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CASE REPORT

Robotic approach to remove four tailgut cyst cases in Brazil: a case series

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ABSTRACT

Tailgut cysts are rare congenital lesions that are remnants of the embryonic hindgut. This abnormality presents with non-specific symptoms or no symptoms; therefore, misdiagnosis is common. Here, we present four cases of tailgut cysts that were successfully removed using a robotic surgical approach. A 42-year-old woman with tenesmus, pain in the right gluteal region, and discomfort in the rectal region during evacuation was referred to our medical center. Another patient was a 28-year-old woman who presented with the same symptoms to our general practitioner. Both patients underwent upper abdominal and pelvic magnetic resonance imaging that revealed a tailgut cyst. Further, a 36-year-old woman was referred with coccyx and hypogastric pain. Magnetic resonance imaging revealed two pararectal cystic formations. She underwent robot-assisted surgery, and after analysis by a pathologist, the conclusion was that the tailgut cyst was associated with scarring fibrosis. A 55-year-old woman with posterior epigastric pelvic pain associated with heartburn underwent robot-assisted surgery to resect a retroperitoneal tumor. These cases highlighted the importance of tailgut cysts in the differential diagnosis of rectal lesions. Surgical treatment is preferred because malignant transformations can occur. The difference between laparoscopic and robotic approaches is the better visualization and stability of the latter, inducing less tissue damage. Robotic resection is a safe procedure, especially in patients with a narrow pelvis, because it reduces tissue damage.

Keywords: Cysts; Rectal neoplasms; Congenital abnormalities; Robotic surgical procedures; Margins of excision

INTRODUCTION

Tailgut cysts are rare congenital lesions that arise from the remnants of the post-anal primitive gut and develop in the retrorectal space. The caudal extension of the hindgut typically regresses during the sixth week of gestation during the embryonic period. Therefore, if regression fails, a tailgut cyst is formed.[1] The retrorectal space is located in front of the sacrum and behind the rectum, and the lateral walls are surrounded by the ureters, iliac blood vessels, sacral nerves, and lateral stalks of the rectum, making the surgical approach difficult.[2]

Most patients are asymptomatic; however, when symptoms appear, they are caused by the local pressure of the cyst on the surrounding structures, such as lower back pain, constipation, and urinary symptoms. The main complications are infection, bleeding, and rare malignant degeneration, such as adenocarcinoma and carcinoid tumors.[3]

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Surgery is the preferred treatment, and the minimally invasive approach can minimize mortality and improve recovery, as it reduces the handling of structures around the lesion. Moreover, robotic surgery is a better option than laparoscopy because it allows better manipulation and visualization of the tailgut cyst, and the risk of nerve injuries may be lower compared to the laparoscopic technique.[4]

As a tailgut cyst is a rare pathology and has a limited number of reports and reviews in the literature, we conclude that this series of cases will contribute to the readers, especially because of the robotic surgical approach.

CASE REPORT

Case 1: A 42-year-old woman presented with tenesmus, right gluteal region pain, and rectal discomfort during evacuation. The patient had undergone liposuction, reduction mammoplasty, subtotal hysterectomy, and laparoscopic treatment for pelvic endometriosis six years previously. An upper abdomen and pelvic magnetic resonance imaging (MRI) were performed, and the main finding was the presence of a presacral retrorectal cystic formation, measuring approximately 7.7 × 6.2cm, composed of cysts of varied sizes and different signal intensities associated with lesions suggesting deep endometriosis. The patient was diagnosed with deep pelvic endometriosis and tailgut cysts. Multidisciplinary treatment involving gynecologists and general surgeons was proposed, and a robotic approach was chosen to treat both diseases better. The patient's condition improved postoperatively.

Case 2: A 36-year-old woman presented with coccyx and hypogastric pain. Pelvic MRI showed two pararectal cystic formations with slightly thicker contents. One of the cysts measured 1.5cm, and the other, which had the most lobulated and septate shape, measured 5.6 × 3.6cm. Pelvic MRI results are shown in figure 1. Both the cysts were in extensive anterior contact with the elevator muscle and posterior contact with the ischiococcygeal muscle. Computed tomography (CT) was also performed, and the major finding was a cystic formation measuring 5.4 × 3.8 × 3.6cm and an estimated volume of 38.4cm^3. Based on these findings, the possible diagnosis was a tailgut cyst. The patient underwent robot-assisted surgery to remove the retroperitoneal tumor; the surgery was successful. The pathologist received a cystic structure for analysis, measuring 4.1 × 2.8 × 1.6cm, and the external surface of the cyst was chestnut and granular. After cutting, the cavity was filled with brownish and pasty material. Tailgut cysts are associated with scar fibrosis and acute, chronic inflammation caused by multinucleated foreign cells. The samples did not meet the histological criteria for malignancy.

