

Hidden in plain sight: A rare subcutaneous tailgut cyst mimicking chronic sacrococcygeal lesions

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ABSTRACT

Tailgut cysts (retrorectal cystic hamartomas) are rare congenital remnants of the embryonic hindgut. They typically present as deep-seated presacral masses, often diagnosed incidentally or after secondary complications. However, subcutaneous tailgut cysts are extremely rare, often leading to misdiagnosis. We report a unique case of a 37-year-old female with a chronic, painful sacrococcygeal lesion, initially misdiagnosed as a recurrent infected sinus. The case underscores the diagnostic challenges, the need to differentiate tailgut cysts from other subcutaneous lesions in this region, and the crucial role of histopathology in achieving an accurate diagnosis.

Key words: Differential diagnosis, Histopathology, Rare case, Sacrococcygeal lesion, Subcutaneous cyst, Tailgut cyst

Tailgut cysts, also known as retrorectal cysts, postanal dermoid cysts, or hamartomas of the hindgut, are rare congenital lesions that arise from remnants of the embryonic hindgut [1]. The incidence of tailgut cysts is estimated to be 1 in 40,000 individuals. While these cysts can manifest across all age groups, they are most frequently observed in middle-aged women [2,3]. These cysts are generally considered a developmental anomaly that occurs when the vestigial structures of the postanal gut or tailgut fail to regress properly during embryogenesis. The tailgut itself is a vestigial structure that extends from the cloaca and is an early precursor to the anorectal region [4]. In most cases, this structure regresses and is absorbed into the surrounding tissues as development progresses; however, when this process does not occur properly, it leads to the formation of a cystic structure known as a tailgut cyst, lined with various epithelial types, including squamous, transitional, and glandular epithelium [5]. No specific risk factors have been identified in the literature [6-9]. Their cellular composition suggests that tailgut cysts share histopathological similarities with other retrorectal developmental cysts, but their unique embryological origin differentiates them from other congenital lesions. While tailgut cysts are classically located in the presacral or retrorectal space, rare occurrences of their presentation in subcutaneous tissue have been documented [10,11], making their diagnosis particularly challenging.

This case report revealed the details of a 37-year-old female with a chronic, painful lesion in the sacrococcygeal region, characterized by serosanguinous discharge and a history of two unsuccessful incision and drainage (I and D) procedures. The lesion's unusual clinical presentation, including its depth of probing and the lack of communication with the anal canal, prompted further investigation and ultimately led to surgical excision and histopathological analysis, confirming the diagnosis of a tailgut cyst. Following complete excision, the patient experienced significant relief from chronic pain, marking a notable improvement in her quality of life.

CASE REPORT

A 37-year-old female presented to our clinic with a long-standing, painful lesion in the sacrococcygeal region. This lesion was associated with serosanguinous discharge that had persisted for several years. Despite undergoing two I and D procedures over the past 8 years, the lesion remained symptomatic and unresponsive to treatment. The patient reported intermittent episodes of pain, discharge, and swelling but did not experience any changes in bowel habits or other gastrointestinal symptoms.

Upon clinical examination, the patient had normal blood pressure, heart rate, and respiratory rate, with the absence of fever. She had local pain and discomfort in the sacrococcygeal region. Physical examination showed a painful, tender fleshy swelling of 3.5 × 2.5 cm surrounded by erythema and mild induration of the skin with mild hyperpigmentation (Fig. 1). The

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lesion appeared wet with mild serous discharge. It was firm and tender to palpation, and its consistency suggested the presence of a subcutaneous mass. Anal examination of the patient was unremarkable. There were no signs of neurological effects like loss of sphincter tone or sensory deficits. No palpable regional lymph nodes were identified. The lesion was firm and tender to palpation, and its consistency suggested the presence of a subcutaneous mass.

Given the location and presentation, a suspected diagnosis of a recurrent perianal abscess or a pilonidal cyst was considered. However, probing the lesion with a blunt instrument revealed a depth of approximately 10 cm, without any communication with the anal canal or the rectum (Fig. 2). This finding excluded the possibility of a perianal abscess or fistula. The absence of any connection to the anal canal, along with the lesion's depth and chronicity, necessitated further surgical intervention. Routine laboratory tests, including complete blood count and inflammatory markers, were unremarkable, and the patient's vital signs remained stable throughout the course of her evaluation.

