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Case Report

Retrorectal Epidermoid Cyst Developing After Fistulectomy: A Case Report

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Background: Retrorectal tumors are rare and often present with nonspecific symptoms, leading to delayed diagnosis. Epidermoid cysts constitute an uncommon subgroup of retrorectal lesions and are typically benign; however, they may rarely undergo malignant transformation or develop secondary to trauma or surgery.

Case Presentation: We report a very rare case of a retrorectal epidermoid cyst in a 66-year-old man that developed following a fistulectomy. The patient presented with an erythematous bulge in the sacrococcygeal region associated with mild tenderness and a palpable mass on the right side of the rectum. Imaging studies revealed a retrorectal cystic lesion. Surgical excision was performed, and the specimen was submitted for histopathological examination, which confirmed the diagnosis of an epidermoid cyst.

Discussion: Retrorectal epidermoid cysts are rare and may be either congenital or acquired. Surgical implantation of epithelial elements following procedures such as fistulectomy may contribute to their development. Despite their benign nature, these cysts carry a risk of recurrence and, rarely, malignant transformation, necessitating complete surgical excision

Conclusion: Retrorectal epidermoid cysts should be considered in patients presenting with presacral masses, particularly those with a history of prior anorectal surgery. Early diagnosis and complete surgical excision are essential to achieve optimal outcomes and prevent recurrence

Keywords: Retrorectal tumor; Epidermoid cyst; Epidermal inclusion cyst; Fistulectomy; Presacral mass; Case report

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Introduction

The retrorectal space (presacral space) is a potential fascial space in the pelvis, located posterior to the rectum and anterior to the sacrum and coccyx. The retrorectal space is a potential anatomical compartment located between the rectum anteriorly and the sacrum posteriorly. It is divided into superior and inferior compartments by the rectosacral fascia, which arises variably from the sacral levels (S2–S4) and fuses with the rectal visceral fascia above the anorectal junction. Inferiorly, the space is limited by the fusion of the presacral and rectal visceral fascia at the level of the anorectal junction, just above the levator ani muscle (1).

Tumors arising in this region are rare and often present with nonspecific clinical manifestations, frequently leading to misdiagnosis and inappropriate surgical interventions (2). These tumors encompass a wide spectrum of histological subtypes. Their estimated incidence is approximately 1 in 40,000 patients, with a female predominance and a mean age at presentation of 30 years (3, 4). Retrorectal lesions may be congenital, inflammatory, neurogenic, osseous, or derived from embryological remnants (2). Congenital lesions are relatively more common, and approximately two-thirds of these are developmental cysts, including epidermoid,

dermoid, enteric, tailgut, and duplication cysts (5). Epidermoid cysts are benign lesions that generally remain asymptomatic unless they become secondarily infected. Although they are slow-growing lesions, they occasionally demonstrate malignant transformation (6).

While epidermal inclusion cysts can occur in various regions of the body, particularly on the trunk, back, scalp, and face, they are rarely encountered in the retrorectal region. Although several theories have been proposed regarding the etiopathogenesis of epidermoid cysts, their exact cause remains unclear. Studies suggest that these cysts most commonly arise spontaneously; however, they may also develop secondary to trauma or surgical procedures through implantation of epithelial elements into the subepithelial tissues (7).

Malignant transformation of an epidermoid cyst into carcinoma is uncommon but has been reported, with squamous cell carcinoma (SCC) being the most frequently described histological subtype (8). We report a rare case of a retrorectal epidermal inclusion cyst developing after fistulectomy in a 66-year-old man. Physical examination revealed an erythematous bulge in the sacrococcygeal region associated with mild tenderness. To our knowledge, only a limited number of cases with similar histopathological features arising in the retrorectal space have been reported, making this case particularly noteworthy. This case report has been prepared in accordance with the SCARE 2025 guidelines (9).

Case Presentation

A 66-year-old man was referred to our colorectal surgery clinic with complaints of a sacrococcygeal mass, discomfort, and difficulty sitting. He denied rectal bleeding, weight loss, or purulent discharge. Eight years before presentation, he had undergone surgical drainage and fistulectomy for a high posterior transsphincteric fistula associated with a posterior anal abscess. Since then, he had remained asymptomatic. He had no significant past medical history or relevant family history. The patient was not taking any medications, and his social and allergy histories were unremarkable.