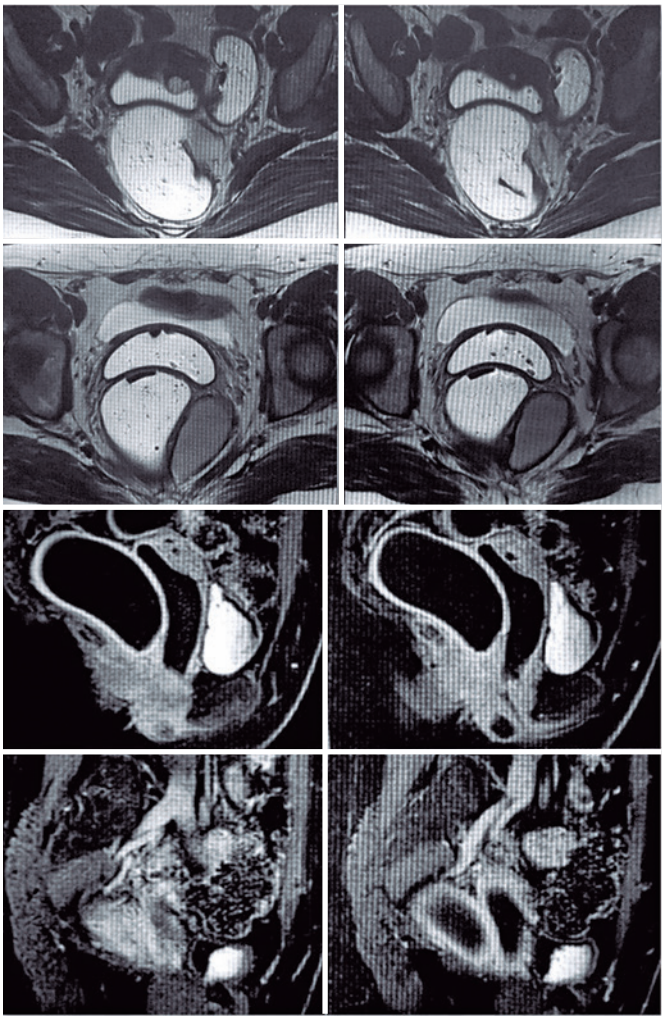

Figure 1. Pelvic magnetic resonance imaging of a woman with tailgut cyst

Case 3: A 28-year-old woman with no associated diseases reported tenesmus, right gluteal pain, and rectal discomfort during evacuation. The patient's previous surgeries included four punctures of the pelvic cyst in early 2013, video laparoscopy for treating gastrointestinal perforation by a foreign body with partial colectomy and protective colostomy in late 2013, and closure of the colostomy in 2014. Magnetic resonance imaging of the upper abdomen and pelvis revealed deep pelvic endometriosis and tailgut cysts. Both diseases were treated simultaneously using a robotic surgical approach. Figure 2 shows intraoperative images of a tailgut cyst before and after robotic surgery.

Case 4: A 55-year-old woman presented at the emergency department with epigastric pain associated with heartburn and posterior pelvic pain. Pelvic MRI

