

Case Report

Laparoscopic Resection Combined with a Transsacral Approach for a Recurrent Tailgut Cyst with a Refractory Fistula

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Tailgut cyst is a rare cystic disease of the anterior sacral surface and the remains of an embryonic tail gut. Tailgut cysts have a potential for malignancy, and complete resection with an adequate surgical margin is necessary. Even if incomplete resection does not result in recurrence of malignant disease, there is a risk of local infection leading to refractory fistulas. The optimal treatment for such refractory recurrent lesions has not been reported. We describe a case in which the combination of laparoscopic and transsacral approaches was effective for resecting a recurrent refractory fistula after incomplete resection of a tail gut cyst.

Key words: tailgut cyst, laparoscopic resection, fistula formation

The treatment strategy for tailgut cysts is complete resection of the cyst, and the usefulness of laparoscopic resection has been reported [1, 2]. However, when residual lesions recur, complete resection is difficult due to adhesions or scarring. Herein we describe a case in which laparoscopic resection combined with a transsacral approach was useful for the treatment of a recurrent tailgut cyst that had formed a refractory fistula without the development of cancer.

Case Presentation

The patient was a 48-year-old woman. She had seen a physician 3 years earlier with complaints of anorectal pain and fever. She underwent computed tomography (CT)-guided drainage with a diagnosis of pelvic abscess. One month after drainage, the infection improved. The patient was diagnosed with a multifocal ischiorectal

abscess and underwent radical resection by a transsacral approach and was finally diagnosed with tailgut cyst. Two months after the surgery, abscess formation was observed. The abscess was also resected by a transsacral approach. The rectal wall was sutured because of communication between the rectum and abscess cavity. Seven months after the surgery, discharge from the wound was detected. Her wound was opened again, and the contents of the cavity were curetted. Negative pressure wound therapy was then started, and the wound shrank; one month later, however, the patient again had pain and discharge (Fig. 1). CT showed fluid collection inside the wound. Magnetic resonance imaging (MRI) showed a T2-low-intensity lesion, which included T1-low-intensity and T2-high-intensity images, inside on the ventral side of the sacrum, suggesting abscess formation (Fig. 2). Blood tests showed WBC of 3,440/ μ L and CRP of 0.12 mg/dL, with no evidence of inflammation, and normal tumor marker

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levels (CEA 3.3 ng/mL, CA19-9 28.9 U/mL). The diagnosis was recurrence after tailgut cyst surgery with abscess formation and a refractory fistula. The decision was made to perform laparoscopic resection combined with a transsacral approach to the abscess and fistula.

Operative findings: The patient was placed in the lithotomy position. A 12-mm umbilical trocar and four 5-mm port trocars were placed. The posterior rectal space was dissected to the level of the endopelvic fascia, and the cystic lesion on the left pelvic wall was exposed. The cyst was dissected from the pelvic plexus; then the lesion was approached in the mid-anterior aspect of the sacrum and dissected without injury to the capsule. The cystic lesion was pursued toward the caudal direction. The adhesions around the cyst were mild and had little influence from the previous surgery. A fistula was also

identified, but the adhesion to the surrounding area was strong, and the fistula was resected as far as possible by curettage. At this point, the connection between the lesion and the fistula orifice was confirmed (Fig. 3). In the transsacral approach, the coccyx was preserved and the fistula duct contiguous to the tailgut cyst was excised. The operation time was 188 minutes, with minimal blood loss. The patient had a good postoperative course and was discharged on the 7th postoperative day without any complications.

Pathological examination showed evidence of a remnant tailgut cyst (Fig. 4). There were no findings of malignancy, and the resection margin was negative. No recurrence of infection or mass formation was observed 12 months after surgery.

Discussion

In this case, a residual lesion of a tailgut cyst was repeatedly infected and formed a fistula. The combination of a laparoscopic and transsacral approach was effective at achieving complete resection of this intractable fistula.

Tailgut cyst is a relatively rare cystic disease that occurs on the anterior surface of the sacrum as a cystic remnant of the embryonic tail gut [3]. The risk of malignant transformation is about 10.2% [4], and patients diagnosed with a tailgut cyst are generally treated with complete resection. However, there have been reports of cases with incomplete resection and repeated infection [5]. In such cases, even if the infec-

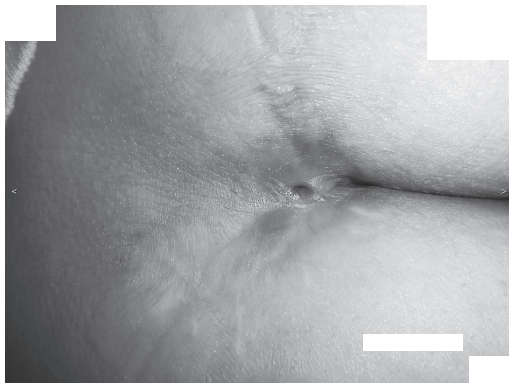


Fig. 1 The sacral skin wound is not healed and continues to exude discharge due to the refractory fistula.

