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Low-grade mucinous neoplasia in a cecal diverticulum: A case report

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ABSTRACT

INTRODUCTION: Low-grade mucinous neoplasia is an uncommon benign tumor that develops in the appendix. The development of mucocele disease has never been reported in a colonic diverticulum. We present a case developing low-grade mucinous neoplasia in a cecal diverticulum.
PRESENTATION OF CASE: A tumor in the ileocecal region was found during a medical examination of a 66-year-old woman. Three months later, the tumor was still present and the patient developed abdominal pain. Laparoscopic ileocecal resection with D2 lymph node dissection was performed. Histopathological examination revealed a low-grade mucinous neoplasm in a cecal diverticulum.
DISCUSSION: Colonic mucoceles reportedly originate from the appendix. There are no previous reports of mucocele disease in a colonic diverticulum worldwide. This report reviews and discusses the management of the appendiceal mucoceles.
CONCLUSION: The incidence of colonic diverticula has recently begun to increase in Japan. The possibility of a mucocele within a colonic diverticulum should be considered in patients with submucosal colonic tumors.

1. Introduction

Low-grade mucinous neoplasia is an uncommon benign tumor that develops in the appendix. A mucocele of the appendix was first described by Rokitansky in 1842 [1]. Mucoceles are encountered in low percentages (0.004–4.1%) of appendectomies [2,3]. However, the development of mucocele disease in a colonic diverticulum has never been reported. We herein present the first case of low-grade mucinous neoplasia in a cecal diverticulum.

2. Presentation of case

A tumor in the ileocecal region was found by abdominal computed tomography (CT) during a medical examination of a 66-year-old woman. The tumor was still present on abdominal CT 3 months later, and she was referred to our hospital for a detailed examination.

Colonoscopy revealed a submucosal tumor (SMT), and the patient experienced abdominal pain. Therefore, she was referred to our surgical department for an operation. Her medical history included a total hysterectomy at 33 years of age, hyperlipidemia, and a pancreatic cyst. Laboratory studies revealed normal levels of carcinoembryonic antigen (CEA) (1.1 ng/μL) and CA19-9 (14.3 U/μL). Abdominal CT revealed an 18- × 36-mm tumor in the ileocecal region (Fig. 1). Colonoscopy revealed an approximately 40-mm-diameter SMT in the ileocecal region. It was difficult to insert the endoscope to the mouth side of the tumor. The orifice of the diverticulum in this region was not identified (Fig. 2). Examination of the biopsy specimens indicated no malignancy.

A gastrointestinal stromal tumor, lipoma, or appendiceal mucocele was suspected, and the possibility of a malignant tumor could not be ruled out. Thus, we performed a laparoscopic ileocecal resection with D2 lymph node dissection. We used five ports: one 12-mm camera port in the umbilical region, one 12-mm port in the left upper abdominal wall, and three 5-mm ports in the right upper abdominal wall and both sides of lower abdominal wall. Intraoperative examination revealed no ascites or adhesion. The tumor was located in the cecum, and there was no continuity between the tumor and appendix (Fig. 3). We could not determine whether the mass was benign or malignant; therefore, we performed ileocecal resection (D2 lymph node dissection). The umbilical wound was extended to 35 mm by an additional incision, and the colon was reconstructed by functional end-to-end anastomosis. The resected tumor measured 35 × 30 mm and was located in the ileocecal region; it was not continuous with the appendix (Fig. 4). Histopathological examination revealed a cystic mass with mucinous collection in the muscularis propria. The colonic lamia propria curved within the muscularis propria; therefore, this fistula was considered to be a cecal diverticulum. Because

