

Small Bowel Volvulus Secondary to Mucinous Cystadenoma of the Appendix: A Case Report

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ABSTRACT:

Small bowel volvulus secondary to mucinous cystadenoma of the appendix: a case report

Objective: Appendiceal cystadenoma (AC) is a rare pathology diagnosed in 0.6% of all appendectomy operations. The purpose of the report was to present a rare case of ileal volvulus due to appendiceal cystadenoma, which was successfully treated with surgery.

Case: A woman aged 76 years was admitted to our emergency service with severe right lower quadrant pain. Abdominal tomography and X-ray showed features of generalized dilatation of bowel segments with periluminal fluid collections. Intraoperatively, we found bowel volvulus and a 2 cm mass at the appendiceal borders and an appendectomy was performed. The volvulated ileal segment was resected and restored with end-to-end anastomosis. Histopathologic examination of the mass revealed mucinous cystadenoma. Our patient was discharged from hospital at postoperative one week with complete clinical recovery.

Conclusion: Obstruction of the appendiceal lumen by a mucinous neoplasia can cause appendicitis followed by secondary volvulus in different parts of the gastrointestinal tract. Our case was further complicated by ileal volvulus, which led to an extremely rare presentation.

Keywords: Appendix, bowel, cystadenoma, volvulus

ÖZET:

Appendikste yer alan müsinöz kistadenomaya bağlı ince bağırsak volvulus vakası

Amaç: Appendikste oluşan kistadenoma appendektomi ameliyatlarının %0.6'sında tespit edilen nadir bir patolojik durumdur. Bu yayının amacı, apendiks kistadenomuna bağlı gelişen ve cerrahi ile başarılı bir şekilde tedavi edilen, nadir görülen ileal volvulus olgusunu sunmaktır.

Olgu: Yetmiş altı yaşındaki kadın hasta acil servisimize şiddetli sağ kadrın ağrısı ile kabul edildi. Yapılan abdominal bilgisayarlı tomografide (Abdominal BT) ve abdominal direkt grafide bağırsak segmentlerinde yaygın dilatasyon ve periluminal sıvı koleksiyonları izlendi. Yapılan cerrahi operasyonda ince bağırsak bölgesinde volvulus ve apendiks bölgesinde 2 cm'lik kitle lezyonu nedeni ile apendektomi uygulandı. Volvüle olarak izlenen ileal segment parsiyel olarak uç-uca anastamoz yapılarak rezeke edildi. Alınan kitle histopatolojik olarak müsinöz kistadenoma ile uyumlu olarak geldi. Hastamız genel durumu düzeldikten sonra bir haftalık yatış periodu sonrası taburcu edildi.

Sonuç: Apendiks lümeninde müsinöz neoplazi ile oluşan obstrüksiyon gastrointestinal sistemin değişik yerlerinden volvulus ile komplike olabilir. Vakamız ileal volvulus ile komplike olan ileri derecede nadir vaka presentasyonu özelliğini göstermektedir.

Anahtar kelimeler: Apendiks, ince bağırsak, kistadenoma, volvulus

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INTRODUCTION

As a rare pathology, appendiceal cystadenoma (AC) is diagnosed in 0.6% of all appendectomy operations. Appendiceal cystadenoma is often deformed and flat in pathologic specimens. This

situation increases the probability of misdiagnosis (1,2). Mucinous cystadenomas (MC) are composed of villous adenomatous changes of the appendiceal epithelium, which are the result of mucin distension of the appendiceal lumen. This condition can present as right iliac fossa pain as with acute

appendicitis; however, about 25% of the cases are asymptomatic (3,4). When such tumors are diagnosed as appendicitis, complete removal is mandatory because of the cystadenocarcinoma presentation (5).

The self-rotation of bowel segments can be a cause of lumen obstruction, which is called volvulus. Small bowel volvulus is a rare disorder reported infrequently in the literature. Small bowel volvulus accounts for 3-6% of all causes of small bowel obstruction (6,7).

Torsion of the bowel secondary to appendiceal mucocele has rarely been reported and preoperative diagnosis is an unusual feature (8-10). The aim of this report was to present a rare case of ileal volvulus due to appendiceal cystadenoma, which was treated successfully with surgery.