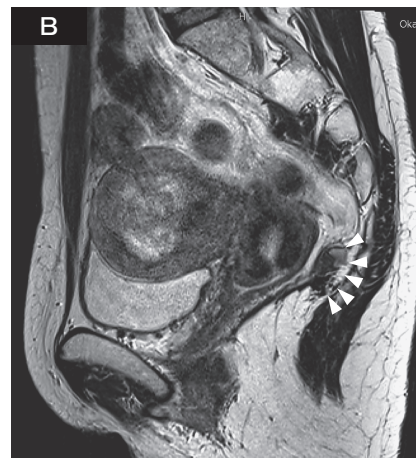
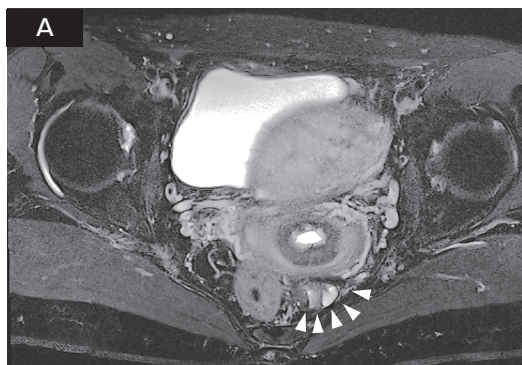


Fig. 2 MRI T2-weighted images show a low-signal intensity lesion with an internal high-signal intensity area on the anterior sacral surface, but communication with the rectum is not evident. **A**, horizontal section; **B**, sagittal section. Red triangles indicate the cystic lesion.

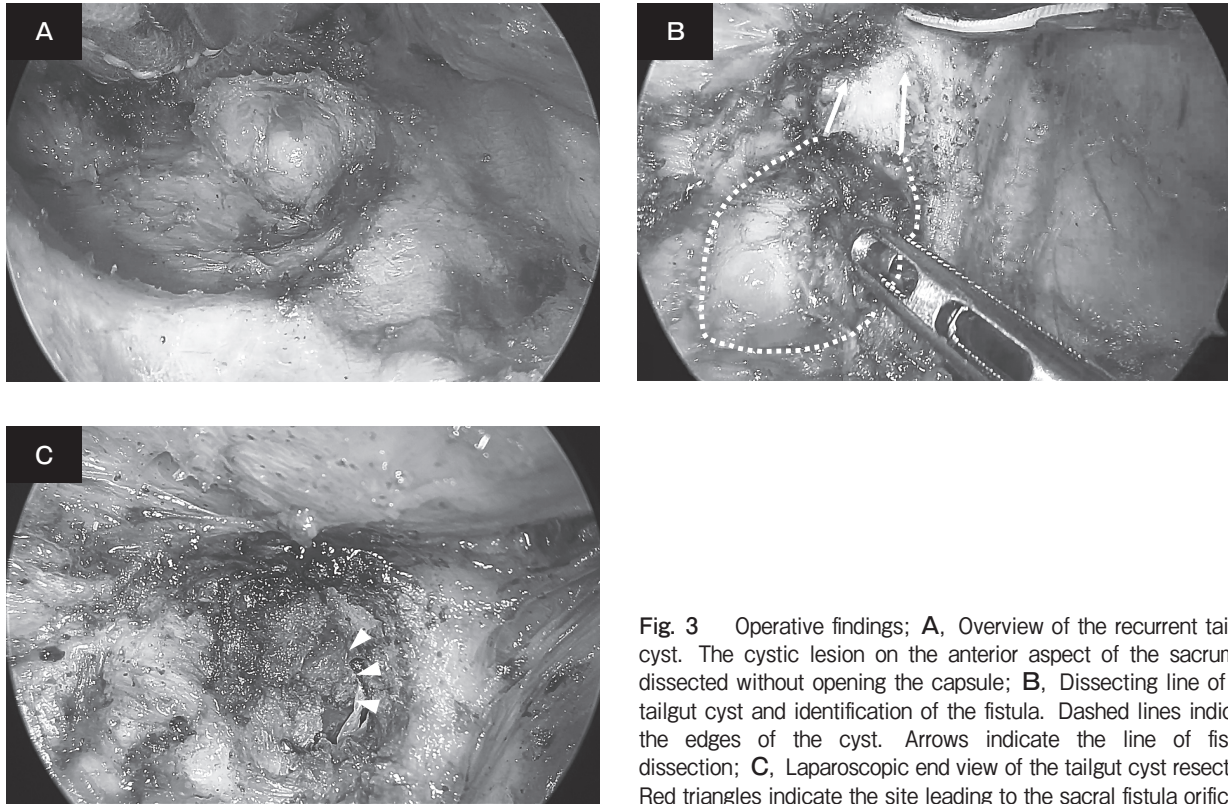


Fig. 3 Operative findings; **A**, Overview of the recurrent tailgut cyst. The cystic lesion on the anterior aspect of the sacrum is dissected without opening the capsule; **B**, Dissecting line of the tailgut cyst and identification of the fistula. Dashed lines indicate the edges of the cyst. Arrows indicate the line of fistula dissection; **C**, Laparoscopic end view of the tailgut cyst resection. Red triangles indicate the site leading to the sacral fistula orifice.

