CASE REPORT

Late-onset congenital lateral dermal sinus tract

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SUMMARY

Cases of laterally located, congenital dermal sinus tracts are extremely rare, with only six having been reported to date. We describe a case of a 14-year-old girl who developed symptoms of this type of sinus tract at an age that was considerably older than is usually reported. At the age of 12 years, the patient exhibited a purulent discharge from a pit on the right buttock. MRI indicated the presence of two tracts running from the right buttock skin to a cystic lesion that had formed on the right ala of the sacral spine. The lesion was surgically resected and successfully reconstructed using a partial iliocostalis lumborum muscle flap, without any functional morbidity. From our experience, such flaps appear to be appropriate treatment choices for lateral congenital dermal sinus tracts that develop late and result in large defects.

Figure 1 Preoperative status of the back. Two pits surrounded by ruddy skin on the right buttock; one was 7 cm from the midline and the other was 12 cm from the midline, below the posterior iliac crest.

BACKGROUND

A congenital dermal sinus tract is a type of closed spinal dysraphism characterised by an epithelium-lined sinus tract extending inward from the dorsal skin surface for a variable distance.^{1,2} The incidence of congenital dermal sinus tract is approximately 1:2500–3000 live-births.³ Such a tract is normally located at the midline of the dorsum. Only six cases of lateral dermal sinus tracts have been previously reported.^{4–7} We report an extremely rare case of laterally located, dermal sinus tracts in a 14-year-old girl, who was successfully treated with surgical excision and reconstruction.

CASE PRESENTATION

The patient's parents provided their consent for the publication of this report. The patient was diagnosed, at birth, with a persistent cloaca, and had undergone several operations. At 12 years of age, a scab and pit appeared on her right buttock that eventually oozed a purulent discharge. The condition was originally diagnosed, at another dermatology department, as a chronic pyoderma. However, the lesion did not resolve after intravenous and oral administrations of several antibiotics. At 13 years of age, the patient was referred to our department. On examination, two pits, surrounded by ruddy skin, were observed on her right buttock; one was 7 cm from the midline, and the other was 12 cm from the midline, below the posterior iliac crest (figure 1).

INVESTIGATIONS

Careful surgical probing using a surgical probe revealed that both pits possessed sinus tracts, running subcutaneously towards the midline. However, the ends of the tracts were unclear. MRI showed 2 sinus tracts running, subcutaneously, from

the right buttock skin towards the right erector spinae muscle. The sinus tracts were interconnected, forming one large tract that ran deep between the upper margin of the gluteus maximus and gluteus medius muscles. The tract eventually reached the dorsal side of the right ala of the sacral spine, forming a cystic lesion (figure 2). The sacral spine and the coccyx were hypoplastic, and the spinal canal was open dorsally; thus, the patient was also diagnosed with spina bifida. A retrospective review of the MRI scan performed at the other hospital, when the patient was 7 years old, showed that the lesion was present at that time; however, it had been overlooked. Based on these imaging findings, she was diagnosed with congenital lateral dermal sinus tracts, spina bifida, and a persistent cloaca.

TREATMENT

As conservative treatment was ineffective, surgical excision was performed when the patient was 14 years old. After the two tracts were stained with crystal violet, they were carefully excised with the surrounding abnormal skin and scars. The skin abnormalities were believed to have been caused by the chronic inflammation. The caudal tract was pulled from the cephalic incision site to avoid a major skin incision. Finally, the tracts and the cystic lesion, near the periosteum of the right sacral ala, were completely excised with the periosteum and surrounding tissue. A bundle of hair shafts was found in the cyst and tracts. Following excision, a large tissue defect (depth, 6 cm; length, 4 cm; width, 3 cm) was present near the sacral ala. Since the defect was too large for simple closure, tissue filling was required to avoid creating a dead space around the spine. Therefore, a partial iliocostalis lumborum muscle flap (10×3.5 cm), above the superficial side of the defect,

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Figure 2 Preoperative MRI. MRI showing the cephalic sinus tract running from the right buttock skin towards the right erector spinae muscle. The tract crossed the upper margin of the gluteus maximus muscle. Finally, the tract reached the dorsal side of the right sacral ala to form a cystic lesion.

was elevated to fill the space (figure 3). Two drains were inserted, above and under the flap. After irrigating the wound with saline, simple closure without flap elevation was performed because the skin deficiency was small.

