Mechanical intestinal obstruction secondary to appendiceal mucinous cystadenoma
A case report and brief review
Zheng-shui Xu, MS, Wei Xu, PhD, Jia-qi Ying, MS, Hua Cheng, MS

Abstract
Background: Appendiceal mucinous cystadenoma can present in various ways, and it is most commonly encountered incidentally during appendectomy, but mechanical intestinal obstruction secondary to an appendiceal mucocele has been rarely reported.

Methods: We report a case of mechanical intestinal obstruction secondary to appendiceal mucinous cystadenoma. After nasogastric decompression and initial aggressive intravenous fluid resuscitation, an emergency operation was performed under the diagnosis of acute mechanical intestinal obstruction.

Results: We performed an appendectomy and intraoperative enteral decompression without anastomoses. The pathologic examination (PE) revealed appendiceal mucinous cystadenoma. After the operation, the patient’s recovery went smoothly, and the patient was discharged on the fifth postoperative day. No tumor recurrence was recorded over an 8 month follow-up period.

Conclusion: Early operative intervention should be recommended to the patient with acute mechanical complete intestinal obstruction, especially the patient who had no previous abdominal surgery. And it is vital to discriminate benign and malignant appendiceal mucocel in determining the extent of surgery.

Abbreviations: AM = appendiceal mucocele, CT = computed tomography, PE = pathologic examination.

Keywords: appendiceal tumor, intestinal obstruction, mucinous cystadenoma

1. Introduction
Mucocele is an uncommon disease of the appendix. Appendiceal mucocele can present in diversified ways, but it is most commonly encountered incidentally during appendectomy.1-3 A case report of mucinous cystadenoma, a particular type of mucocele, at the tip of the appendix that led to a mechanical intestinal obstruction has rarely been described in the literature. Here, we present a case report describing a 76-year-old man who was diagnosed with mechanical complete intestinal obstruction secondary to appendiceal mucinous cystadenoma.

2. Case report
A 76-year-old previously healthy man, who had no history of abdominal surgery, presented to our emergency room with a 38-hour history of intermittent abdominal pain, nausea, and the disruption of bowel movements. His abdomen was mildly distended and tympanic with diffuse slight tenderness, particularly in the periumbilical areas. He had sonorous bowel sounds. A computed tomography (CT) scan of the abdomen revealed dilated small bowel loops with air-fluid levels suggestive of a small bowel obstruction (Fig. 1). The patient’s leukocyte count was 1890, with 92.1% neutrophils. After nasogastric decompression and initial aggressive intravenous fluid resuscitation, an emergency operation was performed under the diagnosis of acute mechanical complete intestinal obstruction. The patient underwent exploratory laparotomy that revealed dilated enteral loops with air-fluid content and uncomplicated mild ascites. In addition, there was an appendiceal mucocele present, measuring 4.0 × 2.5 × 2.5 cm, at the tip of the appendix (Fig. 2). The mucocele had adhered to the right paracolic sulci that formed a loop which the ileum was confined within. The incarcerated ileum was between 2 and 60 cm from a leocecal valve drilled into the loop in a “U” shape (Fig. 3). Because there was no intestinal necrosis, we performed an appendectomy and intraoperative enteral decompression without anastomoses. The pathologic examination revealed appendiceal mucinous cystadenoma (Fig. 4). After the operation, the patient’s recovery went smoothly, and the patient was discharged on the tenth postoperative day. By telephone call following up, the patient indicated that he had been free of abdominal pain, nausea bloating, etc., and no tumor recurrence was recorded over an 8 months period.
Appendiceal mucocele is a relatively rare disease, observed in 0.2% to 0.3% of appendectomies and 8% to 10% of appendiceal tumors. Appendiceal mucoceles are more frequent in women than in men, and are often discovered in patients older than 50 years. Mucocele is an uncommon pathology of the appendix characterized by a cystic dilation of the lumen as a result of an abnormal accumulation of mucus, and it usually results from either benign (cystadenoma) or malignant (cystadenocarcinoma) epithelial proliferation. Clinical manifestations of appendiceal mucoceles are quite diversified. 30% to 64% of patients present right lower quadrant pain, similar to the acute appendicitis, which is the most common manifestation. A quarter of patients are asymptomatic which are more frequently found in benign appendiceal mucocele, while appendiceal mucoceles are discovered incidentally during physical examinations (by palpation), at surgery or during imaging study for other problems. Other clinical manifestations mainly include intussusception, genitourinary symptoms, low gastrointestinal bleeding, and so on. However a bowel obstruction secondary to an appendiceal mucocele has been rarely reported. And the early diagnosis is challenging. Imaging with CT has better apply to establish a preoperative diagnosis in similar cases.

Although it would have been almost impossible to make the right diagnosis prior to the operation in this case, there are still many aspects that are worth reflecting on. First, the patient came into the emergency room with signs and symptoms of acute mechanical intestinal obstruction, and he had no previous abdominal surgery. Therefore, we almost could exclude the diagnosis of adhesive intestinal obstruction. The differential diagnosis at initial presentation included a colonic tumor, benign or malignant small intestinal tumors, and a stricture secondary to other conditions (e.g., enteroceles, volvulus, intussusception, and so on), all of which often required an operation. Second, when we take the medical history of a patient with an unexplained intestinal obstruction, we should be more careful with the patient’s past medical history and potential causes of the symptoms, these may help us make right diagnosis.
we reinquiry the patient’s case history on postoperative day, the patient had had a right lower quadrant abdominal intermittent slight chronic pain symptoms in the past 20 years, but there was no other influence in his work and daily life, so he did not care about the symptoms at all. Now if we integrated the medical history and CT, it seemed that we may have made the right diagnosis before operation. Third, we support the clinical application of total abdomen CT scanning in the case of an unexplained mechanism of abdominal obstruction. An abdominal CT is a rapid, simple, and effective means for diagnosing the location, cause, and degree of obstruction in cases of unexplained intestinal obstructions. It is particularly useful for orienting the clinician to the location of obstruction, and it would help surgeons decide where to make an incision, as an appropriately placed incision is crucial to the success of an operation.