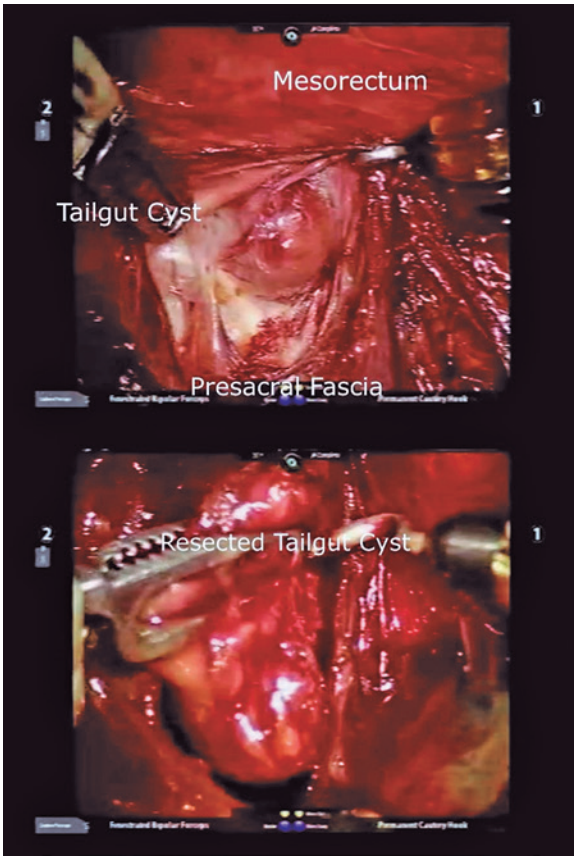

Figure 2. An intraoperative image from before and after the resection of the tailgut cyst by robotic surgery

Table 1. Clinical cases of patients

	Age	Gender	Previous abdominal surgery	Main symptoms	Intra and postoperative complications
Case 1	42	Female	Subtotal hysterectomy Laparoscopy for endometriosis	Tenesmus, pain in the right gluteal region, and rectal discomfort during evacuation	None
Case 2	36	Female	No	Coccyx and hypogastric pain	None
Case 3	28	Female	Partial colectomy + colostomy Colostomy closure	Tenesmus, pain in the right gluteal region, and rectal discomfort during evacuation	None
Case 4	55	Female	No	Epigastric pain associated with heartburn and pelvic posterior pain	None

showed a nonspecific cyst without any solid component measuring approximately 2.0 × 2.1 × 1.6cm below the elevator of any muscle and another multiloculated hematic cyst in the retrorectal region without any solid component measuring approximately 2.4 × 1.8 × 1.2cm. Based on these findings, the possible diagnosis was a tailgut cyst, and the patient underwent robot-assisted surgery to treat the retroperitoneal tumor. The anatomopathological report describes the lesion as a retrorectal tumor consisting of an irregular tissue structure measuring 1.5 × 1.0 × 0.9cm. The cystic cavity was filled with yellowish pasty content, and all materials were subjected to histological analysis to confirm a tailgut cyst without atypical signs of malignancy. The patient progressed well postoperatively. These cases are summarized in table 1.

The study was approved by the Research Ethics Committee of *Centro Universitário FMABC* (CAAE: 67244623.3.0000.0082; # 5.983.185).

DISCUSSION

Epidemiology

Lesions in the retrorectal space are rare and affect a heterogeneous group. The literature has focused predominantly on the surgical approach and histological classification of these pathologies. These reports estimated the incidence of those lesions in 1 from 40.000 to 63.000 during hospitalization.[5] Despite these numbers, the true prevalence of rectal lesions remains unknown because they are often misdiagnosed and asymptomatic. Tailgut cysts are classified as congenital; they arise from remnants of the embryonic hindgut and are mainly located in the presacral space among retrorectal lesions. This space is defined posteriorly by Waldeyer's fascia, anteriorly by the fascia propria of the rectum, inferiorly by the coccygeus and elevator ani muscles, superiorly by the peritoneal reflection between the second and third sacral segments, and laterally by the ureters and iliac vessels.[6] Some cases have described locations other than the presacral region, such as a tailgut cyst located anterior to the rectum, perianally, or in the perirenal region. An example of a presacral tailgut cyst is shown in figure 3.

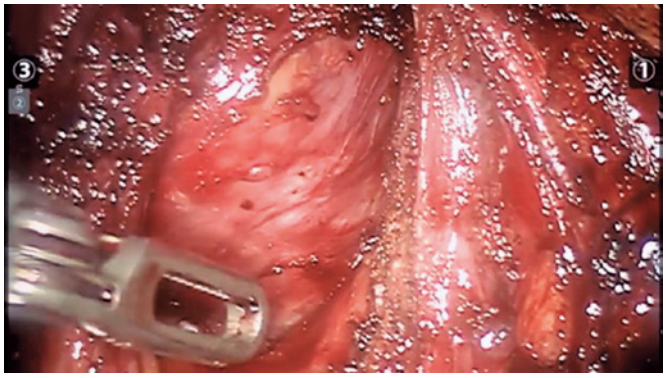

Figure 3. Presacral tailgut cyst

Tailgut cysts can present at any age but are more prevalent in middle-aged women. In a single-center experience (South Korea) of 24 cases of tailgut cysts, a retrospective analysis was made between 2007-2018, and the median age of the patients was 51.5 years (range, 21–68 years) and 18 patients were females (75%); thus, the female-to-male ratio was 3:1.[7]

