

A Rare Case of Retrorectal Tailgut Cyst

Dr. Shahbaz Habib Faridi ¹, Prof. Hasan Harris ², Dr. Bushra Siddiqui ³, Dr Rajashree Kundu ⁴

¹ Assistant professor, Department of Surgery, Jawaharlal Nehru Medical College, AMU, Aligarh, Uttar Pradesh

² Professor, Department of Surgery, Jawaharlal Nehru Medical College, AMU, Aligarh, Uttar Pradesh

³ Assistant professor, Department of Pathology, Jawaharlal Nehru Medical College, AMU, Aligarh, Uttar Pradesh

⁴ Junior resident, Department of Surgery, Jawaharlal Nehru Medical College, Uttar Pradesh

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I. Case Report:

A 25-year-old female came with a complaint of epigastric pain for a year with soft, non-tender abdomen and per rectally, a fluctuant cystic extrinsic swelling behind the rectal wall was found.

Ultrasonography revealed cholelithiasis along with a well-defined, heterogeneous echotexture mass of 68*45 mm lying posterior to the urethra. CECT abdomen (fig 1) showed a well-defined, predominantly hypochoic lesion with foci of calcification in its wall in the right ischio-rectal fossa region extending up to tip of the coccyx inferiorly, levator ani muscle superiorly and external anal sphincter medially.



Fig 1: CECT abdomen

Laparoscopic cholecystectomy followed by cyst excision was planned. After lower midline incision, the vaginal wall was mobilised on the right side (fig 2). Dense adhesions between the cyst and vaginal wall were dissected by Ligasure and rectum was also mobilised. (Fig 3). The rectal wall and presacral fascia were intact.



Fig 2: Intra-op dissection



Fig 3: Dimension

HISTOPATHOLOGY REPORT: HPE revealed a fibro-collagenous cyst wall lined by skin on one side and flattened epithelium on the inner side. Chronic inflammatory infiltrate was also seen. (Fig 4).

