

A Rare Case of Tailgut Cyst Turning into Mucinous Adenocarcinoma: Case Report

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Abstract

A tailgut cyst is an embryological remnant of the posterior intestine and is usually benign in nature. Malignant transformation is very rarely seen. We report a case of a tailgut cyst transforming into a mucinous adenocarcinoma. An 18-year-old gentleman presented to our healthcare center with progressive pain and heaviness in the lower abdomen and lower back for 1 year. Contrast-enhanced computed tomography reveals a cystic mass in the presacral region. The patient underwent transabdominal excision of the cyst in toto, and histopathological evaluation revealed mucinous adenocarcinoma. The patient underwent adjuvant chemo-radiotherapy, and after one year, contrast-enhanced computed tomography revealed no residual or recurrence. Tailgut cysts carry the risk of fecal incontinence, fecal fistula, neurogenic bladder, and malignant transformation. Hence, complete excision followed by adjuvant treatment is advised. Excision can be done by transabdominal, posterior (Presacral/ Pre-coccygeal), or combined approach.

Keywords: Tailgut cyst, Malignant transformation, Mucinous adenocarcinoma of tailgut cyst

INTRODUCTION

Tailgut cysts are rarely seen entities.¹ They are remnants of the posterior intestinal duct embryologically.² They occur mainly in middle-aged, predominantly female individuals, and are usually detected incidentally.³ Most of them are benign in nature; however, malignant trans-

formation has been seen.⁴ They are usually asymptomatic, and if symptomatic, then due to compression from a large mass, i.e., heaviness and pain in the lower abdomen and back, tenesmus, chronic constipation, infection, fecal fistulas, and obstructive urinary symptoms.³

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CASE REPORT

An 18-year-old male with complaints of gradually progressive heaviness, pain in the lower abdomen and back, on-off constipation for one year, and recently worsening lower urinary tract obstructive symptoms. The patient had a history of congenital anal atresia with a diversion stoma followed by reconstruction done during infancy. On palpation, a non-tender, immobile, firm mass of size 10×10 with a smooth surface was present in the hypogastrium. Digital rectal examination reveals a cystic mass along the posterior wall of the rectum, causing luminal narrowing. Contrast-enhanced computed tomography of the abdomen revealed a $14 \times 9 \times 8$ cm presacral homogenous cystic mass, compressing the left ureter, causing proximal hydronephrosis and multiple hemivertebrae (Figures 1 and 2), and no other lymph node was enlarged. Left-side DJ stenting was done pre-operatively to decompress the left kidney ureter system. Intraoperatively (Figure 3), the cystic mass of size $14 \times 9 \times 9$ cm (Figure 4) with serosanguinous fluid was present in the presacral region with positional compression on the rectum and left ureter, and no communication with the rectum, ureter, and rest of the viscera. Subsequently, the patient underwent complete surgical excision of the cystic mass in toto. Histopathology shows some “signet ring” cells with high mitotic activity and atypical hyperplasia infiltrating columnar epithelium in glandular or cribriform pattern in lakes of mucin, suggestive of Tail gut cyst with mucinous adenocarcinoma transformation (Figure 5). The patient received adjuvant chemo-radiotherapy. Follow-up contrast-enhanced abdomen computed tomography after 1 year shows no residual disease.



Figure 1 The sagittal section shows a large homogenous cystic mass in the presacral region (Arrow)

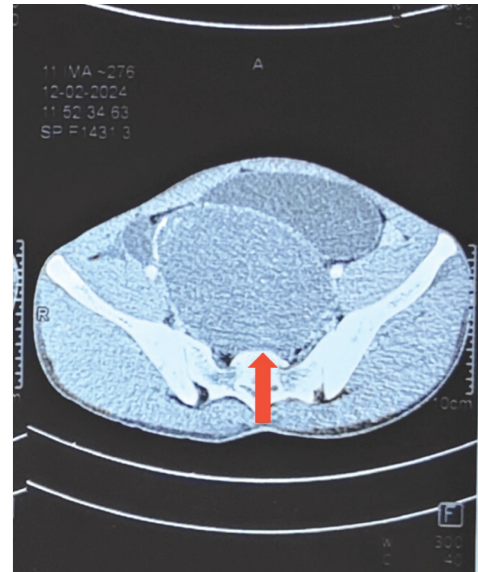


Figure 2 Axial section showing presacral cystic mass (Arrow)

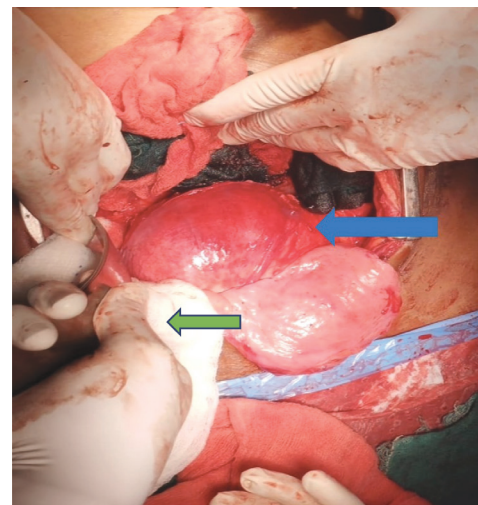


Figure 3 Intra-op showing presacral cystic mass (blue arrow= cystic mass, green arrow=rectum)