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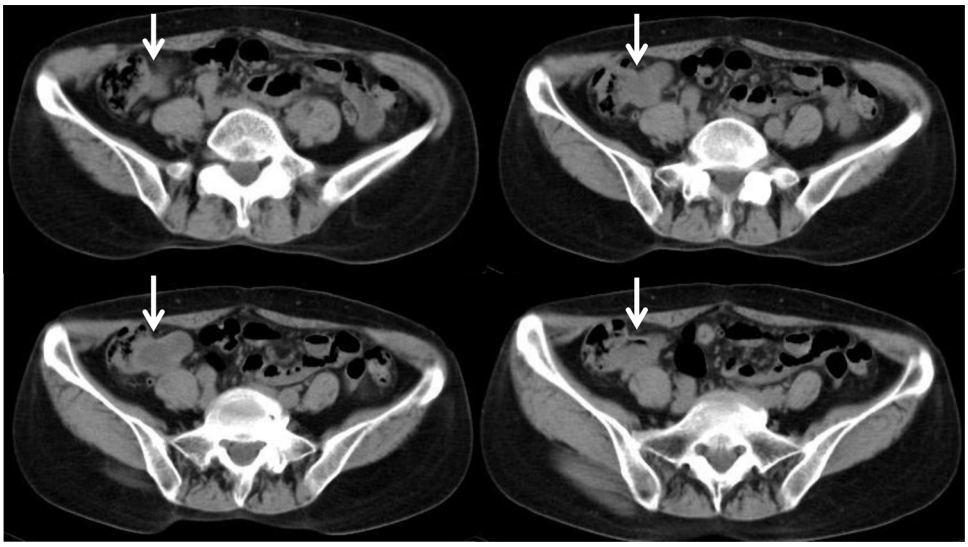

Fig. 1. Abdominal CT revealed an 18- × 36-mm tumor,in the ileocecal region (white arrow).

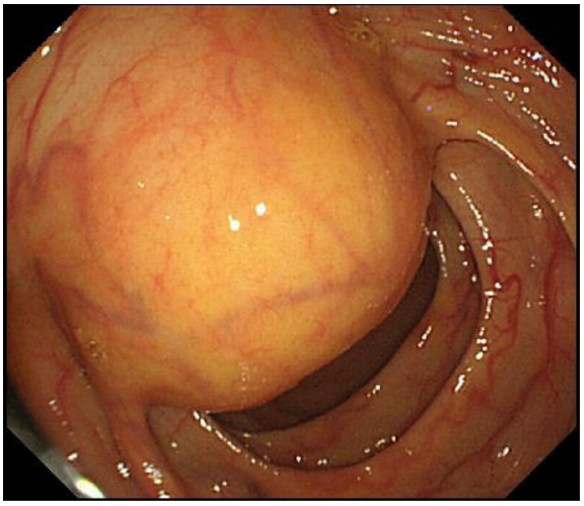

Fig. 2. Colonoscopy revealed a submucosal tumor in the ileocecal region. It was difficult to insert the endoscope to the mouth side of the tumor. The orifice of the diverticulum was not found.

the cystic mass was continuous with the fistula, we considered that the cystic mass was present within the diverticulum. Histopathological examination revealed that the colonic lamia propria curved within the muscularis propria; there was continuity in the colonic lamia propria of the surface and the muscularis propria. The orifice of the diverticulum was present on the surface of the colonic lamia propria, therefore it was considered to be cecal diverticulum. There was a cystic mass with mucinous collection in the muscularis propria. So the cystic mass was present within the diverticulum. Although epithelial neoplastic transformation was evident in the wall of the cystic mass as shown by cells exhibiting increased nuclear size and chromatin, these malignant cells were not present in the cystic tumor. These histological features were consistent with a low-grade mucinous neoplasm (Fig. 5). Because the tumor was not continuous with the appendix, we diagnosed a low-grade mucinous neoplasm in a cecal diverticulum.

The patient recovered uneventfully and was discharged from our hospital on postoperative day 6. She had no evidence of recurrence 12 months postoperatively.

3. Discussion

Colonic mucoceles reportedly originate from the appendix. Mucoceles of the appendix have no specific clinical presentation; they may be discovered incidentally, in patients with chronic abdominal pain, in patients with a palpable mass in the right lower abdomen, by CT or ultrasound examination, or in patients with an intestinal tract invagination or ileus.

In patients with low-grade mucinous neoplasms, laboratory test results are not specific, but some reports have described high serum CEA levels [4]. In our case, The CEA level was normal. Abdominal CT in patients with mucoceles of the appendix shows a well-encapsulated cystic mass that sometimes exhibits mural calcification [5,6]. Colonoscopy reveals the “volcano sign,” which is characterized by localization of the appendiceal orifice at the center of the mound formed by a submucosal mass [7]. Magnetic resonance imaging reveals mucin as a low- and high-density region on T1- and T2-weighted images, respectively [8]. However, establishment of a preoperative diagnosis may be difficult. In our case, a submucosal mass was found in the cecal lumen, but the diverticular orifice was not found in the vicinity of the tumor on colonoscopy. CT showed no calcification in the mass region. Thus, we could not determine preoperatively whether the tumor was benign or malignant.

