BRIEF COMMUNICATION

Unusual Case of Multiple Lateral Spinal Dermal Sinuses in a Child

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1. Introduction

Walker and Bucy (cited by Carrillo et al1) first used the term “congenital dermal sinus” in 1934 to describe a rare form of spinal dysraphism resulting from an incomplete separation of the neural ectoderm from the epithelial ectoderm in the early embryologic development. As is true with other forms of spinal dysraphism, the tract can be located in or near the midline anywhere from the coccyx to the cervical region, with 75% of the tract observed in the lumbar and lumbo-sacral areas.2,3 The tract is most often unique, associated with local skin markers.

In this report, we present an unusual case of multiple and lateral spinal dermal sinuses associated with other skin markers.

2. Case Report

A 9-month-old female baby was referred to our department for skin abnormalities on the right buttock. The baby was born at full gestation to nonconsanguineous parents, and since birth, she was noted to have skin markers suggestive of spinal dysraphism, although she had no history of infection. On admission, a physical examination revealed a healthy child, with growth parameters within the normal range. She had a normal head circumference, and she was able to move all her extremities equally. Examination of the back (Figures 1A–1D) showed a subcutaneous lipoma on the right buttock with a “finger-like” skin tag, deviated gluteal fold, and a small, shallow, almost midline coccygeal dimple. Firm compression of this pit with two thumbs showed that it had a “bottom.” A thorough inspection of the back confirmed the existence of two other dimples: the first was 2 cm lateral to the coccygeal pit and the second was at the base of the dermal finger-like skin tag. Compression of the area surrounding these two latest dimples revealed their indiscernible depth and discharge of whitish matter. However, according to the mother, there was no swelling from these dimples previously.

Magnetic resonance imaging of the brain and the whole spine revealed a subcutaneous lipoma in the right buttock: the sinus tracts were well recognized in the axial T1 and T2 images through the subcutaneous tissue, and they tapered and disappeared over the paravertebral fascia (Figures 1E and 1F). No further abnormalities were detected primarily at the level of the conus and configuration of the filum. Circumferential skin incision around the lateral dimples and the finger-like skin tag was performed. Most of the lipoma was removed, and the sinus tracts were easily identified: they gradually tapered and both ended over the paravertebral fascia. The postoperative course was uneventful.

The histopathologic examination revealed the presence of an epidermoid cyst associated with the dermal tract located at the base of the dermal finger-like skin tag.

3. Discussion

Spinal dermal sinus is a rare form of spinal dysraphism that results from a failure of disjunction early in embryogenesis. The tract lined by stratified squamous epithelium can be
found in or near the midline anywhere from the coccyx to the cervical area; however, most cases are located in the sacral and lumbar areas. The internal ending of the sinus tract varies from one case to another; it reaches the dura mater and thecal sac in 60% of the patients, and it ends in the epidural space in 10–20% of the patients. The tract blindly ends in the subcutaneous tissue in rare cases.4

In 1985, Carrillo et al1 reported two cases of spinal dermal tract that did not originate at the midline. In their first case, the dimple was located on the left buttock; however, the second patient had three dimples on the lower back and the left buttock. To the author’s knowledge, this location—off the midline—has never been reported previously. More recently in 2014, Nishimon et al5 described a case of 14-year-old female with two pits on her right buttock; they were located below the posterior iliac crest at 7 and 12 cm from the midline, respectively. Both tracts ended near the periosteum of the right sacral ala. As noted by Nishimon et al., only six other cases of lateral dermal sinus tracts have been reported in the literature. Laterally placed spinal dermal sinus is exceedingly rare.1,4,6 Some characteristics seem to be important; no patient presented with infection of the central nervous system, except a case of meningitis reported by Carrillo et al,1 in which the infection followed radiography with metrizamide injected through the opening at the skin. In this lateral site, most often the tract ended blindly in the subcutaneous soft tissues (5 out of 7 cases from the literature) similar to our patient. Only two tracts reported by Carrillo et al,1 ended in the spinal canal: outside the dural sac in one case and intradural in the other case. Moreover, in cases of lateral spinal dermal tract previously reported in the literature,1,5,6 four patients had additional dimples; our patient also had two dimples with a coccygeal pit. We also note that dermal sinuses in midline are commonly associated with a dermoid cyst, and those in the paramedian location are associated with an epidermoid cyst’ similar to our case. Our report broadens the clinical literature regarding this uncommon entity.

Conflicts of interest

The authors have no conflicts of interest relevant to this article.

References