Case Report

Image findings of dermal sinus in the lateral buttocks

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Abstract

This case series showed the sonographic and magnetic resonance imaging findings of two patients with dermal sinuses in the lateral buttocks. According to the 2 present cases, and 8 cases previously reported, the distal sides of the sinuses may be connected to internal tissues, such as the subarachnoid, gluteal fissure, peritoneal cavity, or pelvic bone. Since it is difficult to predict the involvement of the distal side of a dermal sinus based on the dermal pit location, ultrasound and magnetic resonance imaging were postulated as a useful modality to aid evaluation. Based on these image findings, complete surgical resection was performed by surgeons.

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Introduction

A congenital dermal sinus is an epithelial-lined tract of variable length that runs from the skin to deeper tissues [1–4]. Although this condition most commonly involves the midline, it sometimes occurs in lateral locations, such as periauricular, peristernal, lateral cervical, or lateral buttock regions [1,2,5–13].

To date, a few cases with dermal sinuses in lateral buttocks have been reported, and the affected side, that is, the proximal or distal side, differed in each of these reports [8–10,12,14]. For correct surgical planning, it is important to evaluate the lesion completely from the proximal to the distal side [8–10,12,14]. Previous case reports have shown dermal sinuses on computed tomography or magnetic resonance imaging (MRI), but no report has visualized them via ultrasound. Recently, ultrasound has been reported to be a useful modality to evaluate the distal side of the dermal sinus [1,6]. Further, it is usually accepted as the first-choice imaging modality for pediatric patients because it carries no risk of irradiation and can be performed at the patient’s bedside [1,2,6,7]. In addition,
sonographers or radiologists may be able to use the findings from the sonographic examination to recommend further examination, such as MRI, for surgeons or physicians to evaluate the distal side of the dermal sinus more accurately [1,6].

In this case report, we describe two cases with different types of dermal sinus in the lateral buttocks. Ultrasonography and MRI provided useful information about the distal side of the sinuses, enabling surgeons to completely resect these lesions. In addition, we review previous case reports and summarize the types of dermal sinuses seen in lateral buttocks to date.

**Case presentation**

**Patient 1**

A 1-year-old female presented with a mass and a dimple on her right lower buttock (Fig. 1A). The mass was not palpable. Ultrasonography revealed a tract leading from the buttock to an infected mass of subcutaneous tissue and a pelvic mass on the right side of the rectum (Fig. 1B and 1C). Therefore, MRI was recommended and subsequently performed. Similar to the ultrasonography results, the MRI also showed a tract that was connected to a cystic mass in the buttock and a cyst-
Fig. 2 – The sonography and MRI of a dermal sinus in the lateral buttock in a 4-year-old female. 
(A) Dermal pit (arrow) located at the upper outer quadrant of the left gluteal area with the top of the intergluteal cleft present above it. 
(B) The axial sonogram shows a hypoechoic cystic lesion (arrows) in the subcutaneous tissue. 
(C) The axial sonogram shows that the fistula (arrow) is running into the iliac crest. 
(D) The axial sonogram shows that distal side (arrow) is not clearly seen due to an acoustic shadow from the iliac bone. 
(E) Coronal T2-weighted image shows that the dermal sinus (arrows) is running cranially into the iliac crest. 
(F, G, H) The axial T2-weighted image (panel F) is at level of line f, panel G is at the level of line g, and panel H is at the level of line h in panel E. The dermal sinus (arrows) originates in the subcutaneous tissue and runs cranially to the top of the iliac crest. The fistula is penetrating between the iliacus muscle and the longissimus thoracis muscle near the sacroiliac joint. The distal side of the fistula (arrows) is running into the unfused anterior side of sacrum (arrowheads).