So how did we determine the extent of surgery for the patient with appendiceal mucocoele? It is vital to discriminate benign and malignant appendiceal mucocoele in determining the extent of surgery. Appendectomy is appropriate therapy for unruptured benign appendiceal mucocoeles, like this case. When benign appendiceal mucocoele protrudes into cecal lumen, partial colectomy may be curative. “If either cecal wall or ileum is invaded by tumor or adequate surgical margins cannot be secured, ileocecal section or right hemicolectomy may be required.”[18] If malignancy cannot be exclusive, right hemicolectomy should be considered.[1,18] In this case, we should exclude the possibility of the intestine necrosis; otherwise, we must resect the highly suspected necrotic intestinal tissue further. The key-point of surgical treatment is to make resection margin clear and keep the appendiceal mucocoele intact. The prognosis for benign appendiceal mucocoele following complete ablation is excellent and the 5-year survival rate is almost 100%,[18] A patient with malignant mucocoele, however, is much poorer, and the 5-year survival rate has association with the degree of extension of the tumor and varies between 30% and 80%.[19]

4. Conclusion

For acute mechanical complete intestinal obstruction, the patient’s past medical history and potential causes of the symptoms should be paid attention to, and an abdomen CT is very necessary; Besides early operative intervention should be recommended to the patient with acute mechanical complete intestinal obstruction, especially the patient who had no previous abdominal surgery. Moreover, it is vital to discriminate benign and malignant appendiceal mucocoele in determining the extent of surgery.

References
[1] Stocchi L, Wolff RG, Larson DR, et al. Surgical treatment of appendiceal mucocoele. Arch Surg 2003;138:585–9.
[2] Ruiz-Tovar J, Teruel DG, Castañeras VM, et al. Mucocele of the appendix. World J Surg 2007;31:542–8.
[3] García Lozano A, Vazquez Terraga A, Castro García C, et al. Mucocele of the appendix: presentation of 31 cases. Cir Esp 2010;87:108–12.
[4] Caliskan K, Yildirim S, Bal N, et al. Mucinous cystadenoma of the appendix: a rare cause of acute abdomen. Ulus Travma Acil Cerrahi Derg 2008;14:303–7.
[5] Louis TH, Felter DF. Mucocoele of the appendix. Proc Bayl Univ Med Cent 2014;27:33–4.
[6] Oppeau RC, Sobinsky J, Basson MD. Appendicitis and benign appendiceal mucocoele presenting as large bowel obstruction. J Gastrointest Surg 2013;17:609–10.
[7] Pickhardt PJ, Levy AD, Rohrmann CA Jr. et al. Primary neoplasms of the appendix: radiologic spectrum of disease with pathologic correlation. Radiographics 2003;23:645–62.
[8] Pickhardt PJ, Levy AD, Rohrmann CA, et al. Primary neoplasms of the appendix manifesting as acute appendicitis: CT findings with pathologic comparison. Radiology 2002;224:775–81.
[9] Carr NJ, McCarthy WF, Sobin LH. Epithelial noncarcinoid tumors and tumor-like lesions of the appendix: a clinicopathologic study of 184 patients with a multivariate analysis of prognostic factors. Cancer 1995;75:757–68.
[10] Lee J, Lim HK, Lee SJ. Ureteral obstruction caused by mucocoele of the appendix: MDCT findings. Austras Radiol 2007;51:559–61.
[11] Kim SH, Lim HK, Lee WJ, et al. Mucocele of the appendix: ultrasonographic and CT findings. Abdom Imaging 1998;23:292–6.
[12] Pickhardt PJ, Levy AD, Rohrmann CA Jr, et al. Primary neoplasms of the appendix: radiologic spectrum of disease with pathologic correlation. Radiographics 2003;23:645–62.
[13] Papazogas B, Koutelidakis I, Tsiaousis P, et al. Appendiceal mucocoele. A retrospective analysis of 19 cases. J Gastrointest Cancer 2007;38:141–7.
[14] Garg PK, Prasad D, Aggarwal S, et al. Acute intestinal obstruction: an unusual complication of mucocoele of appendix. Eur Rev Med Pharmacol Sci 2011;15:99–102.
[15] Mourad FH, Hussein M, Balawaw M, et al. Intestinal obstruction secondary to appendiceal mucocoele. Dig Dis Sci 1999;44:1354–9.
[16] Shang J, Ruan LT, Dang Y, et al. Contrast-enhanced ultrasound improves accurate identification of appendiceal mucinous adenocarcinoma in an old patient: a case report. Medicine (Baltimore) 2016;95:e4637.
[17] Kamitori M, Mahtne K, Hrashima T, et al. Forty-three cases of obturator hernia. Am J Surg 2004;187:549–52.
[18] Park KJ, Choi HJ, Kim SH. Laparoscopic approach to mucocele of appendiceal mucinous cystadenoma: feasibility and short-term outcomes in 24 consecutive cases. Surg Endosc 2015;29:3179–83.
[19] Weber G, Terintehau G, Goudard Y, et al. Mucocele appendiculaire. Feuillets Radiol 2009;49:40–4.