J Ped Surg Case Reports 14 (2016) 38–41

Contents lists available at ScienceDirect

Journal of Pediatric Surgery CASE REPORTS

journal homepage: www.jpscasereports.com

Tailgut cyst in a female infant with a skin dimple at the coccygeal region

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ARTICLE INFO

Article history:
Received 1 July 2016
Received in revised form
29 August 2016
Accepted 29 August 2016

Key words:
Tailgut cyst
Teratoma
Presacral

ABSTRACT

A tailgut cyst is a congenital cystic lesion that is situated at the presacral and postrectal area and is considered to be a remnant of the tailgut that develops in early fetal life and usually regresses later. Approximately 20 pediatric cases of tailgut cyst have been reported. We report an infantile case of tailgut cyst that was complicated with a skin dimple at the coccygeal region. The cyst was completely resected and the pathological diagnosis was mature teratoma. We finally diagnosed it as a tailgut cyst by several clinical findings including the site of the cyst, MRI image, the fact that it was complicated with a skin dimple, and the pathological findings.

A tailgut cyst is a congenital lesion that usually exists at the presacral and postrectal area. It is considered to be a remnant of the tailgut which appears behind the hind gut early in fetal development and usually regresses later [1]. Many adult cases of tailgut cyst have been reported [2] and some of them showed malignant transformation of the tailgut cyst, which reflected a clinically important aspect [3,4].

Conversely, there are few reports of pediatric tailgut cysts and only approximately 20 cases have been reported [5–14]. We report an infant with a tailgut cyst that was accompanied by a skin dimple at the coccygeal region.

1. Case

A female infant was referred to our department for the treatment of a presacral cyst, which had been incidentally found by magnetic resonance imaging (MRI) examination. The patient had a skin dimple at the coccygeal region (Fig. 1a) and MRI had been performed to check for abnormal development of the spinal cord. The spinal cord and spinal canal were normal but there was a multiloculated cyst at the presacral and postrectal area (Fig. 1b,c). The cyst had no calcification and did not contain a fat element. The cyst showed homogeneous hypo-intensity on T1-weighted images and hyper-intensity on T2-weighted images. There was no solid component in the cystic cavity. Barium enema showed a presacral mass that compressed the rectum (Fig. 1d). These images suggested that the cyst was a tailgut cyst.

She underwent surgery at 12 months of age and complete resection of the cyst was performed under general anesthesia. A skin incision was made by the posterior sagittal approach to resect both the skin dimple at the coccygeal region and the cystic lesion. The cyst was situated close to the posterior wall of the rectum (Fig. 2a) but could be resected without injury to the rectal wall. The coccygeal bone was also resected. The puborectal muscle was not enfolded by the cyst (Fig. 2b) and postoperative recovery was uneventful.

Pathological examination showed that the cyst was a unilocular cyst and contained serous fluid. The epithelial layer consisted of various epithelia such as, pseudostratified ciliated columnar, pseudostratified cuboidal and transitional types (Fig. 3a–c). The presence of epithelium was verified by the presence of connective tissues, adipose tissues and smooth muscle fibers (Fig. 3d), and

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http://dx.doi.org/10.1016/j.epsc.2016.08.011

