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CASE REPORT

Unusual Intraperitoneal Location of a Tailgut Cyst: a Case Report

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Abstract
Tailgut cyst, known as retrorectal cystic hamartoma, is a rare congenital lesion derived from the remnants of the embryological tailgut. It is usually located in presacral or retrorectal space. We report the first case of a tailgut cyst presented as an intraperitoneal tumor. In this paper, we present one patient who has undergone surgical excision of tailgut cyst. A 33-year-old woman with no symptoms underwent laparoscopic tumor excision for the definite diagnosis and treatment for a left-sided, cystic, and pelvic mass in computerized tomography (CT). The initial diagnosis was left ovarian benign tumor by radiologic study. Histologic findings revealed a tailgut cyst. After surgery, she was discharged after 3 days without any complications. We report an unusual case of tailgut cyst located in intraperitoneal space. Although it is extremely rare for a tailgut cyst to occur in the intraperitoneal area, physicians may consider tailgut cyst in differential diagnosis of an intraperitoneal cystic mass.

Keywords Cyst · Unusual location

Introduction

A tailgut cyst, known as retrorectal cystic hamartoma, is a rare congenital lesion derived from the remnants of the embryological tailgut. It is usually located in the presacral or retrorectal space [1–3]. However, rarely, some unusual locations have been reported, such as the perirenal area [4, 5], the subcutaneous tissue [6], and anterior to the rectum [1, 2, 7]. We report the first case of a tailgut cyst which presented as an intraperitoneal tumor. To the best of our knowledge, this location has not yet been reported in the published literature.

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Case Report

A 33-year-old woman underwent an emergency caesarean section, and a large pelvic mass was coincidentally detected by the surgeon. After 2 months, she was referred to our hospital. She did not have any symptoms. Routine abdominal and pelvic examinations were not remarkable. Routine laboratory tests including those for a complete blood count, electrolytes, cancer antigen (CA) 125, CA 19–9, carcinoembryonic antigen, and α-fetoprotein were within normal limits. Abdominal computerized tomography (CT) showed a left-sided, well-defined, pelvic cystic mass which was located on the left ovary (Fig. 1). The initial diagnosis was left ovarian benign tumor by radiologic study.

She underwent laparoscopic tumor excision for the definitive diagnosis and treatment. During laparoscopic exploration (Fig. 2), a well-defined 6 × 6-cm, soft, cystic, and multilobulated mass was identified. The entire mass was freely placed in the internal digestive and urogenital organs. There was no communication between the tumor and the rectum. The flexible and long stalk, which was approximately 5-cm long, connected the tumor to the retroperitoneal space near the sacral promontory. Blood vessels were observed in this long stalk. The cyst also did not share the blood supply with

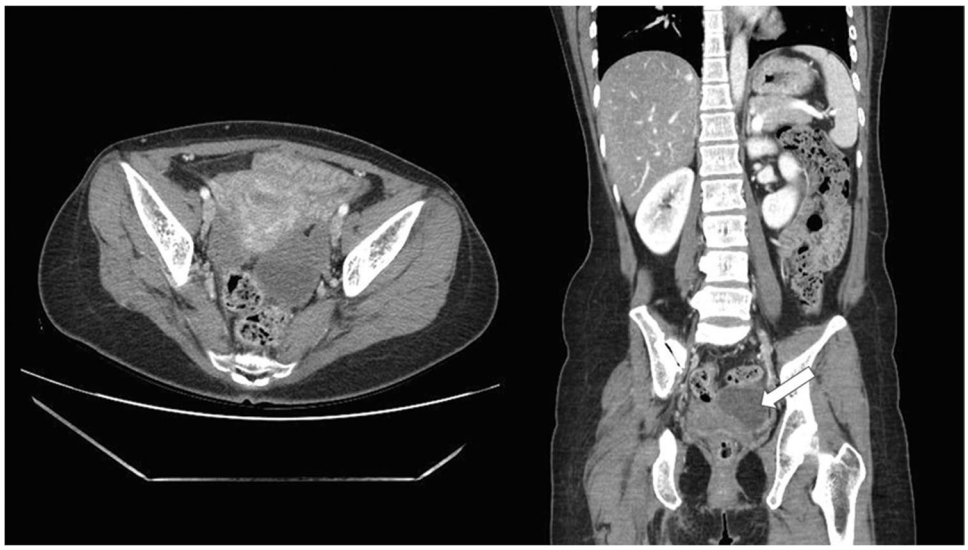

Fig. 1 Computerized tomography shows a left-sided, well-defined, pelvic cystic mass (measuring 6×5 cm), which was located on the left ovary (white arrow)