After considering the clinical presentation and ruling out more common diagnoses, the patient was scheduled for surgical excision of the lesion. Given the chronic nature and superficial location



Figure 1: Clinical picture of an erythematous, discharging, fleshy protruding lesion at the sacrococcygeal region

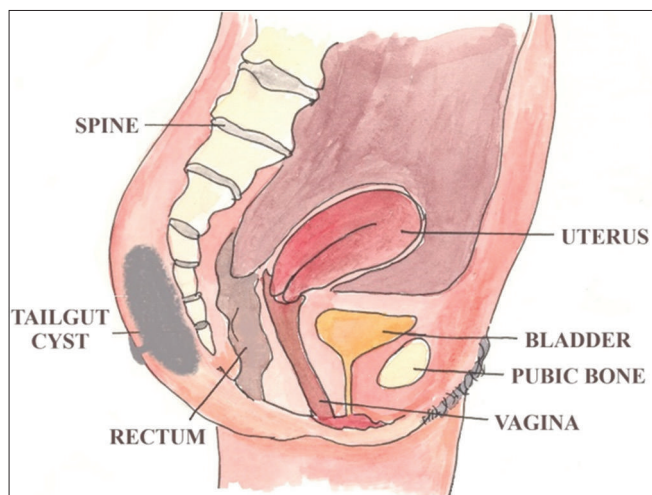


Figure 2: Diagram of sacrococcygeal cyst and its anatomical relations

of the lesion, preoperative imaging studies such as ultrasound or magnetic resonance imaging were not initially performed.

A midline vertical incision was made from the sacrum to the coccyx area to provide adequate exposure to the lesion. Dissection revealed a cyst measuring 10.0 × 4.0 × 3.0 cm, in the subcutaneous plane, located just beneath the skin overlying the sacrococcygeal region. The cyst did not extend into deeper pelvic spaces, which further suggested that it was confined to the subcutaneous layers. The cyst was filled with thick, whitish material, and its walls were noted to be approximately 2 cm thick. Careful dissection was performed to avoid damaging surrounding structures, particularly the sacrum and coccyx.

Macroscopically, the well-circumscribed cystic mass had mucosal characteristics on one side with a white/pink color and a smooth appearance with an irregular surface on the other side (Fig. 3). It was fixed with 10% buffered formalin for 24 h. The entire specimen was processed following staining of the irregular surface with Indian ink. These specimens were dehydrated in ethanol and xylene at room temperature and embedded in paraffin (60°C). Following the preparation of 4-μm sections, they were stained with hematoxylin and eosin at room temperature according to a standard protocol. Two pathologists independently evaluated the histopathological features of all the slides under a microscope.

Microscopic examination revealed a cyst lined by predominantly non-keratinizing stratified squamous epithelium, followed by stratified cuboidal epithelium focally. Disorganized smooth muscle bundles were identified in the cyst wall. There was the absence of two-layer muscle and neural plexus in the cyst wall (Fig. 4). No evidence of necrosis, granulomas, dysplasia, or malignancy was observed. The histopathological findings confirmed the diagnosis of a tailgut cyst. Complete excision provided significant relief from the patient's long-standing chronic pain, improving her quality of life.

DISCUSSION

Tailgut cysts are rare congenital lesions arising from remnants of the embryonic hindgut. They are predominantly located in

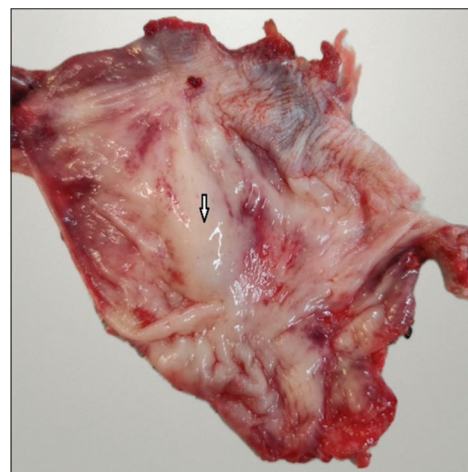


Figure 3: Gross picture of excised cystic mass with a smooth shiny white/pink mucosal surface (arrow)