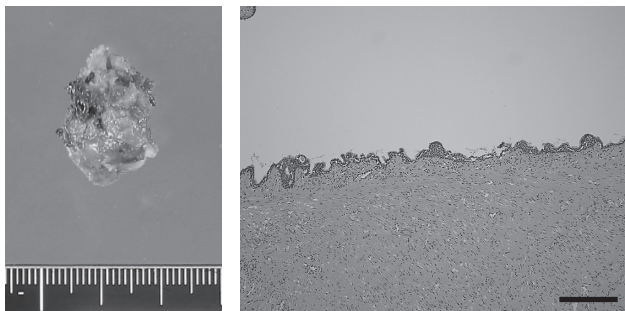


Fig. 4 Hematoxylin-eosin staining of the cyst wall tissue shows a multilayered squamous epithelium, consistent with a tailgut cyst. There is no evidence of malignancy in the resected specimen. Scale bar: 200 μ m.

tion is temporarily controlled by drainage, it will recur after a short period of time. In addition, malignant lesions frequently develop [1, 4, 6], such that complete excision of the abscess cavity including the fistula tract should be considered for recurrent lesions. In this case, because the cranial edge of the cyst was located at a high level of the sacrum, it was difficult to visualize by a transsacral approach; as a result, resection of the cyst had been incomplete.

Surgical approaches include transabdominal, trans-sacral, and combined approaches. One should consider the approach based on the location and size of the lesion. In one study of 76 cases in the U.S., the trans-sacral approach was chosen in 76.7% of cases [4]. In a retrospective study of 24 cases from Asia, the transabdominal approach was selected in 41% of cases and had fewer complications than the trans-sacral approach [7]. The transabdominal approach allows the tumor to be exposed directly behind the rectum, and the positioning of the surrounding vessels and rectum can be confirmed. However, if the tumor is lower than S3, its exposure using this method is difficult; in short, the transabdominal approach is useful for tumors higher than the third sacral level (S3) while the transsacral approach is considered useful for tumors lower than S3 [8].

Recently, the usefulness of laparoscopic surgery as the initial treatment for tailgut cysts has also been reported [1, 2]. However, recurrent lesions such as the present case are rarely reported, so the surgical approach should be considered carefully. In our case the fistula was open in the buttock, so a trans-sacral approach was necessary. The cavity itself was located close to the rec-

tum and had previously communicated with the intestinal tract, so the transabdominal approach was preferable to achieve complete resection. In addition, the laparoscopic approach with high magnification is useful for resecting cysts and fistulas without damaging surrounding organs deep within the body. The reason for using a laparoscopy in this case was that the previous surgical manipulation had not affected the patient and it was easier to identify the recurrent tailgut cyst from this vantage.

Although this patient had a recurrent lesion, malignancy was not suspected based on the preoperative examination findings. There have been several cases of recurrence after initial surgery in Japan, all of them associated with malignant findings. Two patients underwent transsacral partial resection first, and abdominoperineal resection was performed when the malignant tailgut cyst recurred 3 years later [9, 10]. One patient underwent initial surgery for a hemorrhoidal fistula, and transabdominal transsacral tumor resection and sigmoid colostomy were performed when the malignant tailgut cyst recurred 5 years later [11]. These patients had malignant findings on preoperative examinations that suggested invasion into the surrounding organs; therefore, combined resection of the intestinal tract was inevitable. Since there have been few reports of patients with recurrent lesions and no malignant findings, as in the present case, it was difficult to determine the surgical strategy. The treatment plan was based on three points in the following order of priority. First, the cystic lesion needed to be completely resected. The second priority was to prevent damage to the surrounding organs. Finally, the surgical invasiveness to the patient should be minimized. Laparoscopic magnification was very useful for dissecting the cystic lesion on the posterior sacral surface of the rectum without breaching the septal wall and without damaging the surrounding organs. The laparoscopic view facilitated complete resection of the cyst and fistula ducts. In addition, the laparoscopic approach was minimally invasive and reli-

able, with good postoperative pain control.

A laparoscopic approach and an adjunctive transsacral approach were useful for a patient with a tailgut cyst that formed a refractory fistula with repeated recurrences. Laparoscopic resection may be a reasonable treatment modality for deep pelvic cystic lesions and may reduce the patient's burden of surgery.

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