CASE REPORT

A woman aged 76 years was admitted to our emergency service with severe right lower quadrant pain, abdominal distention, and vomiting. The symptoms started gradually one week before admission and increased progressively. She also had weight loss of approximately 5% during this time period. The patient expressed an inability to defecate. Her first examination revealed a blood pressure of 88/45 mmHg; heart rate: 98/min; respiration rate, 23/min; and body temperature: 38.5°C. The patient reported minimal rectal bleeding, which occurred only once. She had a history of diabetes and hypertension and had no history of abdominal operation. There was no organomegaly on abdominal palpation. Additionally, physical examination revealed severe tenderness in the right side of the abdomen. Laboratory test results on admission were as follows; Hb: 11.6 g/dL, white blood cell (WBC): $7.45 \times 10^3/L$, K: 3.27 mmol/L, Albumin: 2.61 g/dL, Cl: 116 mmol/L, Fibrinogen: 437 mg/dL, C-Reactive protein: 26 mg/L, and INR: 1.32 with normal liver function. Abdominal tomography showed features of generalized dilatation of bowel segments with periluminal fluid collections. There was also a conglomeration in the lower right intestinal segments (Figure-1,2).

Dilatation of the bowel segments in the X-RAY Dilatation of the bowel segments in the CT

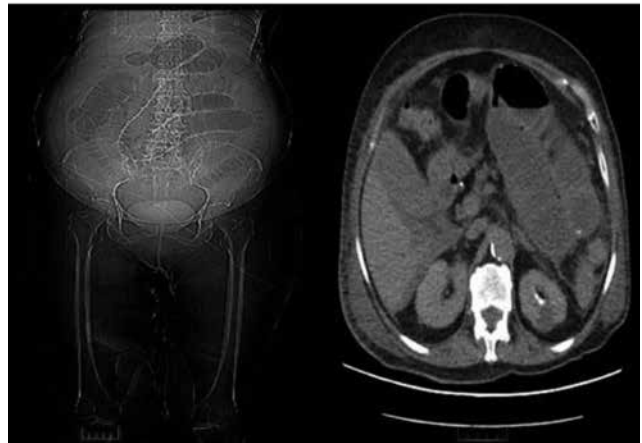


Figure-1: Dilated bowel segments in the CT and abdominal radiologic imaging

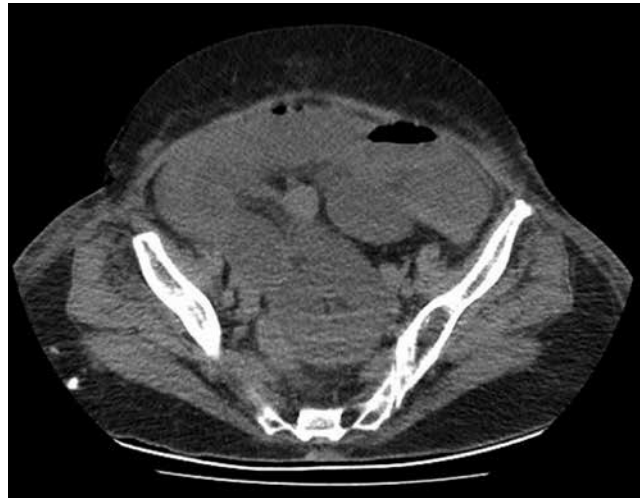


Figure-2: The abdominal tomography of dilated bowel walls in the ileocolonic junction

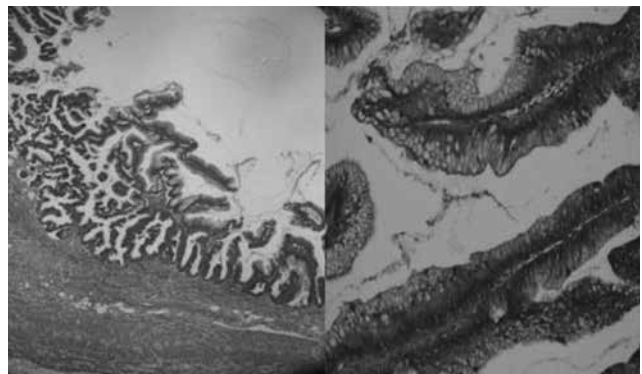


Figure-3: The cystic surface epithelium was composed of columnar cells with basally located elongated hyperchromatic nuclei and cytoplasmic apical mucin.

