



Tailgut Duplication Cyst: A Rare Cause of Refractory Low Back Pain in a Teenage Girl - A Case Report

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Tailgut cyst, or retrorectal cystic hamartoma, is a rare congenital malformation originating from retrorectal or presacral space. It stands as an embryonic vestige of the hindgut's postnatal component. Owing to its rarity, undiagnosed asymptomatic cases and diverse array of clinical presentations that potentially mimic other diseases, correct diagnosis is often delayed. It can present at any age but is most prevalent in middle-aged females, often remaining asymptomatic and eluding detection during digital rectal examinations. Postoperative histopathological examination is the definitive diagnostic method. This case report highlights an unusual presentation of a tailgut cyst as an underlying etiology for lower back pain in a 17-year-old female patient, emphasizing the importance of considering it as a potential differential diagnosis for intractable lower back pain, especially in young females. Timely recognition of this condition is pivotal for precise management and better patient outcomes.

Key words: Tailgut cyst, retrorectal mass, hamartoma, presacral space

INTRODUCTION

The retrorectal space, also known as presacral space, is a complex area in the human body that can harbor variety of masses, both benign and malignant.¹ These masses can arise from congenital, embryological, inflammatory, osseous, neurogenic, or neoplastic origins.²⁻⁴ One particularly rare congenital malformation that can develop in this space is the tailgut cyst, which represents an embryological remnant of the postnatal component of the hindgut.^{2,3} Due to their rarity, diverse clinical presentations and potential to mimic other conditions, tailgut cysts are often diagnosed late.⁵ It can occur at any age but most prevalent in middle-aged females. They are frequently asymptomatic.^{2,6} Computed tomography (CT) scan and magnetic resonance imaging (MRI) are the cornerstone of investigation.^{1,2} Complete surgical resection of the mass is the treatment of choice.^{1,2} Postoperative histopathological examination remains the definitive mode of diagnosis.⁷ This case report presents the unique occurrence of a tailgut cyst as a rare cause of refractory lower back pain in a 17-year-old female patient.

CASE REPORT

A 17-year-old female patient sought medical attention with complaints of diffuse abdominal and lower back pain persisting for 8 months. Over the past 3 months, she had experienced acute pain during defecation and occasional perrectal bleeding. Her initial symptoms were occasional low back pain, primarily around the coccyx, which eventually progressed to diffuse abdominal pain radiating to the right inguinal region. Digital rectal examination revealed a soft, tender bulging mass of approximately 3–4 cm in close proximity to the rectal wall. Hematological tests showed anemia (hemoglobin: 9 g/dL), whereas other biochemical parameters were within normal range.

Imaging studies, including ultrasonography (USG) and contrast-enhanced CT of the abdomen and pelvis, revealed a well-defined multilobulated cystic lesion measuring 6.0 cm × 5.8 cm × 4.0 cm in the presacral and precoccygeal space on the left side of the rectum. This cystic mass displaced

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the rectum to the right and was closely associated with the rectal wall, sacrum, and coccyx [Figure 1]. MRI of the pelvis further confirmed the presence of this large, multiloculated cystic mass that caused mild compression of the rectum to the right. All these radiological findings strongly indicated the presence of a retrorectal cystic mass, necessitating histopathological analysis for a definitive diagnosis.

After ruling out common causes of lower back pain through hematological, biochemical, and radiological investigations, the patient underwent surgical excision of the mass using a posterior Kraske technique as a transsacrococcygeal approach to gain access to the retrorectal space. The excised specimen, upon gross pathological examination, revealed a 6.5 cm × 4.5 cm × 4 cm hard, uneven, rubbery, brownish-red tissue mass composed of multiple cysts filled with white jelly-like material [Figure 2a and b]. Microscopic examination of multiple sections showed multilocular cysts predominantly lined by stratified squamous epithelium, with adnexal structures, some lined by pseudostratified ciliated columnar epithelium, and containing mucinous and keratinous debris. The sections also displayed fibrofatty tissue, smooth muscles, fibrocollagenous tissue, nerve fibers, areas of granulation tissue formation, and dense chronic inflammatory infiltrate with hemorrhage. Importantly, no malignancy was detected, confirming the diagnosis of a benign tailgut cyst [Figure 3a and b]. The postoperative recovery period was uneventful, and the patient was discharged 2 days postsurgery. Subsequent pelvic MRI follow-up at 3 months showed no signs of recurrence.

DISCUSSION

The retrorectal space, which lies between the rectum anteriorly and sacrum and coccyx posteriorly, can give rise to a wide array of diseases, including inflammatory, neoplastic, congenital, solid, and cystic lesions.¹⁻⁴ These lesions have various names in surgical, pathological, and radiological literature, such as tailgut duplication cyst, myoepithelial rectum hamartoma, and retrorectal cystic hamartoma.⁴ The first recorded case of a tailgut cyst by Middeldorpf dates back to 1885 in a female newborn, and its embryological origin is associated with the failure of the tailgut to regress during fetal development.^{4,8}

Tailgut cysts are exceptionally rare, with an estimated incidence of 1 in 40,000 cases, and can present at any age, although they are most commonly detected in individuals aged 30–60, predominantly affecting females with a female-to-male ratio of approximately 5:1.^{1,2,5} This case stands out as a rare occurrence of a tailgut cyst in a 17-year-old female patient.

