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Case Report

Retrorectal cystic hamartoma: A case report

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ABSTRACT

Introduction: and importance: Retrorectal cystic hamartoma (RCH) is a rare congenital lesion of the presacral space, which is part of the vestigial cystic tumors often benign and predominantly in women. Generally asymptomatic, the appearance of symptoms such as pain or neurological disorders should raise suspicion of degeneration.

Case presentation: We report an unusual observation of a 62-year-old patient admitted for perineal pain evolving for 2 months associated with tenesma and chronic constipation. The digital rectal examination found a posterior bulge at 4 cm from the anal margin, without intraluminal lesion. Rectosigmoidoscopy had noted posterior extrinsic compression but no rectal tumor. Pelvic CT and MRI had shown a solidocystic formation of the retro-rectal and presacral spaces, related to an enteric cyst. The operation was performed by abdominal approach and the surgical exploration had found a bilobed cystic formation. The cystic mass was removed and the anatomo-pathological examination concluded that it was a cystic hamartoma with no sign of malignancy.

Clinical discussion: Retrorectal tumors develop in the space bounded anteriorly by the propria fascia of the rectum and posteriorly by the presacral fascia overlying the sacrum. Common in children and then often malignant, inversely, in adults, they are rare and most often benign tumors. They are generally asymptomatic with a predominance of females, unlike our observation where the patient was male with a symptomatology dominated by perineal pain and constipation. The discovery is incidental in the majority of cases, however, in some cases, these cysts may be revealed by complications. The lesion can be explored by transrectal or suprapubic ultrasound, MRI and CT scan. Rectoscopy and fistulography may complete the exploration in case of diagnostic doubt. The resection must be thorough and in monobloc because of the risk of recurrence and the approach depends on the location and the size of the lesion.

Conclusion: RCH is a rare benign lesion whose morphological characteristics seem quite stereotyped. A detailed postoperative anatomopathological examination allows the diagnosis to be made and, above all, to look for a site of malignant transformation. This is why a complete surgical removal is necessary to prevent recurrence.

1. Introduction

Retrorectal cystic hamartoma (RCH) is a rare, often benign, asymptomatic entity with a female predilection. The positive diagnosis is currently based on the contribution of modern means of imaging. The main differential diagnoses are retrorectal teratoma, retrorectal epidermal cyst and rectal duplication. The treatment is essentially surgical and the prognosis is usually favorable. The work has been reported in line with the SCARE criteria [1].

2. Case report

Patient aged 62 years, admitted for perineal pain evolving for 2 months associated with tenesma and chronic constipation without externalized digestive hemorrhage, the abdominal clinical examination did not reveal any palpable mass while the digital rectal examination found a posterior bulge at 4 cm from the anal margin, without intraluminal lesion. Rectosigmoidoscopy had noted posterior extrinsic compression but no rectal tumor. Pelvic CT and MRI (Fig. 1, Fig. 2) had shown a solidocystic formation of the retro-rectal and presacral spaces, related to an enteric cyst. The operation was performed by abdominal approach and the surgical exploration had found a bilobed cystic

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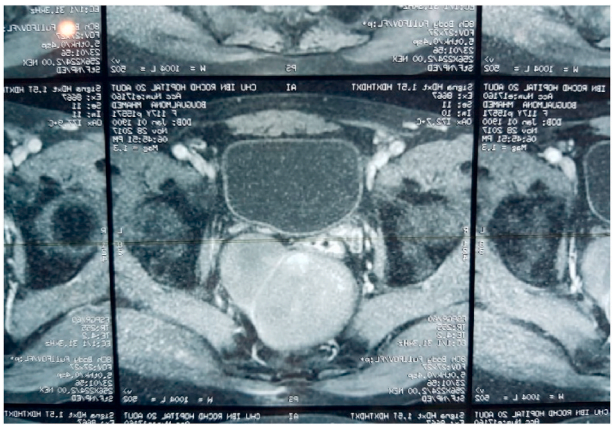


Fig. 1. MRI showing a retrorectal tissue mass in T1 hyposignal.

