



Case Report

Retrorectal Cystic Hamartoma: Case Report

Marwah Sami M Hussain¹, Feras Alsannaa², Nahla Arab^{3*}

¹General Surgery Registrar, Department of General Surgery, Prince Sultan Military Medical City, Kingdom of Saudi Arabia

²Consultant General Surgeon, Department of General Surgery, Prince Sultan Military Medical City, Kingdom of Saudi Arabia

³Consultant Colorectal Surgeon, Department of General Surgery, Prince Sultan Military Medical City, Kingdom of Saudi Arabia

***Corresponding author:** Nahla Arab, Consultant Colorectal surgeon, Department of General Surgery, Prince Sultan Military Medical City, Riyadh, Kingdom of Saudi Arabia

Citation: Hussain MSM, Alsannaa F, Arab N (2020) Retrorectal Cystic Hamartoma: Case Report. Ann Case Report 14: 519. DOI: 10.29011/2574-7754.100519

Received Date: 06 November, 2020; **Accepted Date:** 13 November, 2020; **Published Date:** 19 November, 2020

Abstract

Introduction: The retrorectal space is a space with wide differential diagnosis: congenital cysts, primary tumors and metastasis. CT and MRI have become the best diagnostic modalities for retrorectal space lesions. Retrorectal cystic hamartoma is the remnants of the embryonic hindgut, rare condition, appear at any age, usually asymptomatic. Surgical resection is through standard Kraske or abdominal approach are the most commonly described.

Case Report: A 36-year-old male, presented with complain of progressing lower back pain for one-year associated with tenesmus and proctalgia during evacuations. PR: showed an extrinsic, nontender mass, on the posterior wall. Patient underwent a complete excision of presacral cyst and coccygectomy through posterior approach.

Conclusion: Retrorectal hamartoma is a rare condition but should be considered as a possible differential in any case of a perirectal cyst, irrespective of age. Complete surgical excision is indicated when diagnosed.

Keywords: Case report; Presacral tumors; Retrorectal tumor; Retrorectal cystic hamartomas, Tailgut cyst

Introduction

The retrorectal space is a space with wide differential diagnosis: congenital cysts, primary tumors and metastasis. CT and MRI have become the best diagnostic modalities for retrorectal space lesions. Retrorectal cystic hamartoma is the remnants of the embryonic hindgut, rare condition, appear at any age, usually asymptomatic. Surgical resection is through standard Kraske or abdominal approach are the most commonly described.

Case Report

This is a 36-year-old male, medically free who was presented to our center with complain of lower back pain for one-year progress in the last 3 months, associated with tenesmus and proctalgia during evacuations. He had a history of constipation, no history of per rectum bleeding or urinary complaints. No history of fever, No history of documented weight loss. General physical examination revealed no abnormal findings. Digital rectum examination showed an extrinsic, non-tender mass, on the posterior wall with smooth mobile rectal mucosa over it. Routine

laboratory investigations were within the normal range. Abdomen and pelvic CT with rectal contrast (Figure 1) showed: 3.2 x 3.2 x 5 cm well define rounded cystic lesion at the right ischia-anal fossa, pushing the rectum. There is no clear communication between the cyst and the rectum. Pelvic MRI (Figure 2) showed: the cystic lesion is included in the rectoanal fascia, inseparable from the coccyges.

The patient was undergoing general anesthesia in a prone jackknife position, complete excision of the presacral cyst, and coccygectomy through the posterior (Kraske) approach. An incision over the lower portion of the sacrum and coccyx down to the anus, lone star retractor was applied, the anococcygeal ligament was transected and the levator ani muscles were retracted laterally (Figure 3). To provide good exposure, excision of the coccyx was necessary. The cyst was retrieved completely (Figure 4). Adequate reconstruction of the perineum, suction drain inserted, and wound closure. The patient postop has uneventful recovery and was discharged in 5th day with regular follow up for the wound and histopathology which came as retrorectal cystic hamartoma (Tailgut cyst), Foci of benign mucinous and serous acini are seen. Negative for malignancy. Follow up in the clinic after 6 months, the patient was asymptomatic with no signs of recurrence in follow-up pelvic MRI.

