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A Case Report of Congenital Dermal Sinus Tract with Dermoid Cyst and Lipoma

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Abstract

Congenital dermal sinus tract (CDST) is a rare entity of spinal dysraphism with an incidence of 1 in 2500 live births. The presumed etiology relates to a focal failure of disjunction resulting in a persistent adhesion between the neural and cutaneous ectoderm. CDST is commonly associated with other pathologies such as myelomeningocele, split cord malformation, tethered cord and inclusion tumors. The authors report a case of a 2-year-old girl discharging lumbosacral CDST. The preoperative MRI showed the dermal sinus tract with 2 intradural lesions from L1 to L3. The tract was surgically excised followed by complete excision of the cyst and lipoma. The dermal sinus tract was attached to the medullary cone and was cut there, allowing "en bloc" removal of the dermal sinus tract. There were two distinct types of tumor. One cystic is located posterior to the medullary cone and the other is located laterally to the medullary cone, whose appearance suggests a lipoma. Both tumors were removed completely. The dura was closed in a tight manner. The pathological examination confirmed the lipoma and Dermoid Cyst. In the postoperative course, the child had a motor deficit in the lower limbs, which recovered completely after 3 months of physiotherapy.

Keywords

Dermal Sinus, Spinal Dysraphism, Myelomeningocele

1. Introduction

Congenital dermal sinus tract (CDST) is a congenital anomaly with an incidence of 1 in 2500 live births [1]-[3]. This pathology is thought to occur when the neural

ectoderm fails to separate completely from the cutaneous surface ectoderm between the third and eighth week of gestation [4] [5]. Clinical presentations include skin abnormalities, infections, and neurologic deficits [1] [6] [7]. Other forms of spinal dysraphysm, such as lipomyelomenigocele, tethered cord and split cord malformation may be accompanied by CDST. Approximately half of CDST is associated with dermoid or epidermoid tumor [8]. The treatment of CDST includes complete resection of the tract using intradural exploration and the treatment of the associated abnormalities such as other forms of spinal dysraphysm or inclusion tumor. In this paper, the authors report a case of CDST that presented with a dermoid cyst and lipoma.

2. Case Report

A 2-year-old girl was referred to our department. She had a history of lumbar skin anomaly since her birth. A week prior to her admission to our department, her mother noticed a clear liquid leaking through the lumbar skin anomaly, requiring a consultation with the pediatrician who referred her to us. At clinical examination, the child was alert with GCS at 15. Body temperature was 37.6˚C. There was a lumbar dimple with ostium (**Figure 1**) without other skin abnormalities such as hypertrichosis. Neurological examination was normal, particularly with no motor weakness and no sensory and reflex changes. The CT scan (**Figure 2**) performed shows a spinal tumor at L1 to L3 level with dermal sinus tract. Magnetic resonance imaging (MRI) (**Figure 3**) performed shows two intradural tumors at L2 to L4 level with dermal sinus tract.

Surgical resection of the tract was decided. In the operative room, the child was placed in a prone position. An elliptical skin incision was made centered around the tract. The dissection was carried out in the subcutaneous space to avoid entering the dermal sinus tract (**Figure 4**). Laminectomy from L2 to L3 was performed and dura was opened. The dermal sinus tract was attached to medullary cone and was cut there, allowing "en bloc" removal of the dermal sinus tract (**Figure 5**). There were two distinct types of tumor. One cystic is located posterior to the medullary cone and the other is located laterally to the medullary cone, whose appearance suggests a lipoma (**Figure 5**). Both tumors were removed completely. The dura was closed in a tight manner. The pathological examination revealed the fibrous lined by keratinizing squamous epithelium and proliferation of mature adipocytes, which are compatible with dermoid cyst and lipoma. In the postoperative course, the child had a motor deficit in the lower limbs, which recovered completely after 3 months of physiotherapy. After 18 months of follow-up, the child presents no complications and walks normally.

