Lateral Buttock Congenital Dermal Sinus Tract
—Case Report—

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Abstract

A 6-month-old female presented with purulent discharge from a dimple in the right lateral buttock. A subcutaneous abscess was palpated on the right paravertebral region at the L5-S1 level. She had low-grade fever with laboratory findings of leukocytosis and elevation of C-reactive protein levels. Klebsiella and Enterococcus species were cultured from the pus. Computed tomography (CT) clearly showed a tract traversing the subcutaneous tissue and connecting to the abscess. Magnetic resonance (MR) imaging showed no abnormality in the spinal canal. The diagnosis was infected congenital dermal sinus (CDS) in the right buttock. After normalization of body temperature and laboratory findings in response to antibiotic treatment, the dermal sinus tract was surgically removed. Intraoperative findings showed that the tract gradually tapered and ended at the subcutaneous abscess space over the lumbosacral fascia. Histological examination confirmed the lesion was dermal sinus. Although laterally placed CDS in the buttocks is extremely rare with only 5 previous cases reported, lateral CDS should be included in the differential diagnosis of a dimple in the buttocks. CT as well as MR imaging should be performed to evaluate suspected lateral CDS.

Key words: congenital dermal sinus tract, lateral buttock, infection, etiology

Introduction

Congenital dermal sinus (CDS) is a type of spinal dysraphism with an approximate incidence of 1 in 2500 to 3000 live births.5,8) The sinus ostium is located in the midline of the posterior of the body from the occiput to the lumbosacral region, but most cases of CDS are found between the lumbar and sacral spine. Patients with CDS can present with skin findings, infection, and space-occupying lesions caused by inclusion tumors such as dermoid, epidermoid, and tethered cord syndrome.1,5) Internal endings of the sinus tract vary in each case, with some tracts penetrating the spinal canal. A tract which penetrates the dural sac may reach around the conus medullaris. The tract may be associated with dermoid/epidermoid tumors. The sinus tract blindly ends in the subcutaneous tissue in some cases.2,3,5) The most accepted etiology of CDS involves non-disjunction theory; a failure of separation between the neuro- and cutaneous-ectoderm in the midline results in persistent ectodermal tissue between the skin and neural elements between the third and fifth weeks of gestation.1,5,9) Therefore, the dimple will usually be found in the midline along the spinal axis.3–5) Laterally placed CDS in the buttocks is extremely rare, with only 5 cases reported.2,3,7)

We describe a sixth case of lateral CDS in the gluteal region presenting with infection, and illustrate the neuroimaging and surgical findings of this rare entity.

Case Report

A 6-month-old female presented with purulent discharge from a dimple in the right buttock. She had low-grade fever. Laboratory examinations showed leukocytosis and slight elevation of C-reactive protein levels. Physical examination revealed discharge of pus from a dimple in the right buttock. The right paravertebral soft tissue was inflamed and subcutaneous fluid collection was palpable at the L5-S1 level (Fig. 1). Klebsiella and Enterococcus species were cultured from the pus. The pediatrician suspected infected CDS, and the patient was referred to our department.

Magnetic resonance (MR) imaging showed subcutaneous fluid collection at the right paravertebral L5-S1 level, but no abnormality in the spinal canal. The sinus tract was slightly recognized in the right paramedian sagittal T1-weighted image (Fig. 2A). Computed tomography (CT) clearly showed the sinus tract, which was depicted as isodense to the skin, traversing the subcutaneous tissue and its connection to the abscess (Fig. 2B, C). The patient was first treated with antibiotics under a diagnosis of infected congenital dermal sinus. Surgery was performed af-
normalization of body temperature and laboratory findings.

A circumferential skin incision around the dimple was made. The sinus tract was easily identified in the subcutaneous fat tissue, and the skin incision was extended along the tract, which ascended to the cranial side and close to the midline structure. The tract gradually tapered and ended at the subcutaneous abscess over the lumbosacral fascia. The sinus tract was totally removed and the abscess irrigated (Fig. 3). Histological examination showed that the inner surface of the tract was lined with stratified squamous epithelium with hairs and adnexa of the skin. Inflamed granulation tissue was also found. The postoperative course was uneventful, and no infection recurred.

Discussion

Lateral CDS in the buttock is an extremely rare lesion, with only 5 previous cases (Table 1). Three patients were male and 3 were female, aged from 6 months to 5 years. Five of six lesions were found on the left side of the buttocks. All but one patient presented with focal infection of the dermal sinus tract. Surgical findings disclosed that the internal endings of two tracts were located in the spinal canal, whereas four tracts ended blindly in the subcutaneous soft tissues. Four patients had concomitant skin stigmata along the midline of the lumbosacral region, three patients had additional dimple(s), and one had abnormal pigmentation in the intergluteal cleft. However, in all cases including ours, the ostia of the lateral CDS had no abnormality such as focal pigmentation or hypertrichosis. These concomitant skin stigmata along the spinal axis seem to suggest that lateral CDS is also derived from failed closure of the midline. However, why the dermal sinus

![Photograph showing a dimple in the right buttock (thick arrow). The right paravertebral soft tissue was inflamed and swollen due to subcutaneous fluid collection (arrows).](image1)

![Right paramedian sagittal T1-weighted magnetic resonance image showing the sinus tract was slightly recognized (arrow). A: Axial computed tomography (CT) scan at the S1 level showing a subcutaneous abscess over the right paravertebral muscle (arrows). C: Axial CT scan at the S4 level clearly showing the dermal sinus tract traversing the subcutaneous tissue (arrow).](image2)