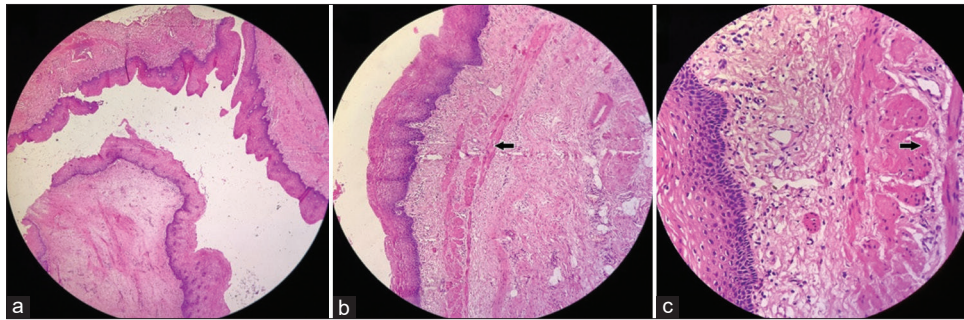


Figure 4: Microscopic findings: (a) An overview of the cyst wall architecture (Hematoxylin and eosin [H and E] stain, $\times 4$). (b) The cyst wall is covered with non-keratinizing squamous epithelium and shows the presence of disorganized smooth muscle bundles (arrow) (H and E stain, $\times 10$). (c) The cyst wall shows the presence of disorganized bundles of smooth muscle (arrow) without neural plexi (H and E stain, $\times 40$)

the presacral space, with subcutaneous presentations being exceptionally uncommon. In a comprehensive review by Hjernstad and Helwig, 53 cases of tailgut cysts were documented over 35 years, underscoring their rarity [1]. More recent literature continues to emphasize the infrequent nature of these lesions. For instance, a study by Patel *et al.* analyzing a multi-institutional database further confirmed the rarity of tailgut cysts and highlighted the variability in their clinical presentation [12].

The subcutaneous manifestation of tailgut cysts, as observed in our patient, is particularly rare. A case reported by Li *et al.* described a subcutaneous tailgut cyst in a 55-year-old woman, highlighting the diagnostic challenges due to its atypical location [10]. Similarly, a case by Haydar *et al.* detailed a tailgut cyst incidentally detected in a 42-year-old female, emphasizing the importance of considering this diagnosis even in asymptomatic patients [13]. Comparing our case with these findings, our patient also presented with a subcutaneous lesion, further adding to the limited number of reported cases in this unusual location. Notably, the age range of reported subcutaneous cases appears variable, suggesting that this presentation is not limited to a specific age group. A more recent case report by Ganapathy *et al.* described a subcutaneous tailgut cyst in a 38-year-old male, further supporting the possibility of subcutaneous presentation and providing additional insights into the clinical characteristics and diagnostic pathways in such instances [14].

The rarity of subcutaneous tailgut cysts often leads to misdiagnosis, as they can mimic more common lesions in the sacrococcygeal region. In our case, the lesion was initially misdiagnosed as a recurrent infected sinus, leading to multiple ineffective treatments. A study conducted by Malutan *et al.* similarly emphasized the diagnostic significance of considering tailgut cysts in the differential evaluation of perirectal tumors, particularly in the context of patients with a documented history of multiple unsuccessful procedures and surgical interventions [15]. This aligns with our experience, where the chronic nature and lack of response to standard treatments for more common conditions should have raised suspicion for a less frequent entity like a tailgut cyst. A recent study by Samdani *et al.* discussed the diagnostic challenges of sacrococcygeal masses and highlighted the importance of a thorough clinical and radiological evaluation to avoid misdiagnosis [16].

Several differential diagnoses must be considered for a subcutaneous cystic lesion in the sacrococcygeal region, including:

pilonidal disease, epidermoid and dermoid cysts, recurrent perianal abscess or fistula, and enterogenous cyst. Histopathology remains the gold standard for diagnosis, as imaging alone may not always distinguish between these entities. Importantly, although tailgut cysts are benign, they carry a small risk of malignant transformation, emphasizing the necessity of complete surgical excision [17]. Although rare, cases of tailgut cysts undergoing malignant change to adenocarcinoma or neuroendocrine tumors have been reported [18]. Consequently, surgical excision remains the definitive treatment, preventing future complications and allowing for histological confirmation of benignity.

CONCLUSION

This case highlights the importance of considering tailgut cysts in the differential diagnosis of persistent sacrococcygeal lesions. Subcutaneous tailgut cysts, though extremely rare, can mimic other common lesions, leading to misdiagnosis and ineffective treatment. A high index of suspicion, combined with histopathological confirmation, is crucial for timely and definitive management. Complete surgical excision is curative and prevents potential complications, as well as significantly alleviates chronic pain and improves the patient's quality of life. The rarity of these cysts emphasizes the need for increased awareness among clinicians to ensure timely identification and management.

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