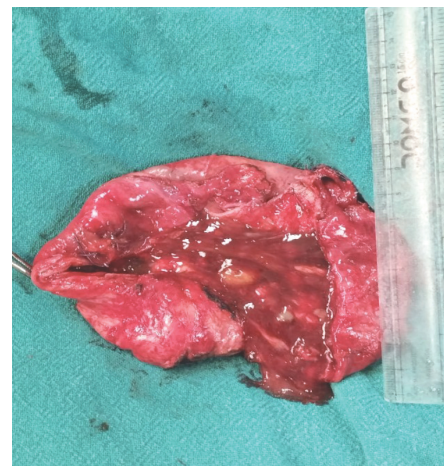


Figure 4 Cystic mass of size $14 \times 9 \times 8$ cm with serosanguinous fluid in it

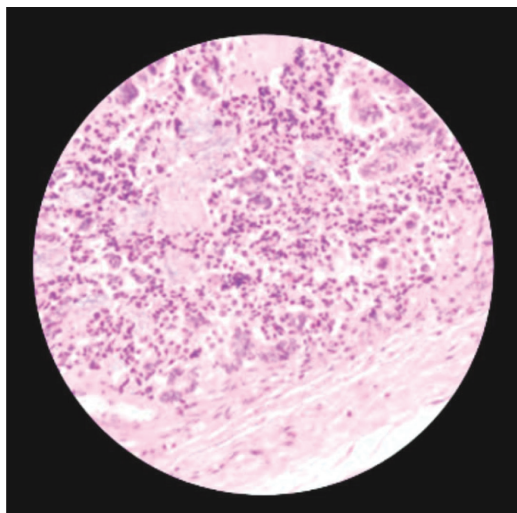


Figure 5 Histopathological evaluation showing feature of tailgut cyst with mucinous adenocarcinoma

DISCUSSION

We present a rare case of mucinous adenocarcinoma developing from a tailgut cyst. Approximately more than 28 cases of Tailgut cyst adenocarcinoma have been reported. Tailgut cysts are congenital pathologies that develop from the remnant of the posterior intestinal duct in the presacral space. They are usually thin-walled cysts that can be single or multiloculated in nature and contain all three germinal layer tissues. They may be asymptomatic or present with non-specific symptoms due to compression by a large mass.² It can cause several complications, i.e., fecal incontinence, internal fecal fistula, bowel obstruction, neurogenic bladder, or malignant transformation.^{5,6} Most Tailgut cysts are benign, but malignant transformation has been reported, the most common being the adenocarcinomas, and others include neuroendocrine, endometrioid, adenosquamous, and squamous cell carcinomas.⁷ On Magnetic resonance imaging (MRI), Cystic tumors with smooth, well-defined boundaries and no infiltrative or gadolinium enhancement are generally considered benign. In contrast, cysts with thickened and irregularly enhancing cyst wall boundaries, which inflammatory changes may even surround, are usually malignant.⁸ Core needle biopsy or FNAC is not recommended in resectable tumors due to the risk of the possibility of tumor seeding.⁹ When imaging shows malignant features, serum carcinoembryonic antigen (CEA) acts as a surrogate tumor marker, and its level decreases post-treatment and helps identify recurrence.

The treatment of choice is complete R0 resection due to the risk of malignant transformation. The surgical approach depends on tumor location, which includes a posterior approach for tumors extending below S4, and for tumors above S4, the abdominal or abdominal-perineal approach is advisable.⁹ When tailgut cysts show a feature of malignancy, treatment should include adjuvant radiation and chemotherapy.^{4,10-12} Capecitabine and oxaliplatin (CapeOX) chemotherapy has been used in previous case reports.⁸ Strict patient follow-up is advised by radiological investigation, including (Magnetic resonance imaging, contrast-enhanced computed tomography) and serum CEA levels, which help detect recurrence.¹² Compared with neuroendocrine tumors, adenocarcinomas arising from TCs have a poorer prognosis and carry a risk of local recurrence and metastasis.¹³

CONCLUSION

Tailgut cysts are usually asymptomatic and present with non-specific symptoms or are detected incidentally. They are usually benign in nature, but malignant transformation (the most common being Adenocarcinoma) has been seen. Tailgut cyst malignancy diagnosis is pre-operatively done by radiological investigation, especially by Magnetic resonance imaging (MRI) and high serum carcinoembryonic antigen (CEA). The malignancy nature is identified by histopathology. Complete excision in toto is recommended, followed by adjuvant treatment. Follow-up is necessary to detect recurrence or residual using radiological investigation and tumor markers.

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