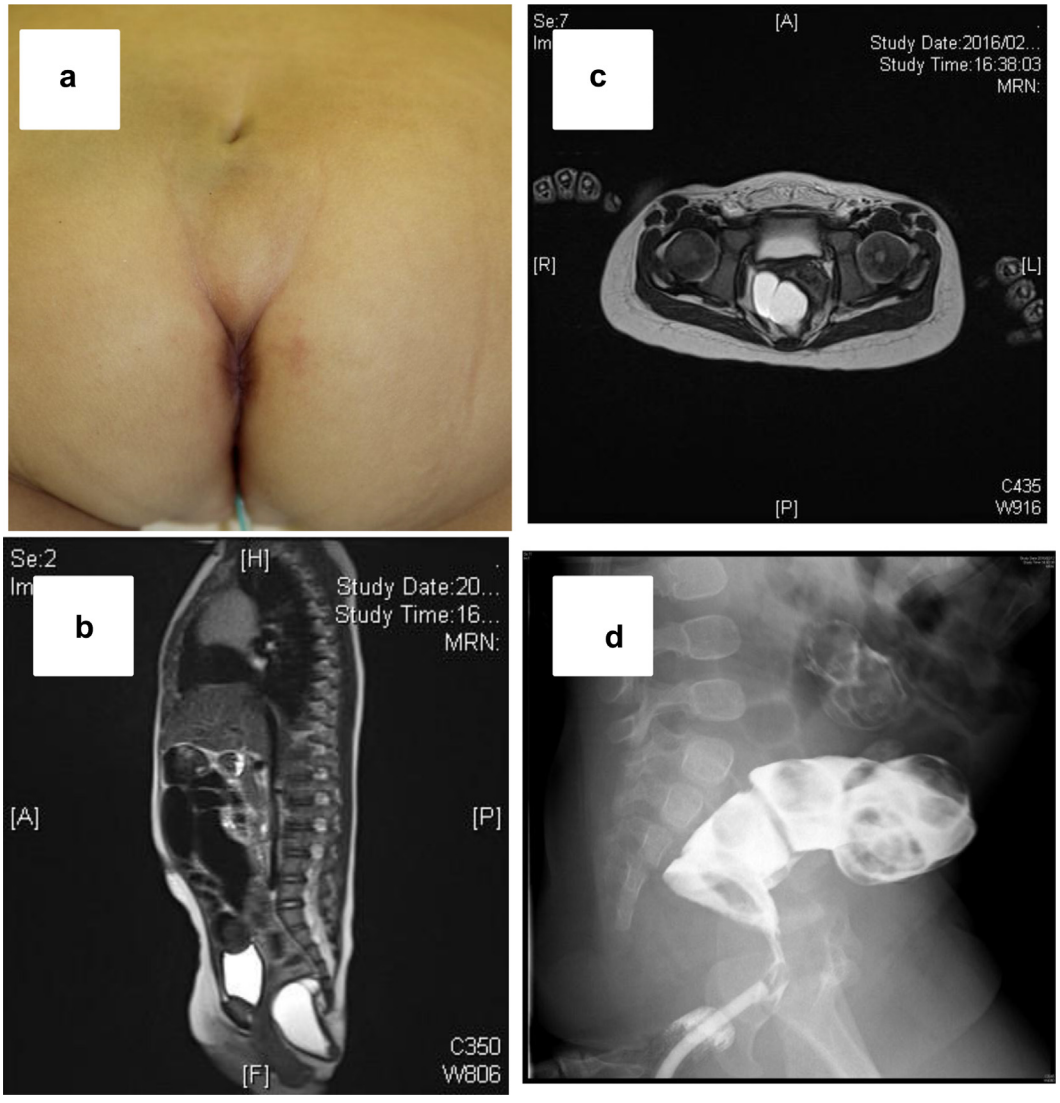

Fig. 1. a) A skin dimple was seen at the coccygeal region and a mild bulge was detected around the cranial area of the anus. b) MRI imaging. A sagittal section showed a hyperintensity mass in T2-weighted imaging. The mass was situated in the presacral and postrectal area. c) MRI imaging. A cross section showed a multilocular cyst at the presacral area which was slightly deviated to the right side. d) Barium enema showed a presacral mass that compressed the rectum.

some glial tissues and bronchial glands were scattered in the connective tissues (Fig. 3c,d). Skin adnexa was not seen in the epithelium. No serosa was seen around the cyst wall. The pathological diagnosis was mature teratoma. The skin dimple was also resected. Examination showed that it was only a skin dimple and did not have the microscopic characteristics of a pilonidal sinus.

