



Appendiceal intussusception to the cecum caused by mucocele of the appendix: Laparoscopic approach

Said Ait Laalim^{a,*}, Imane Toughai^a, El bachir benjelloun^b, Karim Hassani Ibn Majdoub^a, Khalid Mazaz^a

^a Department of General Surgery, B. CHU Hassan II. Fez. Morocco

^b Department of General Surgery, A. CHU Hassan II. Fez. Morocco

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ABSTRACT

INTRODUCTION: Appendiceal intussusception is a very rare disease that is found in only 0.01% of patients who have undergone an appendectomy. Clinical symptoms vary and some patients are asymptomatic. Laparoscopic surgery for appendiceal tumors is still controversial. We present a case of intussusception of the appendix with a mucinous cystadenoma treated by laparoscopic surgery.

PRESENTATION OF CASE: We report a case of 47 year-old-women patient who presented with a six month history of intermittent right lower abdominal pain. Abdominal computer tomography CT showed appendiceal mass with intussusception. A laparoscopic right hemicolectomy was performed because the tumor involved the entire appendix. Histopathological examination revealed mucocele due to mucinous cystadenoma of appendix.

DISCUSSION: Appendiceal intussusception to the cecum caused by mucocele of the appendix is extremely rare. It is very difficult to diagnose the presence of an intussuscepted appendix pre-operatively and investigations will usually include colonoscopy and CT scan. An appendicular intussusception should not be reduced by colonoscopy. Laparoscopic surgery for appendiceal tumors is still controversial; the main concerns to be addressed are the adequacy of resection and intraperitoneal rupture of the tumor. Our patient successfully underwent laparoscopic surgery without any complications.

CONCLUSION: Appendiceal intussusception to the cecum caused by mucocele of the appendix is a rare cause of abdominal pain and difficult to diagnose. The laparoscopic surgery for appendiceal tumors is safe, feasible, and even may be beneficial.

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1. Introduction

Appendiceal intussusception to the cecum is an uncommon type of intussusception, with an incidence of approximately 0.01% in patients undergoing appendectomy.¹ In adults it is usually associated with pathological origins. Intussusception caused by appendiceal mucocele is extremely rare and these tumors are rarely associated with clinical manifestations. The role of laparoscopic resection in the management of appendiceal mucocele is not well defined in the literature. We report the case of intussusception of the appendix caused by low-grade appendiceal mucinous neoplasm, for which a formal right hemicolectomy was successfully performed using a laparoscopic approach.

2. Case report

47-Year-old women with no notable medical or surgical history presented with a two days history of pain in the right side of the abdomen, nausea and vomiting. He reported having had similar but

much less severe episodes during the previous 6 months. Physical examination revealed tenderness in the right lower quadrant. There is no palpable masse, and no guarding or rebound tenderness was observed. Ultrasound showed a pathologic mass, 5 cm of diameter in the right side of the abdomen. Abdominal CT revealed swelling of the appendix (15 mm) evocative of an appendiceal mucocele with appendiceal intussusception to the cecum and secondary distal ileum invagination (Fig. 1). The laparoscopic exploration confirmed the diagnosis of appendiceal mucocele intussusception (Fig. 2). The tumor was seen to be involving the entire appendix up to the base. So the decision to perform a formal right hemicolectomy was made. A laparoscopic right hemicolectomy was performed without directly handling the tumor. Postoperative recovery was uneventful. Pathological diagnosis was low grade appendiceal mucinous neoplasm. The patient was followed up for 24 months and has had no recurrence so far.

3. Discussion

Appendiceal intussusception is a very rare disease that is appearing often as acute appendicitis, making it very difficult clinical or radiological preoperative diagnosis. In 1963, Collins concluded in a 40-year prospective study involving 71,000 cases of appendectomy, that the incidence of appendiceal intussusception

* Corresponding author at: 6, Hay el amal, narjiss 30000, FES, Morocco.
Tel.: +212 661488803.

E-mail address: aitlaalims@yahoo.fr (S.A. Laalim).

