MRI evaluation of not complicated Tailgut cyst: Case report

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A B S T R A C T

INTRODUCTION: The Tailgut cyst (cystic hamartoma) is an uncommon congenital disease of presacral retrorectal space and is embryologically part of some forms of enteric cysts. It is a benign malformation, although cases have been described in neoplastic degeneration.

PRESENTATION OF CASE: A caucasian 24 year old female presented shortly after childbirth with hypogastric abdominal discomfort associated with rectal tenderness, bleeding and moderate urinary symptoms for about three weeks. No previous similar episodes were reported. The patient was not suffering from haemorrhoids or inflammatory disease of the gastrointestinal tract.

Clinical examination revealed no significant abnormalities or in the perianal area and gluteal surface. Digital rectal examination was suspicious of the presence of a presacral retrorectal mass. However, it could not exclude a trans-spincteric perianal fistula. There was no fistulous communication with the exterior and the pain seemed to be more pronounced in the rectum.

MRI, which has a diagnostic accuracy of 76–100% for the detection of any perianal fistulae, was performed and demonstrated the presence of a retrorectal cystic hamartoma (Tailgut cyst).

DISCUSSION: The most common retrorectal space cystic lesions includes epidermoid cysts, dermoid cysts and enteric cysts. It presents with pelvic pain, and sometimes with local abscess, secondary to a sinus cyst. There can also be a communication between Tailgut cyst and fistula; in the absence of primary infection may develop postinflammatory fibrosis. Radiological investigation is carried out by TRUS, CT and MRI. During MRI, on T1-weighted images, the signal intensity may change from hypointense to hyperintense as protein concentration increases, as well as in the case of bleeding. On T2-weighted images, signal intensity of mucinous fluids can decrease from highly hyperintense to hypointense with increasing protein concentration and viscosity.

CONCLUSION: MRI is a non-invasive useful imaging investigation with high diagnostic accuracy when a retrorectal cyst is suspected. Despite its rarity, Tailgut cyst should be considered, both for acute complications, like infection or bleeding, and for the risk, however infrequent, of neoplastic degeneration.

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1. Introduction

A Tailgut cyst (cystic hamartoma) is an uncommon congenital disease of presacral retrorectal space and is embryologically part of the forms or enteric cysts. It’s a benign malformation, although cases have been described in neoplastic degeneration.1,2 It consists of several variants of the intestinal epithelial tissue, most often by columnar epithelium.3 Usually asymptomatic, rarely it causes local rectal compression, constipation and urinary disorders.4–6 The main complications are bleeding, infection and malignant degeneration.5–8

Magnetic Resonance Imaging (MRI) is a useful technique to evaluate pelvic disorders because of its multiplanar imaging capability and its good soft tissue contrast.2

Computed Tomography (CT) and Transrectal Ultrasound (TRUS) may be useful.

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2. Presentation of case

A caucasian 24 year old female presented shortly after childbirth with hypogastric abdominal discomfort associated with rectal pain, bleeding and moderate urinary symptoms. No previous similar episodes were reported. The patient was not suffering from haemorrhoids or inflammatory disease of the gastrointestinal tract. Laboratory tests, including serological markers (HCG, CA-125, CEA, CA 19.9) were negative.

No significant abnormalities were observed in the perianal area and gluteal surface during clinical examination. Digital rectal examination was suspicious of the presence of a presacral retrorectal mass. However, it could not exclude a trans-sphincteric perianal fistula. There was no fistulous communication with the exterior and the pain seemed to be more pronounced in the rectum.

MRI, which has a diagnostic accuracy of 76–100% for the detection of any perianal fistulae, was performed. It was decided by mutual agreement with the patient not to perform TRUS.

MRI exam (Fig. 1a and b; Fig. 2a–c) identified a cystic elongated mass with seromucinous content, formed by small cysts grouped
together in a honeycomb pattern with several internal septa, which appears hypointense on T1-weighted images and hyperintense on T2-weighted and on T2-weighted images with fat signal suppression (Sair).