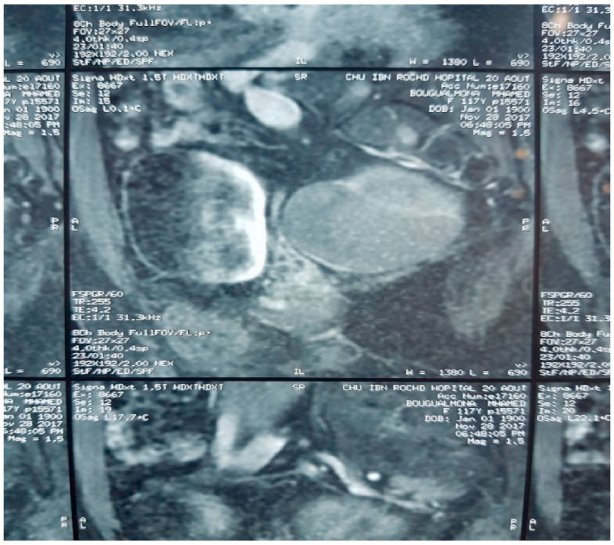
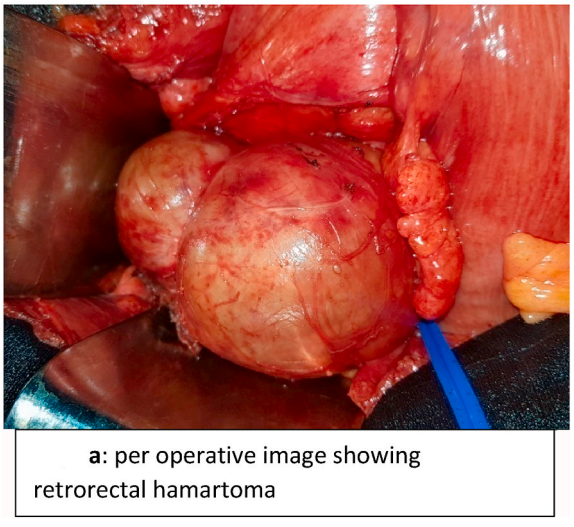


Fig. 2. MRI showing a retrorectal tissue mass in T1 hyposignal.

formation (Fig. 3a and b) measuring approximately 10 cm on its long axis, with a clean wall adjoining the posterior face of the mesorectum with the presence of a separation border. The cystic mass was removed.



a: per operative image showing retrorectal hamartoma

Fig. 3a. per operative image showing retrorectal hamartoma.

b: Surgical specimen after surgical resection

Fig. 3b. Surgical specimen after surgical resection.

The opening of the specimen had shown a thick beige pasty appearance (Fig. 4). Anatomopathological examination concluded that it was a cystic hamartoma with no sign of malignancy (Fig. 5).

3. Discussion

Retrorectal tumors develop in the space bounded anteriorly by the propria fascia of the rectum and posteriorly by the presacral fascia overlying the sacrum, it extends superiorly to the peritoneal reflection of the rectum and inferiorly to the pelvic floor formed by the elevator muscles of the anus. Common in children and then often malignant, they are usually obvious with an exophytic development. Inversely, in adults, they are rare and most often benign tumors. In a set of 341 studies including 1708 patients, cystic hamartomas represented 346 cases [2]. They are generally asymptomatic with a predominance of females [3], unlike our observation where the patient was male with a symptomatology dominated by perineal pain and constipation resulting from the mass effect applied by the tumor. The discovery is incidental in the majority of cases. However, in some cases, these cysts may be revealed by complications such as infection; rectal, anal or skin fistulization; or even malignant degeneration [3]. Imaging by transrectal or suprapubic ultrasound shows the cystic nature of the lesion, its fluid content and its retrorectal location. MRI and CT scan enable to specify the site of the lesion, its benign or malignant nature and its locoregional extension thus

Fig. 4. The opening of the specimen had shown a thick beige pasty appearance.

