Perforated acute appendicitis resulting from appendiceal villous adenoma presenting with small bowel obstruction: a case report

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Abstract

Background
Acute appendicitis and complete small intestinal obstruction are the most common surgical emergencies. Mechanical obstruction induced by acute perforated appendicitis has been recognized in the literature, but it is difficult to find cases of perforated appendicitis caused by a benign tumor. Less than 1% of all appendectomy specimens contain tumors, of which less than 10% are villous adenoma [2]. To our knowledge, acute appendicitis caused by villous adenoma is extremely rare.

Case Presentation
This is the first report describing appendiceal villous adenoma, an occasional cause of perforated acute appendicitis, presenting an unusual appearance with complete intestinal obstruction.

Conclusions
The present report describes this uncommon case and reviews the literature pertaining to its incidence, symptoms, diagnosis and treatment.
Background
Intestinal obstruction is a common problem in clinical practice. The common causes are mechanical obstruction or ileus [3]. Although postoperative adhesion accounts for up to 75% of small bowel obstruction [4], it is notable that acute appendicitis has also been a rare cause of small bowel obstruction [1,5]. Acute appendicitis results from obstruction of the appendiceal lumen, and the most common causes of obstruction are fecaliths and lymphoid follicular hyperplasia. In contrast, acute appendicitis induced by obstruction with a benign tumor is very uncommon. In particular, villous adenoma is an extremely rare appendiceal neoplasm. Early recognition expedites diagnosis, and surgery is very important. Here, we present a case of complete small intestinal obstruction caused by acute appendicitis with perforation resulting from obstruction by a villous adenoma.

Case Presentation
A 78-year-old woman visited our emergency department with several hours’ history of diffuse abdominal pain and distension.

On physical examination, the patient had a blood pressure of 140/70 mmHg, oral temperature of 37.1 ºC, pulse rate of 108/min and respiratory rate 22/min. Abdominal examination revealed distension with tympanic percussion and local tenderness, especially over the left lower abdominal region. No obvious muscle guarding or rebound pain was noted. Laboratory investigation revealed a white blood cell count of 15.5 × 10^3/µL (95% neutrophils) and C-reactive protein (CRP) of 35.20 mg/dL (reference range, < 0.5 mg/dL). An abdomen plain film showed dilated bowel loops in the left abdomen (Figure 1). An abdominal computed tomography (CT) scan revealed small bowel distension with air-fluid level and a transitional region at the left lower abdomen (Figure 2A & 2B). The patient was treated surgically with exploratory
laparotomy. During surgery, a ruptured inflamed appendix and an adhesion band were found (Figure 3A & 3B). The histopathological report on the surgical specimen indicated acute suppurative appendicitis and rupture with a villous adenoma that was confined to the mucosa of the appendix and did not extend to the resection margins (Figure 4). The patient was discharged on the 10th postoperative day in a stable condition.

**Discussion**

Intestinal obstruction is a common clinical problem. Depending on the level of the obstruction, it is classified as small bowel obstruction (SBO) or large bowel obstruction (LBO). The common causes of SBO are benign lesions while LBO is often caused by malignancy [4]. SBO is more common than LBO.

The most common presentations of SBO are abdominal pain, vomiting, constipation and even fever. The abdominal physical examination shows abdominal distention, tenderness or guarding. The most common causes of SBO are adhesion and hernia, which account for more than 80% of all cases. The causes are divided into mechanical or nonmechanical based on the mechanism of obstruction. The etiologies of mechanical obstruction include extrinsic, intrinsic or intraluminal lesions and the nonmechanical causes are adynamic ileus or spastic ileus. The extrinsic lesions are adhesions, while hernias, appendicitis, diverticulitis, neoplasms, Crohn’s disease and tuberculosis are the common intrinsic lesions. Intraluminal lesions include gallstones, foreign bodies and bezoars [4].

SBO are also classified into simple and complicated obstructions. Simple obstructions include intermittent, incomplete or partial obstruction where the lumen contents can pass through the lesion site. Complicated obstructions include closed loops or strangulation.
Acute appendicitis has been recognized as a cause of SBO, which in such cases may be mechanical or the result of ileus [5]. The obstruction may result from adhesions caused by periappendicular inflammation or ileus, because of generalized or localized peritonitis caused by a perforated acute appendicitis. Luminal obstruction is the pathological hallmark of acute appendicitis. The most common cause of this obstruction is the fecalith, followed by enlarged lymph nodes and tumors.

