

RECURRENT MENINGITIS ASSOCIATED WITH CONGENITAL PARAVERTEBRAL DERMAL SINUS TRACT*

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SUMMARY: Andıran N, Coşkun T, Özaydın E, Seçmeer G. (Department of Pediatrics, Hacettepe University Faculty of Medicine, Ankara, Turkey). Recurrent meningitis associated with congenital paravertebral dermal sinus tract. Turk J Pediatr 1998; 40: 121-125.

Recurrent meningitis is a rare but important event that needs to be searched for a predisposing factor. Congenital dermal sinuses occurring in the midline are among the etiological factors. Here, an 18-month-old boy with three attacks of recurrent meningitis due to a paramedian dermal sinus tract is presented. *Klebsiella* was the cultured causative agent. The lesion was suspected on physical examination and demonstrated by lumbosacral magnetic resonance imaging. To the best of our knowledge, this is the first case in English-language literature of the paravertebrally located dermal sinus tract resulting in recurrent meningitis. Therefore, a careful physical examination, especially including the paravertebral region beside the midline, is essential. Magnetic resonance imaging is a non-invasive descriptive method in the evaluation of congenital dermal sinus tracts. *Key words: dermal sinus tract, magnetic resonance imaging, recurrent meningitis.*

Recurrent meningitis (RM) is a rare event defined as any reappearance of clinical and laboratory signs and symptoms of bacterial meningitis after adequate and successful treatment of a preceding meningitis, regardless of the time of reappearance.

RM is generally associated with a predisposing factor. The presence of an acquired or congenital anatomical communication between cerebrospinal fluid (CSF) and a mucocutaneous site, or defects in host immunity, especially in humoral and complement systems, predisposes to recurrent meningitis¹. It constitutes an important problem in both the determination of cause and the treatment.

Congenital dermal sinuses, mostly occurring at lumbosacral and occipital regions in the midline, are among the important etiological factors of RM. Today, magnetic resonance imaging (MRI) makes it possible to provide detailed anatomical configurations of the sinus tracts and the associated cyst preoperatively^{2,3}.

Here, a case with a right paramedian sacral dermal sinus tract and an associated dermoid cyst is reported with MRI findings.

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Case Reports

An 18-month-old male patient was referred to our hospital because of RM. He had been admitted and treated at his local hospital for two attacks of purulent meningitis three and five months previously. Physical examination revealed a well developed and well nourished child in nonapparent distress, with minimal nuchal rigidity, a temperature of 38 °C, pulse rate of 130/min, and respiratory rate of 26/min. There was no obvious dimple, or hyperpigmented or hairy lesion along the spine or in the occipital region. An approximately 0.5 cm in diameter hard subcutaneous lesion, 2 cm away from the midline to the right at about L5-S1 paravertebral localization, was palpable. Leukocyte count was 8900/mm³ and the peripheral smear revealed 60 percent polymorphonuclear leukocytes, 7 percent bands, 27 percent lymphocytes and 6 percent monocytes. Lumbar puncture disclosed a cloudy CSF with a leukocyte count of 2,000/mm³, with almost all cells in the sediment being polymorphonuclear. The CSF contained a protein of 142 mg/dl, a glucose of 38 mg/dl simultaneous with a blood glucose level of 106 mg/dl.

Blood, urine and CSF cultures were obtained and a combined therapy with intravenous cefotaxime and vancomycin was instituted. Forty-eight hours after admission, klebsiella sensitive to cefotaxime was identified in the CSF culture. Immunological studies showed normal serum immunoglobulin levels and CH50. Plain radiographs of the skull and the spine were normal, but in order to exclude a spinal anomaly, a lumbosacral MRI was done. A right paramedian dermal sinus tract at S2 level and a lumbosacral dermoid cyst at L5-S2 level were detected (Figs. 1 and 2). Having controlled the infection with 10 days' antibiotic treatment, the patient underwent an exploratory surgery. The dermal sinus tract and intradural dermoid cyst containing hair were completely excised. Histopathological examination confirmed the diagnosis. After surgical intervention, the patient had an uneventful recovery with no neurological sequelae.



Fig. 1: The sagittal T1-weighted section of the lumbosacral magnetic resonance image showing the dermoid cyst at L5 to S2 level.



Fig. 2: The right paramedian dermal sinus tract demonstrated in the transverse section of the lumbosacral magnetic resonance image (between the two arrows).

Discussion

An incomplete separation of neuroectoderm and epithelial ectoderm between the third and fifth weeks of intrauterine gestational development results in the formation of a dermal sinus tract. The common form of this congenital anomaly is a tract that extends from the skin to the conus medullaris ending in a cyst: epidermoid, if lined by stratified squamous epithelium or; more commonly, dermoid, if the lining also comprises skin appendages like hair follicles and sebaceous glands⁴. An infected dermal sinus tract that extended into the dura was first reported by Moise⁵ in 1926. They can present as a dimple in the skin or as associated cutaneous manifestations such as a hyperpigmented patch, a hairy nevus or hemangioma^{4,6}. However, these defects may be unremarkable without any cutaneous stigmata and can be easily overlooked by clinicians. Although these sinus tracts are mostly located on the midline, in our patient, it was located at paravertebral position without any remarkable sign. To the best of our knowledge, this is the first paravertebrally located dermal sinus tract case reported in the English language literature.

Dermal sinus tracts have been found in patients ranging in age from early childhood to 25 years, and 60 percent were discovered due to the presenting meningitis⁷. In most cases of lumbosacral defects, the delayed diagnosis of the fistula to the spinal canal leads to RM. Lieb et al.⁸ found developmental anatomical abnormalities as the underlying cause in 23 of 25 patients with RM; the remaining two patients had traumatic CSF leaks and no immunodeficiency was found. All 25 of their cases had anatomical lesions. This was perhaps noticed because the authors are neurosurgeons. Kline⁹ studied 47 cases gleaned from the literature, including children and adults, with predisposing factors including congenital CSF fistula (in 55% of cases), traumatic or surgical CSF fistula (in 17%), immunodeficiency (in 21%) and unknown causes (in 6%). The rate of 21 percent immunodeficiency cited by Kline seems an overestimate, since

it is deducted from the literature review, because immunodeficiencies are more enthusiastically reported than cases of developmental or traumatic anatomical lesions without special features. Therefore, in the case of RM, a developmental or traumatic anatomical defect must be primarily suspected and investigated.

The most common etiological agents for meningitis in these patients include *S. aureus*, *S. epidermidis*, *S. pneumoniae*, *H. influenzae*, *E. coli*, proteus species, *Salmonella*, anaerobic microorganisms and polymicrobes^{2, 3, 8, 10-16}. Another distinct feature of the present case was the detection of *Klebsiella* as the cultured microorganism of the meningitis; to our knowledge, the first reported case of such in English language literature.

The lumbosacral MRI of the patient enabled us to discover the dermal sinus tract and the associated dermoid cyst preoperatively. This was a noninvasive investigation, obviously superior to the previous methods such as sinography or myelography in the evaluation of dermal sinus tracts.

Clinicians should be suspicious and make a careful physical examination of patients who present with RM in order to not overlook an underlying insidious lesion located at paraspinal regions beside the midline. Enthusiasm in the identification of the microorganism is vital for the treatment of choice.

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