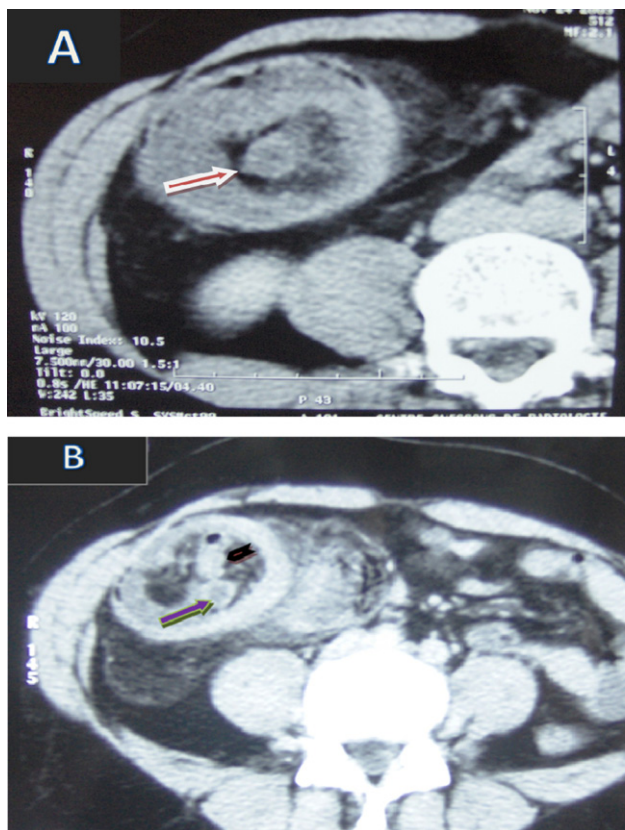


Fig. 1. (A) Abdominal CT revealed an appendix surrounded by a rim of digestive structures displaying a “target-like” sign, typical of appendiceal intussusceptions. (B) Abdominal CT showed a well-encapsulated cystic mass protruding in the cecum lumen evocative of an appendiceal mucocele (the “cup-and ball” pattern) (arrow) and secondary distal ileum invagination (arrowhead).

was 0.01%.¹ Appendiceal intussusception is caused by irregular appendiceal peristalsis developed by local irritation, and is more likely to occur in mobile mesoappendix wide appendicular lumen, and thin and mobile appendix.² The clinical features include a range of signs and symptoms, and in some cases, even asymptomatic patients were described in the literature. As seen in our case the presenting symptoms and physical exam findings were not specific for intussusception caused by appendiceal mucocele. Abdominal pain and distention along with changes in bowel movements are symptoms associated with a long list of differential diagnoses, including gynecological causes in female patients. However, the medical

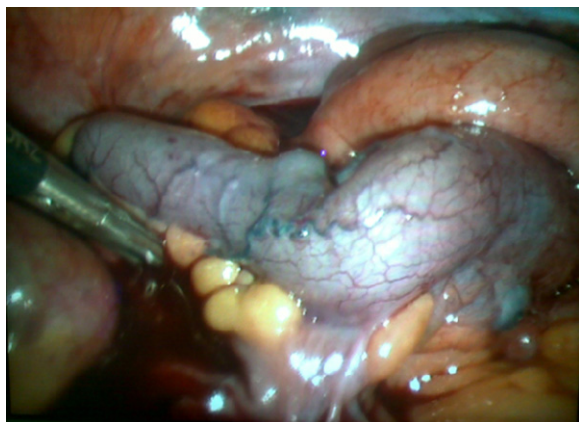


Fig. 2. Perioperative photograph showing an intra cecal intussusception appendiceale.

history of similar but much less severe episodes during the previous 6 months suggested the possibility of intussusception.

Radiologic evidence helps in narrowing down and focusing attention to a subset of possible etiologies. Ultrasound is the method of choice for children with suggestive signs and symptoms, the classic imaging of intussusception being of “onion skin-like lesion”. CT is the most commonly used diagnostic modality. It shows a subhepatic cecum and an appendix surrounded by a rim of digestive structures displaying a “target-like” sign, typical of appendiceal intussusception. It was also associated with a well-encapsulated cystic mass protruding in the cecum lumen evocative of an appendiceal mucocele (the “cup-and ball” pattern).^{3,4} Colonoscopy is another method that can show signs suggestive of this condition, since it allows the direct view of a vegetating image inside the cecum, which must be differentiated from a neoplastic process.⁵ In our patient the CT scan has clearly shown the picture of appendiceal mucocele intussusception.