We made the final diagnosis of tailgut cyst clinically because (1) the cyst was situated at the presacral and postrectal area; (2) the cystic lesion was complicated with a skin dimple at the coccygeal region; (3) the MRI image was compatible with a tailgut cyst, i.e. there was no fat and calcified tissue and the lesion was a simple multiloculated cyst [15,16]; (4) pathological findings were compatible with a tailgut cyst in that there were various types of epithelia and the presence of epithelia was verified by the presence of unorganized fibrous tissues and there were no nerve plexus, serosa, and skin adnexa. These pathological findings were typical characteristics that strongly suggested tailgut cyst, referring to the past several literatures of tailgut cyst [1,2,5–14]. Also, the tissues of the lesion mainly consisted of mesoderm and endoderm tissues and very few ectoderm tissues were seen; these characteristics might not be compatible with a sacrococcygeal teratoma.

2. Discussion

A tailgut cyst is a rare congenital anomaly that is usually diagnosed in adults incidentally or with some symptoms such as local pain, local mass lesion and difficulty of defecation [1]. Approximately 100 adult cases have been reported [2] and some of them were complicated with malignant transformation of the lesion [3,4].

The tailgut develops early in fetal life beyond the anal point and regresses later in most individuals [1]. However, in a very few cases the tailgut remains at the presacral region for an unknown reason and forms a cystic lesion postnatally. It is quite interesting that cases of tailgut cyst are mainly reported in adults [2] although the lesion is congenital and only ~20 pediatric cases have been reported thus far [5–14]. In some pediatric cases a tailgut cyst was found along with other congenital anomalies such as an imperforated anus [10], anal stenosis [7] and abnormal spinal cord [14]. In our patient, MRI was performed to check for developmental anomalies of the spinal cord and the cystic lesion was incidentally found. Improvements in imaging modalities will increase early detection of tailgut cyst in the future [15,16].

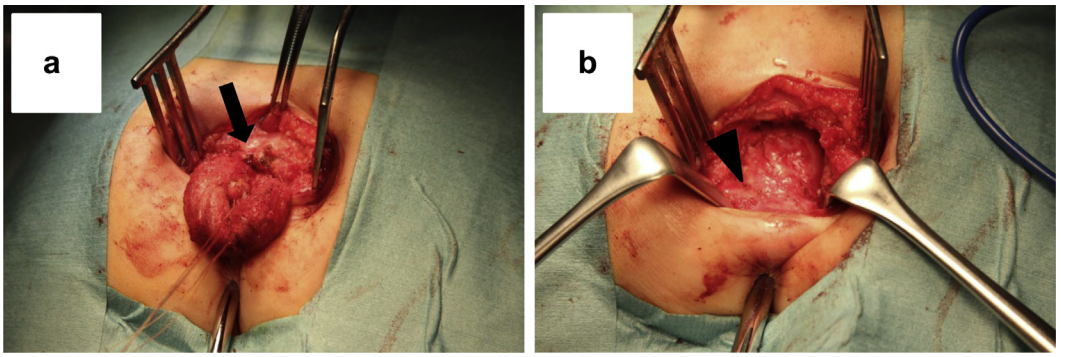

Fig. 2. a) Operative findings. A posterior sagittal incision was made, and the skin dimple and the cystic lesion were resected. The coccygeal bone (arrow) was also resected. b) After resecting the cystic lesion, the posterior wall of the rectum was exposed and the puborectal muscle (arrowhead) was secured.

A tailgut cyst has the following characteristics: 1) the cyst is situated at the presacral and postrectal area; 2) the epithelial layer consists of many kinds of epithelia such as squamous, pseudostratified columnar, goblet columnar, ciliated columnar, transitional, and cuboidal epithelia; 3) the presence of an epithelial layer is usually verified by the presence of a smooth muscle layer and connective tissues but such tissues do not have a well-organized layer and do not contain any nerve plexus and differentiated neuron cells. However, the definitive diagnosis of a tailgut cyst is still controversial even today. Considering the early developmental stages, it is difficult to make a differential diagnosis of cystic lesions around the presacral area, and there are many possible diagnoses such as a dermoid cyst, epidermoid cyst, enterogenic cyst, teratoma, duplication cyst and inflammatory pseudocyst. Sometimes it is very difficult to make a definitive diagnosis. Indeed, Hjermstad et al. [1] commented that the pathological diagnosis of a tailgut cyst was made in only 2 out of the 53 cases that they summarized from past reports. We believe that the diagnosis of a tailgut cyst should be based on the total clinical findings including the localized space, associated anomalies, imaging examination, and pathological findings.

