Case Report
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Mucocele appendicular, review of the bibliography and presentation of clinical cases

Abstract

An appendicular mucocele is defined as an obstruction of the appendix caused by intraluminal mucus. It is a rare condition whose etiology is not fully understood. Given its minimal symptoms and the nonspecific nature of laboratory findings, the diagnosis is usually incidental. Treatment always involves appendectomy, and right hemicolectomy and adjuvant chemotherapy may be necessary. We present four clinical cases of appendiceal mucinous neoplasms with different clinical presentations and outcomes.

Keywords: appendiceal mucocele, appendicitis, peritonitis

Volume 14 Issue 2 - 2026

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Received: July 10, 2026 | **Published:** July 22, 2026

Introduction

Mucinous tumors can originate in various sites, including the appendix, ovary, colon, pancreas, and gallbladder. They are the second most common tumors affecting the appendix after carcinoid tumors. The appendiceal mucocele was first described in 1842 by Rokitansky.[1] It is a rare entity accounting for a small proportion of appendiceal lesions. Although there is some disagreement on this point, most published studies agree that both sexes are affected, with a slight predominance in females; the average age ranges from 40 to 70 years.[2]

Clinical presentation is generally nonspecific and may manifest as pain in the right iliac fossa mimicking appendicitis; however, in most cases, the diagnosis is incidental.[2,3] The definitive diagnosis is made through histology and immunohistochemistry; imaging-based diagnosis is rare given the characteristics of the lesion.[3] Treatment is always surgical, involving appendectomy, and may require right colectomy and adjuvant chemotherapy in cases of peritoneal pseudomyxoma, a complication that occurs in up to 15% of cases.[4]

Clinical cases

We present 4 clinical cases of appendicular mucocele with different clinical presentations.

Case 1

A 67-year-old woman with hypertension, hypothyroidism, and diabetes. During a routine gynecological physical examination at a polyclinic, a solid, irregular, and mobile right parauterine mass was detected. A contrast-enhanced computed tomography (CT) scan of the abdomen and pelvis revealed a well-defined ovoid mass measuring 90 × 40 mm with homogeneous, water-density content and no enhancement (Figure 1). The requested tumor markers were CA 125: 8.2, CEA: 15.6, and CEA 9-9: 32. Given the clinical and imaging findings, the gynecology team recommended exploratory laparoscopy to perform a right adnexectomy.

Intraoperatively, a whitish appendicular mass was identified, approximately 10 cm in size with an healthy and healthy adnexa (Figures 2 & 3). The general surgery team was summoned and performed an appendectomy; the intact surgical specimen was removed and placed in a bag. The patient had a favorable postoperative course and was discharged from the hospital after 48 hours. The delayed pathological report indicated a low-grade mucinous appendiceal neoplasm. No involvement of the appendiceal base. pTNM (AJCC 9th Edition) pTis (5).