Mucocele of the appendix is a descriptive term for several pathological processes of which the commonest being mucinous cystadenoma. The presence of a mucinous cystadenoma can lead to malignant transformation and spontaneous or iatrogenic rupture. This can result in implantation of mucin-producing cells inside the peritoneal cavity, and subsequent pseudomyxoma peritonei.⁶ Patients with benign mucinous cystadenomas have an excellent prognosis, with 5-year survival rates of greater than 90%. However, those with malignant cystadenocarcinoma and pseudomyxoma peritonei have a 5-year survival rate of 25%, signifying the need for early recognition, management and followup.⁷ No current consensus exists as to the diagnostic criteria or treatment of appendiceal tumors.⁸ An appendiceal intussusception should not be reduced to avoid exposing the peritoneal cavity to these mucin-producing cells.⁹ There are some cases of intussusception reported in the literature reduced by colonoscopy, suggesting that colonoscopic reduction by non-operative air infusion could be a possibility.¹⁰ Such procedure may be of a big risk of dissemination, peritoneal seeding, venous embolism and perforation leading to an unexpected outcome.¹⁰ A radically removed appendix is curative in most cases of appendiceal tumors. However, a right hemicolectomy should be considered for patients with malignant mucinous lesions or if a benign tumor involves the base of the appendix. The latter was the case in our patient making it impossible to achieve a tumor-free margin at surgery, so we proceeded with a formal right hemicolectomy. Also, if a mucocele is more than 2 cm in size, it is more likely to be neoplastic. In our patient, the tumor size was 7 cm × 3 cm.¹¹

The indications for the laparoscopic approach to the resection of these tumors have not yet been established definitively. As evidence of the benefits associated with laparoscopic appendectomy accumulates, an increasing number of resections for appendiceal tumors are being performed via laparoscopy. Despite growing evidence favoring the laparoscopic approach, Gonzalez et al. reported a case of laparoscopic mucocele resection that was followed by early peritoneal progression, forcing them to conclude that this entity was a contraindication to laparoscopic resection.¹² As the technique of laparoscopic appendectomy evolves, the feasibility of resecting appendiceal neoplasms via this approach should also be assessed.¹³ Several cases of laparoscopic treatment of appendiceal mucoceles have been reported with good results.^{13–16} Data from some studies indicate that laparoscopic appendectomy for the management of appendix neoplasms is associated with long-term results comparable to those obtained with open appendectomy.¹⁷ Our case report adds to the existing data regarding the safety of laparoscopy for cases of appendiceal neoplasm. We did not touch or handle the lesion during any part of the procedure. The wound protector that we applied will prevent port-site seeding.

The application of laparoscopic surgery has been more widely adopted in the treatment of gastrointestinal diseases, and in some cases laparoscopic surgery is currently the standard treatment. In the surgical treatment of appendiceal mucoceles, however, open surgery is currently the standard surgical treatment due to its well-known safety and easy accessibility. The authors are also aware of the possibility of rupture during laparoscopic dissection, and the subsequent fatal complication of pseudomyxoma peritonei. Nevertheless, if we can handle the tissue with minimal manipulation, and can secure enough safety by using various laparoscopic instruments, laparoscopic resection of an appendiceal mucocele might be an alternative surgical option to open surgery.

4. Conclusion

Appendiceal intussusception to the cecum caused by mucocele of the appendix is a rare cause of abdominal pain and difficult to diagnose. The laparoscopic resection of mucocele of the appendix is feasible in spite of the danger of malignancy, provided necessary precautions are taken.

Conflicts of interest

None.

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None.

Ethical approval statement

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.

Authors' contribution

Imane Toughai has contributed in the care of the patient. El bachir benjelloun helped in drafting the article. The other authors participated in the literature research.

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