Tumors of the appendix are very uncommon and are usually diagnosed at operation or autopsy. In previous classical studies of appendectomy specimens, most appendiceal tumors were benign [6,7] and consisted of leiomyomas, neuromas and lipomas. Reports of villous adenoma are extremely rare. In general, villous adenoma is a neoplasm that most often occurs in the rectum and sigmoid colon [8]. Although such a benign neoplastic lesion located in the appendix is extremely rare, when found, acute appendicitis is its most common form of clinical presentation [9].

The clinical manifestations of intestinal obstruction induced by primary appendiceal rupture are the same as those induced by mechanical intestinal obstruction: cramping abdominal pain occurs in paroxysms and the patient is relatively comfortable in the intervals between the pains. Vomiting is almost invariable, and the symptoms usually precede the obstruction. When the two diseases coexist, the clinical manifestations of SBO usually dominate the clinical symptoms and mask the usual presentation of appendicitis [10]. Therefore, sudden onset complete small bowel obstruction with adhesion band formation in patients without surgical history must be carefully investigated to identify the underlying problems behind this simple disease.

In our patient, no typical presentations of acute appendicitis such as local tenderness, local rigidity over the right lower quadrant abdomen or McBurney’s point tenderness were found on the initial examination. The probable reasons for these
atypical symptoms are listed below. First, the peritoneum contained the perforated appendix within the sac, preventing widespread inflammation. Thus, in this patient, the presentation was of local tenderness, and no muscle guarding or peritoneal signs were found. Second, the coexistence of the two diseases might change the typical pathogenesis of this condition. In general, when atypical symptoms are found initially, we must be aware of the possibility of occult reasons for the intestinal obstruction.

Most villous adenomas are found incidentally during the histopathological examination after appendectomy. Because villous adenoma has a propensity to develop to adenocarcinoma, further therapy may be considered, depending on whether the resection line of the appendix stump is clear. Appendectomy is the treatment of choice for those cases in which the lesion does not reach the line of resection. In neoplastic lesions where involvement reaches the resection line, cecectomy is advisable to assure adequate excision of the tumor [11]. However, some reports have suggested that a right hemicolecetomy or ileocecal resection with lymph node dissection should be performed when carcinoma is diagnosed preoperatively or during surgery [12].

Colon cancer (adenocarcinoma) synchronous with appendiceal adenoma has been noted. Thus, colonoscopic examination may play an important role in patients with incidentally discovered appendiceal tumors, and is particularly indicated in patients in their 6th–8th decades [13].

**Conclusions**

This is the first report of an unusual presentation of a perforated appendicitis induced by intraluminal villous adenoma. No previous report of a similar presentation (complete intestinal obstruction) was found in the literature. Acute appendicitis and complete intestinal obstruction are surgical emergencies. However, in the elderly, a
complete mechanical intestinal obstruction without obvious previous surgical history or malignancy, the possibility of occult reasons or another coexisting disease such as acute appendicitis or hernia must be considered. The postoperative specimen needs aggressive follow-up. It must be considered whether a perioperative frozen specimen of the appendectomy should be regularly arranged for patients in their 6th–8th decades.

Consent
Written informed consent was obtained from the patient for publication of this case report and the accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Abbreviations
SBO: Small bowel obstruction

LBO: Large bowel obstruction

CT : Computed Tomography

Competing interests
The authors declare that they have no competing interests.

Authors’ contributions
YG Chen and YL Chen was involved in writing, reviewing, editing and finalizing the manuscript. HM Chang managed the whole operative procedures. YG Chen and HM Chang participated in the care of the patient and assisted in drafting the manuscript. CH Hsu was the specialist in the gastroenterology involved with the case and also reviewed, edited and finalized the manuscript. All authors have read and approved the final version
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**Figures**

**Figure 1**
Abdominal roentography taken during the admission screen revealed dilated bowel loops with air accumulation in the whole abdomen.

**Figure 2**
Abdominal computed tomography. (A) Axial view shows the small bowel distended with air-fluid levels, swelling of the wall of the intestine and fluid accumulation over the lower region. (B) Coronal view reveals a transitional region at the left lower abdomen.
**Figure 3**
The perioperative view showed (A) adhesion between the bowel loop and peritoneum because of pus peel formation. (B) Appendicitis with perforation was noted.

**Figure 4**
The appendix tissue reveals villous adenoma with moderate to severe dysplasia and acute suppurative appendicitis. (Hematoxylin and eosin stain, original magnification, $\times 400$.)

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