Radiology Case Reports 17 (2022) 2559–2562

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journal homepage: www.elsevier.com/locate/radcr

Case Report

Recurrent retrorectal tailgut cyst mimicking deep pelvic abscess: A diagnostic dilemma☆

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ARTICLE INFO

Article history:
Received 20 March 2022
Revised 21 April 2022
Accepted 24 April 2022

Keywords:
Tailgut cyst
Pelvic abscess
Presacral tumor
Retrorectal cyst
Case report

ABSTRACT

Tailgut cysts are congenital anomalies that are rare and arise from incompletely regressed primitive hindgut. These are more commonly found in women and are usually asymptomatic. When symptoms develop, these can present with pain, infection, hemorrhage, difficulty in defecation, and rarely malignant change. We report a middle-aged married woman who presented with deep-seated perineal pain for a couple of months, which increased during defecation and sexual intercourse. Although abdominal examination was unremarkable barring deep tenderness in the hypogastrium, rectal and vaginal examinations suggested a tender pelvic swelling. An abdominal ultrasonographic examination diagnosed a cystic swelling in the pelvis extending until the Levator ani muscles. Considering her symptoms, a pelvic abscess was diagnosed and transvaginal drainage was done. Due to persistence of symptoms and recurrence after a month, she was further investigated and was diagnosed to have a presacral benign cystic tumor based on CT and MRI scans of the pelvis. The lesion was completely excised through a combined abdomino-perineal approach and histopathological report suggested a benign tailgut cyst. That a cystic presacral swelling with features of inflammation can be confused with a deep pelvic abscess is hereby highlighted in this report. An MRI scan is diagnostic of these lesions. Failure to differentiate it from a pelvic abscess may result in drainage, which may be of concern if the lesion is malignant.

☆ Competing interests: The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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https://doi.org/10.1016/j.radcr.2022.04.041

Introduction

Tailgut cysts are rare congenital malformations occurring in the presacral space [1]. The presacral space is bound by the fascia propria of the rectum anteriorly, the presacral fascia posteriorly, peritoneal reflection superiorly, rectosacral fascia inferiorly, and the internal iliac vessels, sacral nerve roots, pyriformis muscle, and ureters laterally [2,3]. Primitive endodermal remnants from the primitive hindgut beyond the cloacal membrane, which fail to regress in the embryonic life, form tailgut cysts [4].

It usually manifests in the middle age and female: male ratio is 5:1 [1]. Asymptomatic cases are often missed on rectal examination due to small size and deep location [1]. Symptoms are due to mass effect on adjacent organs such as polyuria, dysuria, pain on defecation and intercourse, rectal fullness, constipation, and low back ache. Long standing cases may rarely develop malignancy [2,5]. If infected, these may mimic pelvic abscess. We present a case of an infected tailgut cyst that was mistaken as a pelvic abscess and was drained transvaginally but recurred again, which was excised through a combined abdomino-perineal approach. The diagnostic dilemma in such cases especially when dealing with features of infection is highlighted and its management is discussed.

Case report

A 43-year-old woman presented with complaints of constant dull pain in the perianal region and buttocks for 2 months that aggravated during defecation and sexual intercourse. It increased in the next 7 days and was associated with dysuria. She denied any history of fever, vomiting, abdominal distension or constipation, melena, or foul-smelling discharge per rectum. Her menstrual history was normal without any history of menorrhagia or dysmenorrhea. She was initially managed with broad spectrum antibiotics but symptoms did not abate completely after 5 days of therapy. A gynecological consultation and an ultrasound (USG) examination of pelvis diagnosed her having a deep pelvic abscess. It was drained through the transvaginal route under real time USG guidance and antibiotic cover. But her symptoms did not abate and the swelling recurred within a month when a surgical consultation was taken for a probable pelvic tumor.

