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Case Report

Surgical Management of Tailgut Cysts: A Report of Two Cases

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A tailgut cyst is an uncommon congenital developmental cyst that arises from the postnatal remnants of the primitive gut. These cysts are generally located in the retrorectal space. The rarity of this lesion and its anatomical position frequently lead to difficulties in its diagnosis and surgical management. Complete surgical resection remains the cornerstone of treatment, and numerous surgical approaches to achieve this objective have been described in the literature. We present 2 cases of tailgut cysts that were resected using a trans-sacral approach. We discuss the surgical management of these cases, including the operative approach that was adopted. Two cases of tailgut cysts that presented as incidentally detected retrorectal tumors are described here. A pelvic magnetic resonance imaging scan revealed a retrorectal multicystic tumor consistent with a tailgut cyst. The patients underwent surgery via a trans-sacral approach with coccygectomy. The histopathologic diagnosis for each patient was a tailgut cyst with no evidence of malignancy. Surgical approaches for tailgut cysts depend on the exact locations of the cysts and suspicions regarding the malignancies of the cysts. En bloc organ resection, including coccygectomy, is required if malignancy is suspected. We endorse the trans-sacral approach as a feasible, easy to perform, minimally invasive, and safe option for treating relatively low-lying tailgut cysts.

Key words: Tailgut cyst – Retrorectal hamartoma – Coccygectomy

A retrorectal cystic hamartoma, which is also known as a tailgut cyst, is an uncommon congenital developmental cyst that arises from the postnatal remnants of the primitive gut and is generally located in the retrorectal space.[1,2] In 1953, Hawkins *et al*[3] histologically classified developmental cysts into three categories: dermoid cysts, epidermoid cysts, and mucus-secreting cysts.

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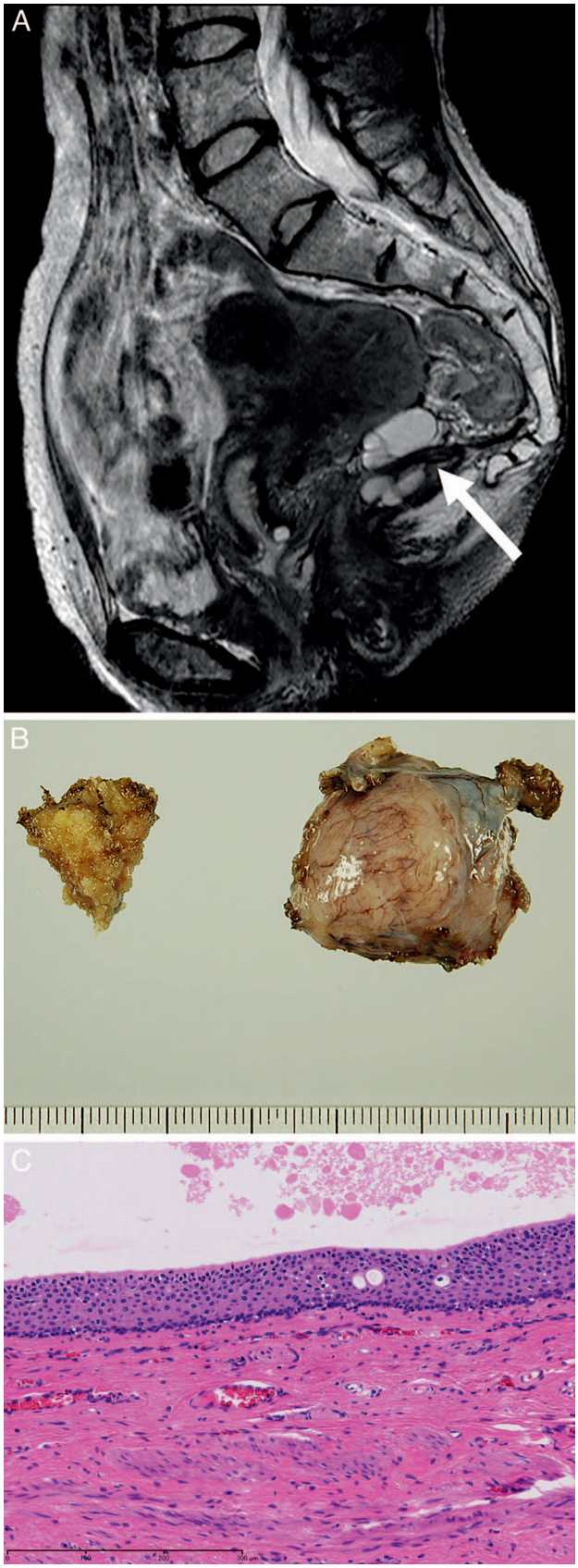

