



TAIL GUT CYST- A RARE FINDING- CASE REPORT AND REVIEW OF LITERATURE

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ABSTRACT

Introduction: Tailgut cysts are rare congenital entity that arises from the embryological hindgut and usually located in presacral space. Here we report a case of tail gut cyst which is an infrequent condition.

Case report: Here, we report a nineteen year old male with lower abdominal pain. The relevant investigations were performed. The CT abdomen revealed a well-defined cystic lesion in the pelvis in lower presacral region and posterior to rectum with small calcified foci suggestive of tail gut cyst with scalloping of sacrum. MRI pelvis suggested large well defined unilocular cystic lesion in the retro-rectal and pre-sacral region extending from S2 to mid coccygeal level showing thin imperceptible walls few tiny projections and small rounded non enhancing nodule along its posterior wall with thin focal calcification seen in its wall with no evidence of malignancy and no signs of osseous destruction. The patient was explored and intraoperatively findings revealed a cyst of size 8 cm * 5 cm behind the rectum and in front of sacrum which was densely adhered to the posterior wall of rectum but free from sacrum. The mass was resected as a whole and sent for histopathological exam which was suggestive of tailgut cyst. **Conclusion:** Tailgut cysts is infrequent condition which manifests as chronic lower abdominal pain. It is crucial that such entity be diagnosed and surgically excised early in its presentation. It is important to rule out malignancy in the post operative biopsy.

KEYWORDS : Tailgut cyst; Presacral tumours; Retrorectal space

INTRODUCTION

Presacral tailgut cysts also known as retrorectal cystic hamartomas are very infrequent entity which are considered to originate from remnants of the hindgut during development of embryo^(1,2).

The usual location of tail gut cyst is in presacral space, anteriorly close to the rectum, posteriorly bound by the sacrum, superiorly covered by the peritoneal reflection, inferiorly bound by the levator ani and pelvic floor, and laterally related to the iliac vessels and ureters^(3,4). These cyst can be located in the unusually in the prerectal, perianal, and perirenal regions^(5,7). This entity is difficult to diagnose but should be diagnosed and treated early in the course.^(5, 8-12) The preferred mode of treatment is to surgically excise the. Cyst due to its malignant potential, secondary infection and perianal fistula formation.

Case Report

We present a case 19 year male who came to the hospital with the complaints of pain in the lower abdomen since 2 weeks which was insidious in onset and dull aching in character. There were no complaints of bowel or bladder disturbances.

Ultrasonography revealed a well defined cystic Lesion measuring 5.9cm *6.7 cm *6.9 cm seen in midline in posterior aspect of urinary bladder in close relation to prostate showing echogenic debris within it and eccentric calcification foci with no vascularity seen in it.

Contrast enhanced CT abdomen(Fig 1) showed well defined hypodense /cystic lesion in the pelvis in lower presacral region and posterior to rectum with small calcified foci suggestive of tail gut cyst with scalloping of sacrum. MRI pelvis (Fig 2) suggested large well defined unilocular cystic lesion in the

retro-rectal and pre-sacral region extending from S2 to mid coccygeal level showing thin imperceptible walls few tiny projections and small rounded non enhancing nodule along its posterior wall with thin focal calcification seen in its wall Tailgut Cyst with no evidence of malignancy and no signs of osseous destruction. The physical exam showed a normal anal tone with no intraluminal mass but revealed a fullness in the posterior rectal wall

The intraoperative findings revealed a cyst of size 8 cm * 5 cm behind the rectum and in front of sacrum which was densely adhered to the posterior wall of rectum but free from sacrum. (Figure 3). The mass was resected as a whole and sent for histopathological exam suggestive of tailgut cyst. (Figure 4)

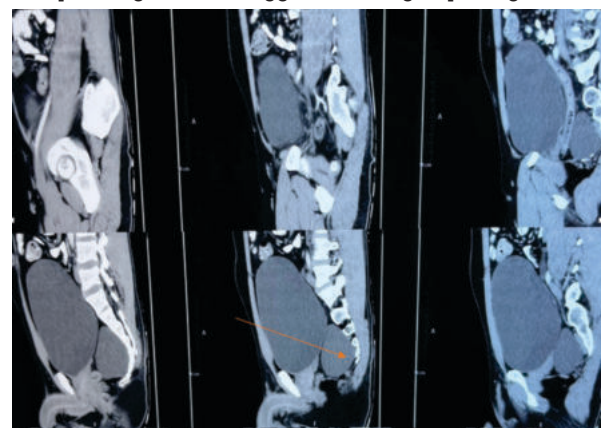


Figure 1- Coronal section of contrast enhanced CT Abdomen showing (Orange arrow)well defined hypodense /cystic lesion in the pelvis in lower presacral region and posterior to rectum with small calcified foci with scalloping of sacrum

