

CASE REPORT

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Non-inflamed appendix with LAMN causing closed-loop small bowel obstruction

Hein Maung, Matthew Marino, Fidel Touma

ABSTRACT

Introduction: Low grade appendiceal mucinous neoplasms are generally encountered incidentally in appendectomies for suspected appendicitis. This case is a documentation of a rare occurrence of LAMN presenting with small bowel obstruction.

Case Report: An 83-year-old male presented to the Emergency Department with a three day history of nausea, vomiting, and obstipation and profoundly dehydrated. Computed tomography revealed a high grade small bowel obstruction with 2 transition points in the right iliac fossa, concerning for closed loop obstruction, necessitating an urgent laparotomy.

Conclusion: While acute appendicitis is a common cause of emergency abdominal pathology, when coupled with small bowel obstruction, a neoplastic cause should be considered as part of the differential diagnosis.

Keywords: Appendicitis, LAMN, SBO

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INTRODUCTION

Small bowel obstruction due to an appendiceal inflammation or adhesion is an uncommon presentation, with patients presenting with symptoms of gastrointestinal obstruction, with or without systemic sepsis [1]. Appendicitis causing small bowel obstruction was first described in the literature by Naumov et al. in 1963, and in this case we discuss an even rarer occurrence of this presentation with the additional presence of a low grade mucinous neoplasm [2].

CASE REPORT

An 83-year-old man presented with three days of nausea, vomiting, and obstipation. He had a past surgical history of an open umbilical and inguinal hernia repairs over 10 years prior. He didn't have any medical comorbidities, and was independent at home with his wife.

On physical examination, he was profoundly dehydrated with decreased skin turgor, pallor, dry mucous membranes and was in rapid atrial fibrillation, with a low blood pressure of 103/60, and afebrile. His abdomen was distended, tympanic, and was generally tender.

Hemoglobin was 11.3 g/dL; white blood cell count was $14.5 \times 10^9/L$; C-reactive protein was 98 mg/dL; Creatinine 170 $\mu\text{mol/L}$. Computed tomography (CT) of his abdomen demonstrated a high-grade small bowel obstruction (SBO) with two transition points within the right iliac fossa. The intervening bowel was edematous, concerning for an ischemic closed loop.

A nasogastric (NG) tube was inserted for draining copious amounts of bilious fluid over 1 liter, and he was resuscitated in the emergency department (ED) prior

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to the operating theater for an emergency laparotomy. Turbid free fluid was found on entry. A 50-centimeter loop of ischemic, non-viable small bowel was found incarcerated underneath a ring formed by the appendix 15-centimeters from the terminal ileum, forming a closed-loop SBO. The appendix was non-inflamed and had a prominent thickening in its mid-portion, the tip of which was embedded within the mesentery of the terminal ileum. The band was released and appendectomy performed. The ischemic small bowel was resected and a stapled side to side anastomosis performed. The patient made an uneventful recovery and was discharged post-operative day 7.

Histology revealed a low-grade mucinous neoplasm (LAMN) within the mid-portion of the appendix over 1-centimeter from the nearest margin, with no invasive component and no perforation or extravasated mucin. The patient was discussed at a multidisciplinary team meeting where it was deemed no further treatment was necessary (Figures 1–3).

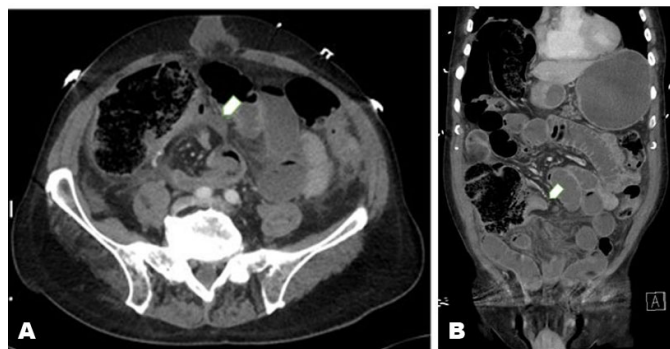


Figure 1 (A and B): Computed tomography (CT) with intravenous contrast (axial view and coronal view). Arrow shows the appendix forming a band through which terminal ileum passes, forming a closed loop obstruction.