Subsequently, Hjermstad and Helwig[4] referred to mucus-secreting cysts as tailgut cysts, and the latter term has become widely used.

Tailgut cysts are usually reported in middle-aged women. These cysts, which are generally asymptomatic, are typically found during routine physical examination or at childbirth; therefore, diagnoses of these cysts are often delayed. Although developmental cysts, including tailgut cysts, are often asymptomatic, patients may present with symptoms resulting from local effects of the masses. These symptoms can include constipation, rectal fullness, lower abdominal pain, and dysuria. The initial presentation of tailgut cysts may also be delayed until the onset of complications such as infections with fistulization, bleeding, or malignant degeneration.[4] Complete surgical excision is recommended not only to establish diagnoses, relieve symptoms, and avoid complications, but also to exclude the possibility of an underlying malignancy.[5]

A large variety of surgical approaches for facilitating tailgut cyst removal have been reported in the literature; in particular, abdominal (anterior), trans-sacral (posterior), and combined abdomino-sacral approaches have been well described.[5] The investigation modalities, treatment strategies, and surgical approaches for tailgut cysts continue to evolve.[6,7]

In this report, we present 2 cases of tailgut cysts resected using a trans-sacral approach. In addition, we review the relevant literature on the surgical resection of tailgut cysts and discuss the surgical management of these cysts, including the surgical approach used.

Case 1

A 45-year-old woman presented with an incidentally detected retrorectal tumor during an evaluation for chronic constipation. A multidetector row computed tomography (MDCT) scan of the pelvis with contrast revealed a well-defined 39×33 mm^2

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Fig. 1 (A) A pelvic MRI revealed a $40 \times 38 \times 30$ mm^3 retrorectal multicystic tumor of high intensity on a T2-weighted image (arrow). (B) A macroscopic view of the resected coccyx (left) and the cystic tumor (right). The tumor wall was smooth, and the tumor contained a collection of mucinous fluid. (C) Histologically, the $27 \times 23 \times 18$ mm^3 unilocular cyst contained stratified columnar epithelium. The cyst wall contained fibrous tissue and a small number of scattered smooth muscle fibers.

homogeneous cystic mass in the left presacral area. A pelvic magnetic resonance imaging (MRI) scan with contrast revealed a $40 \times 38 \times 30$ mm^3 retrorectal cystic tumor in the left presacral area with intermediate to high signal intensity on T1- and T2-weighted images and a 25×15 mm^2 cystic mass in the precoccygeal space with high signal intensity on T1-weighted images (Fig. 1A).

The patient underwent surgery via a trans-sacral approach with excision of the coccyx. This procedure was performed with the patient in the jack-knife position. Intraoperative findings indicated that the tumor was adherent to the rectum without invasion of the rectal wall. As appropriate, a transanal finger was used to identify the posterior rectal wall to facilitate the dissection and prevent rectal injury. Primary closure with simple sutures was used to close the surgical wound. A drain was placed in the retrorectal space. The patient's postoperative recovery was uneventful, and she was discharged from the hospital after 4 days.

Histologically, the $27 \times 23 \times 18$ mm^3 unilocular cyst contained stratified columnar epithelium. The cyst wall contained fibrous tissue and a small number of scattered smooth muscle fibers. The histological findings were consistent with the diagnosis of a tailgut cyst. No heterologous elements suggestive of a teratoma were observed. There was no evidence of malignant change or infiltration (Figs. 1B and 1C).

Case 2

A 41-year-old man with a left ureteral stone presented with an incidentally detected retrorectal tumor. A physical examination indicated the presence of left lower abdominal pain. A pelvic MRI with contrast revealed a $48 \times 25 \times 15$ mm^3 retrorectal multicystic tumor in the presacral area with intermediate to high signal intensity on T1- and T2-weighted images.