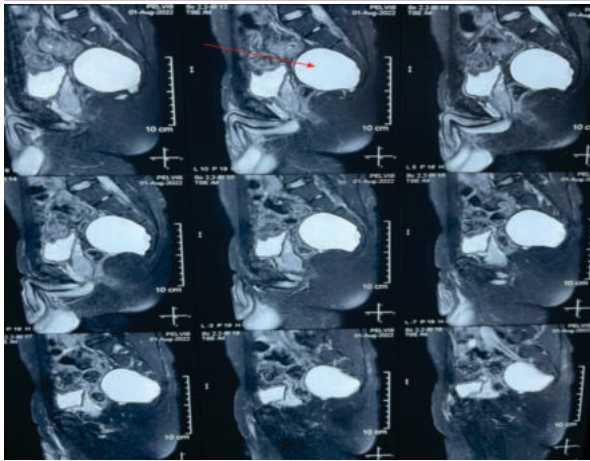


Figure 2 – MRI abdomen showing (Red arrow) large well defined unilocular cystic lesion in the retro-rectal and pre-sacral region extending from S2 to mid coccygeal level showing thin imperceptible walls few tiny projections and small rounded non enhancing nodule along its posterior wall with thin focal calcification

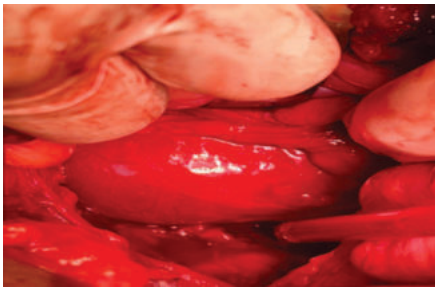


Figure 3- Intraoperative finding of cystic mass of 8cm * 5 cm behind the rectum and anterior to sacrum

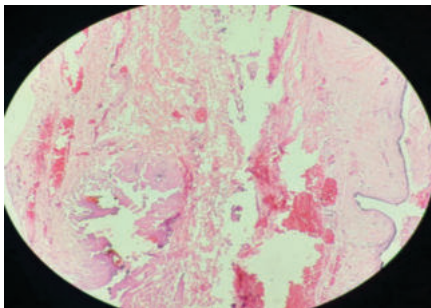


Figure 4- Histopathological findings showing cyst lined with pseudostratified ciliated epithelium with wall composed of collagen and muscle tissue with congested blood vessels with no evidence of dysplasia or malignancy

DISCUSSION

The retro rectal lesions are less common entity in adults and the incidence from patients of Mayo Clinic have been found to be 1 in 40,000^[8]. Out of all retro rectal tumours, tailgut cysts arise from failure to obliterate the embryological remnant of hindgut^[9].

Tailgut cysts are uncommon but more predominant in female population. Patsouras and colleagues found the female to male ratio to be 7 : 1. Tailgut cysts are prevalent mostly in the fifth decade of life; however, they can occur at any age.^[10-13] The signs and symptoms of the tailgut cysts are often nonspecific and mostly diagnosed in the radiological investigations^[13]

Most common presenting symptoms are rectal pain, constipation, tenesmus, painless per rectal bleeding, dysuria, lower abdominal pain, back pain, and neurological compressive symptoms due to pressure on the sacral plexus.

Unusual symptoms are recurrent perianal infections, perineal pain, perianal swelling and fistula, and sacrococcygeal sinus.^[11,13]

The gold standard diagnostic tool to detect the tailgut cyst is pelvic MRI which can find unilocular, multilocular or septated cyst^[14]. A typical tailgut cyst on T1 weighted MRI has low signal intensity and on T2-weighted images has high signal intensity. In a few cases due to mucinous material, high protein content or blood, T1 weighted high signal intensity images can be seen^[15-17]. MRI is gold standard for the surgical planning and also differentiates between benign and malignant tail gut cyst. A typical malignant hind gut cyst is heterogeneous with both solid and cystic content and irregular borders.

