

Thoracic Dorsal Dermal Sinus with Secondarily Infected Intramedullary Dermoid Cyst

Abstract

Dorsal dermal sinus (DDS) is an uncommon type of occult spinal dysraphism most often located in the lumbar region. Patients present either due to secondary infection or compression of neural structures by an associated dermoid or epidermoid cyst. We report a rare case of 2-year-old child who presented with progressive paraparesis with magnetic resonance imaging of spine showing a thoracic DDS with an infected intramedullary dermoid cyst. Partial excision of the dermoid cyst and resection of the sinus opening was done with partial clinical improvement postsurgery.

Keywords: *Dermoid cyst, dorsal dermal sinus, intramedullary, secondary infection, thoracic region*

Introduction

Congenital dorsal dermal sinus (DDS) is a disorder of abnormal neurulation in which there is focal nondisjunction of surface and neural ectoderm resulting in an epithelium-lined tract extending inward to the spinal canal from the skin surface with an open skin defect. Dermoid, epidermoid, teratoma, and posterior arch defects of the vertebral column can be seen in the association of DDS. The patients become symptomatic either by infection due to bacterial ascent through the sinus tract or because of compression of neural structures by an associated dermoid or epidermoid tumor. There are <10 case reports with magnetic resonance imaging (MRI) findings demonstrating an infected intramedullary dermoid cyst in thoracic region with DDS.

Case Report

A 2-year-old female child presented with progressive weakness of both lower limbs since 1 year, sinus and pus discharge in the midline in upper back, incontinence of bowel and bladder since 6 months. She was born out of a normal vaginal delivery and cried immediately after birth. She had a dimple which was identified at birth, however not further investigated. She had normal development up to 1 year. On physical examination, the child's skin over the upper thoracic region showed

a midline dimple with redness, mild swelling, and seropurulent discharge [Figure 1a and b]. Pus was sent for culture, and she was treated with intravenous vancomycin. MRI revealed an oblong well-circumscribed intramedullary cystic lesion extending from C7 to D5 level and a sinus tract connecting it to the skin surface. The intramedullary lesion was hypointense on T1w [Figure 2a] and hyperintense on T2-weighted images [Figure 2b] with smooth, thick peripheral enhancement [Figure 2c] suggestive of an abscess. Spina bifida was noted at D3-D5 clearly evident on computed tomography images [Figure 3a and b].

The child was taken up for surgery. The sinus opening was dissected from the surrounding soft tissue and sinus tract was followed. Spinous process of D1, D3 was excised, laminectomy of D1, D2, and D3 was done with sinus tract carefully dissected and followed till spinal cord. Myelotomy done after opening the dura. 4 cc of pus was aspirated from the sinus opening and sent for gram stain, acid-fast bacilli, culture sensitivity, and fungal culture. The dermoid cyst was present inside the spinal cord with gelatinous material and hair was visible [Figures 4 and 5]. The dura and pia mater were adherent to each other due to inflammation. All the gelatinous material and hair were removed carefully. The wall of the cyst (capsule) could only be partially removed due to the fear of spinal

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Figure 1: Clinical photographs (a and b) of the patients skin over the upper thoracic region showing a midline dimple with ostium and seropurulent pus discharge (blue arrows)

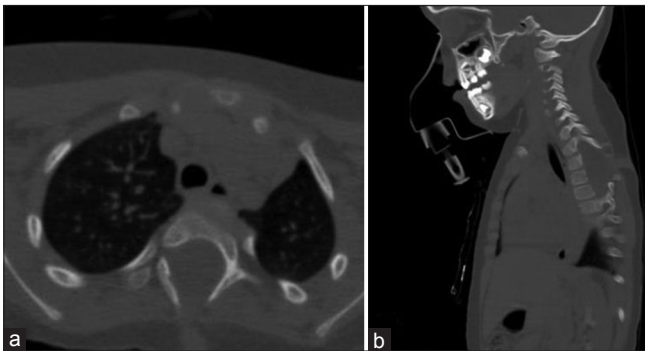


Figure 3: Computed tomography axial (a) and sagittal (b) images in bone window demonstrates spina bifida at D3-D5 level

cord injury. The postoperative course was uneventful, with partial improvement in neurological function.