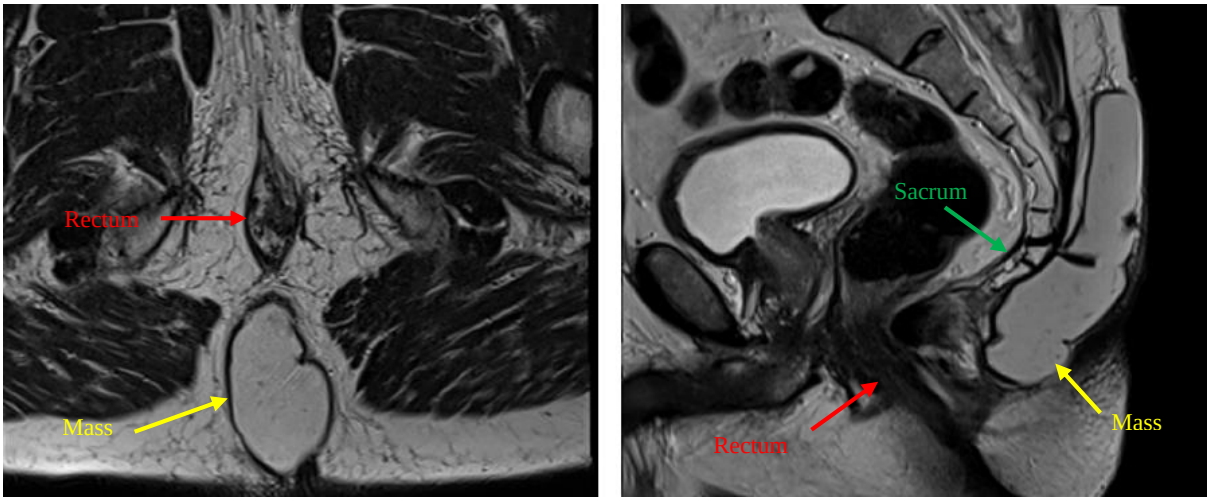

Fig. 1. Magnetic resonance imaging (MRI) revealed a well-defined, ovoid cystic lesion measuring 120 × 60 × 25 mm within the retrorectal/presacral space, located in the midline inferior to S3 level. On axial images, the lesion demonstrates homogeneous internal signal intensity with a thin, smooth, and regular wall. No definite solid component or internal nodular enhancement is identified. On sagittal images, the lesion appears as a large cystic structure with high T2 signal intensity, consistent with fluid content. It is closely related anteriorly to the rectum and posteriorly to the sacrum. Inferior extension toward the perineum is noted. No obvious invasion of adjacent structures is identified on the provided description.

Physical examination revealed an erythematous bulge in the sacrococcygeal region associated with mild tenderness. Digital rectal examination demonstrated a palpable mass on the right side of the rectum without evidence of a perianal fistula or abscess. The remainder of the physical examination was unremarkable, and laboratory investigations were within normal limits.

MRI demonstrated a well-defined, ovoid cystic lesion measuring 120 × 60 × 25 mm in the retrorectal (presacral) space, located in the midline inferior to the S3

level. The lesion showed homogeneous signal intensity with a thin wall and no solid component or nodular enhancement. It appeared hyperintense on T2-weighted images, consistent with fluid content, and was situated anterior to the sacrum and posterior to the rectum, with inferior extension toward the perineum. No evidence of invasion into adjacent structures was seen. The patient underwent surgical excision under spinal anesthesia through a trans-sacral approach. Intraoperatively, the lesion was found to extend close to the rectal mucosa without invading it. The rectal mucosa remained intact, and the lesion was completely excised en bloc with its capsule while preserving the rectal wall.

Gross examination revealed a completely cystic mass with a smooth brown wall containing yellow gelatinous material.On sectioning, the cyst was unilocular and contained white/brown, cheesy keratinous material. No solid nodules or papillary excrescences were identified.

Histopathological examination demonstrated a cyst wall lined by stratified squamous epithelium with a well-defined granular layer. The cyst lumen contained lamellar keratin. Infiltration by chronic inflammatory cells was also observed. No skin adnexal structures (hair follicles, sebaceous glands) were identified in the cyst wall.There were no evidence of dysplasia or malignancy.Overall, the findings were consistent with an epidermal inclusion cyst with superimposed chronic inflammation, a lesion that is rarely encountered in the retrorectal region.

The patient was discharged on the first postoperative day and was subsequently followed regularly in the colorectal surgery outpatient clinic. No evidence of recurrence was observed during the two-year follow-up period

Discussion

The retrorectal space, also known as the presacral space, is a potential anatomical compartment located posterior to the rectum and anterior to the sacrum and coccyx. Lesions arising in this region are often asymptomatic, which may delay diagnosis. However, some patients present with symptoms resulting from compression of adjacent structures. Common manifestations include chronic constipation, pelvic discomfort, and a sensation of rectal fullness or heaviness(10). In certain cases, a mass may be detected during digital rectal examination. Therefore, a detailed medical history, careful physical examination, and appropriate imaging studies are essential for establishing an accurate diagnosis(11).