3. Discussion

Dermal sinus tract is a congenital anomaly with an incidence of 1 in 2500 live births [1]. The anomaly results from the failure of the neuroectoderm to separate from the cutaneous ectoderm at the end of neurulation [6] [8]. CDST is usually

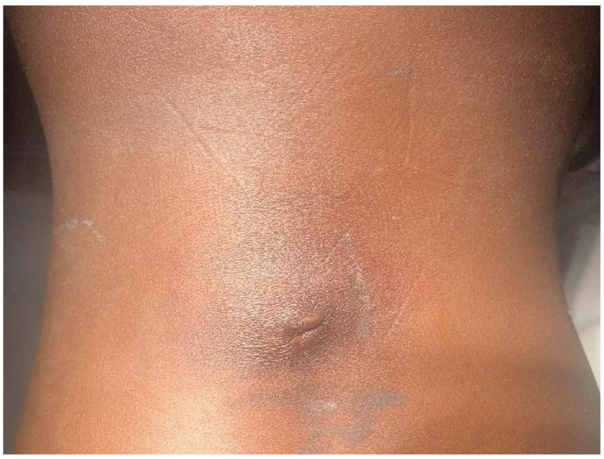

Figure 1. Lumbar dimple with ostium.

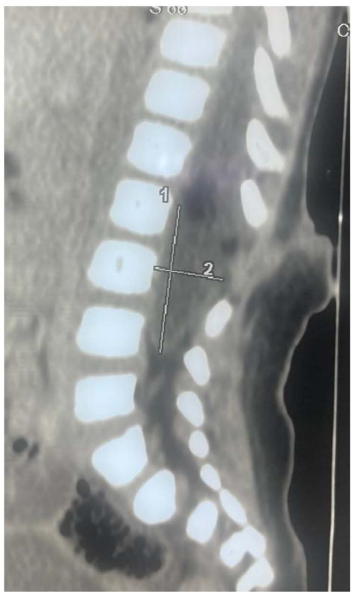

Figure 2. CT scan showing spinal tumor with dermal sinus tract.

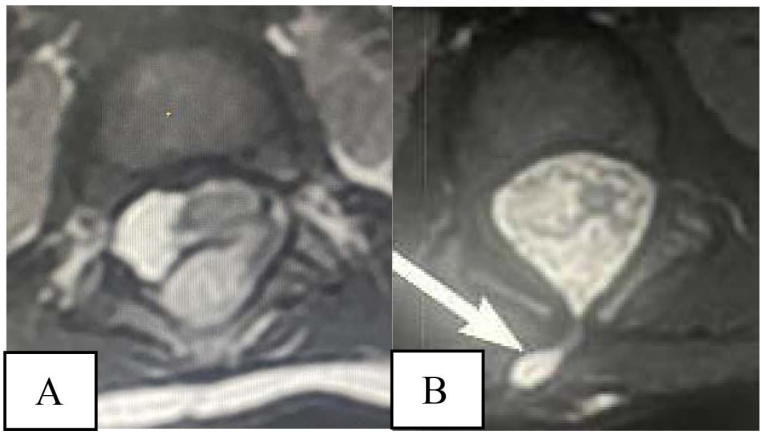

Figure 3. MRI showing spinal tumors (A) with dermal sinus tract (B).

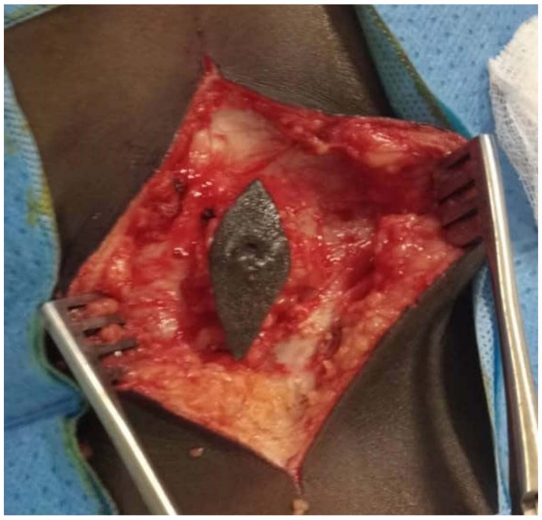

Figure 4. An elliptical skin incision with dissection of the subcutaneous space.