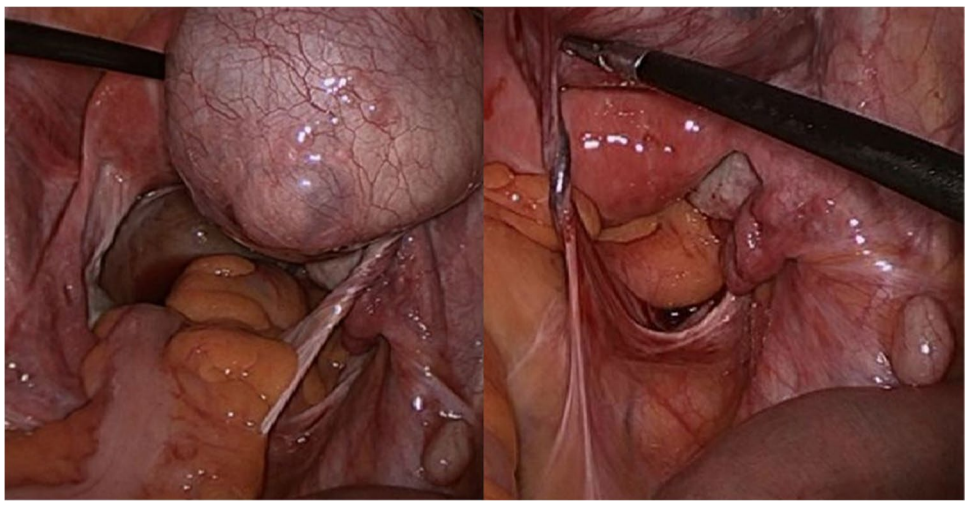

Fig. 2 At laparoscopic exploration, a well-defined 6×6 cm, soft, cystic, and multilobulated mass is seen as freely placed from the internal digestive and urogenital organs. The long stalk connects the tumor to the retroperitoneal space near the sacral promontory

the rectum. The uterus, ovaries, and fallopian tubes were unremarkable.

Macroscopic examination revealed a multilobulated, pinkish, and ovoid cyst measuring 7×6×5.0 cm in cross diameter (Fig. 3). The outer surface of the mass showed a pinkish, smooth, and thickened fibrous wall. The cyst contained turbid, brown, and serous fluid. In microscopic examination, hematoxylin and eosin staining showed that the cyst was lined by glandular epithelium and had fascicles of smooth muscle bundles in the cystic wall (Fig. 4a). Immunohistochemical stain for smooth muscle actin showed a strong positivity in the disorganized fascicles of the smooth muscle bundles (Fig. 4b). Histologic findings revealed a tailgut cyst. Three days post-surgery, she was discharged without any complications.

Discussion

A tailgut cyst is a rare congenital multi-cystic tumor derived from the remnants of the tailgut, a primitive gut temporarily present in the caudal portion of the embryo [1]. Tailgut cysts occur predominantly in middle-aged

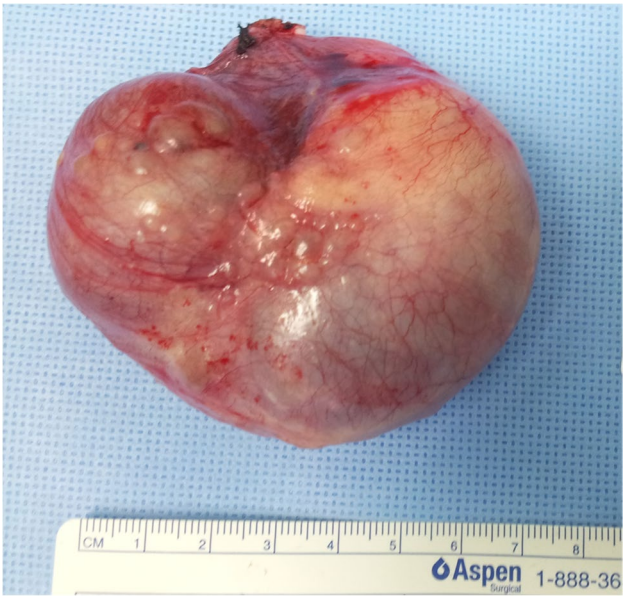

Fig. 3 Macroscopic examination reveals a multi-lobulated, pinkish, and ovoid cyst measuring 7×6×5.0 cm in cross diameter. The outer surface of the mass shows a pinkish, smooth, and thickened fibrous wall. The cyst contains turbid, brown, and serous fluid

