Laparoscopic approach to tailgut cyst (retrorectal cystic hamartoma)

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Abstract

Retrorectal cystic hamartomas, or tailgut cysts, are complex congenital cystic lesions which arise from embryologic tissues. Fewer than 200 cases have been reported worldwide, with women outnumbering men by 3:1. They are asymptomatic in 50% of the cases; the remainder present with back pain or mass effect as the most common symptoms. Malignant transformation rarely occurs. Guided biopsy is controversial, while surgery is the therapy of choice. We report the case of a 31-year-old woman complaining about perineal and vague lower abdominal pain, who was submitted to magnetic resonance imaging, which revealed a multilocular cystic, well-circumscribed retrorectal mass. Subsequently, laparoscopic excision was successfully accomplished. Operative time was 175 min. Intra- and post-operative course was uneventful. Hospital stay was 75 h. While any malignancy suspicion should lead to open surgery, given the risk of rupture, we support the benefits of laparoscopy may also be applied.

Keywords: Congenital disease, cysts, presacral space, retroperitoneal tumour, retrorectal hamartoma

INTRODUCTION

Tailgut cysts are an uncommon[1] and complex entity in adults and children, which arise from embryological tissues. If they show malignant transformation, retrorectal cystic hamartomas (RCHs) are more frequently diagnosed as adenocarcinoma or carcinoid. Complete surgical excision is the treatment of choice, and definitive diagnosis is based on pathological examination of the specimen.[2]

The most commonly described surgical techniques are through standard Kraske or open abdominal approach. Laparoscopic surgery of RCHs has recently been reported and allows complete excision with low morbidity. Potential benefits should include less post-operative pain, shorter hospital stay, less blood loss, faster recovery time, fewer complications and better cosmesis.[3]

We communicate the excision of a tailgut cyst utilising a conventional laparoscopic procedure. The authors would like to underscore that, despite this technique has been applied to various surgical procedures, very few RCH excisions have been published.

CASE REPORT

A 31-year-old female presented abdominal discomfort and perineal and vague lower abdominal pain. Her medical
history and her physical examination were irrelevant. Digital rectal examination revealed an extraluminal tumour bulging from the posterior rectal wall. The magnetic resonance imaging (MRI) examination showed a multilocular, well-defined cystic mass of $6 \times 5 \text{ cm} \times 5 \text{ cm} \times 8 \text{ cm}$ in the presacral space [Figure 1]. It displaced the rectum and the inferior aspect of the levator ani anteriorly and showed no evidence of invasion or distant metastases. Surgical access was gained with a 12-mm supraumbilical port followed by one 10-mm and three 5-mm ports in both the right upper/lower and left upper/lower quadrants. The mesorectal dissection from the sacral promontory was continued up to the level of puborectalis sling and the levator ani muscles [Figure 2]. The cyst was accidentally opened before last adhesion was excised and approximately 2 ml of mucoid fluid were aspirated. Before specimen removal, rectal integrity and haemostasis of the operative field were ensured.

Operative time was 175 min. A drain was placed and removed 48 h after surgery. The patient was discharged 75 h after surgery without specific analgesia requirements (only on-request conventional analgesia) and continued with oral antibiotic treatment during 4 days. Cosmetic appearance of the incisions was very satisfactory. The postoperative period was uneventful. No evidence of recurrence has been observed after a follow-up of 18 months, and the patient has hence recovered nicely and remains asymptomatic.

A multilocular, well-circumscribed cyst of $65 \text{ cm} \times 55 \text{ cm} \times 86 \text{ mm}$ in size filled with mucoid fluid was sent to pathological examination. No evidence of malignancy was found. Pathologic diagnosis was benign tailgut cyst with negative lateral and vertical margins.

DISCUSSION

There is an important lack of information on retrorectal cystic hamartoma natural history and biological behaviour: fewer than 200 cases have been published, with women outnumbering men by 3:1. Nearly half the patients are asymptomatic, which explains the frequent delay in clinical diagnosis; the remainder present with symptoms as back pain, rectal fullness, perianal pain and rectal bleeding. Malignant transformation is rare (2%–13%), and the largest series of 53 cases was published by Hjermstad with only one case of malignancy. Preoperative biopsy is controversial, due to risk of spreading dysplastic cells, bleeding and infection, especially with the improvements in the imaging technique. MRI has evolved to be the cornerstone of the evaluation of the tailgut cyst, as it provides excellent anatomic detail, visualises soft-tissue planes and evaluates relationships with adjacent structures and local invasion. Complete en bloc resection with clear surgical margins is the therapy of choice to prevent infection and malignant change. Indeed, incomplete resection can lead to recurrence. Historically, multiple techniques have been reported. Laparoscopic resection, with the reduction of surgical trauma, has recently been reported as a valid alternative to standard Kraske or other abdominal procedures. Potential advantages should include fewer complications, faster recovery of bowel function, shorter hospital stays, less post-operative pain, less blood loss, and improved cosmesis results.

In conclusion, laparoscopic approach of tailgut cysts is feasible, even when tumours contact the elevator ani and coccygeus muscle. Due to the potential risk of recurrence and spreading dysplastic cells if incidentally opening the cyst, other approach should be considered if preoperative studies show malignancy signs.
Declaration of patient consent
The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patient understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest
There are no conflicts of interest.

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