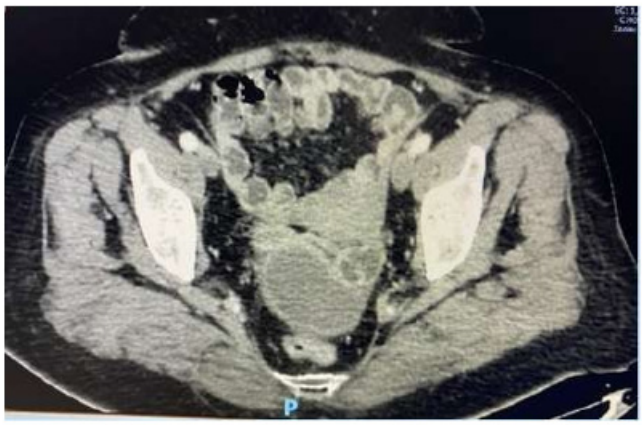

Figure 1 CT showing cystic pelvic mass

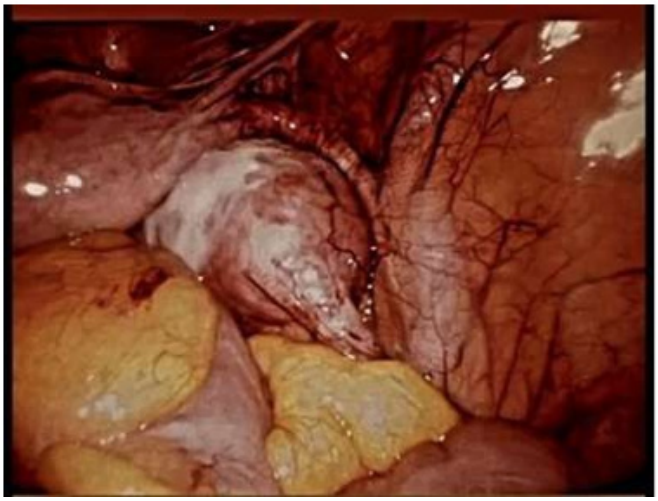

Figure 2 Laparoscopic appendiceal mucinous mass

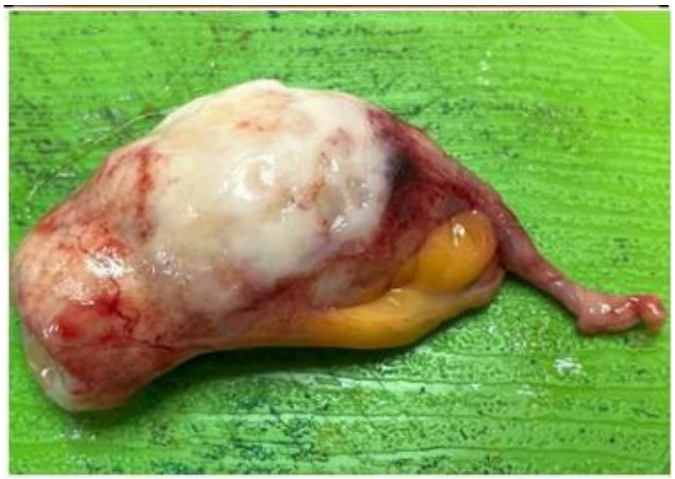

Figure 3 Resected appendiceal neoplasm specimen

Case 2

A healthy 46-year-old man presented to the emergency department with pain in the right iliac fossa (RIF) that occurred while at rest; the pain was constant, of low intensity, and radiated to the hypogastrium, with no fever, vomiting, or other accompanying symptoms. A CT scan revealed a 70-mm structure in the RIF, containing fluid, with sparse thin septa and small calcifications; a mesenteric cyst was suspected (Figure 4).

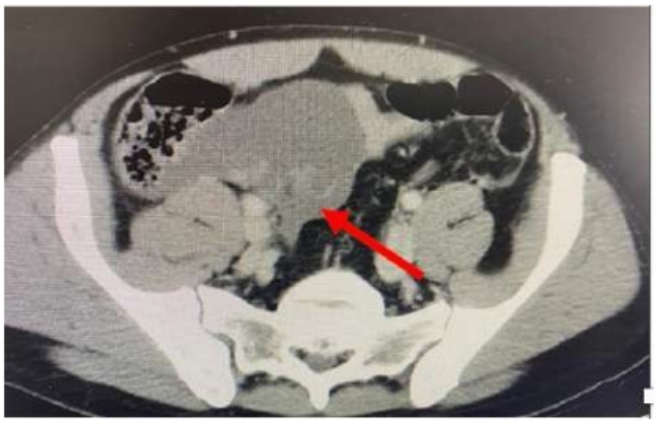

Figure 4 CT showing right iliac cyst

Tumor markers were requested: CEA 6.8, CA19-9 6.4. An exploratory laparoscopy was performed, revealing abundant mucoid contents in the pelvis and a perforated cecal appendix with an intact appendix base. An appendectomy was performed, the specimen was removed in a bag, the mucoid contents, and thorough peritoneal lavage (Figure 5). The pathology report indicated mucosecretory mucosal hyperplasia. No malignancy.

Case 3

A 70-year-old woman with hypertension. She presented to the emergency department with a 6-day history of constant lower right quadrant (LRQ) pain radiating to the hypogastrium, without accompanying symptoms. On examination, she had a distended abdomen that was tender to palpation in the LRQ without guarding. Blood tests were unremarkable. A CT scan revealed a distended cecal appendix, 36 mm in diameter, with hypodense contents and high-density images at its base. The adjacent fat was altered (Figure 6).

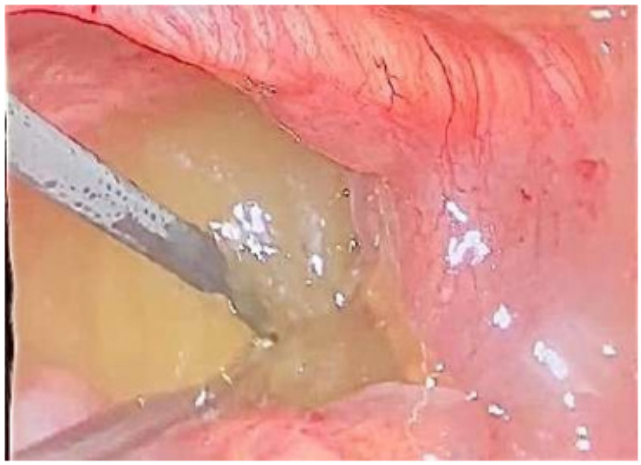

Figure 5 Laparoscopic perforated appendiceal mucocele.