Malignant transformation

In 1932, Ballantine reported the first case of a tailgut cyst with malignant transformation.[8] Malignant transformation of tailgut cysts includes neuroendocrine carcinoma, squamous carcinoma, adenocarcinoma, endometrioid carcinoma, and sarcoma. The first three are the most common.[9]

The incidence of malignant transformation of tailgut cysts differs among studies. In 1988, a large body of literature reported a malignant transformation rate of only 2% in 53 cases. However, more recent studies have reported rates between 13% and 40%.[10-12] A systematic review of 144 articles, including case reports and case series, reported that 32.1% of cases were malignant degeneration of tailgut cysts, and symptomatic cases were associated with this malignant transformation.[13]

Histological examination after resection is essential to diagnose the malignant transformation of a tailgut cyst.[14] The biopsy alone is not the best choice because the specimens usually obtained do not have epithelial tissues or malignant foci, and malignant cells may leak into the peritoneal cavity during biopsy. On MRI, malignant cysts tend to appear heterogeneous, with solid and cystic contents, irregular borders, or cyst calcification.[15,16] Most presacral carcinoids are immunohistochemically investigated using antibodies against general neuroendocrine markers such as chromogranin A, synaptophysin, and neuron-specific enolase.

The pathogenesis of malignant transformation in tailgut cysts remains unknown. However, a study published in 2010 showed that ghrelin and estrogen are important for the origin and development of the malignant transition of tailgut cysts.[14] Before this study, neoplasms exclusively composed of ghrelin-producing cells and associated with high circulating ghrelin levels were described only in the stomach and pancreas.[10] Therefore, the expression of ghrelin found in tailgut cysts that undergo malignant transformation into well-differentiated neuroendocrine tumors may be hypothetically explained by the possible activation of the ghrelin gene, since this hormone is not usually found in endocrine cells of the hindgut. Although this expression may indicate the foregut phenotype of the tumor, it is important to consider that hormones may also be responsible for the malignant transformation of tailgut cysts.

Diagnostic workup

For retrorectal cysts, differential diagnoses such as dermoid or epidermoid cysts, rectal duplication cysts, neurogenetic cysts, teratomas, anterior sacral meningoceles, leiomyosarcomas, cystic lymphangiomas, pyogenic abscesses, neurogenic cysts, sacral chordomas, and tailgut cysts should be considered.[7] In the literature, some cases of patients who underwent unnecessary operations before the correct diagnosis were reported. In a large series of 53 cases collected over 35 years, Hjermstad and Helwig found that 51 out of 53 cases were not given the correct initial diagnosis of tail gut cysts.[17]

The gold standard of the currently available imaging modalities is pelvic MRI for detecting unilocular, multilocular, and small peripheral cysts, which typically demonstrate low signal intensity on T1-weighted images and high signal intensity on T2-weighted images.[18] On CT, tailgut cysts present as thin-walled, unilocular, or multilocular, non-enhancing lesions with low density in the retrorectal space. This imaging modality helps to determine whether the cysts involve the sacrum or adjacent structures. MRI appears to have an advantage over CT because contrast resolution is superior for soft tissues, which provides good delineation of the anatomic extent of the tumor and superior tissue characterization.

Notably, MRI and CT alone might lead to the misdiagnosis of tailgut cysts with malignant transformation. In some cases, a tailgut cyst was diagnosed as endometrioma. The final diagnosis remains histopathological; however, biopsy is not advertised and is controversial because it carries the risk of seeding tumor cells. In cases of cysts with mixed solid and cystic components, a percutaneous preoperative parasacral biopsy should be performed to determine the surgical strategy and whether postoperative adjuvant therapy is needed for the patient.[19]

Tailgut cysts usually have a nonspecific clinical presentation, and up to 50% of patients are asymptomatic and are likely to be identified accidentally by MRI and CT.[20] In symptomatic cases, the symptoms are usually due to the mass effect and include constipation, obstructed defecation, lower abdominal pain, back pain, rectal pain, tenesmus, painless rectal bleeding, dysuria, urinary frequency, and even lower limb neurological problems. Owing to the wide range of symptoms, the diagnosis of tailgut cysts is complicated by the rare complications of bleeding, infection, and malignant transformation.[21]