In our case the lesion was located at the presacral and postrectal area and the cyst had compatible findings to a tailgut cyst in imaging examination [15,16] and pathological analysis. Moreover,

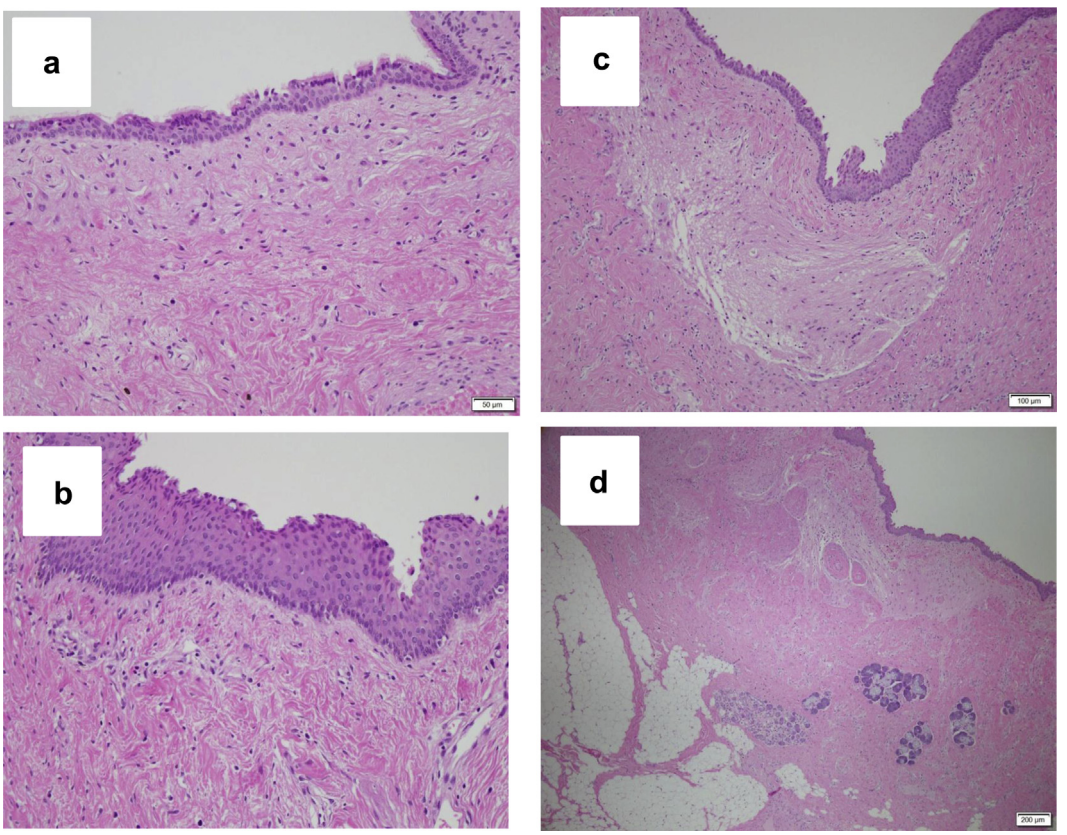

Fig. 3. Pathological findings of the cyst. Several different types of epithelia lined the cyst wall. a) Pseudostratified ciliated columnar epithelium. b) Transitional epithelium. c) The presence of pseudostratified ciliated columnar epithelium and transitional epithelium was verified by the glial tissue and connective tissue. d) The presence of pseudostratified ciliated columnar epithelium was verified by the presence of an unorganized connective tissue layer. Several bronchial glands were seen in the unorganized connective tissue layer.

the patient had a skin dimple at the coccygeal region, which was stressed as an indicator of an underlying tailgut cyst in some reports [1,8]. We finally concluded that in our case the cyst was a tailgut cyst clinically although the pathological diagnosis was mature teratoma.

Conflicts of interest

None.

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