The patient underwent surgery via a trans-sacral approach with excision of the coccyx. The procedure was performed with the patient in the jack-knife position. Intraoperative findings indicated that the tumor was adherent to the rectal wall and the levator ani without invasion. As appropriate, a transanal finger was used to identify the posterior rectal wall to facilitate the dissection and prevent rectal injury. Primary closure with simple sutures was used to close the surgical wound. A drain was placed in the retrorectal space. The patient's postoperative recovery was uneventful, and he was discharged from the hospital after 7 days.

Histologically, the resected mass was a $48 \times 25 \times 15$ mm^3 multilocular cyst containing stratified columnar epithelium and multilayer squamous epithelium. The cyst wall contained fibrous tissue and scattered smooth muscle fibers. The findings were consistent with the diagnosis of a tailgut cyst. There was no evidence of malignant change or infiltration.

Discussion

The 2 described cases of tailgut cysts were surgically resected using a trans-sacral approach with coccygectomy. The histologic findings confirmed that both of the extracted masses were tailgut cysts without malignant transformation. The patients had uneventful postoperative courses and were discharged on postoperative days 4 and 7. They remained healthy after being discharged and were symptom-free during the postoperative follow-up period.

Complete surgical resection is established as the traditional treatment of choice for tailgut cysts because complete resection eliminates the potential for recurrence, hemorrhage, infection, compression, and malignant transformation. Historically, three different types of surgical approaches have typically been used in the tailgut region: an anterior (transabdominal) approach, a posterior (intersphincteric, trans-sphincteric parasacrococcygeal, trans-sacral, trans-sacrococcygeal, trans-anorectal and trans-vaginal) approach, or a combination of the two.[8]

We have listed the advantages and disadvantages of the different surgical approaches for tailgut cysts and indicated the ideal cases for each approach (Table 1). The selection of an appropriate approach, which is key to successful treatment, is determined by the morphology, location, and size of the retrorectal lesion and the lesion's relationship with adjacent structures. It has been argued that an anterior (abdominal) approach should be reserved for relatively high lesions (above S3 or the sacral promontory) and that a posterior approach is required if the retrorectal mass is below S3 or the sacral promontory. When a tumor extends both above and below S3, a combined abdominosacral approach may be required.[9]

Therefore, the surgical approach used for a tailgut cyst depends on the exact location of the cyst, the infection status of the cyst because infections can cause adherence of the cyst to surrounding organs

Table 1 Various approaches for tailgut cysts

Approaches	Advantage	Disadvantage	Ideal case of each approach
Anterior approach	Good direct visualization of important structures, such as iliac vessels and the ureters	Poor access to the caudal component especially in narrow pelvis	Above S3 or the sacral promontory Oncological resection in cases involving suspected malignancy
Posterior approach	Good access to the caudal component	Absence of control over pelvic vessels and the potential injury to the lateral pelvic nerves	Below S3 or the sacral promontory In case of relatively small, noninfected and low-lying lesions
Combined approach	Better surgical exposure compared with an anterior or posterior approach alone	Potential drawback is the greater morbidity that can occur in case of iatrogenic injury to the lateral pelvic nerves	A tumor extends both above and below S3 Cysts with diameters > 5 cm Inflammation persist or adjacent structures and organs are involved Suspect of malignant transformation

(the bladder, ureters, and/or rectum), and whether or not the cyst is expected to be malignant. En bloc organ resection, including coccygectomy, is required if malignancy is suspected. An examination of specific preoperative pelvic MDCT or MRI findings relevant to the aforementioned parameters is important to the selection of an appropriate surgical approach.

An anterior approach ensures the visualization of important structures, such as the iliac vessels and the ureters, and therefore allows for better oncologic resection in cases involving suspected malignancies.[10] A posterior approach provides ready access to the caudal component. However, the major disadvantages of a posterior approach are an absence of control over pelvic vessels and the potential for injury to the lateral pelvic nerves.[11] A posterior approach should be avoided for higher tumors (above S3), due to the resulting lack of access to pelvic vessels in the event of intraoperative bleeding. A combined approach may be selected based on the location or size of a cyst.[12] In particular, Hjermstad and Helwig[4] proposed that a surgical procedure that combines anterior and posterior approaches should be performed to remove cysts with diameters > 5 cm because the use of a combined approach instead of an anterior or posterior approach alone provides better surgical exposure and helps to prevent intraoperative injuries to organs or vessels near the cyst.

