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Appendiceal Mucocele Spontaneously Drained into the Cecum: Report of a Case

Çekuma Spontan Olarak Drene Olmuş Olan Apendiks Mukoseli: Olgu Sunumu

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ABSTRACT

We aimed to present a case of appendiceal mucocele who was admitted with abdominal pain, operated under elective conditions and found to have been spontaneously drained from the root of the appendix to the colon in surgical exploration. A 41-year-old male patient was admitted to the emergency department with abdominal pain. In clinical examination; tenderness, rebound and defense in the lower right quadrant of abdomen and leucocytosis were detected. Computerized tomography scan showed the appearance of a cystic dilated appendix. After resolution of the physical examination findings based on the antibiotic treatment, we decided to perform elective surgery. In surgical exploration, ileocecal resection was performed by determining the mucocele of the appendix that had been spontaneously drained from the root of the appendix into the cecum lumen. On the sixth day after the operation, he was discharged without complications. Histopathological examination revealed high-grade appendiceal mucinous neoplasia. Our patient's follow-up in the first year after the surgery revealed no recurrence. The patient is disease-free at the end of his first postoperative year. The mucocele of the appendix is a cystic neoplasia containing mucin and its perforation during surgery may result in the development of pseudomyxoma peritonei in the late period. In our case, the fact that the lesion was spontaneously drained into the lumen of the colon reduced the risk of the mucocele rupturing and spreading into the abdomen during surgery.

Keywords: Appendix, neoplasms, mucocele, cecum, general surgery

ÖZ

Karın ağrısı şikayeti ile başvuran, elektif şartlarda ameliyat edilen, cerrahi eksplorasyonda apendiks kökünden kolona spontan drene olmuş olduğu saptanan apendiks mukoseli olgusunu sunmayı amaçladık. Karın ağrısı şikayeti ile acil polikliniğe başvuran 41 yaşında erkek hastada sağ alt kadranda hassasiyet, defans, *rebound* bulguları ve lökositoz saptandı. Bilgisayarlı tomografide kistik dilate apendiks görünümü izlendi. Antibiyotik tedavisine başlanan hastanın fizik muayene bulgularının gerilemesinin ardından elektif ameliyat kararı alındı. Cerrahi eksplorasyonda; apendiks kökünden çekum lümeni içerisine spontan drene olmuş olan apendiks mukoseli saptanarak ileoçekal rezeksiyon uygulandı. Takiplerinde komplikasyon gelişmeyen hasta ameliyat sonrası altıncı günde taburcu edildi. Histopatolojik inceleme sonucunda yüksek dereceli apendiseal müsinöz neoplazi saptandı. Hastamızın ameliyat sonrası birinci yılındaki takipleri nükssüz devam etmektedir. Apendiks mukoseli, müsin içeren kistik bir neoplazi olup cerrahi sırasında perfore olması geç dönemde psödomiksoma peritonei gelişimi ile sonuçlanabilir. Olgumuzda lezyonun kolon lümenine spontan olarak drene olmuş olması, ameliyat sırasında mukoselin rüptüre olarak karın içerisine yayılması riskini azaltmıştır.

Anahtar Kelimeler: Apendiks, neoplaziler, mukosel, çekum, genel cerrahi

Introduction

Appendiceal mucocele is a rare formation that causes obstructive dilatation in the appendix by the accumulation of mucoid material in the lumen (1). We aimed to present the case of appendiceal mucocele who was admitted with abdominal pain, operated under elective conditions in our clinic and found to have been spontaneously drained from the root of the appendix to the colon during the exploration.