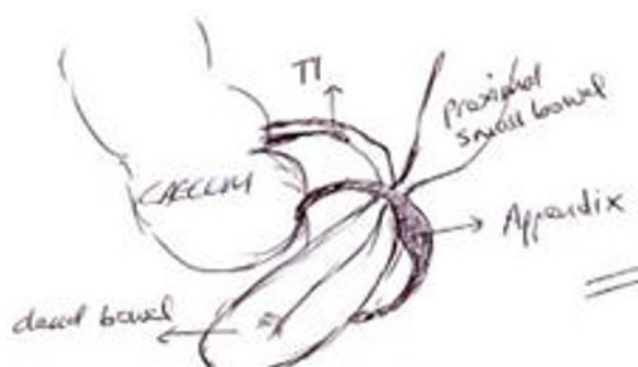


Figure 2: Schematic of the operative findings.

DISCUSSION

Small bowel obstructions are a common cause of surgical admission. The most common causes of SBO are adhesional, accounting for 75% of all SBO [3]. The appendix as a cause of SBO has now been well reported in

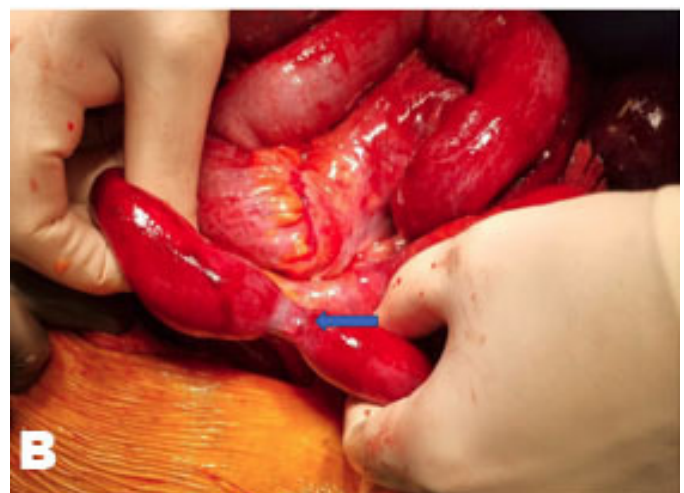


Figure 3: Intraoperative photograph showing the appendix causing a band around ischemic small bowel (A) and the indentation on the small bowel caused by the appendix (B).

the literature; however, almost exclusively in the context of appendicitis [4–6]. An “appendiceal tie” strangulating the small bowel via appendicitis has been documented in the literature and was first described by in 1963 [2, 5, 7]. A classification system for SBO in the context of appendicitis has been suggested, with mechanisms via adynamic (paralytic ileus), mechanical without strangulation, strangulation, and finally mesenteric ischemia [8]. A literature review in 2017 by Makama et al. identified 45 patients with obstruction due to appendicitis: (A) Appendix laid across bowel loops bound by adhesions; (B) Herniation through a ring or gap formed by appendix tip attached to base; (C) Tip attached to bowel causing torsion; (D) Kinking of bowel; (E) Complex knotting [1].

In our case, SBO was caused by the mechanism of strangulation with the tip of the appendix embedded in the mesentery of small bowel, forming a “ring” for small bowel to herniate through. The underlying cause of the adhesion is speculated to more likely have been secondary to previous tip appendicitis rather than the LAMN itself, given its location—the LAMN was mid appendix, while the tip was embedded in the mesentery of the ileum, without any signs of acute inflammation.

Low-grade mucinous neoplasm of the appendix is different from most malignant tumors, with a mechanism of growth with a pushing margin of low-grade dysplasia, in contrast to infiltrative growth, and disseminates via perforation leading to pseudomyxoma peritonei (PMP). Two distinct clinicopathological types of LAMN lesions were identified, with type 1 lesions confined to the appendiceal lumen, while type 2 lesions involved appendiceal wall or peri-appendiceal tissue with or without perforation of the appendix, and thus were more likely to be referred for hyperthermic intraperitoneal chemotherapy (HIPEC).

A literature search on PubMed identified 4 cases of appendiceal LAMN causing SBO via adhesions in different locations:

- (1) Small bowel mesentery to small bowel mesentery causing partial obstruction,
- (2) Ileocecal intussusception,
- (3) Adhesions from appendix to small bowel +/- spread of mucin/nodules to peritoneum [3].

In this case, the appendiceal tip had embedded in mesentery without perforation, inflammation, or high-grade features of LAMN. This case would make it a total of 6 documented cases of this rare pathology.

CONCLUSION

While acute appendicitis is a common cause of emergency abdominal pathology, when coupled with small bowel obstruction, a neoplastic cause should be considered as part of the differential diagnosis.

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Author Contributions

Hein Maung – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Matthew Marino – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

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Written informed consent was obtained from the patient for publication of this article.

Conflict of Interest

Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.

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