On examination, her vital parameters were stable. Deep tenderness was present in the lower abdomen without any muscle guard or palpable lump. Perineal examination was unremarkable. A digital rectal examination revealed a tender palpable fullness posteriorly in the mid-and lower rectum. Vaginal examination revealed fullness of posterior fornix and a tender cervix. Her hematological, biochemical and urine investigations were unremarkable. An USG examination revealed a well-defined cystic lesion with few internal echoes in the pouch of Douglas extending to the right and posterior to rectum. USG-guided aspiration revealed 200 ml of thick yellowish fluid that was sterile on culture. A contrast-enhanced CT-scan of the abdomen and pelvis showed a large multiloculated cystic lesion of size 11.3 × 12.8 × 15.7 cm in the presacral space (Figs. 1A and B). It was extending up to the level of L5-S1 intervertebral disc and was displacing bowel loops superiorly and the base of urinary bladder and cervix upward and anteriorly. An MRI of the pelvis revealed a multiloculated cystic lesion (11 × 13 × 16 cm^3) in the presacral space (Figs. 2A and B). It appeared hypointense on T1 and hyperintense in T2 with few T1 hyper- and T2 hypointense loculi and was extending superiorly until S1 vertebra and inferiorly into right ischiorectal fossa and abutting lower rectum, right puborectalis muscle and external anal sphincter, and was in close proximity to sacrum, coccyx, pyriformis muscle and the internal iliac vessels postero-laterally on the right side. A diagnosis of either a tailgut cyst or a neuro-enteric cyst was suggested, although chronic pelvic abscess, endometrioma, dermoid cyst, and tumors of genitourinary tract were also considered.

Before surgical exploration, an initial diagnostic laparoscopy was done, which was unremarkable. Abdominal exploration was done through a lower midline laparotomy, which revealed a 10 x 12 cm cystic lump in the presacral area. It was densely adhered to the vaginal and rectal walls anteriorly, pelvic wall and iliac vessels laterally and the sacrum posteriorly. The cyst extended through the Levator ani hiatus into the right ischiorectal space. Meticulous dissection was done to prevent damage to important pelvic viscera and nerves. Excision of the cyst was finally completed through a combined abdomino-perineal approach (Fig. 3). Histological examination proved it to be a tail-gut cyst with no evidence of malignancy. Her recovery was uneventful and she was discharged on the sixth postoperative day. She followed up after 2 weeks and then 3 monthly, and has not developed any recurrence of symptoms or disease in 1 year.

Discussion

In embryonic life, the hindgut grows into a primitive tail distal to the cloacal membrane at sixth week of gestation. This endodermal extension, known as the postanal gut or tailgut, completely disappears by 8 weeks [5]. Its persistence leads to tailgut cyst formation. The true incidence is not well known as only few large series exist. However, according to Mayo Clinic data, it is likely at 1 in 40,000 population [5]. Most of these presacral cystic tumors are asymptomatic due to the deep location and slow growing nature. However, when these present with symptoms, the clinical diagnosis is often confused with a number of other conditions such as chronic pelvic abscess, endometrioma, dermoid cyst, epidermoid cyst, chordoma, and teratoma [1,4,6]. Only a histological examination can confirm its nature.

MRI and CT-scan are cornerstones of diagnosis. The tailgut cyst is usually seen as a multilocular cyst in the presacral space or a large cystic mass associated with smaller peripherally located cysts that shows low signal intensity on MRI T1-weighted images and high signal intensity on T2-weighted images [7,8], although findings may vary depending on its content. In the present case, imaging showed a well-defined multiloculated cyst lesion in the presacral region. CT scan showed no calcification/ fat density within the lesion. Further, no bony

