

Tailgut cyst: a case report in a female neonate

Abstract

Tailgut cyst, or retrorectal cystic hamartoma, is a rare congenital lesion found in the presacral space. The lesion has been infrequently reported in the literature. We report a 16days female neonate presented with urine retention.

Keywords: tailgut cyst, presacral space

Volume 2 Issue 2 - 2015

Alhasani AA, Shamaon RS

Basrah Medical College, Iraq

Correspondence: Alhasani AA, Basrah Medical College, Basrah, Iraq, Tel 00964 7722771445, Email abbasalhasani@yahoo.com

Received: August 20, 2014 | **Published:** April 18, 2015

Introduction

Tailgut cyst, or retrorectal cystic hamartoma, is an uncommon lesion found in the presacral space. The lesion is believed to arise from the embryonic postanal gut.¹⁻³ It is mostly found within the retrorectal or presacral space⁴ and rarely in the perirenal area.^{2,3,5} the subcutaneous tissue in the anorectal region⁶ and more rarely in the prerectal location. Microscopically, the tailgut cyst is characterized by the presence of a cysts lined with varying types of epithelium; columnar, mucin secreting epithelium predominates, while other areas of squamous and transitional epithelium often coexist.⁷ The differential diagnosis of cystic masses in this space includes sacrococcygeal teratoma, dermoid cysts and other complex ovarian masses, rectal duplication cysts, anterior meningoceles, cystic lymphangiomas and abscesses.^{8,9}

There is an approximately 3:1 female preponderance of cases reported.⁴ Uncomplicated lesions are typically asymptomatic, and patients usually come to medical attention when the cyst becomes infected or when the lesion is incidentally discovered either radiologically or during physical examination. Perineal pain, pain with defecation, painless rectal bleeding, urinary frequency, and change in stool caliber have been reported. A few rare cases of malignant change associated with a tailgut cyst have been reported, but none in children to our knowledge.^{4,10,11} Few cases have been reported in children. The largest series by Hjermsstad et al.⁴ at the Armed Forces Institute of Pathology describes 53cases of the entity collected during a 35-year period. Five cases demonstrating the CT and sonographic findings of tailgut cysts were described by Johnson et al.¹² at the Armed Forces Institute of Pathology. There are only a handful of reports of the MR findings of this entity in adults and only a few cases in the pediatric population.¹³

Case report

A 16days-old girl presented with acute attack of irritability and failure to pass urine over 10hours. She was born at term by normal vaginal delivery to a healthy mother and uneventful pregnancy and was well in the first few days of life. Careful clinical evaluation showed irritable neonate with moderately distended abdomen.

The patient underwent urinary catheterization to evacuate the urinary bladder, after which, she got dramatic symptomatic relief and a small palpable mass become readily palpable in the suprapubic area arising from the pelvis. Complete laboratory workup including haematological and biochemical tests were done and were within

the normal limits. Alfa fetoprotein was within normal limit as well. The patient then sent to abdominal and pelvic ultrasonography that showed a cystic lesion in the retrorectal space causing dilated urinary bladder and rectum, but normal proximal urinary tract. MRI was used to get more details and to exclude anterior sacral meningocele, sacrococcygeal teratoma, tethered spinal cord, or other occult abnormality. The lesion was a well-demarcated oval-shaped cyst which was located at the presacral and causing marked extra luminal pressure on both bladder neck and rectum and in turn resulted in a proximal dilatation of both organs.

After an adequate preparation, the patient underwent explorative laparotomy for the definite diagnosis and treatment. Intra-operative finding (Figure 1) showed that the cyst was unilocular and tense with marked thick wall dilated urinary bladder and rectum but was easily separated from them and from the sacrum, complete excision was performed via suprapubic transverse incision. Histopathologically, the fibrous cyst wall was lined by a single layer of mesothelial cells with smooth muscles and adipose tissue. Postoperatively, she got a smooth recovery time. Urinary catheter was removed 2days later and the patient was sent home on the 3rd postoperative day. The patient's recovery remained uneventful for 2years after the operation.

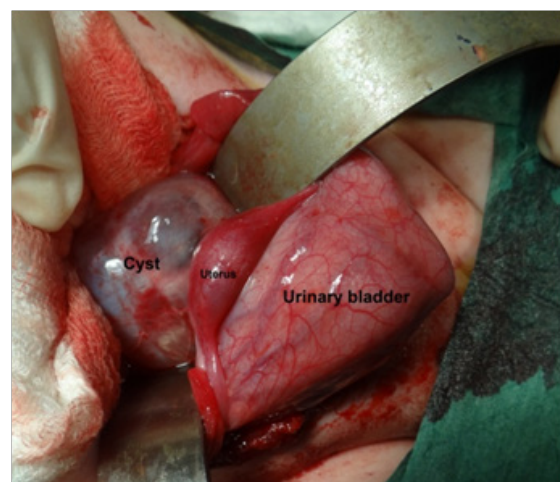


Figure 1 Intra-operative finding cyst.