women but can occur at any age [2]. Half of these patients are asymptomatic, and these tumors are usually found incidentally during physical examinations or pelvic imaging studies. Most patients with symptoms complain of lower abdominal pain, rectal bleeding, rectal fullness, constipation, and pain on defecation [1, 2]. Tailgut cysts are usually benign lesions, with few cases of malignant transformation reported in the literature. A complete surgical excision is required for accurate diagnosis and treatment.

Tailgut cysts are usually located in the presacral or retrorectal space. In rare cases, tailgut cysts can be located in other sites such as the prerectal [1, 2, 6], perirenal [4, 5], and the perianal skin [6]. Most of the tailgut cysts are located in the extraperitoneal space. The intraperitoneal location of the tailgut cyst was confirmed in our patient. To the best of our knowledge, no cases with intraperitoneal location have been reported in the published literature.

Piura et al. mentioned two histological criteria for the diagnosis of tailgut cysts as follows [8]. (1) The lining epithelium of the lumen surfaces of the cysts must contain transitional and/or glandular-type (columnar) epithelium with or without a stratified squamous component. (2) The underlying stroma must consist of fibrous connective tissue containing scattered discontinuous bundles of smooth muscle fibers, without a well-defined muscular layer and serosa found in the gastrointestinal wall. These criteria differentiate tailgut cysts from epidermoid, dermoid, and rectal duplication cysts [8]. In our case, there are two distinct points that confirmed the diagnosis of a tailgut cyst. First, the luminal surface of the cyst was lined by glandular epithelium. Second, the underlying stroma consists of connective tissue and disorganized fascicles of smooth muscle bundles without myenteric plexus.

Conclusion

We report an unusual case of a tailgut cyst located in the intraperitoneal space. Surgical excision is recommended for the treatment of tailgut cysts to confirm the diagnosis and exclude malignant tumor. Although it is extremely rare for a tailgut cyst to occur in the intraperitoneal area, physicians may consider a tailgut cyst in the differential diagnosis of an intraperitoneal cystic mass.

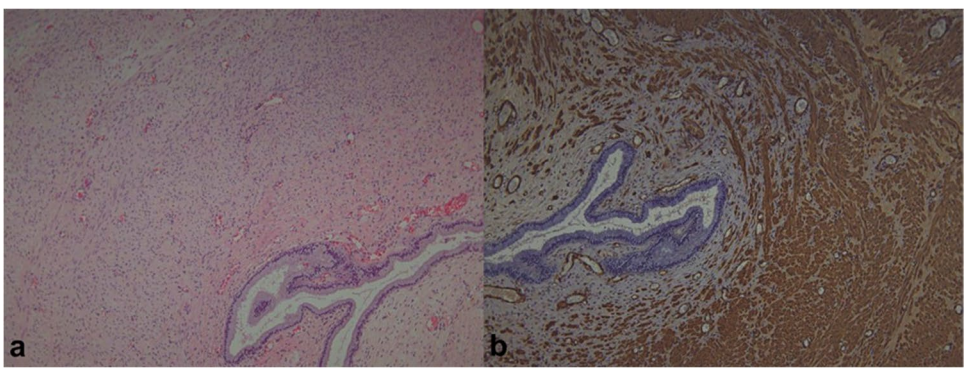

Fig. 4 **a** Pathologic examination demonstrates that one cystic mass is lined by glandular epithelium and has fascicles of smooth muscle bundles in the cystic wall. (H&E stain, × 100). **b** Immunohistochemical staining for SMA shows a strong positivity in the disorganized fascicles of the smooth muscle bundles (SMA stain, × 100)

Declarations

Ethics approval and consent to participate All applicable international, national, and/or institutional guidelines were followed. Before patients were enrolled in this study, informed consent was obtained.

Conflict of interest The authors declare no competing interests.

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