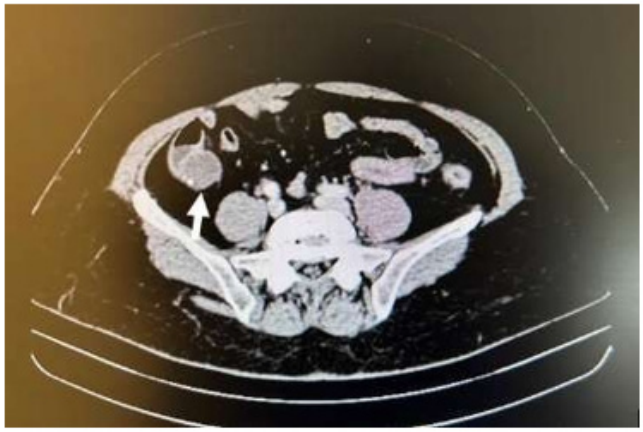

Figure 6 CT showing appendiceal mucocele

A laparotomy was performed, revealing an appendiceal mucocele appendicular involving the appendix base. Given the involvement of the appendix base, an appendectomy with cecectomy and ileocolic anastomosis was performed (Figure 7). Pathology reported a low-grade mucinous neoplasm of the appendix with clear margins. Lymph nodes 0/11. pT3N0 (5) Good postoperative course; the patient was discharged on the 5th day.

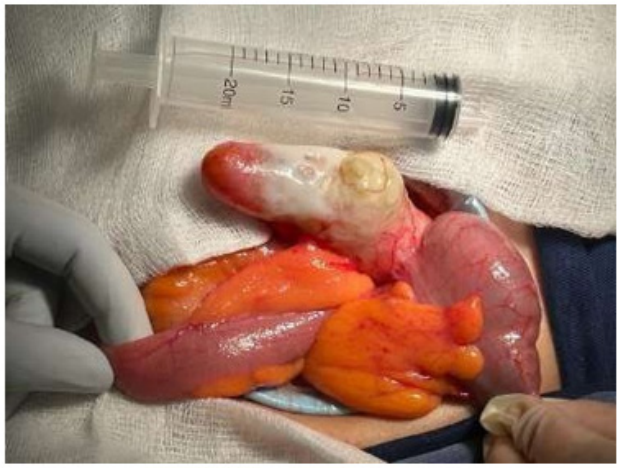

Figure 7 Appendectomy with partial cecectomy specimen

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Case 4

24-year-old male presenting with symptoms that have been present for 48 hours, including intense, colicky pain in the lower left quadrant of the abdomen, accompanied by a fever of up to 38. Blood tests reveal leukocytosis of 14,000 and C-reactive protein of 40. The patient has a distended abdomen, tenderness and guarding in the lower left quadrant, with tenderness extending to the lower left quadrant and hypogastrium. A CT scan of the abdomen and pelvis revealed an abscessed appendicular plastron.

An exploratory laparoscopy was performed, revealing an appendiceal plastron with an intact base, a perforated appendiceal apex with mucus protruding, and one of the walls of the plastron formed by the lateral surface of the sigmoid colon, which was perforated. The procedure was converted to a laparotomy, and an appendectomy and Hartmann's procedure were performed (Figure 8).

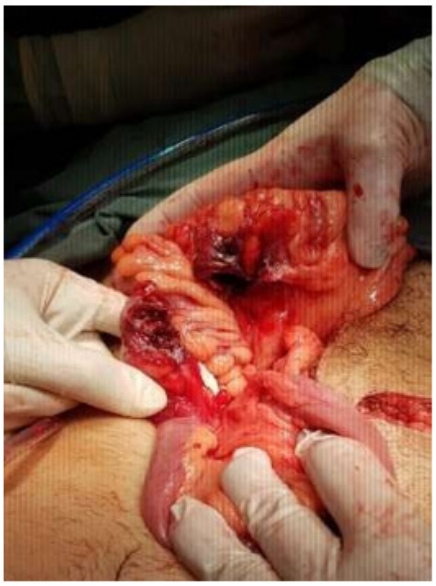

Figure 8 Hartmann procedure with appendectomy

The pathology report indicates a low-grade mucinous neoplasm of the appendix. pT3N0[5] 6 months later reconstruction reconstruction is performed of the traffic with no evidence of macroscopic abnormalities. Good subsequent outcome.