There was no communication with the rectum and no signs of oedema or inflammatory hyperemia, or any fistula through the rectal wall.

Then presence of a before Tailgut cyst was suspected. The patient was operated on and histological examination confirmed the suspicion detected in MRI.

3. Discussion

The most common retrorectal space cystic lesions includes epithelial cysts, dermoid cysts and enteric cysts.

Tailgut cyst (also called cystic hamartoma or mucin-secreting cyst), like rectal duplication cyst, is included on enteric cysts classification.\(^9\)

They are more common in middle-aged women but they may affect individuals of all ages, male female ratio of 3:1.

A Tailgut cyst is an uncommon developmental lesion thought to arise from the embryonic postanal gut, well described by Hjermstad et al. at the Armed Forces Institute of Pathology in a series of 53 cases.\(^10,11\)

It often consists of columnar cells, squamous, transitional, or a combination histology. They are mucin-secreting cells.\(^9\)

The clinical presentation is variable. Usually half of the cases are asymptomatic and are discovered only incidentally.\(^11\) It often depends on the size and the relationships with the adjacent structures.

Infections are the most frequent (40–50%). It presents with pelvic pain, and sometimes with local abscess, secondary to a sinus cyst. There can also be a communication between Tailgut cyst and fistula; in the absence of primary infection may develop postinflammatory fibrosis.\(^5–7\)

Rectal bleeding is less frequent in Tailgut cyst, and it may occur more commonly in rectal duplication, which produce gastric ectopic tissue type; bleeding is most often secondary to infection.\(^5–12\)

Malignant degeneration has been described but is very uncommon and represents about 7% of enteric Tailgut cyst.\(^1,2,13\)

Radiological investigation is carried out by TRUS, CT and MRI. TRUS may help to see multilocular cystic lesions in the presence of a fluid with a mucinous character.\(^1\)

CT shows a more or less conspicuous retrorectal mass with water or soft tissues density, depending on the cystic content.\(^1,14,15\)

During MRI, on T1-weighted images, the signal intensity may change from hypointense to hyperintense as protein concentration increases, as well as in the case of bleeding. On T2-weighted images, signal intensity of mucinous fluids can decrease from highly hyperintense to hypointense with increasing protein concentration and viscosity (Fig. 1a and b: Fig. 2a–c).\(^16–18\)

In the case of cystic degeneration, the images appear with low signal both in T1 than in T2-weighted images. Septa appear hypointense in T2-weighted images.

It is important to diagnose Tailgut cyst because they may undergo a malignant degeneration, although in very low percentage (<5%).\(^2\)

Tailgut cyst should be distinguished from other presacral cysts and masses, such as epidermoid cyst, dermoid cyst, anal gland cyst, cystic meningocele and lymphangioma, but especially from rectal duplication cyst.\(^11,13\) Rectal duplication cyst is uncommon (5% of duplication cysts); the differential diagnoses from Tailgut cyst, could be made according to three histological criteria: the rectal duplication cysts are in continuity or contiguity with the rectum, have a mucosal lining similar to the rectal mucosa (with islands of ectopic tissue) and are located between two layers of smooth muscle.

However, it may develops, less than Tailgut cyst, to malignant degeneration.

Several authors argue the importance of differential diagnosis with ovarian cancer.\(^19\)

Usually cystic lymphangioma or Tailgut cyst is multiloculated, unlike other retrorectal diseases.

MRI is suitable for preoperative evaluation of Tailgut cyst and improves tissue characterization because of its high contrast resolution between different tissue compartments.\(^20\) However, biopsy histology, which is still the gold standard, is often necessary.

4. Conclusion

MRI is a non-invasive useful imaging investigation with high diagnostic accuracy when a retrorectal cyst is suspected. Despite its rarity, Tailgut cyst should be considered, both for acute complications, like infection or bleeding, and for the risk, however infrequent, of neoplastic degeneration.