Historically, coccygectomy has been recommended as a routine treatment for tailgut cysts because the coccyx was thought to harbor cellular remnants that could evolve to form a recurrent cyst.[13] However, current data support the theory that the remnants of the postanal gut are contained within the cyst itself rather than the coccyx.[14] Therefore, coccygectomy provides no additional advantage in cases of tailgut cysts unless en bloc resection is mandated for malignancy, the tailgut cyst is severely adhered to surrounding structures, or better surgical exposure is required.[15] In the 2 cases described here, a retrosacral approach with coccygectomy was selected because the coccygectomy improved the intraoperative field of view and thereby enhanced the surgical procedure.

Recently, a laparoscopic approach for tailgut cyst removal has been introduced; this approach is particularly useful for low-lying cysts.[16] The main advantage of this approach is the magnifying effect it provides in a narrow pelvis. In selected cases involving small and radiologically uncomplicated lesions, the laparoscopic approach is a feasible option for experienced surgeons.

Patients were examined 1 month after surgery and annually during the first 5 years after surgery. MRI was performed if any sign of potential recurrence was detected by clinical examination. An annual digital examination to assess for recurrence has been recommended for cases involving tailgut cysts.[13]

We reviewed 10 previous reports with >10 cases of tailgut cysts (Table 2).[4,5,13,18–24] Most of the patients presented with symptoms resulting from local effects of the masses, such as pain and constipation. Almost all of the reports noted that

Table 2 Literature review of previous reports with >10 cases of tailgut cysts

Author	Year of publication	Adult patients, n	Presenting complain, n	Mode of diagnosis	Preoperative histology	Type of surgery	Complications, n	Malignancy, n
Uhlig *et al*[5]	1975	16	N/A	N/A	N/A	N/A	N/A	N/A
Jao *et al*[18]	1985	16	N/A	CT	None	N/A	N/A	N/A
Hjermstad and Helwig[4]	1988	53	26	N/A	N/A	N/A	N/A	4
Lev-Chelouche *et al*[19]	2003	12	9	CT	None	Ant: 9, post: 3	3 (N/A)	0
Grandjean *et al*[20]	2008	30	N/A	MRI	None	Ant, 2; post: 26; com: 2	2 (local recurrence)	3
Aflalo-Hazan *et al*[21]	2008	11	9	MRI	None	Ant, 1; post, 8; com, 2	N/A	1
Mathis *et al*[13]	2010	31	17	MRI	None	Post, 8; others, 23	8 (N/A)	4
Chéreau *et al*[22]	2013	47	37	CT, MRI, EUS	None	Post, 42; ante or com, 5	4 (local infection, hematoma)	9
Macafee *et al*[23]	2012	13	N/A	CT, MRI	None	N/A	N/A	N/A
Patsouras *et al*[24]	2015	17	16	MRI	None	Post, 16; com, 1	5 (local infection, rectum injury, severe anal pain)	1

Ant, Anterior approach; com, combined approach; EUS, endoscopic ultrasound; N/A, not applicable; post, posterior approach.

preoperative MRI or CT is useful for making a diagnosis of a tailgut cyst. The literature has not previously recommended preoperative biopsies in order to avoid the risk of tumor seeding and infection. Each surgical approach was determined by consideration of the location of the cyst, potential infection of the cyst, potential adherence to surrounding organs, and suspicions of malignancy. Some studies reported surgical complications, such as local recurrences and hematomas. Malignant transformation was observed in 5.8% to 19% of cases.

In conclusion, we described 2 cases of tailgut cysts that were surgically resected using a retrosacral approach with coccygectomy. We recommend the trans-sacral approach as a feasible, easy to perform, minimally invasive, and safe option for treating relatively low-lying (below S3) tailgut cysts. Relative to other approaches, this approach incurs no additional costs and is associated with extremely low predicted morbidity.

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