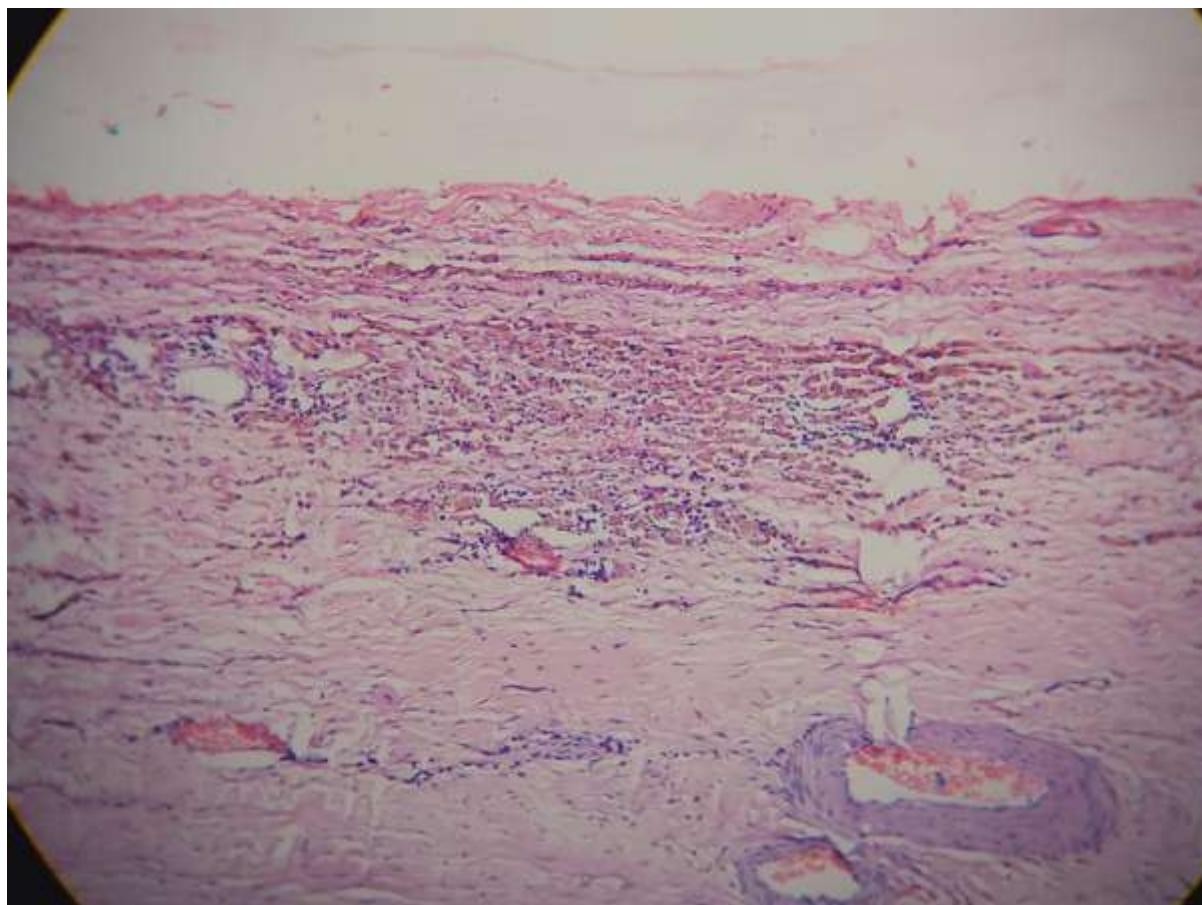


Fig 4: Histopathology; H&E staining; 100x Zoom

II. Discussion:

Incidence of tailgut cyst is 1:40,000 (Mayo Clinic).^[1] The retrorectal space is bounded anteriorly by the mesorectum, posteriorly by the sacrum, superiorly by the peritoneal reflections and inferiorly by the recto-sacral fascia. Symptoms vary from none to prolapsing rectal mass and defecation difficulties (obstipation)^[2].

The cyst may also recur following excision, according to Mayo Clinic Data^[3]. Congenital lesions account for two thirds which include developmental (tailgut, neuroenteric, dermoid, epidermoid, teratoma), chordomas (notochord remnants) and anterior sacral meningoceles. Lovelady and Docketry classified presacral cysts as congenital or acquired tumour.^[4]

Middeldorpf first described a retrorectal mass in 1885 (rectal duplication cyst).

Embryologically, tailgut cysts arise from remnants of embryonic hindgut. The human embryo actually possesses a true tail at the age of 35 days gestational age (8 mm size) into which the primitive hindgut extends. Hence this part is called 'tailgut' and it completely regresses by the age of 56 days gestational age (35 mm size).

Tailgut, if and when fails to regress, gives rise to cystic remnants known as tailgut cyst.

Largest case series was published by Hjermstad and Helwig which included 53 tailgut cysts (1950-1985, American Armed Forces Institute of Pathology).^[5] In 2013, Sami Akbulut studied tumour development of tailgut cysts which showed a female preponderance (male: female=1:3) and age ranging from 4 to 73 years. There was no association between age and cyst size. Chief complaints were low back pain, rectal pain and pain during defecation, rectal bleed, change in stool consistency and urinary frequency and average duration of symptoms was 7.5 months. Most asymptomatic lesions were found incidentally. Most lesions were multicystic with average diameter being 3.9 cm. Epithelial lining and cyst content varied (from clear fluid to dense mucus). One patient had mucinous adenocarcinoma associated with the tailgut cyst highlighting its malignant potential and need of surgical excision even in an asymptomatic scenario.

Most cysts are per-rectally palpable as fluctuant masses. No risk factor has been identified. Transrectal ultrasound delineates the layers of the rectal wall. CECT usually shows a well defined, thin-walled, uni or

multilocular, non enhancing lesion in the retrorectal space. Calcification, if present, should arouse the suspicion of malignancy.^[6]

MRI is the investigation of choice with T1 and T2 showing low and high signal intensities respectively. Irregular wall thickening, intermediate signal intensity before contrast on both T1 and T2 with enhancement post-contrast point towards malignancy.

Biopsy is debated. Posterior paravertebral route is preferred. If the sample is found to be malignant, then en bloc resection of the biopsy tract is indicated. The malignant degeneration in larger case series was 2%.

The various approaches are posterior, anterior abdominal and combined approach.^[7] Posterior approach, easier for low-lying lesions (below S3), needs bowel preparation and a pre-operative briefing about possible stoma. Various routes are— trans-sacrococcygeal, transperineal, transrectal, transsacral and transsphincteric and positions are—jack knife, lithotomy or lateral position. After incision (Parasacrococcygeal/curvilinear/horizontal) is made to access the presacral space, ano-coccygeal ligament and gluteus maximus are divided. Coccygectomy is done in cases of malignant cysts. Cyst should be dissected off rectum and its lateral attachments with a finger in the rectum. Unilateral preservation of S2-S4 is adequate for normal bladder and bowel function. Drainage tract/old biopsy tract/scar should be resected to avoid recurrence.

Transabdominal approach, suitable for high lying cysts (above S3), allows direct visualisation of middle sacral artery, presacral veins, nerves, rectum and the uterus after lower midline incision and rectal mobilisation.

Cysts more than 4-5 cm, best approached by combined approach. Transrectal routes are usually avoided. There is no standard recommendation for follow up.

III. Conclusion:

Surgical excision of tailgut cysts is mandated irrespective of symptoms. MRI is the gold standard investigation. Both anterior and posterior approaches are acceptable as per recommendations.

CONFLICT OF INTEREST: None

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