![Photograph of the surgical specimen. A probe is inserted in the lumen of the dermal sinus tract. B: Photomicrograph showing the sinus tract was lined by stratified squamous epithelium. The surrounding tissue showed fibrosis and inflammation. Hematoxylin and eosin stain, original magnification ×100.](image3)

Table 1 Summary of the clinical features of cases of lateral congenital dermal sinus

<table>
<thead>
<tr>
<th>Author (Year)</th>
<th>Age</th>
<th>Sex</th>
<th>Side</th>
<th>Presentation</th>
<th>Internal ending of the tract</th>
<th>Concomitant skin stigmata</th>
</tr>
</thead>
<tbody>
<tr>
<td>Carrillo et al. (1985)</td>
<td>22 mos</td>
<td>M</td>
<td>left</td>
<td>suppuration</td>
<td>intraspinal canal, outside the dural sac</td>
<td>none</td>
</tr>
<tr>
<td></td>
<td>2 yrs</td>
<td>M</td>
<td>left</td>
<td>suppuration</td>
<td>intraspinal canal, intradural with lipoma</td>
<td>two sinuses, lumbosacral lipoma</td>
</tr>
<tr>
<td>Ikwueke et al. (2008)</td>
<td>3 yrs</td>
<td>F</td>
<td>left</td>
<td>skin finding</td>
<td>extraspinal canal, the gluteal fascia</td>
<td>pigmented patch, midline</td>
</tr>
<tr>
<td>Qi et al. (2010)</td>
<td>3 yrs</td>
<td>F</td>
<td>left</td>
<td>suppuration</td>
<td>extraspinal canal, the tip of coccyx</td>
<td>sinus, postanal</td>
</tr>
<tr>
<td></td>
<td>5 yrs</td>
<td>M</td>
<td>left</td>
<td>suppuration</td>
<td>extraspinal canal, the tip of coccyx</td>
<td>sinus, postanal</td>
</tr>
<tr>
<td>Present case</td>
<td>6 mos</td>
<td>F</td>
<td>right</td>
<td>suppuration</td>
<td>extraspinal canal, paravertebral fascia</td>
<td>none</td>
</tr>
</tbody>
</table>

F: female, M: male.

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tract formed distantly from the midline has not been determined.

Spinal cord formation during embryogenesis consists of two stages: primary and secondary neurulation. Secondary neurulation is characterized by the fusion of bilateral neural folds in the midline and formation of the neural tube between the third and fifth gestational weeks. Secondary neurulation to form the filum terminale and conus medullaris is characterized by the fusion and subsequent partial regression of the caudal cell mass. Therefore, most of the spinal cord is derived from primary neurulation, and a part of the caudal tip of the spinal cord is derived from secondary neurulation. The etiology of dermal sinus, which is speculated to be persistent cutaneous ectoderm in the subcutaneous layer, is generally accepted as separation failure between the neuro- and cutaneous ectoderm.

The dimples above the gluteal cleft are considered to be derived from midline disjunction of the primary neurulation and the tract will ascend subcutaneously. In contrast, dimples in the gluteal cleft and lower sacral segment are thought to be derived from secondary neurulation and the tract will not ascend but ends beside the fascia along the lower sacral spine. In the present case, the spinal level of the lateral CDS was S4, but the surgical findings showed the tract ascending and directing to the midline and then ending above the fascia at the S1 level. Thus we speculate that our case of lateral CDS was derived from a disorder of primary neurulation.

The etiology of lateral occurrence of CDS has not been elucidated, but may involve abnormal canalization of the caudal cell mass. However, this theory does not sufficiently explain why the tract is formed laterally. A “zipping error theory” has been proposed to explain the formation of “paramedian” (not lateral) CDS. It is an accepted concept that the neural tube is formed by the closure of bilateral neural folds in the midline, like closing a zipper. Therefore, if aberrant lipomatous tissue occupies a part of the trajectory for the closure line of neural folds, a redundancy of neural fold(s) may be produced around the lipomatous tissue. Like a zipping error, a part of the redundant neural fold will remain laterally to the midline and may result in off-midline CDS. This zipping process accounts for primary neurulation, so this “zipping error theory” can be applied to off-midline CDSs between the cervical and upper sacral spine. Since our case of lateral CDS was derived from a disorder of primary neurulation, this zipping error theory may be applicable to the etiology of our case.

MR imaging is the first choice of modality to diagnose CDS. CDS typically appears as a low intensity tract that ascends in the subcutaneous tissue with high intensity on both T1- and T2-weighted images. This modality is especially useful for detecting low-placed conus and associated anomalies such as inclusion tumors or split cord malformations. However, MR imaging may not clearly demonstrate small tracts, especially in a neonate or with a deviated course like our case. Only 40% of tracts were detected in the preoperative MR images. MR imaging was described in only one previous case of lateral CDS. In our case, CT showed that the tract, which appeared isodense to the dermis, traversed the subcutaneous fat layer, which appeared as low density. Although CT does not allow evaluation of the extension of the tract into the spinal canal, we should consider CT in patients with suspected CDS.

The present case of lateral buttock CDS demonstrates that infected lateral CDS might be misdiagnosed as benign skin lesion such as atheroma or skin acne. Lateral CDS should be considered in the differential diagnosis of a dimple in the lateral buttock region.

References