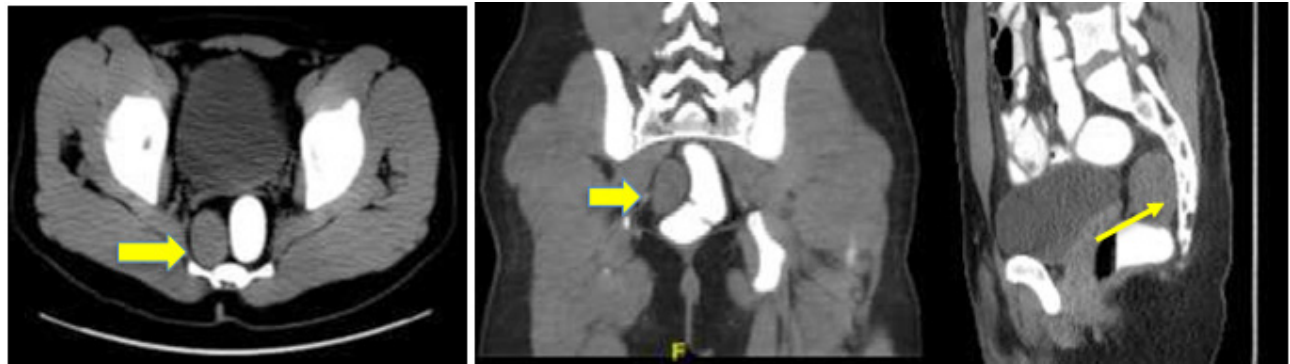


Figure 1: There is 3.2 x 3.2 x 5 cm well define rounded cystic lesion (thick arrow) seen at the right ischia-anal fossa. It pushed the contrast filled rectum to the left side. There is no clear communication between the cyst and the rectum. No clear fat plan between the cyst and coccygeal (narrow arrow).

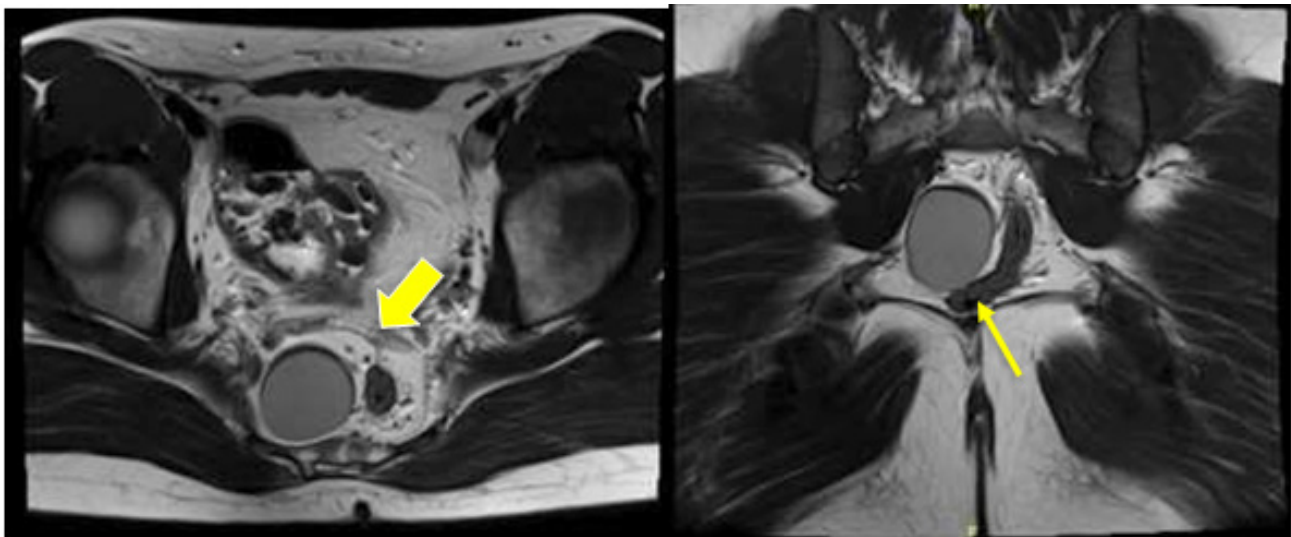


Figure 2: MRI pelvis (T2WI) A- axial, B- coronal, the cyst shows intermediate high signal intensity. The lesion is clearly included in the recto anal fascia (thick arrow) and inseparable from the rectum and show stalk (narrow arrow).



Figure 3: Intraoperative picture, Posterior /Kraske procedure.



Figure 4: Unilocular presacral cyst, smooth surface measuring 5 x 4 x 3 cm.

Discussion

The retro-rectal space is a potential space bounded superiorly by the peritoneal reflection, anteriorly by the rectum, posteriorly by the sacrum, and inferiorly by coccygeus muscle and the levatorani. A great diversity of differential diagnosis of lesions in retrorectal space can be, due to the presence of multiple embryologic remnants and miscellany of tissue types within this space. Including congenital cysts (rectal duplication cyst, cystic teratoma, dermoid, and epidermoid cyst), primary tumors of neurogenic and osteogenic, metastasis, and inflammatory processes [1,2]. Retrorectal cystic hamartoma (Tailgut cyst) arises from the remnants of the embryonic hindgut, which normally regresses during the seventh or eighth week of the embryonic phase [3]. It is more common lesions in female, children and young adults, however, can be detected at any age, including infancy. Hamartoma is slowly growing, usually asymptomatic; but symptoms result from mass effect (fulness, back pain, and tenesmus) or secondary infection (perianal pain and rectal discharge).