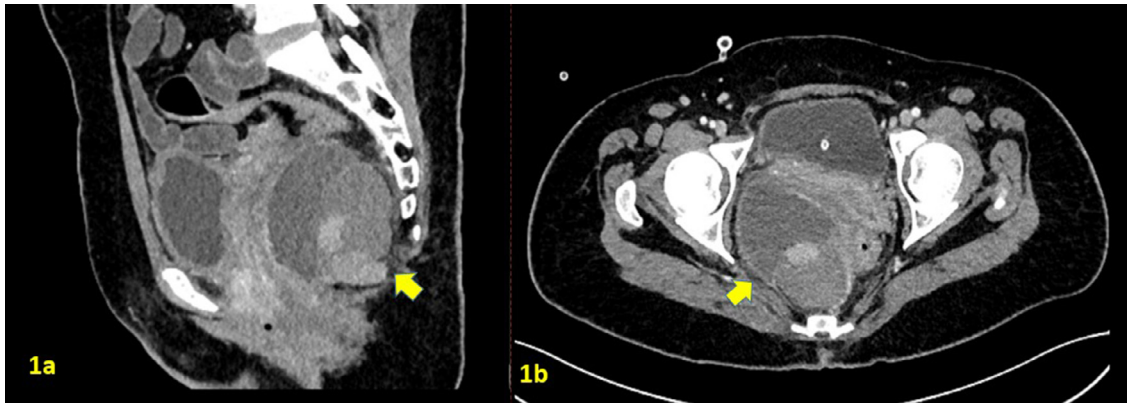

Fig. 1 – Contrast-enhanced CT-scan pelvis – (A) sagittal section showing multiloculated presacral cystic lesion (yellow arrow) abutting and displacing the cervix anteriorly, and (B) axial section showing displacement of rectal anteriorly and to the left.

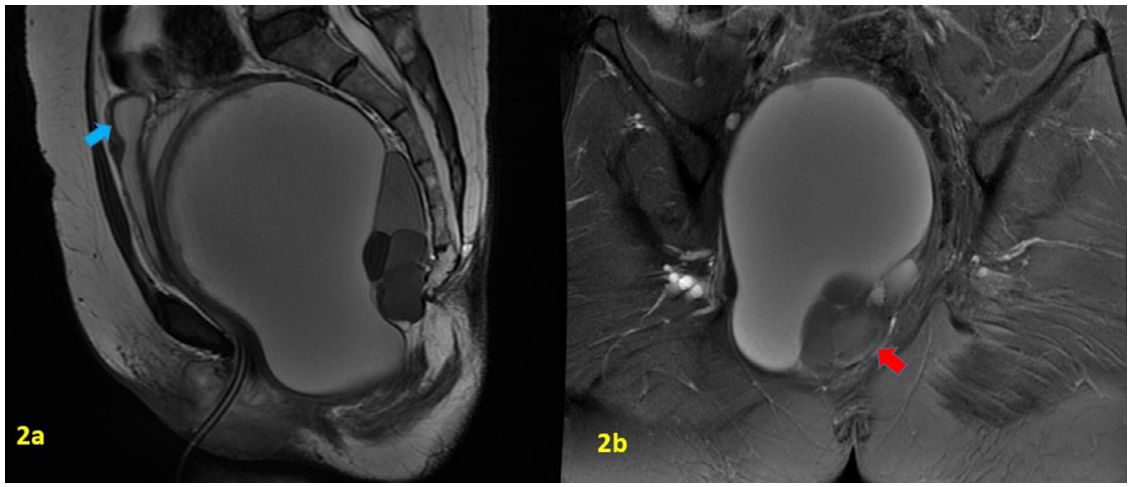

Fig. 2 – MRI pelvis – (A) sagittal section-T2 sequence showing multiloculated presacral cystic lesion with variable intensity in the loculi, displacing the urinary bladder anteriorly and upwards (blue arrow), (B) axial section showing displacement of the rectum towards left side (red arrow).