Discussion

Appendicular mucocele is a rare condition with an incidence of approximately 2%. It is characterized by cystic dilation of the lumen of the ileocecal appendix due to an obstructive etiology, leading to retrograde accumulation of mucoid material.[2] Using the classification of *the Peritoneal Surface Oncology Group International* (PSOGI), we can distinguish between neoplastic and non-neoplastic lesions; the latter correspond to those that do not exhibit dysplasia or hyperplasia, among which are the simple mucocele or retention cyst. On the other hand, neoplastic lesions can be classified as polyps, appendiceal mucinous neoplasms, adenocarcinomas,and pseudomyxoma.[6] The serrated polyps of the appendix are hyperplastic lesions, with or without dysplasia. They typically have *KRAS* mutations but lack *BRAF* mutations.[6]

Peritoneal pseudomyxoma is a rare malignant tumor that may result from the perforation of an appendiceal tumor (whether malignant or low-grade), leading to the spread of mucin throughout the peritoneal cavity (6,4). Peritoneal pseudomyxoma may present with more insidious symptoms, characterized by a gradual increase in abdominal girth due to intraperitoneal mucin accumulation.[7] In 25–50% of patients, the diagnosis is incidental, discovered during imaging studies, surgical procedures, or endoscopic examinations performed for other reasons.[3] Mucinous appendiceal adenocarcinoma is characterized by infiltration of the muscularis propria and can be classified as well-, moderately, or poorly differentiated. The presence of signet-ring cells indicates a poorly differentiated tumor.[6]

The symptoms, as mentioned, are usually nonspecific and may include abdominal pain, nausea, vomiting, and/or fever; therefore, various differential diagnoses are typically considered, such as acute appendicitis, adnexal, or urinary tract disorders.[2] In the cases discussed, most patients presented with pain in the right lower quadrant or hypogastrium, and only one case was accompanied by fever.

Regarding laboratory tests, the latest PSOGI recommendations advocate for baseline measurement of CEA and CA 19.9, as these markers are elevated in most patients with advanced mucinous appendiceal tumors (67%) and are associated with an increased risk of recurrence and death.[4] Only one of our patients presented CEA positive.

Abdominal ultrasound is generally insufficient for diagnosis; however, contrast-enhanced CT of the abdomen and pelvis is the most commonly used imaging method for diagnosis and, in many cases, can reveal appendiceal abnormalities similar to appendicitis (as reported in cases 3 and 4) or adnexal mass (as in case 1).[3]

Appendectomy is always the cornerstone of treatment, given the unknown risk of progression from a benign to a malignant lesion and the risk of rupture with pseudomyxoma formation.[4,7] The procedure should be tailored to the findings: appendectomy is recommended for benign lesions confined to the cecal appendix, while cecal resection or hemicolectomy is recommended when the appendicular base is involved, regional lymphadenopathy is present, or pseudomyxoma is detected.[4]. Regarding treatment, appendectomy with clear margins is sufficient for patients with mucinous appendiceal neoplasia.[4]

The choice of surgical approach is controversial in the literature. It has been suggested that the laparoscopic approach may increase the risk of rupture and peritoneal seeding; therefore, some authors advocate conversion upon intraoperative diagnosis; however, studies comparing the two approaches have suggested that laparoscopic resection without effusion or rupture is feasible and appropriate, provided that the specimen is removed in a bag, as was done in Case 1.[7]

There is no consensus on the management of perforation with local effusion, with or without positive margins. If the effusion consists solely of acellular mucin without epithelial cells, the recurrence rate is estimated to be between 3% and 7%.[4] This low recurrence rate has led many authors to support a close-follow-up approach for selected patients, since no additional benefits were observed with right hemicolectomy compared to appendectomy alone.[4,8]

Prognosis

Regarding prognosis, the survival rate for simple mucoceles and cystadenomas after surgery is excellent, with a 10-year survival rate of 90–100%.[2] Cystadenocarcinomas respond well to resection. Peritoneal pseudomixoma has a 5-year survival rate of 25%.[7]

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Conclusion

Appendicular mucocele is a rare condition with nonspecific clinical and laboratory findings that may mimic appendicitis, appendicular plastron, or an ovarian or cecal mass. As soon as a diagnosis is made or suspected, surgical treatment should be performed given the risk of perforation and peritoneal contamination.

Laparoscopic appendectomy is sufficient for patients without involvement of the appendiceal base; however, colonic resection may be necessary for those with base involvement or penetration. Tumor markers are useful for predicting disease-free survival and overall survival. In patients with negative tumor markers, resection with clear margins, and histopathology showing low-grade lesions, the prognosis is very favorable.

Acknowledgements

None.

Conflicts of interest

The authors declare that there are no conflicts of interest.

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