OUTCOME AND FOLLOW-UP

Microscopic examination showed that the inner surface of the dermal sinus tract was lined with stratified squamous epithelial

cells; a bundle of hair shafts and keratinised material filled the tract. An abscess had formed adjacent to the tract, and the surrounding tissue showed fibrosis (figure 4). No recurrence or inflammation of the wound was noted postoperatively. Moreover, apparent functional deficiency of the iliocostalis lumborum muscle was not observed, and good posture and all-directional movement of the back were completely preserved.

DISCUSSION

Congenital dermal sinus tracts are generally located at the midline of the back. Only six cases of lateral dermal sinus tracts have been reported.⁴⁻⁷ Further, in our case, the symptoms did not manifest until the patient was 12 years old, which is considerably older than the usual age of symptom manifestation; in the six previously reported cases, the age of onset was between 6 months and 5 years of age.⁴⁻⁷ Although the present patient exhibited a complete dermal sinus tract on an MRI performed at the age of 7 years, it had been overlooked. However, the patient did not have back-related symptoms until she was 12 years old, which may have been because the pit was too small to be detected from the surface.

Although surgical probing did not provide a definitive diagnosis, MRI revealed that the disease involved congenital lateral dermal sinus tracts. Generally, probing or staining of the tracts must be performed carefully to avoid pushing the tract contents into the dural space. If the tracts had continued to the dural space, they might have caused meningitis or intradural abscesses. In our case, since preoperative MRI showed that the tracts did not continue to the dural space, staining and surgical excision could be performed safely, with a lower risk of harming the intradural space.

Figure 3 Intraoperative photographs. The tracts run deeply and medially (left upper). Note that the cyst is filled with crystal violet beyond the lateral edge of the iliocostalis lumborum muscle. The partial iliocostalis lumborum muscle flap measures 10×3.5 cm and is designed above the superficial side of the defect (right upper). The asterisk indicates the bottom of the defect (dorsal side of a sacral ala). The elevated iliocostalis lumborum flap (left lower). The flap is moved into the defect (right lower).

Figure 4 Pathological findings. The inner surface of the dermal sinus tract is lined with stratified squamous epithelium. A bundle of hair shafts and keratinised material fills the tract. Scale bar: 200 μ m.

The operation was performed when the patient was 14 years old, when the subcutaneous tissue had thickened, and the distance from the skin to the bottom of the sinus tract was >8 cm. The sinus tracts may have enlarged as well. Thus, the final defect, after excision of the tracts, was too large to allow simple closure, as was reported in previous studies. To fill the defect and avoid creating a dead space around the spine, a partial iliocostalis lumborum muscle flap was elevated and used.

The iliocostalis lumborum muscle is the most lateral component of the erector spinae muscle, which is a large musculotendinous mass that differs in size and composition at different vertebral levels. The dorsal rami of the spinal nerves are responsible for motor innervation of the muscle, allowing extension, standing and side flexion of the lumbar vertebra. Since the iliocostalis lumborum muscle is a part of the large erector spinae muscle, its use as a muscle flap usually does not cause a functional deficiency in the movement of the back because the residual muscle may show a compensatory function. Thus, such

a flap may be an effective choice for treating the large defects created by late-onset, lateral congenital dermal sinus tracts.

Learning points

- ▶ A congenital dermal sinus tract may occur laterally, rather than exclusively along the midline.
- ▶ Congenital lateral dermal sinus tracts may be asymptomatic for many years, preventing their recognition until adolescence.
- ▶ Large defects, caused by surgical excision, should be filled with tissue to avoid creating a dead space around the spine.
- ▶ The partial iliocostalis lumborum muscle flap is an effective choice for treating such defects without major functional morbidity.

Competing interests None.

Patient consent Obtained.

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