The preoperative biopsy in tailgut cyst is controversial topic. Hall et al.^[18] have found that ultrasound-guided needle biopsy can be used for preoperative diagnostic modality for tailgut cysts but this can cause seeding of tumour cells.^[19] The cysts with mixed component can be biopsied by percutaneous Para sacral approach so as to plan the surgery accordingly, for prognosis of the disease and requirement of chemotherapy^[12].

Tailgut cysts are usually lined with different types of epithelium like cuboidal, columnar within same cyst hence have high preponderance for malignant transformation commonly adenocarcinoma and carcinoid tumours types. However, other variety of tumours such as neuroendocrine cancer, endometrioid cancer, adenosquamous carcinoma, squamous cell carcinoma, and sarcoma can also be found.^[11,19,20] There are different studies showing rates of malignant transformation in tailgut which in the largest series in the literature in 1988 was 2%.^[10] According to Patsouras et al. the malignant transformation rate was 6% and other studies found the rate to be 13 to 40%.^[12-14]

There are other entities like neurogenic cyst, sacral chordoma, leiomyosarcoma, cystic lymphangioma, pyogenic abscess, dermoid, epidermoid cyst, rectal duplication cyst, neuroenteric cyst, teratoma, anterior sacral meningocele which needs to be differentiated from tailgut cyst.^[19] The differential diagnosis for prerectal cysts can be simple cyst of the seminal vesicle, bladder diverticulum, prostatic utricle, simple cyst of the prostate, rectal duplication cyst.^[21]

The surgical approach is planned according to the proximal extension of the cyst, size, infective component, proximity with important structures like bladder, uterus and rectum and presence of malignancy which involves en bloc resection.^[12,13] The most commonly used approach is the posterior approach.^[22,23] The other approaches commonly used for the tailgut cyst are the anterior approach or laparoscopic approach. In case of prerectal cysts meticulous dissection is needed to prevent trauma to adjacent structures including the vagina, uterus, bladder, and rectum.

McCarroll and Moore demonstrated^[24] transanal minimally invasive surgery technique for tailgut cyst surgery. Kildušis and Samalavičius^[25] managed a case with open transrectal approach. There are several studies which demonstrate removal of coccyx so as to prevent nidus for recurrence.^[4,26] There are several research evidences which do not recommend removal of coccyx and recommend preservation of the coccyx until it has to be removed in case of en bloc resection for malignant cases.^[27] The avoidance of injury to the rectal wall is important and is facilitated by packing rectum with soft paraffin gauze so as to identify intraoperatively along with preoperative adequate bowel preparation.

The recurrence rates are in range of 0%–16%, mostly due to incomplete excision.^[13] A regular follow up is suggested for a patient with typical symptoms and low malignancy risk. An

exact diagnosis cannot be obtained till surgical removal and histopathological analysis.^[28]

CONCLUSION

Surgical excision is mandatory regardless of symptom status because of the risk of malignancy. Complete surgical excision is gold standard treatment which can be tailored according to the patient and the surgeon's preference.

Ethical Approval

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Conflict of Interest

The authors report no potential conflicts of interest relevant to this article.

REFERENCES:

- Vega Menéndez D, Quintáns Rodríguez A, Hernández Granados P, Nevado Santos M, García Sabrido JL, Rueda Orgaz JA, et al. Tailgut cysts. *Cir Esp* 2008;83:53-60.
- Dahan H, Arrivé L, Wendum D, Docou le Pointe H, Djouhri H, Tubiana JM. Retrorectal developmental cysts in adults: clinical and radiologic-histopathologic review, differential diagnosis, and treatment. *Radiographics* 2001;21:575-84.
- Lev-Chelouche D, Gutman M, Goldman G, Even-Sapir E, Meller I, Issakov J, et al. Presacral tumors: a practical classification and treatment of a unique and heterogeneous group of diseases. *Surgery* 2003;133:473-8.
- Wolpert A, Beer-Gabel M, Lifschitz O, Zbar AP. The management of presacral masses in the adult. *Tech Coloproctol* 2002;6:43-9.
- Gips M, Melki Y, Wolloch Y. Cysts of the tailgut. Two cases. *Eur J Surg* 1994;160:459-60.
- Sidoni A, Bucciarelli E. Ciliated cyst of the perineal skin. *Am J Dermatopathol* 1997;19:93-6.
- Sung MT, Ko SF, Niu CK, Hsieh CS, Huang HY. Perirenal tailgut cyst (cystic hamartoma). *J Pediatr Surg* 2003;38:1404-6.
- Jao SW, Bear RW Jr, Spencer RJ, Reiman HM, Ilstrup DM. Retrorectal tumors. Mayo Clinic experience, 1960-1979. *Dis Colon Rectum* 1985;28:644-52.
- Prasad AR, Amin MB, Randolph TL, Lee CS, Ma CK. Retrorectal cystic hamartoma: report of 5 cases with malignancy arising in 2. *Arch Pathol Lab Med* 2000;124:725-9.
- Killingsworth C, Gadacz TR. Tailgut cyst (retrorectal cystic hamartoma): report of a case and review of the literature. *Am Surg* 2005;71:666-73.
- Hjermstad BM, Helwig EB. Tailgut cysts. Report of 53 cases. *Am J Clin Pathol* 1988;89:139-47.
- Mathis KL, Dozois EJ, Grewal MS, Metzger P, Larson DW, Devine RM. Malignant risk and surgical outcomes of presacral tailgut cysts. *Br J Surg* 2010;97:575-9.
- Patsouras D, Pawa N, Osmani H, Phillips RK. Management of tailgut cysts in a tertiary referral centre: a 10-year experience. *Colorectal Dis* 2015;17:724-9.
- Yang DM, Park CH, Jin W, Chang SK, Kim JE, Choi SJ, et al. Tailgut cyst: MRI evaluation. *AJR Am J Roentgenol* 2005;184:1519-23.
- Kim MJ, Kim WH, Kim NK, Yun MJ, Park YN, Lee JT, et al. Tailgut cyst: multilocular cystic appearance on MRI. *J Comput Assist Tomogr* 1997;21:731-2.
- Lim KE, Hsu WC, Wang CR. Tailgut cyst with malignancy: MR imaging findings. *AJR Am J Roentgenol* 1998;170:1488-90.
- Moulopoulos LA, Karvouni E, Kehagias D, Dimopoulos MA, Gouliamos A, Vlahos L. MR imaging of complex tail-gut cysts. *Clin Radiol* 1999;54:118-22.
- Hall DA, Pu RT, Pang Y. Diagnosis of foregut and tailgut cysts by endosonographically guided fine-needle aspiration. *Diagn Cytopathol* 2007;35:43-6.
- Gönül İI, Bağlan T, Pala I, Menteş B. Tailgut cysts: diagnostic challenge for both pathologists and clinicians. *Int J Colorectal Dis* 2007;22:1283-5.
- Song DE, Park JK, Hur B, Ro JY. Carcinoid tumor arising in a tailgut cyst of the anorectal junction with distant metastasis: a case report and review of the literature. *Arch Pathol Lab Med* 2004;128:578-80.
- Jang SH, Jang KS, Song YS, Min KW, Han HX, Lee KG, et al. Unusual prerectal location of a tailgut cyst: a case report. *World J Gastroenterol* 2006;12:5081-3.
- Hannon J, Subramony C, Scott-Conner CE. Benign retrorectal tumors in adults: the choice of operative approach. *Am Surg* 1994;60:267-72.
- Singer MA, Cintron JR, Martz JE, Schoetz DJ, Abcarian H. Retrorectal cyst: a rare tumor frequently misdiagnosed. *J Am Coll Surg* 2003;196:880-6.
- McCarroll RH, Moore LJ. Transanal minimally invasive surgery for resection of retrorectal cyst. *J Surg Case Rep* 2018;2018:rjy021.
- Kildušis E, Samalavičius NE. Surgical management of a retro-rectal cystic hamartoma (tailgut cyst) using a trans-rectal approach: a case report and review of the literature. *J Med Case Rep* 2014;8:11.
- Izant RJ Jr, Filston HC. Sacrococcygeal teratomas. Analysis of forty-three cases. *Am J Surg* 1975;130:617-21.
- Böhm B, Milsom JW, Fazio VW, Lavery IC, Church JM, Oakley JR. Our approach to the management of congenital presacral tumors in adults. *Int J Colorectal Dis* 1993;8:134-8.
- Au E, Anderson O, Morgan B, Alarcon L, George ML. Tailgut cysts: report of two cases. *Int J Colorectal Dis* 2009;24:345-50.