defect was seen in the sacrum. The MRI showed locules with variable signal intensity, largest locule showed T1 hypointense and T2 hyperintense signal. The lesion showed no significant restriction or blooming. No communication with the neural foramina was observed. The imaging differential diagnoses of presacral cystic space occupying lesion usually includes dermoid cyst, anterior sacral meningocele, ischio-rectal abscess, and epidermoid cyst. Dermoid cyst is a teratoma of cystic nature containing mature cyst tissue component such as tufts of hair, cartilage or teeth along with areas of fat deposition and calcifications within the lesion. In the present case, the lack of calcification and fat made the diagnosis of dermoid cyst less likely. Anterior sacral meningocele is a meningeal cyst in presacral region with congenital defect in anterior aspect of sacrum. CT bone window shows sacral defect on anterior aspect, which was not present in our case. Ischiorectal abscess is a close differential diagnosis. An ischiorectal abscess is usually irregular in outline with thick enhancing wall. On MRI it shows restricted diffusion and peri-focal inflammatory changes. An ultrasonological examination may not be adequate to differentiate it from an abscess, which happened in the present case and prompted a transvaginal drainage. Epidermoid cyst has low signal intensity on T1 weighted image and high signal intensity with island of low intensities on T2 weighted image and usually shows diffusion restriction. In the present case, no significant diffusion restriction was observed. Although fine needle cytology or biopsy have been attempted in presacral cystic masses, complete surgical excision with histopathological examination is usually recommended for its comprehensive management because needle biopsy carries a chance for spillage of cells in the pelvic cavity leading to a probable recurrence, or seeding of biopsy tract in cases of malignancy [9].

A number of surgical approaches ranging from transabdominal (laparotomy or laparoscopic), trans-sacral, intersphincteric, trans-sphincteric, parasacrococcygeal, or a combined abdomino-perineal approach have been described [2]. Transabdominal approach is preferred for higher lesions, and if a malignancy is suspected. The laparoscopic approach is less invasive and reduces risk of surgical site infection. Resection

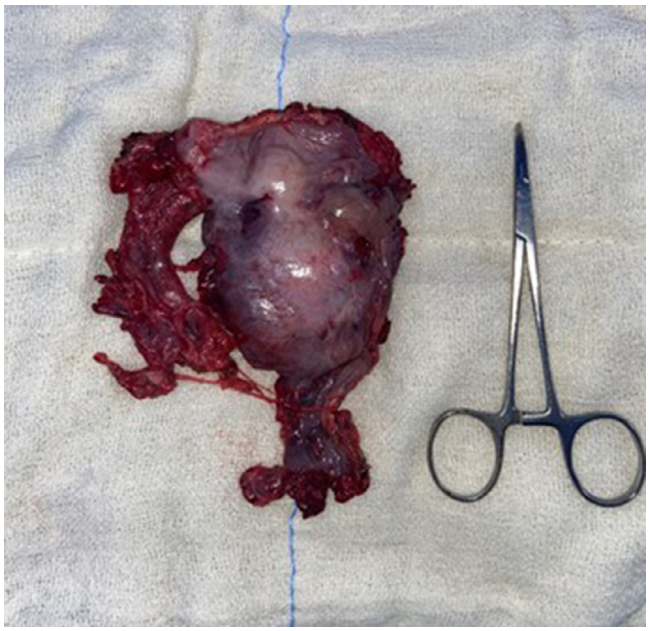

Fig. 3 – Resected specimen of the presacral cystic lesion (after drainage of content).

is indicated in all cases due to the chance of future complications and rarely malignant changes [2,4]. For benign cases, complete excision is curative. Recurrence may occur after incomplete excision, which makes subsequent surgeries more difficult and thus increases morbidity. In the malignant lesions, wide excision and at times pelvic exenteration, are indicated for cure [9,10].

Conclusion

Tailgut cyst, although rare, should be considered as a differential diagnosis of presacral cystic masses. CT/MRI scans are needed to establish the diagnosis and guide necessary treatment. Management should not be planned solely based on USG findings. Every effort should be made to avoid diagnostic confusion with a pelvic abscess which might prompt the surgeon for a tube drainage that can be of great concern in presence of malignancy.

Patient consent

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

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