Treatment modalities and prognosis

Complete surgical mass removal is the best treatment for rectal tumors because of the risk of recurrence, bleeding, chronic infection, or malignant transformation. The most used surgical approaches are anterior (trans-abdominal), posterior (inter-sphincteric, trans-sphincteric parasacrococcygeal, trans-sacral, transsacrococcygeal, trans-anorectal and trans-vaginal), or a combined anterior and posterior approach.[22] The choice of surgical approach depends on factors such as the size and extension of the cyst, the presence of bleeding or infection, adherence to neighboring structures, and the absence of malignant transformation. There are limited reports on recurrence after surgical extraction of tailgut cysts, and the literature suggests a range of 0–16% for complete excision.[23] Thus, the risk of cyst recurrence is low, and patients are mostly seen annually for digital rectal examination.

Minimally invasive treatment/robotic approach

Because surgery is the main recommendation in the presence of tailgut cysts, minimally invasive techniques can minimize morbidity and enhance recovery compared with laparoscopic or open techniques. An example of the placement of the robotic port, the Da Vinci Si, is shown in figure 4.

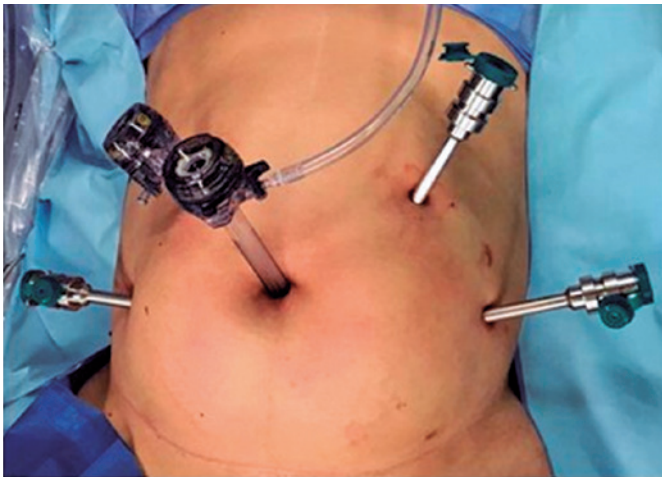

Figure 4. Placement of the robotic ports Da Vinci Si Surgical System

The abdominal laparoscopic approach is the most commonly used technique for resecting tailgut cysts. However, some conditions, such as a narrow pelvis and large cyst dimensions, can lead to technical complications such as tissue damage and nerve injuries. A single-center study of 24 cases of tailgut cysts reported that one patient had sexual dysfunction and another had pelvic floor dyssynergia and lower limb weakness after the laparoscopic procedure.[7] Another study highlighted that the laparoscopic approach does not prevent injury to the nerve fibers but is a better choice than the open surgical approach.[24]

An alternative to reduce the risk of tissue damage is the robotic approach, which helps surgeons manipulate the cyst and reduces the risk of nerve injury. Moreover, because the robotic platform provides precise maneuverability and improved dexterity with wristed instruments, the tailgut cyst can be completely removed, thereby reducing the risk of recurrence.[24] If this possibility is available in a medical center, it is the preferred technique because it reduces the complications of tailgut cyst resection.

CONCLUSION

Minimally invasive treatments, such as the robotic approach for tailgut cysts, are safe and reduce the risk of tissue damage. This paper presents four patients who underwent robotic resection of a tailgut cyst. This is the first reported case of robotic excision of such a lesion in Brazil.

AUTHORS' CONTRIBUTION

Bruno Mirandola Bulisani, Murilo Rocha Rodrigues, and Felipe Martin Bianco Rossi: conceptualization, data curation, investigation, methodology, resources, supervision, validation, and visualization. Luiz Guilherme Lisboa Gomes: conceptualization, data curation, investigation, methodology, resources, supervision, validation, writing – original draft, and writing – review and editing. Milena Arruda de Oliveira Leite: investigation, methodology, writing – original draft, and writing – review and editing. Ricardo Moreno, Renato Barretto Ferreira da Silva, Luiz Carlos Benjamin do Carmo, and Jaques Waisberg: conceptualization, investigation, methodology, project administration, supervision, validation, and visualization.

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