Conflict of interest

None.

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None.

Author Contributions

Luca Saba: Study design.
Federica Fellini and Luca Saba: Data collections.
Luca Saba, Carlo Bocchi, and Federica Fellini: Data analysis.
Luca Saba, Massimo De Filippo, Giorgio Concar, and Cristina Rossi: Writing.

Consent

I have obtained written consent from the patient and I can provide this should the Editor ask to see it.

References

1. Lim KE, Hsu WC, Wang CR. Tailgut cyst with malignancy: MR imaging findings. AJR Am J Roentgenol 1996;167:1488–90.
2. Johnson AL, Ros PR, Hjermstad BM. Tailgut cysts: diagnosis with CT and sonography. AJR Am J Roentgenol 1986;147:1309–11.
3. Campbell W, Wolff M. Retrorectal cysts of developmental origin. Am J Roentgenol Radium Ther Nucl Med 1973;117(2):307–13.
4. Leborgne J, Guiberteau B, Lehur F, Le Geh M, Néel JC, Nombalska MF. Les tumeurs kystiques vestigiales rectofugales de l’adulte. Chirurgie 1989;115:565–71.
5. Williams LS, Rojiani AM, Quisling RG, Mickle JP. Retrorectal cyst-hamartomas and sacral dysplasia: MR appearance. AJNR Am J Neuroradiol 1989;16(9):1043–5.
6. La Quaglia MP, Feins N, Ezaklis A, Hendren WH. Rectal duplications. J Pediatr Surg 1990;25:980–4.
7. Mboyo A, Monek O, Massicot R, Martin L, Destuynder O, Lemouel A, et al. Cystic rectal duplication: a uncommon cause of neonatal intestinal obstruction. Pediatr Surg Int 1997;12:452–4.
8. Abel ME, Nelson R, Prasad ML, Pearl RK, Orsay CP, Abcarian H. Parasaracococcysal approach for the resection of retrorectal developmental cysts. Dis Colon Rectum 1985;28:855–8.
9. Levine E, Batmizky S. Computed tomography of sacral and perisacral lesions. Crit Rev Diagn Imaging 1984;21:307–74.
10. Marco V, Autonell J, Farre J, Fernandez-Layos M, Doncel F. Retrorectal Cyst-hamartomas: report of two cases adenocarcinomas developing in one. Am J Surg Pathol 1982;6:707–14.
11. Hjermstad BM, Helwig EB. Tailgut cysts: report of 53 cases. Am J Clin Pathol 1988;89:139–47.
12. Stockmann JM, Yune VT, Jenkins AL. Duplication of the rectum containing gastric mucosa. JAMA 1960;173:1223–5.
13. Crowley LV, Page HG. Adenocarcinoma arising in presacral enterogenous cyst. Arch Pathol 1960;69:64–6.
14. Piura B, Rabinovich A, Sinelnikov I, Delgado B. Tailgut cyst initially misdiagnosed as ovarian tumor. Arch Gynecol Obstet 2005;272(4):301–3.
15. Ottery FD, Carlson RA, Gould H, Weese JL. Retrorectal cyst hamartomas: CT diagnosis. J Comput Assist Tomogr 1986;10:260–3.
16. Liessi G, Cesari S, Pavanello M, Butini R. Tailgut cysts: CT and MR findings. Abdom Imaging 1995;20:256–8.
17. 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.
18. Aflalo-Hazan V, Rouset P, Mourra N, Lewin M, Azizi L, Hoeffel C. Tailgut cysts: MRI findings. Eur Radiol 2008;18:2586–93.
19. Mouloupoulos LA, Karvouni E, Kehagias D, Dimopoulos MA, Goulamou A, Vlahos L. MR imaging of complex tail-gut cysts. Clin Radiol 1999;54:118–22.
20. 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.