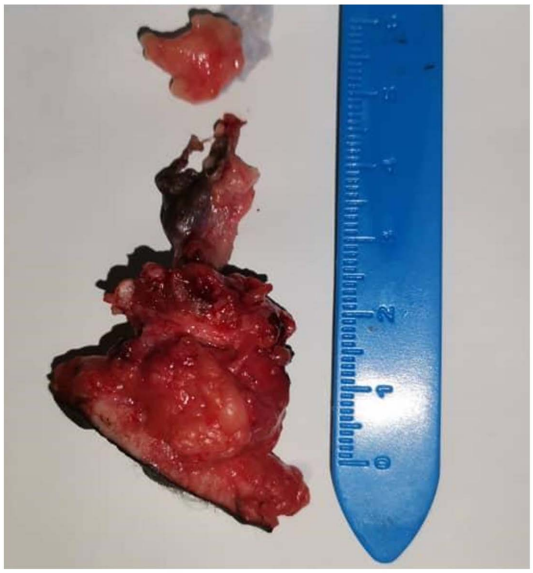

Figure 5. "en bloc" removal of the dermal sinus tract with dermoid cyst and lipoma.

seen in the midline but may be found in the paravertebral location [9] [10]. CDST may involve any level of neuroaxis but is frequently located in the lumbar or lumbosacral region [1] [7]. In our case, the CDST was located in the lumbar region at L1 to L3 level. CDST in cervical and thoracic regions is reported to be in less than 1% and 10% of cases, respectively [6]. CDST may end in any tissue plane but reach the dura and thecal sac in 60% of cases, as in our case. However, they can also end in the epidural space at 10% to 20% [1] [6]. Many enter the vertebral canal under or through a bifid lamina. Skin abnormalities are frequently associated with CDST such as hypertrichosis, hemangioma, dimples, ostium, erythemas, abnormal pigmentation, ostium or subcutaneous lipomas [4] [6]. The combination of 2 or more

of these skin abnormalities strongly suggests the DST [3]. In our case, the child presented with dimple and ostium (**Figure 1**). DST may be associated with other complaints such as neurologic impairment, orthopedic anomalies and infections [1] [6] [7]. Most children with DST have intact neurological function at birth [6]. However, Jindal *et al.* [6] reported that most patients in their study presented with neurologic deficits. The chance of neurologic deficit development is higher in patients who present at older ages [8] due to the relatively high rates of associated pathologies such as tethered cord, infection, and inclusion tumors. CDST provides a portal of entry for bacterial agents into the intraspinal compartments which can cause meningitis or abscess formation [11] [12]. Ackerman *et al.* [8] had 10% of infectious complications. Infection is the most serious risk associated with CDST. Dermal sinus tract can be accompanied by other pathologies such as myelomeningocele, tethered cord, lipomyelomeningocele, split cord malformation, filum abnormalities, and inclusion tumors. In our case, CDST was associated with two different types of tumor (**Figure 3**) confirmed by the pathological finding. According to the literature, half of all CDST are associated with dermoid or epidermoid tumor [8]. The coexistence of CDST with other tethered cord and intradural tumors was reported by Ackerman *et al.* [8] in 79% of patients. MRI is the first choice modality for diagnostic of CDST. The sinus tract appears typically as a high-intensity tract that ascends into the subcutaneous tissue with high intensity on both T1 and T2 weighted images (**Figure 3**). MRI is also used to identify any associated anomalies such as inclusion tumors. In our case, CT scan (**Figure 2**) provides evidence of spinal dysraphism but was not able to evaluate well the anomalies such as associated tumors. The MRI (**Figure 3**) showed two intradural tumors at L1 to L3 level with dermal sinus tract. The main differential diagnosis for CDST is dermal-sinus-like stalks (DSLS). Patients with DSLS tend to be older at the time of the first symptom, do not have a history of meningitis, lack dermoid or epidermoid tumors along their tract, and are histologically of pure mesodermal origin [3].

Treatment of CDST requires complete excision of the tract [1] [8]. The dura should always be opened, intradural extension of the lesion evaluated, and any associated pathologies treated at the same time. Surgical treatment should be applied as a prophylactic measure as soon as possible to prevent complications secondary to infection or other pathologic conditions, such as an inclusion tumor mass.

4. Conclusion

Early diagnosis and referral of DST allows for timely neurosurgical intervention, which can decrease morbidities caused by the development of complications secondary to infection, tethered cord, or tumor.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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