Appendiceal mucoceles are treated by surgical resection. Approximately 90% of appendiceal mucoceles are benign [9]; therefore appendectomy or ileocecal resection without lymph node dissection is sufficient. As previously described, however, preoperative definitive diagnosis may be difficult; thus, ileocecal resection or right hemicolectomy with lymph node dissection is often performed. In our case, we performed laparoscopic ileocecal resection with D2 lymph node dissection because of the possibility of mucinous adenocarcinoma. Recently, the laparoscopic resection for appendiceal mucoceles has been increasing. This approach is effective and minimally invasive, but it is important for the surgeon to prevent spillage of the cystic fluid [10]. Pseudomyxoma peritonei may develop if cystic fluid spills into the intraperitoneal cavity.

Some reports have shown an association between mucoceles and diverticula of the appendix [2]. Diverticula of the appendix are uncommon, reportedly being found in 1–2% of appendectomies [2,11]. They may be classified as true diverticula or pseudodiverticula. Collins [12] reported that pseudodiverticula of the appendix are found in 1.370% of appendectomies, while true diverticula are

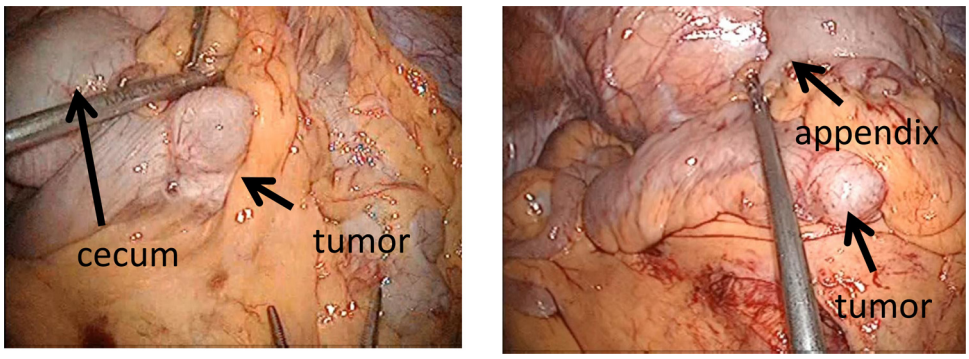

Fig. 3. The submucosal tumor was located in the ileocecal region. There was no continuity between the proximal edge of the appendix and the tumor.

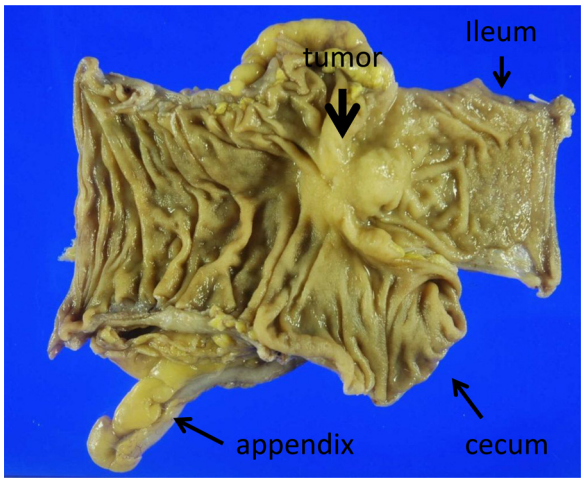

Fig. 4. Macroscopic view of the resected specimen shows the submucosal tumor, which measured 35 × 30 mm, in the ileocecal region (arrow). The tumor was not continuous with the appendix.

found in 0.014% of appendectomies. Thus, pseudodiverticula of the appendix are far more common. Preoperative diagnosis of an appendiceal diverticulum is difficult. These diverticula are detected after appendectomy or incidentally by barium enema examination [13]. Approximately 5.6–48.0% of appendiceal diverticula are associated with mucoceles [2,14,15]. In our case, a low-grade mucinous neoplasm was present in a cecal diverticulum. In Japan, there are no reports that cecal diverticula was associated with mucoceles. We searched PubMed using the keywords diverticulum + mucinous neoplasia, diverticulum + mucinous cystadenoma, and diverticulum + cystadenocarcinoma, but we found no reports of a mucocele in a colonic diverticulum. We believe that this is the first reported case worldwide.

The mechanism of mucocele formation in a cecal diverticulum is unknown. However, one proposed mechanism for mucocele formation in the appendix involves an aseptic obstruction process caused by the root twisting or bending, leading to gross mucinous accumulation. In our case, a similar mechanism may occur in the cecal diverticulum.

