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A rare retrorectal presentation of a bronchogenic cyst: A case report

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ABSTRACT

INTRODUCTION: Bronchogenic cysts are rare abnormalities and a retrorectal presentation is exceptional. Its natural history is not known, but malignant transformation is quite rare. Retrorectal bronchogenic cysts are usually asymptomatic.

PRESENTATION OF THE CASE: We present the case of a 36-year-old young man with a past medical history of HIV seropositivity who underwent a procedure to excise a sacral coccyx cyst at another surgical center in February 2009. A histological examination confirmed it was a sacral cyst that was resected in sano. The patient presented with a recurrence of the cyst, and this report describes the combined surgical procedure using a double sacrococcygeal and abdominal approach.

DISCUSSION: A complete excision without cyst rupture is recommended to reduce the risk of local recurrence and malignant transformation, as previously reported. Resection can be performed using multiple approaches depending on the cyst's location

CONCLUSION: Herein, we report the case of a retrorectal bronchogenic cyst in a 36 years old man who was initially treated for a pilonidal cyst. A double surgical approach (abdominal and Kraske) resulted in complete resection with no recurrence.

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1. Introduction

Herein, we report here the case of a 36-year-old young man with a past medical history of HIV seropositivity who had previously had a sacrum coccyx cyst excised at another surgical center in February 2009. A histological examination confirmed it was a sacral cyst, which was resected in sano.

Its local recurrence led to an iterative excision in February 2010, and the patient was sent to our unit in July 2010 because of the persistence of a large sacrococcygeal wound that was infected. A pelvic MRI was performed (Fig. 1) and revealed the existence of a cavity below the medial caudal region that was in contact with the subcutaneous cellular tissue and nearly in contact with the ilio-coccygeal muscles.

The persistence of the lesion despite prolonged care led to another operation in October 2010 that allowed for the incomplete resection of the cystic cavity of the sacrococcygeal region extending upward in front of the sacrum.

A histological examination revealed a vestigial cyst whose histological appearance resembled a bronchogenic cyst, with no sign of malignancy.

The patient was further treated to promote rapid healing, and a control MRI from April 2011 showed stellate scar tissue formation in the caudal region. A new MRI was performed 5 months later (Fig. 2) and revealed a fluid deposit that was under 2 cm in diameter. It was assumed to be a cyst recurrence, whereas the clinical examination was negative. In March of 2012, a sacrococcygeal supuration reappeared, and the MRI showed minimal extension of the previously described fluid deposit (20 × 13 mm) between the anterior face of the lower portion of the sacrum and the rectal posterior wall.

The patient underwent surgery again using the sacrococcygeal approach in May 2012. The lesion volume was greatly underestimated during the preoperative workup because the cystic lesion was determined to be approximately 10 cm high and 5 cm wide and also contained mucus. Despite the excision of the coccyx and the last two sacral vertebrae, the lower half of the cyst could be removed without wounding the pre-sacral veins. Further excision to remove the remaining upper half of the lesion was performed one month later using infraumbilical median laparotomy, which required an important retro-rectal dissection projecting downward from the previous sacral section. The resection was macroscopically complete and the postoperative course was uneventful. There was no recurrence after three years.

A histological examination revealed a retro-rectal bronchogenic cyst with chronic ulcerated inflammatory lesions. We determined the cyst's epithelium to be of the respiratory type, containing rare

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Fig. 1. Pelvic MRI: A sagittal view showing the retrorectal cyst (white arrow).

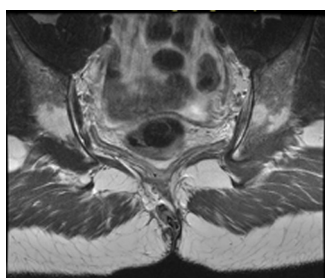


Fig. 2. Pelvic MRI, liquid collection (<2 cm) between the sacrum and rectal posterior wall, cyst recurrence.

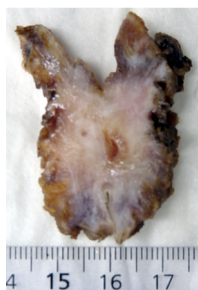


Fig. 3. Excision of the lower half of the lesion using the sacrococcygeal approach (6.5 × 4 × 2.5 cm).

pseudostratified, ciliated mucous cells. We did not observe any cartilage nodules or malignant elements (Figs. 3–5).

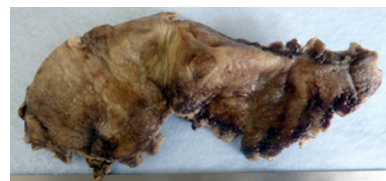


Fig. 4. Removal of the upper half of the lesion using median laparotomy (14 × 6.5 × 0.7 cm).