Our patient was a young male. He was denying any complain before one-year, when lower back pain started, associated with tenesmus and proctalgia during evacuations. No history of perianal producers or discharge. The diagnosis is based on radiological investigations. A biopsy is not necessary for the diagnosis, because of a high risk of its local dissemination, possible complication, and specimens often had low diagnostic value (not decisively). The radiological modalities: Transrectal US is not usually requested, may show a multilocular cystic mass with internal echoes from gelatinous material or inflammatory debris. Computed Tomography (CT) features are a well-marginated, presacral mass with water or soft-tissue density and the relation to the rectum. If infection or malignant transformation occurs, CT may show a loss of discrete margins and involvement of adjacent structures [4].

Magnetic resonance has evolved to be the cornerstone, and determine the anatomic details its relation to the adjacent structures and deciding the best surgical approach, features are hypointense in T1 and hyperintense in T2 if uncomplicated cysts (without infection, inflammation, or malignant change) [5]. The radiological findings in our presented case were typical findings of uncomplicated cyst in CT with water density, pushing the rectum. Moreover, in MRI was hypointense in T1 and moderate hyperintense in T2. Complete surgical resection of the lesion is the best therapeutic option once the diagnosis is established, even in asymptomatic patients to avoid the potential for symptoms, recurrence, infection, or rare malignant degeneration as adenocarcinoma and carcinoid [6]. However, determining the most appropriate approach is challenging: abdominal, perineal, trans-anal, or combined approaches.

A posterior, trans-sacral procedure (Kraske) provides excellent surgical access and permit complete excision and coccygectomy if needed for good exposure or complete excision of a potential communication route and consequent recurrence. The major disadvantages that should be consider for the posterior approach are the potential for injury to the lateral pelvic nerves, and the absence of control over pelvic vessels [7-9]. As illustrated in our report: we approached our patient by posterior procedure due to low position of the cyst lead to easy access and the inseparable from the rectum found in MRI which require a delict dissection and careful examination to rectum. We performed coccygectomy to ensure a complete excision especially for the loss of fat plan between the cyst and coccygeal in CT scan. The abdominal approach, either laparotomy or laparoscopy, is useful if the mass is upward located in the abdomen cavity (above the S4), and achieve an excellent exposure of pelvic structures, iliac vessels, and ureters.

Laparoscopy gives the advantage of a fine mesorectum dissection, low morbidity, less post-operative pain, shorter hospital stays, less blood loss, faster recovery time, fewer complications, and better cosmesis [10]. The combined approach is the procedure of choice for larger lesions that extend above and below S4, beginning with a modified lithotomy position, then repositioned in the jacke-knife or left in the modified lithotomy position. The endoanal approach is of high cost and difficult learning curves [11]. The TAMIS (Transanal Minimally Invasive Surgery) reliable alternative to endoanal at a lower cost.

Conclusion

Retrorectal hamartoma is a rare condition but should be considered as a possible differential in any case of a perirectal cyst, irrespective of age. Complete surgical excision is indicated when diagnosed.

Conflicts of Interest

There were no conflicts of interesting in writing of this case report.

References

1. Mills SE, Walker AN, Stallings RG, Allen MS Jr (1984) Retrorectal cystic hamartoma. Report of three cases, including one with a perirenal component. Archives of pathology & laboratory medicine 108: 737-740.
2. Prasad AR, Amin MB, Randolph TL, Lee CS, Ma CK (2000) Retrorectal cystic hamartoma: report of 5 cases with malignancy arising in 2. Archives of pathology & laboratory medicine 124: 725-729.
3. Johnson AR, Ros PR, Hjermstad BM (1986) Tailgut cyst: diagnosis with CT and sonography. AJR American journal of roentgenology 147: 1309-1311.
4. Peter P, George U, Peacock M (2010) Retrorectal hamartoma: A 'tail' of two cysts! The Indian journal of radiology & imaging 20: 129-131.
5. Mouloupoulos LA, Karvouni E, Kehagias D, Dimopoulos MA, Gouliamos A, et al. (1999) MR imaging of complex tail-gut cysts. Clinical radiology 54: 118-122.
6. Piura B, Rabinovich A, Sinelnikov I, Delgado B (2005) Tailgut cyst initially misdiagnosed as ovarian tumor. Archives of gynecology and obstetrics 272: 301-303.
7. Hobson KG, Ghaemmaghami V, Roe JP, Goodnight JE, Khatri VP (2005) Tumors of the retrorectal space. Diseases of the colon and rectum 48: 1964-1974.
8. Buchs N, Taylor S, Roche B (2007) The posterior approach for low retrorectal tumors in adults. International journal of colorectal disease 22: 381-385.
9. Ghosh J, Eglinton T, Frizelle FA, Watson AJ (2007) Presacral tumours in adults. The surgeon: journal of the Royal Colleges of Surgeons of Edinburgh and Ireland 5: 31-38.
10. Udwadia TE (2011) Single-incision laparoscopic surgery: An overview. Journal of minimal access surgery 7: 1-2.
11. Van den Boezem PB, Kruyt PM, Stommel MW, Tobon Morales R, Cuesta MA, et al. (2011) Transanal single-port surgery for the resection of large polyps. Digestive surgery 28: 412-416.