Case Report

Forty-one-year-old male patient was admitted to the emergency department due to increased abdominal pain complaints lasting about five months. The patient had no known history of chronic disease, and physical examination showed signs of abdominal tenderness, defense, and rebound in the lower right quadrant. Leukocyte count as 17000/mm^3 and c-reactive protein value as 8.66 mg/dL were revealed in the

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laboratory tests. Computed tomography (CT) showed the appearance of cystic dilated appendix associated with cecum and lumen filled with fluid, no appearance consistent with rupture or intra-abdominal fluid collection (Figure 1). Having obtained signed informed consent from the patient, he was admitted to the General Surgery Department with a referral diagnosis of appendiceal mucocele and he was treated with antibiotics. After the abdominal pain, physical examination and laboratory findings declined, we decided to perform the operation under elective conditions during his hospitalization. Surgical exploration revealed a lesion consistent with cystic neoplasia, which originated from the appendix, showed adhesion to the anterior wall of the abdomen and meso of the ileum, with low luminal pressure. When the dissection was continued, we determined that this lesion was an appendiceal mucocele of approximately 15x6 cm which had been spontaneously drained from the root of the appendix into the cecum lumen (Figure 2). Frozen section examination was performed of the peritoneal area of the adhesion to the anterior abdominal wall and it was reported that fat and connective tissues containing chronic inflammatory elements were observed and no mucin or glandular structures were seen. Extensive ileocecal resection with safe surgical margins was performed in the case with no spread that could have been caused by luminal opening, perforation or intra-abdominal implantation. The patient with no complications during his postoperative follow-up period was discharged with surgical recovery on his sixth postoperative day. Histopathological examination of the specimen revealed high-grade appendiceal mucinous neoplasia confined to the mucosa with no signs of invasion. No lymphovascular invasion, perineural invasion or tumor deposit were observed in the patient with negative surgical margins and signs of reactive hyperplasia were observed in 42 dissected lymph nodes. Our patient's first year follow-up after the surgery has been continuing without any late-term complications or recurrence.

Discussion

Appendiceal mucocele, first described by Rokitansky in 1842, is a clinical entity describing neoplasia with malignant potential and containing mucin (1,2). In various series, it has been reported to occur in 0.2-0.7% of appendectomies (3).

Most of the patients are asymptomatic, and symptoms such as abdominal pain, intra-abdominal mass and weight loss are most commonly seen in symptomatic patients. Eight percent of patients are admitted with clinical features of acute appendicitis (4). Diagnosis is usually incidental during surgical exploration with the prediagnosis of acute appendicitis in emergency cases or made by ultrasonography and/or computed tomography in cases investigated under emergency or elective conditions.

Four different histopathological types of mucocele are present; 29% are simple mucocele, 31-34% are epithelial hyperplasia or musinous cystadenoma and 5% are mucinous cystadenocarcinoma (3). Median age of patients diagnosed with malignant musinous adenocarcinoma is 60 and five-year survival is around 58% (5). According to pathological terminology reported in a current consensus study, lesions that show architectural features of low-grade mucinous neoplasia and do not show infiltrative invasion but contain high-grade cytological atypia are classified as high-grade appendiceal mucinous neoplasia (6). Histopathological examination of our case showed high-grade appendiceal mucinous neoplasia confined to the mucosa, which showed no signs of invasion, and no findings of carcinoma.

Appendiceal mucoceles are usually asymptomatic (7). In the study of Lien et al. (8), when the external diameter of the appendiceal mucocele was 15 mm or greater, the sensitivity and specificity of CT were determined as 83% and 92%. These lesions detected in CT are usually located in the lower right abdominal quadrant and characterized by the appearance of an encapsulated cystic mass with a diameter of 2-20 cm contains calcification by 50% (7). In our case who was admitted with abdominal pain, an encapsulated cystic dilated lesion was observed in the lower right abdominal quadrant.

The standard treatment for the mucocele of the appendix is resection with safe surgical margins. Some factors such as the size of the mucocele,