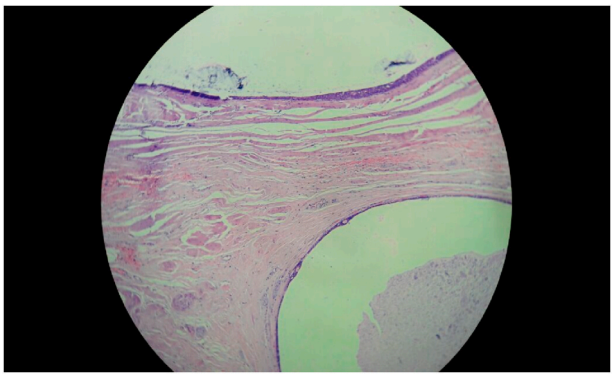


Fig. 5. Histological section showing the cyst wall covered with a flattened squamous as well as a cylindrical secretory lining.

allowing postoperative surveillance. On MRI, hamartoma is classically shown as a T1 hyposignal and a T2 hypersignal, and as a T1 hypersignal when it has a mucoid or hemorrhagic content. In addition, the presence of certain radiological signs, notably parietal thickening and the presence of intracystic vegetations, points to malignant degeneration [4]. Rectoscopy and fistulography may complete the exploration in case of diagnostic doubt or suspicion of rectal duplication [5]. Rectoscopy coupled with echo-endoscopy would have a specificity of 100% [6]. Puncture of the lesion was controversial for a long time; many authors consider it useless because of the risk of infection, skin fistula and tumor dissemination in the case of carcinoma [7]. Retrorectal cystic hamartoma is a benign tumor with a low but existent potential of malignancy, this tumor should be resected in its entirety and in monobloc because of the risks of recurrence even in asymptomatic patients [8]. The approach depends on the location of the cyst. Lesions located below S3 and without extension to the pelvic viscera are approached via the perineal route. Typically, the ventral position and Kraske's approach are used. Abdominal development beyond the S3 root requires an isolated or combined, simultaneous or delayed abdominal approach [9]. The laparoscopic approach has been shown to be effective only after careful selection of patients and after eliminating the possibility of a malignant origin of the lesions [10]. Trans-anal resection, which is dangerous when the cyst is degenerated, is reserved for cysts of less than 4 cm [11]. For benign cystic hamartomas, sequelae after surgery are rare, because dissection is facilitated by a natural peri-cystic cleavage plane; the risk is mainly that of local recurrence occurring in 10–15% of cases. In case of degenerated tumor, metastasis is frequent. In adults, their prognosis is poor despite mutilating surgery with bone and nerve sacrifices, eventually associated with radio chemotherapy, with a median survival of less than two years [12].

4. Conclusion

RCH is a rare benign lesion that deserves to be known and whose morphological characteristics seem quite stereotyped. A detailed postoperative anatomopathological examination allows the diagnosis to be made and, above all, to look for a site of malignant transformation. This is why a complete surgical removal is necessary to prevent recurrence.

Ethical approval

As per international standard written ethical approval has been collected and preserved bythe author(s).

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Author contribution

Othmane El Yamine: Writing the paper and operating surgeon. ***Fatine Amine***: Corresponding author writing the paper and operating surgeon. ***Rachid Boufettal***: Writing the paper. ***Driss Erreguibi***: writing the paper. ***Amal Hajri***: Study concept. ***Saad Rifki El Jay***: Study concept. ***Farid Chehab***: Correction of the paper.

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The following information is required for submission. Please not that failure to respond to these question/statements will mean your submission will be returned. If you have nothing to declare in any of these categories then this should be stated.

Consent

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.

Registration of research studies

This is a case report.

Guarator

Dr amine fatine.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Declaration of competing interest

The authors declare having no conflicts of interest for this article.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.amsu.2021.102362.

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