Most cases are asymptomatic and may go unnoticed during digital rectal examinations.² Symptoms can range from an

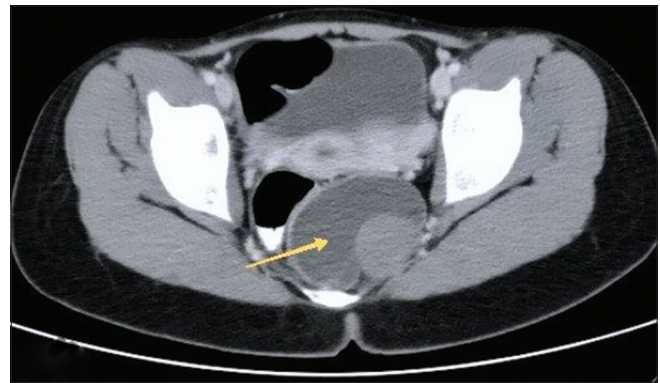


Figure 1: Axial section of contrast-enhanced computed tomography scan of abdomen and pelvis showing a large well defined nonenhancing soft-tissue density cystic component lesion (shown by yellow arrowhead) in the presacral and precoccygeal space, displacing rectum to the right

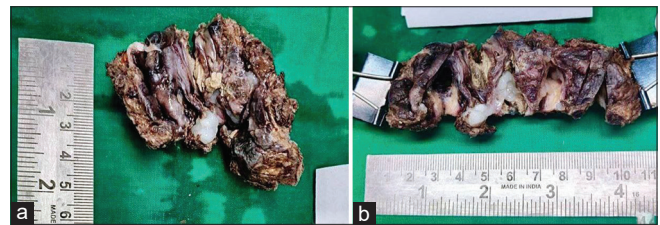


Figure 2: (a) Gross image of complete surgical resection showing an irregular, firm, rubbery, brownish-red tissue mass. (b) Macroscopic examination of the specimen showing a mass comprising of multiple cysts. On cutting open, the cyst revealed a smooth, glistening lining with white jelly-like material oozing out from inside

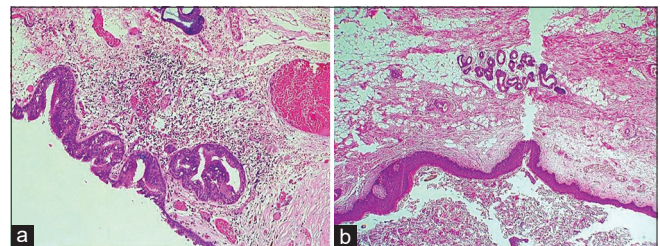


Figure 3: (a) Photomicrograph showing cyst wall lined by stratified squamous epithelium and granulation tissue, along with few mucinous glands (H and E, ×45). (b) Photomicrograph showing cyst wall lined by stratified squamous epithelium with keratinous debris and underlying adnexal structures (H and E, ×45)

incidental mass discovery to more severe clinical presentations, including polyuria, changes in stool caliber, palpable rectal masses, infected tailgut cysts with fistulas or pelvic abscesses, abdominal pain, urinary retention, defecation difficulties, and perrectal bleeding. Uncommon presentations such as sciatica have also been reported.^{2,6}

In diagnosing retrorectal cystic lesions, it can be challenging to differentiate tailgut cysts from other conditions such as duplication cysts, dermoid cysts, epidermoid cysts, cystic hamartomas, sacrococcygeal teratomas, cystic

lymphangiomas, and abscesses.^{4,6} Imaging modalities such as USG, CT scans, and MRI play crucial roles in reaching a differential diagnosis, but the final confirmation requires histopathological examination, as in this case.^{4,6,7}

Although fine-needle aspiration under endosonographic guidance can be attempted, it carries risks, and complications may make subsequent surgical excision more challenging.^{1,2} Surgical removal is the recommended treatment for both symptomatic and asymptomatic cases to prevent complications and malignant transformation.^{2,3} Various surgical approaches exist, each with its advantages and disadvantages. Complete resection, including all attachments, is essential to prevent recurrence.¹⁻³

Malignant transformation of tailgut cysts is rare but possible (as seen in 2%–13% cases), with adenocarcinoma being the most common histological type.^{3,5} This case did not exhibit any malignant changes, aligning with the majority of tailgut cysts.^{2,3,5}

CONCLUSION

Diagnosing tailgut cysts remain a significant challenge due to their rarity, variable clinical presentations, and potential to mimic other conditions. This case report underscores the importance of considering tailgut cysts as a differential diagnosis when evaluating refractory lower back pain, especially in young females. Early diagnosis and prompt management involving an interdisciplinary team of surgeons, radiologists, and pathologists would be most optimal to achieve better patient outcomes.

Declaration of patient consent

The authors certify that they have obtained appropriate consent forms from the legal guardians of the patient. In the form, the guardians have given the consent for the images and other clinical information of the patient to be reported in the journal. The guardians understand that the name and initials of

the patient will not be published and due efforts will be made to conceal the identity, but anonymity cannot be guaranteed

Data availability statement

The data that support the findings of this study are available from the corresponding author on reasonable request.

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Conflicts of interest

There are no conflicts of interest.

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