Discussion

The retrorectal or presacral space is a potential space bounded by the rectum anteriorly and the sacrum and coccyx posteriorly. The presacral space is a region of complex embryology, thus leading to a wide range of potential pathologies including inflammatory, neoplastic, and congenital solid and cystic masses. Many terms exist in the surgical, pathological, and radiological literature to describe these lesions in the presacral space that are believed to be remnants of the post-anal gut. These terms include retrorectal cystic hamartoma,^{14,15} myoepithelial hamartoma of the rectum,¹³ and the preferred term, tailgut cyst.⁴ The first description of this lesion is given to Middeldorpf,¹⁶ who in 1885 described a female infant with a presacral tumor lined by epithelium.

Tailgut cysts are rare congenital lesions in the presacral space characterized by the presence of cystic lesion lined by various types of epithelium. In order to render the diagnosis, Hjermsstad et al.⁴ in their review required the presence of columnar or transitional epithelium to exclude epidermoid or dermoid cysts (which contain only squamous epithelium), and required the absence of myenteric plexus and serosa to exclude bowel duplication cysts. The lesion has been described in neonates as young as 4 days, and in adults through the eighth decade of life, with the average age of presentation in the fourth decade.⁴ Although a rare entity, tailgut cyst should be included in the differential diagnosis of a presacral cystic mass.

Acknowledgements

None.

Conflict of interest

The author declares no conflict of interest.

References

1. Killingsworth C, Gadacz TR. Tailgut cyst (retrorectal cystic hamartoma): report of a case and review of the literature. *Am Surg*. 2005;71(8):666–673.
2. Sung MT, Ko SF, Niu CK, et al. Perirenal tailgut cyst (cystic hamartoma). *J Pediatr Surg*. 2003;38(9):1404–1406.
3. Kang JW, Kim SH, Kim KW, et al. Unusual perirenal location of a tailgut cyst. *Korean J Radiol*. 2002;3(4):267–270.
4. Hjermsstad BM, Helwig EB. Tailgut cysts. Report of 53 cases. *Am J Clin Pathol*. 1988;89(2):139–147.
5. Mills SE, Walker AN, Stallings RG, et al. Retrorectal cystic hamartoma. Report of three cases, including one with a perirenal component. *Arch Pathol Lab Med*. 1984;108(9):737–740.
6. Rafindadi AH, Shehu SM, Ameh EA. Retrorectal cystic hamartoma (tailgut cyst) in an infant: case report. *East Afr Med J*. 1998;75(12):726–727.
7. Lu NH, Tseng MJ. Laparoscopic management of tailgut cyst: case report and review of literature. *J Minim Invasive Gynecol*. 2010;17(6):802–804.
8. Pappalardo G, Frattaroli FM, Casciani E, et al. Retrorectal tumors: the choice of surgical approach based on a new classification. *Am Surg*. 2009;75(3):240–208.
9. Woodfield JC, Chalmers AG, Phillips N, et al. Algorithms for the surgical management of retrorectal tumours. *Br J Surg*. 2008;95(2):214–221.
10. Liessi G, Cesari S, Pavanello M, et al. Tailgut cysts: CT and MR findings. *Abdom Imaging*. 1995;20(3):256–258.
11. Lim K, Hsu W, Wang C. Tailgut cyst with malignancy: MR imaging findings. *AJR*. 1998;170:1488–1490.
12. Johnson AR, Ros PR, Hjermsstad BM. Tailgut cyst: diagnosis with CT and sonography. *AJR Am J Roentgenol*. 1986;147(6):1309–1311.
13. Podberesky DJ, Falcone RA, Emery KH, et al. Tail gut cyst in a child. *Pediatr Radiol*. 2005;35(2):194–197.
14. Edwards M. Multilocular retrorectalcystic disease—cyst hamartoma. *Dis Colon Rectum*. 1961;4(2):103–110.
15. Marco V, Autonell J, Farre S, et al. Retrorectal cyst-hamartoma. Report of two cases with adenocarcinoma developing in one. *Am J Surg Pathol*. 1982;6(8):707–714.
16. Middeldorpf K. Zur Kenntniss der angeborenen sacral geschwulste. *Virchows Arch Pathol Anat*. 1885;101:37–44.