Immediate surgery was performed because of the rapid deterioration of the patient. Upper and lower limited midline incisions were performed after general anesthesia. The intraoperative examination revealed that the ileum had rotated totally on itself, the mesentery and appendix were at the center of the rotation. The rotated ileal segment had a pale red appearance, which could be a sign of strangulation. When the ileum was re-rotated, it was observed that an ischemic segment with necrosis started 30 cm proximal to the ileocecal valve and stretched 60 cm proximally. This segment was resected and restored with a hand-made end-to-end anastomosis. The anastomosis was performed with a single-layer silk suture. We found a 2 cm mass lesion at the appendiceal borders and appendectomy was also performed. With the exception of minimal ascites, the other abdominal organs were observed as normal during the intraoperative examination. All operation specimens were sent for histopathologic examination.

The histopathologic examination revealed hemorrhagic necrosis within the ileal segments that elongated up to the serous borderline. There were also adenomatous changes including cystic mucinous pathologic appearances (mucinous cystadenoma) on the appendix (Figure-3).

The patient continued with medication and nutritional treatment regimens in her postoperative period. No postoperative complications developed and the patient was discharged on the 7th postoperative day. The patient was monitored monthly in our department during the following 6 months. There were no complications in that time period.

DISCUSSION

Small bowel volvulus (SBV) is an infrequent disorder in the adult population. This disorder occurs more often in African and Asian countries (11,12). The presentation of volvulus due to appendiceal mucinous tumor is an extremely rare disorder. Our presentation underwent urgent surgery and dramatically recovered after an exact diagnosis. Following an intensive search of the literature, we

believe that this is the first case reported, of small bowel volvulus due to an appendiceal mucinous cystadenoma. We suspect that the obstructed inflammation of the appendiceal lumen led to a sudden malrotation of the ileal segment, followed by ischemia of this intestinal loop. However, a small number of SBVs due to appendicitis have been presented in the literature. Ben Ely et al. (13) reported 8 patients with intestinal malrotation who presented with appendicitis. All patients were evaluated retrospectively with computerized tomography (CT) scans, which showed a complete malrotation with right-sided duodenum and jejunum, and left-sided colon. All eight patients had an abnormal superior mesenteric artery – superior mesenteric vein relationship and a dysplastic uncinat process of the pancreas (13). In contrast, our patient presented after an obstructed appendix due to cystadenoma with normal anatomic structures, with no congenital abnormality or malrotation.

Preoperative diagnosis of appendiceal mucocoele is difficult. The presentation can be appendicitis or abdominal mass. Other unusual presentations include intestinal intussusception, intestinal obstruction, and obstructive uropathy (14). Rudloff and Malhotra (15) reported a 720° torsion of a gangrenous mucocoele around the proximal part of the appendix. The evaluation of the pathologic sample revealed a hemorrhagic, gangrenous appendix, which was thin-walled and filled with an opaque, mucoid substance. Mucocoeles of the appendix are categorized histopathologically as representing hyperplasia, mucinous cystadenoma, or mucinous cystadenocarcinoma (16). Our presentation included a benign mucinous cyst that had additional neoplastic differentiations. The mucinous cyst caused an elongation of the obstruction throughout the middle ileal segments, which presented as volvulus and ischemia.

There have been volvulus presentations secondary to appendiceal mucinous cysts in other parts of the gastrointestinal tract. One report presented a mucinous cystic neoplasm that resulted with a volvulus in the cecum. The tumor elongated from the appendix and was detected in the cecum, at the border of the appendix and cecum (17). The

cystic neoplasm in that case report was greater in diameter than our presentation. The cyst extended to the distal side of the gastrointestinal tract and caused volvulus in the cecum. In our case, the traction force extended to the proximal tract, which caused an intraperitoneal aberrant rotation and intestinal obstruction. Our cystic disorder was smaller than in this report.

Chong et al. (18) presented a case of strangulated small bowel due to cystadenoma of the vermiform appendix, which was treated surgically with a limited right hemicolectomy and ileocolonic anastomosis. In our case, we found an obvious

volvulus and elongated strangulation on the ileal segments. We resected the strangulated ileal segments using a hand-made end-to-end anastomosis and an appendectomy was performed later during the same operation.

In summary, mucinous cysts, which include neoplastic and non-neoplastic subtypes, may lead to acute abdominal presentations. Obstruction of the appendiceal lumen by mucinous neoplasia may cause appendicitis followed by secondary volvulus in different parts of the gastrointestinal tract. Our case was further complicated by ileal volvulus, which led to an extremely rare presentation.

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