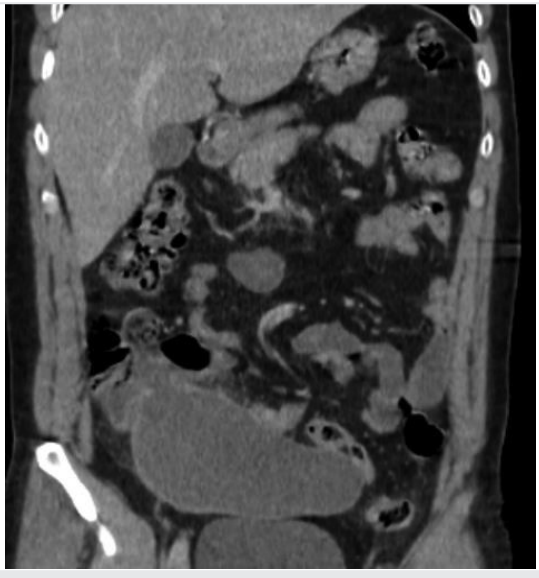

Figure 1. Coronal cross-section image of the appendiceal mucocele in computed abdominal tomography

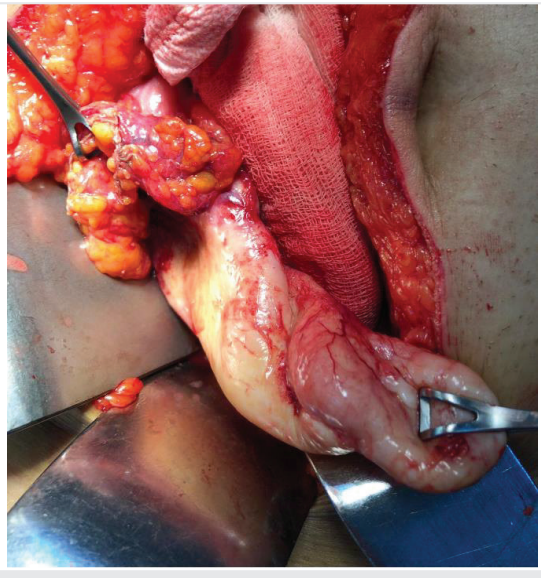

Figure 2. Surgical image showing easy manipulation of the appendiceal mucocele due to low luminal pressure

involvement of the root of the appendix and histopathological findings may affect the extent to which the resection should be and there is still no consensus on the breadth of optimal surgery (9). If there is only one simple mucocele confined to the appendix and if the root of the appendix is intact, appendectomy is sufficient while cecum resection is suggested in the presence of root involvement. On the other hand, ileocecal resection or right hemicolectomy is recommended if there is a high suspicion of malignancy in patients with mucocele involvement to the cecum wall and/or ileum (10). However, it has been reported in few studies that right hemicolectomy may not offer a survival advantage compared to other surgical methods (9). Laparoscopic surgery is a viable method however, open surgery may be preferred due to the risk of perforation (1,9). During open surgical exploration, ileocecal resection was performed in our case after visualizing that the lesion started from the base of the cecum and extended along the entire appendix.

In cases of appendiceal mucocele, it is necessary to make sure that the lesion is not perforated during surgery. Because, peritoneal implantation of mucinous material in perforated patients may result in pseudomyxoma peritonei in the late-term follow-up (7). In our case, it was seen that mucocele content was spontaneously drained providing internal drainage to the cecum and also decompressed the luminal pressure in the lesion and facilitated easier manupilation during surgery and reduced the risk of perforation.

Conclusion

Appendiceal mucocele is a rare cystic neoplasia containing mucin and its treatment is surgical. Its perforation during surgery may result in the development of pseudomyxoma peritonei in the late period. In the case presented, spontaneous drainage of the lesion to the lumen of the colon reduced the risk of the mucocele rupturing into the abdomen during surgery.

Ethics

Informed Consent: Written informed consent was obtained from the patient.

Peer-review: Externally and internally peer-reviewed.

Authorship Contributions: Surgical and Medical Practices - T.E., M.S.Ö., O.A.; Concept- T.E., A.G., M.S.Ö., Ö.E., O.A.; Design- T.E., F.A., Ö.E., O.A. ; Data Collection or Processing- T.E., A.G., F.A., M.S.Ö., Ö.E.; Analysis or Interpretation- T.E., Ö.E., O.A.; Literature Search- T.E., A.G., F.A., M.S.Ö. ; Writing- T.E., A.G., F.A., Ö.E.

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