tic mass adjacent to the rectum (Fig. 1D-1F). The right and left ovaries could be detected in the pelvis. The aforementioned cystic masses showed high signal intensity in diffusion weight images (figure not shown). Therefore, these lesions were thought to be a congenital dermal sinus tract and cyst. Based on the radiographic findings, the surgeons used 2 surgical approaches: a laparoscopic approach for resection of the pelvic mass, and a prone position approach for resection of the dermal sinus in the lateral buttock. Surgical resection was performed and the surgical proof that the dermal sinus and fistula were covered with squamous cells without any malignant cells was obtained. The dermal sinus ran into the cystic mass located in the subcutaneous tissue, and the distal sinus, which originated in the cystic lesion, penetrated the piriform muscle. The distal side was connected to the anterior coccyx. The pelvic cystic lesion and subcutaneous dermal sinus were located close to one another but were not found to be pathologically linked.
Patient 2

A 4-year-old female presented with a dimple on her upper left buttock (Fig. 2A), which had been visible since birth. Although she had undergone pit excision at the age of 3, infection in the buttock had been recurrent. Ultrasonography revealed a sinus tract, whose distal side was oriented towards the sacroiliac joint (Fig. 2B-2D); therefore, MRI was recommended and subsequently performed. Lesions with high signal intensity in diffusion weight images (figure not shown) were visible and a funicular-structured stripe adjacent to the anterior sacrum (Fig. 2E-2H) was seen. Therefore, the lesions were thought to indicate a congenital sinus tract. Based on the radiographic findings, the surgeons selected the prone position approach for resection of the dermal sinus in the lateral buttock. Surgical resection was performed and surgical proof that the dermal sinus was covered with squamous cells was obtained. The dermal sinus originated in the subcutaneous tissue and ran cranially to the top of the iliac crest. The fistula penetrated between the iliacus muscle and the longissimus thoracis muscle; the distal side of the fistula comprised fibrous tissue that was connected to the anterior sacrum.

The location of the dermal pit is typically described by dividing the gluteal area into 4 equal parts, and is based on the association between the dermal pit and the intergluteal cleft, as summarized in Table 1 and Fig. 3, where the red ovals mark the dermal pits connected to the spinal lesion and the blue ovals mark the nonconnected pit.

Discussion

This is the first case series that focuses on sonographic and magnetic resonance images of a dermal sinus in the lateral buttocks. In both cases, the distal side of the sinus passed through a different route and connected to different tissues. Therefore, ultrasound was considered a useful modality to accurately evaluate the distal side of the sinus, such that further examination could be recommended to the surgeon or physicians based on the sonographic findings. Surgeons would, therefore, be able to perform the appropriate surgical resection in each case based on the sonographic and MRI findings.

In the current cases and previous reports, ultrasound was considered a useful modality to evaluate the complications or visualize the distal side of a dermal sinus [1,6]. However, it became difficult to achieve accurate visualization if the distal side was associated too closely with the pelvic bone or ran near/into the pelvic cavity. Therefore, in such cases where radiologists cannot completely evaluate the distal portion of the tract, further examination, such as MRI, might be recommended. Based on these image findings, surgeons can select the appropriate surgical approach and perform complete resection of these lesions.

Although the etiology of the dermal sinus remains unclear, there are some hypotheses regarding its etiology. First, the

Review of previous case reports and our patients

To date, a few case reports have been published regarding dermal sinuses in the lateral buttocks [8-12,14]. We have summarized these previous cases and both of our cases (total 10 cases) in Table 1. Eight patients were female and 2 were male, with an age range of 6 months to 22 years. Five lesions were located on the right buttock and 5 lesions were observed on the left buttock. Spinal anomalies were detected in 2 cases.
Table 1 – Review of previous and current cases.