Discussion

The complex of upper thoracic DDS beginning from the skin, passing through the tissue layers, communicating with the intradural intramedullary dermoid cyst and spina bifida observed in this child is rare occurrence. Wang *et al.*^[1] observed five cases out of which only one had a dermoid cyst at D6-D7 level in association with DDS. Morimoto *et al.*^[2] reported a 1-month-old child who presented with a dimple over the lumbosacral junction discharging pus having DDS in association with intramedullary abscess and dermoid at D12-S1 level. In asymptomatic patient, the physical examination and MR imaging help in diagnosing at an earlier stage and preventing future complications. On clinical examination, midline cutaneous dimple overlying the spine is the hallmark of DDS. Cases with too small sinus orifice need careful close examination for detection. In a child with repeated episodes of unexplained meningitis, it is very important to inspect the midline skin.^[1] The sinus tract begins at the skin dimple and usually tracks cephalad through the soft tissues to traverse the dorsal dura. Patients can become symptomatic as a result of either infection or associated mass lesions such as epidermoid or dermoid like in our case. Progressively enlargement of the inclusion tumor

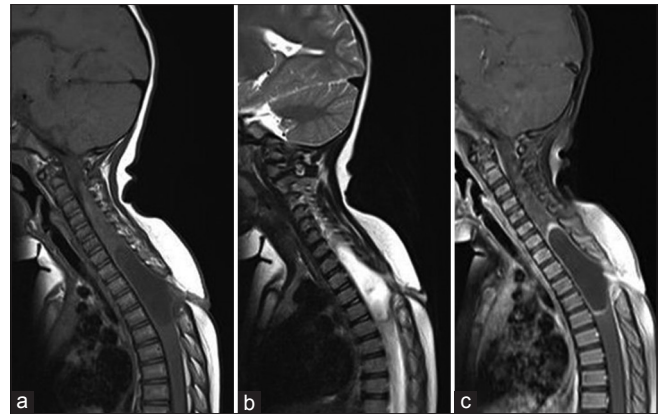


Figure 2: Magnetic resonance imaging of spine sagittal images demonstrating an oblong well circumscribed intramedullary cystic mass lesion extending from upper border of C7 to upper border of mid-D5 and a sinus tract connecting it to the skin surface. The mass is hypointense on T1-weighted images (a) and hyperintense on T2-weighted images (b) with thin peripheral smooth wall enhancement (c)

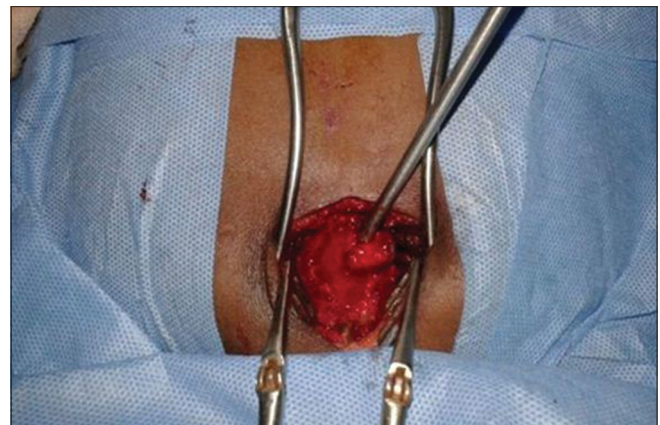


Figure 4: Midline vertical incision showing well-encapsulated mass

along the spinal canal will cause compression of adjacent neural structures. The most common organisms responsible for meningitis are said to be *Staphylococcus aureus* and *Escherichia coli*. Aseptic and recurrent meningitis occurs secondary to irritation caused by excess cholesterol crystals produced from the cells proliferating in the dermal channel.^[3] Drainage of debris or purulent material from the sinus tract may be observed as in our case.

Sinus tract is best demonstrated by MRI.^[3] Good soft-tissue resolution of MRI images helps in rapid and accurate identification of the extent of the lesions and delineation of the extraspinal portion of the sinus tract and associated inclusion tumor and also defines the degree of the spinal cord compression.

There is no role for conservative management in dermal sinus. Complete excision of the dermal sinus along with intraspinal portions and associated dermoid cysts should be promptly done.^[3-6] Whenever possible, the intradural inclusion tumors should be completely excised. Most often even though the tumor may be adherent to the neural element, it can still be easily freed from the neural elements, however, in our case,



Figure 5: Postoperative specimen (a and b) which is well circumscribed and encapsulated. Few hair strands are noted which are separated from the tumor and placed separately

the tumor could only be partially excised due to fear of spinal cord injury. Complete excision is nearly impossible, in cases with ruptured or infected tumors because of dense arachnoid adhesions. In cases with the scarred capsular wall, it is not necessary to attempt complete removal likely in our case; careful bipolar coagulation is known to prevent recurrence and reduce the risk of neurological impairment. Therefore, early prophylactic surgery has to be performed, because it is easier to intervene and gives better outcome unlike in our case where the patient already had complications. Even in cases with no evident infection, it is advisable to use broad-spectrum antibiotics, as in our case, where vancomycin was used even though the pus was sterile. When symptoms of infection develop, cultures should be obtained.^[7]

The occurrence of DDS, infected dermoid cyst, and spina bifida in upper thoracic region is extremely rare. Early clinical suspicion, careful clinical examination, and MR imaging are helpful in the diagnosis, early management, and to prevent complications.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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