2. Discussion

Bronchogenic cysts are rare, presacral tumors [1,2]. Their natural history is not known, but malignant transformations are quite rare [3]. Retroperitoneal bronchogenic cysts occur at equal rates in men and women. According to the literature, 82% have been identified on the left side of the retroperitoneal region [4]. In our case, the lesion was situated medially. Retrorectal bronchogenic cysts are usually asymptomatic. When symptoms appear (sacral or rectal pain, constipation, acute retention, among others), those clinical manifestations usually occur when the cyst is compressing adjacent organs or has become secondarily infected [5]. A communicating sinus can often be found in the posterior midline of the anal canal, so these cysts are sometimes mistaken for fistulas. The histology of retrorectal tumors is highly variable [3,6,7–9] and 30–50% of them are malignant [6] or eventually become malignant during evolution [10,11]. Bronchogenic cysts may contain some normal bronchial elements including bronchial glands, smooth muscle, mucoid plaques, and cartilage [9].

Symptomatic cases are generally handled surgically for either diagnostic or treatment purposes in order to limit the risk of secondary infections. The differential diagnosis of bronchogenic cysts is not always easy. When presenting with a complex architecture, bronchogenic cysts can be confused with cystic teratomas, bronchopulmonary sequestrations, cysts of urothelial and mullerian origin, and other foregut cysts [6,12]. In our case, the homogeneous content of the lesion and the absence of solid material eliminated the diagnosis of dermoid cyst.

The most useful imaging modalities are CT and MRI. These types of cysts often appear as well-circumscribed, homogeneous lesions with high CT values ranging from 30 to 100 HU [13]. According to MRI findings, they exhibit a long T2-weighted relaxation time. However, plain X-rays of the pelvis and sacrum can be helpful by showing soft tissue shadows, sacral deformities, lysis, or an extension of the sacral space.

As previously reported, a complete excision of the cyst without rupture is recommended to reduce the risks of local recurrence and the risk of malignant transformation [12]. Resections can be performed using multiple approaches depending on the cyst's location. Lesions located below S3 can be resected using the perineal

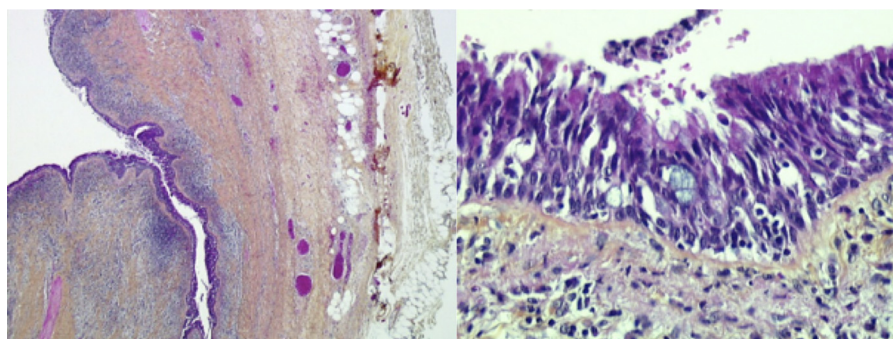


Fig. 5. HES stain (hematoxylin eosin saffron), ×25 magnification (left), ×400 magnification (right).

approach [13], corresponding to the Kraske procedure [14]. Bone sectioning allows for better exposition, but also increases the risk of post-operative osteitis. Others have described a parasacrocygeal approach without bone sectioning [15]. Another transanal way to excise small cysts (less than 4 cm) has been proposed [16]. For lesions located beyond the S3 roots or larger lesions, resection requires an isolated or combined open-abdominal approach. [17]. Laparoscopy can also be a viable approach as it is less aggressive and results in a shorter hospital stay [18].

3. Conclusion

We report here the case of a retrorectal bronchogenic cyst in a 36-year-old man who was initially treated for a pilonidal cyst. A double-surgical approach (abdominal and Kraske) allowed for complete resectioning with no recurrence.

Conflicts of interest

No conflict of interest.

Funding

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Ethical approval

This is a retrospective case with no modification on standard practice. No need of ethical approval.

Consent

The patient gave his written consent.

Author contribution

Arnaud Pasquer, Filbert Djeudji, Valérie Hervieu, Maud Rabeyrin, Xavier Barth contributed to the redaction of this manuscript and proof reading.

Filbert Djeudji and Xavier Barth performed the surgery.

Valérie Hervieu and Maud Rabeyrin performed the pathological analysis of the cyst.

Guarantor

Arnaud pasquer.

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