% neurologic abnormalities in which infection is not uncommon [8]. Intradural infection with arachnoid adhesion can contribute to tethered cord [4,9]. Infection, which usually presents with fever of unknown origin, may lead to morbidity if not treated proper and early. In this reported case, the DST incorporated into the dura. The tract terminated and adhered to the arachnoid membrane. There was no neural elements involvement, inclusion tumor, low-setting conus, tight filum terminali, and intradural infection. This is why the patient had an uneventful recovery with no residue neurological deficit.

The early diagnosis of DSTs mainly depends on a highly suspicion. Skin stigmata, such as a sinus ostium, hypertrichosis, abnormal pigmentation, subcutaneous lipoma, local infection or induration, and repeated meningitis of unknown origin are important clues. Because DST usually transgresses dura, probing or squeezing of the tract is not recommended. Image modalities for diagnosis of spinal DSTs include plain x-ray, sonography, computed tomography (CT) and magnetic resonance imaging (MRI). MRI is the first choice diagnostic study and preoperative evaluation because MR images provide significant information on the neural structures, level of conus, and other associated anomalies and the lesion. Plain x-ray and CT may not show abnormal findings. Sonography of the spinal cord may detect sinus tract, position of conus, and other associated anomalies in young infants [10]. Early investigation with sonography in cases of spinal DSTs allows early surgical intervention [11] which in turn, prevents neurologic deficits or neurological deteriorations. There are favorable outcomes in the prophylactic surgical removal of DSTs. Early surgery may maintain neurological functions as well as bladder function [12]. The gold standard of surgical interventions of DST includes complete resection and intradural exploration [13]. At surgery, the dura should always be opened for the complete resection of the intradural part of the tract and other associated intradural lesions. In cases with active infected lesions, surgery is recommended 3-4 weeks after the infection has been controlled with antibiotics. However, the anatomy of neural element may be disturbed due to extensive arachnoid scarring in case of infection that adds the difficulty of surgery and the potential risk neural tissue damage [14]. Urgent operation provides excellent outcome in the presence of rapidly progressing neurological deficits, in spite of evident meningitis [15]. The surgical result depends on the presence of pre-existing neurological deficits and the severity of infection.

The presence of cutaneous stigmata over the midline neural axis should not be considered as benign, unless proved otherwise. Early diagnosis of spinal DSTs needs highly vigilance of physicians in treating patients with cutaneous anomalies. We should try to find skin stigmata of DST in treating patient with meningitis or a history of meningitis. Using sonography or MRI, early diagnosis of spinal DST is feasible. A definitive operation with intradural exploration and complete excision of the lesion should be undertaken to obviate future complications.

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