| Case | Reference          | Age   | sex | Side | Proximal side                                                                 | Distal side                                 | Accompanying anomaly or disease               |
|------|--------------------|-------|-----|------|--------------------------------------------------------------------------------|---------------------------------------------|-----------------------------------------------|
| 1    | Bruce et al. [12]  | 22 years | F   | L    | Upper outer quadrant of the left gluteal area below the top of the intergluteal cleft | Duplication cyst and peritoneal cavity | Hemivertebrae Duplication cyst                |
| 2    | Carrillo et al. [8] | 22 months | M   | L    | Upper inner quadrant of the left gluteal area below the top of the intergluteal cleft | The subarachnoid                           | Bone defect at the posterior arch of L5       |
|      |                    |       |     |      |                                                                                 |                                             | Meningitis due to the infection from dermal fistula |
| 3    |                    | 2 years | F   | R and midline | Lower outer quadrant of the left gluteal area below the top of the intergluteal cleft | Subcutaneous tissue | Intraspinal lipoma Spinal bifida at L5      |
|      |                    |       |     |      |                                                                                 |                                             |                                               |
| 4    | Ikwueke et al. [14]| 3 years | F   | R    | Upper outer quadrant of the right gluteal area above the intergluteal cleft    | Gluteal fascia                             | Light brown pigmented patch on the left buttock |
| 5    | Qi et al. [10]     | 3 years | F   | L    | Upper outer quadrant of the left gluteal area above the top of the intergluteal cleft | Tip of the coccyx                          |                                               |
| 6    |                    | 5 Years | M   | L    | Upper outer quadrant of the left gluteal area below the top of the intergluteal cleft | Tip of the coccyx                          | Recurrent infection of buttock               |
| 7    | Yamaguchi et al. [9]| 6 months | F   | R    | Upper outer quadrant of the right gluteal area above the top of the intergluteal cleft | Lumbosacral fissure                        | Inflamed subcutaneous tissue                 |
| 8    | Nishimon et al. [11]| 14 years | F   | R    | Upper outer quadrant of the right gluteal area above the top of the intergluteal cleft | Periosteum of right sacral ala             | Persistent cloaca Infection                  |
| 9    | Current patient 1  | 1 year  | F   | R    | Upper outer quadrant of the right gluteal area above the top of the intergluteal cleft | Anterior sacrum                           | Pelvic dermoid cyst Inflamed subcutaneous tissue |
| 10   | Current patient 2  | 4 years | F   | L    | Upper outer quadrant of the left gluteal area above the top of the intergluteal cleft | Periosteum of left sacral ala             | Inflamed subcutaneous tissue                 |

F: female, M: male, R: right, L: left.
Proximal side is described based on the 4 equal parts of the gluteal area and based on the association between dermal pit and intergluteal cleft.

congenital midline dermal sinus is thought to occur because of a defect in neurulation in the third to fifth week of gestation [13]. In cases where the dermal sinus is present on the midline and is associated with the buttocks, it is important to clarify the association between the dermal pit and the intergluteal cleft, in order to predict the connection of the pathology to the intraspinal space [15]. In previous cases where the dermal pit was located near the midline of the buttocks, an association between the dermal sinus and the spinal lesion tended to develop [8]. On the other hand, regardless of the pit location being above or below the intergluteal cleft, associations between the distal or proximal sides of the sinuses and the intraspinal lesion were varied [8–12,14]. Second, a dermal sinus not present at the midline may result from either a branchial anomaly [16,17], or from disorderly fusions between 2 dermal or soft tissues near the dermal fistula [6,18]. In the current case 1 and in some previous cases [9–11,14], the distal side of the sinus was not associated with any vertebral anomalies, and
was connected to various tissues. Therefore, it is usually difficult to accurately predict the involvement of the distal side of a dermal sinus in the lateral buttocks based only on the location of the dermal pit, and ultrasound and MRI may be useful modalities to evaluate the distal side. In addition, the results of previous cases and the current cases did not allow us to clarify the etiology of this disease.

**Conclusion**

This is first case series that focuses on sonographic and magnetic resonance images of dermal sinuses in the lateral buttocks. The distal sides of the sinuses in both cases were connected to different tissues, due to which the extent of the distal side could not be predicted based on the location of the dermal pit alone. We found ultrasound to be an effective, accurate, and useful modality to evaluate the distal side of the dermal sinus in the lateral buttocks, and were able to recommend further examinations, such as MRI, based on the sonographic findings. Based on these image findings, the surgeons were able to select the appropriate surgical approach for complete resection of the dermal sinus of the lateral buttocks.

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