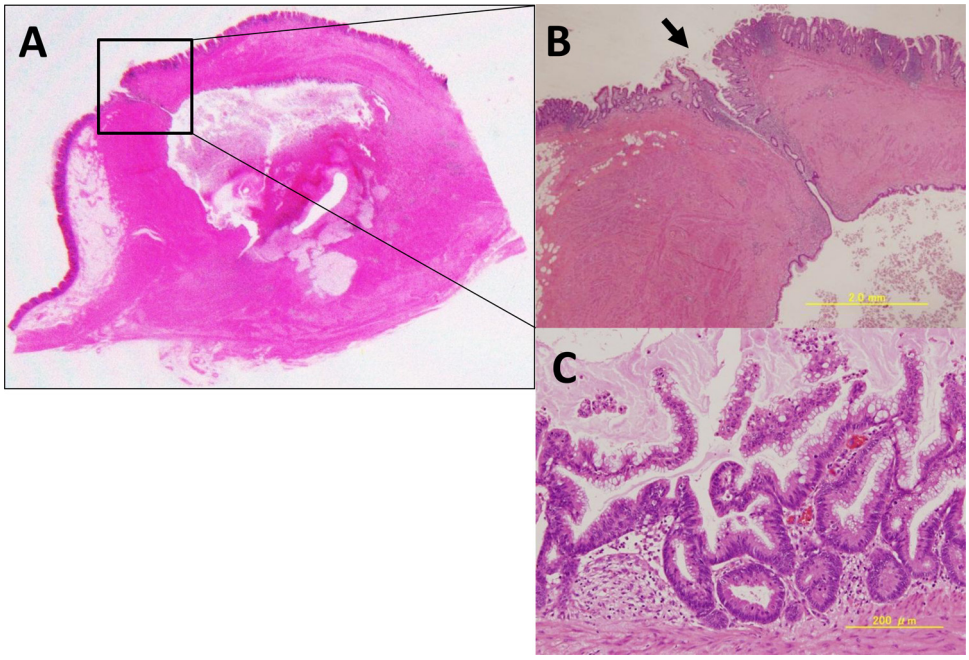

Fig. 5. (A) Loupe image. The cystic mass was present with mucinous collection in the muscularis propria. (B) The colonic lamia propria curved within the muscularis propria; therefore, this fistula was considered to be a cecal diverticulum. There was continuity in the colonic lamia propria of the surface and the muscularis propria. The orifice of the diverticulum was present on the surface of the colonic lamia propria (arrow), therefore it was considered to be cecal diverticulum. (C) Although epithelial neoplastic transformation was evident in the wall of the cystic mass as shown by cells exhibiting increased nuclear size and chromatin, these malignant cells were not present in the cystic tumor.

The incidence of colonic diverticula in Japan has recently been increasing. The prevalence of these diverticula in Japanese individuals aged <40 years is approximately 20%, while that in Japanese individuals aged >80 years is approximately 60%. The incidence of colonic diverticula tends to increase with age [16]. Geographically, the prevalence in the Western population is much higher than that in the Asian population because of the lower amount of alimentary fiber in the Western diet. Additionally, Western countries have a high prevalence (approximately 70%) of left-sided colonic diverticula. Right-sided colonic diverticula are rare in Western populations, but more common in Asia. In Japan, the prevalence of right-sided diverticula is approximately 70% [17,18]. However, with the recent Westernization in Japanese eating habits, the incidence of left-sided or bilateral colonic diverticula has been increasing.

In the present case, a low-grade mucinous neoplasm developed in a cecal diverticulum. This disease should be considered when an SMT is found in the colon.

4. Conclusion

Mucocele disease of the appendix is uncommon. Moreover, there are no previous reports of mucocele disease in a colonic diverticulum worldwide. The incidence of colonic diverticula has recently been increasing in Japan. The possibility of a mucocele in a colonic diverticulum should be considered in patients with a colonic SMT.

Conflict of interests

The authors declare that they have no conflict of interests.

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None

Ethical approval

This paper was not a research study, so ethical approval not required.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images

Author contributions

KT made substantial contribution to conception and drafted the manuscript. KN conducted a literature search and made the contribution for acquisition of data. KT, KN, TS and HR performed the operation. KT, KN, KY and MK reviewed the manuscript and gave final approval for publication. KT was revising it critically for important intellectual content. All authors read and approved the final manuscript.

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