Magnetic resonance imaging (MRI) plays a pivotal role in the evaluation of retrorectal lesions. Owing to its excellent soft-tissue resolution, MRI enables accurate assessment of lesion size, internal characteristics, anatomical extent, and relationships with adjacent pelvic structures. In addition, MRI may help identify features suggestive of malignant transformation, such as irregular wall thickening, mural nodules, or invasion of surrounding tissues. In the present case, MRI revealed a large cystic lesion extending from the intergluteal cleft into the retrorectal space, which was instrumental in preoperative assessment and surgical planning(12).

Retrorectal cystic lesions encompass a broad spectrum of congenital and acquired entities. The differential diagnosis includes epidermoid cysts, dermoid cysts, tailgut cysts, enteric duplication cysts, anterior sacral meningoceles, abscesses, and other presacral cystic masses. Dermoid cysts differ from epidermoid cysts by the presence of skin appendages such as hair follicles, sebaceous glands, and sweat glands within the cyst wall. Tailgut cysts originate from remnants of the embryonic hindgut and may be lined by multiple epithelial types. Enteric duplication cysts are characterized by a gastrointestinal epithelial lining and a well-developed smooth muscle layer. Furthermore, infected cystic lesions may mimic presacral abscesses clinically and radiologically. As imaging findings may overlap, definitive diagnosis often relies on histopathological examination following complete surgical excision(12).

Epidermal inclusion cysts, also referred to as epidermoid cysts, represent a relatively uncommon subtype of cystic lesions occurring in the retrorectal region. Although these lesions are generally benign, they carry a small risk of malignant transformation, most commonly into squamous cell carcinoma, as well as a potential risk of recurrence(13).

Complete surgical excision remains the treatment of choice for retrorectal epidermoid cysts. Removal of the cyst in its entirety, including the capsule, is crucial to minimize the risk of recurrence and to allow definitive histopathological evaluation for the exclusion of malignancy (14). In our patient, complete excision was successfully achieved, and histopathological examination

confirmed the diagnosis of an epidermoid cyst without evidence of malignant transformation.

Studies suggest that epidermoid cysts most commonly arise spontaneously; however, they may also be acquired. In some cases, these lesions develop secondary to trauma or surgical procedures as a result of implantation of epithelial elements into the subepithelial tissues(7). Acquired epidermoid inclusion cysts following surgical interventions have been described in various anatomical locations and are believed to result from traumatic implantation of epidermal tissue into deeper structures.

In the present case, the epidermoid cyst developed several years after a fistulectomy. Although a definitive causal relationship cannot be established, the previous fistulectomy may have contributed to the development of an acquired epidermoid inclusion cyst through implantation of epithelial elements during surgery. This case highlights the importance of considering acquired epidermoid cysts in the differential diagnosis of retrorectal cystic lesions, particularly in patients with a history of prior perianal surgery who present with a delayed cystic mass in the sacrococcygeal region. This observation is consistent with previous reports suggesting that trauma or surgical procedures may contribute to the development of epidermoid inclusion cysts in uncommon anatomical locations.

Conclusion

Retrorectal epidermoid cysts are rare lesions that may be either congenital or acquired. They should be considered in the differential diagnosis of presacral masses, particularly in patients with a history of prior anorectal surgery. Although uncommon, these lesions carry a potential risk of malignant transformation and recurrence. Complete surgical excision, including removal of the cyst capsule, remains the treatment of choice and is essential for preventing recurrence and establishing a definitive diagnosis. Further studies are needed to better clarify the etiology and pathogenesis of retrorectal epidermoid cysts, particularly those occurring after surgical interventions.

Limitation: The main limitation of this report is that a definitive causal relationship between the previous fistulectomy and the subsequent development of the cyst cannot be established. In addition, the absence of imaging before the initial fistulectomy limits the ability to determine whether the lesion was acquired or pre-existing.

Ethical Statement: This study was conducted in accordance with the ethical principles of the Declaration of Helsinki. Ethical approval was obtained from the Ethics Committee of Mazandaran University of Medical Sciences. Written informed consent was obtained from the patient for participation and publication of the case details and accompanying images. All patient identifiers were removed to ensure confidentiality. This work has also been reported in line with the SCARE 2025 guideline (9).

Declaration

Availability of data and materials

The datasets used and analyzed during the current study are available from the corresponding author on reasonable request

Conflict of Interest

The authors declare that they have no conflicts of interest related to this publication. No financial or non-financial benefits have been received from any party directly or indirectly related to the subject of this article.

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Authors' contribution

M.A contributed to the clinical management of the patient, data collection, and drafting of the manuscript. P.K supervised the surgical procedure. Both authors read and approved the final manuscript and are accountable for all aspects of the work.

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Ethics approval and consent to participate

N/A

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