Case Report

Minimally invasive surgery for appendiceal intussusception caused by mucocele of the appendix: case report and review of the literature

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Abstract: Appendiceal intussusception caused by mucocele of the appendix is extremely rare. In the current study, a 32-year-old woman was admitted to the department of general surgery of our hospital, complaining of persistent right, lower quadrant pain without an obvious cause for 17 hours. Physical examination indicated significant pain and tenderness in the right, lower abdominal quadrant. Blood analysis indicated that leukocyte count, the percentage of neutrophils and the serum C-reactive protein were increased. Abdominal and pelvic computed tomography revealed a well-encapsulated cystic mass surrounded by the caecum and intussusception. The appendiceal intussusception caused by mucocele of the appendix was revealed during the laparoscopic exploration. Appendectomy and partial cecectomy were conducted using the laparoscopic approach. Postoperative pathological examinations showed ileocecal intussusception and chronic inflammation, appendiceal mucocele and acute suppurative appendicitis. The patient showed satisfactory recovery that was observed during 15-months of follow-ups. This case highlights that laparoscopic appendectomy and partial cecectomy may be a beneficial, minimally invasive approach for appendiceal intussusception caused by mucocele of the appendix.

Keywords: Intussusception; appendiceal mucocele; laparoscopic surgery; minimally invasive; case report

Submitted Sep 26, 2019. Accepted for publication Nov 22, 2019.
doi: 10.21037/jgo.2019.12.01
View this article at: http://dx.doi.org/10.21037/jgo.2019.12.01

Introduction

Appendiceal intussusception is an uncommon type of intussusceptions, accounting for only 0.01% of appendiceal pathologies (1). The clinical symptoms vary and can mimic various chronic and acute abdominal conditions (2). Appendiceal intussusception caused by mucocele of the appendix is extremely rare. Resection of appendiceal mucocele is recommended when identified since there is potential for the mucocele to harbor a neoplasm (3). Although laparoscopic appendectomy is now commonly used for acute appendicitis, the safety and available of laparoscopic surgery for appendiceal intussusception caused by mucocele of the appendix is still controversial and the report is few (4,5). Here we present a case of appendiceal intussusception caused by mucocele of the appendix, for which the resection of appendix and part of caecum was successfully undertaken using the laparoscopic approach.

Case presentation

A 32-year-old woman presented to the Emergency Department of our hospital on April 24th, 2018, due to persistent right, lower quadrant distending pain without an obvious cause for 17 hours. She felt nauseous and

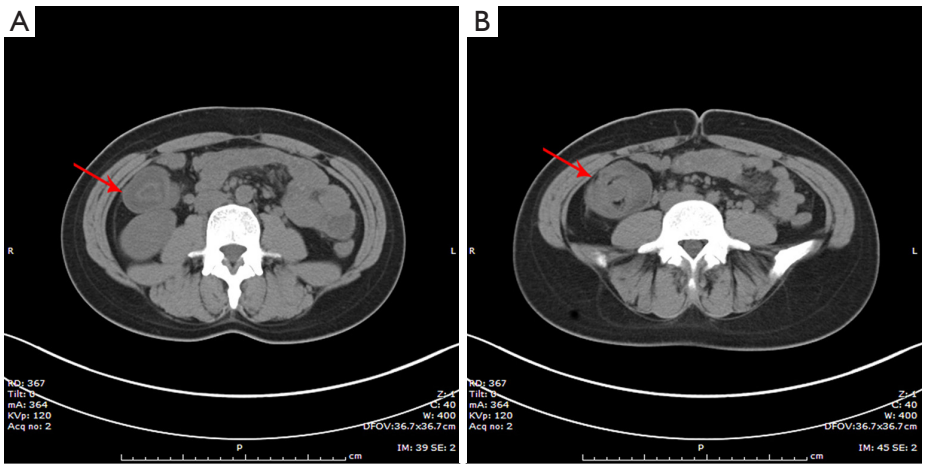

Figure 1 CT images. As the arrows (→) indicate, abdominal CT showed: (A) a "target-like" sign, the appendix surrounded by a rim of digestive structures, typical of appendiceal intussusceptions and (B) a well-encapsulated cystic mass surrounded by the caecum, just as the "cup-and ball" pattern, which is the appearance of the appendiceal mucocele surrounded by the caecum.

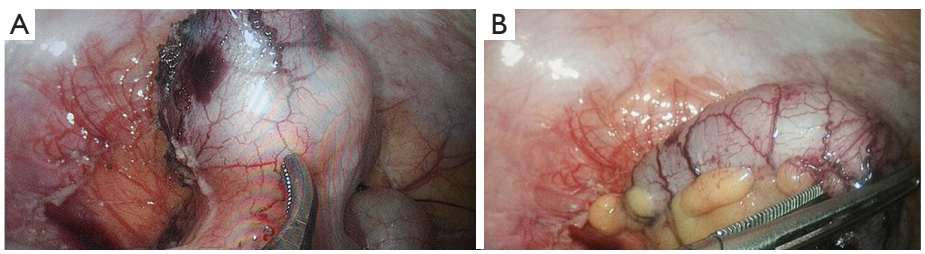

Figure 2 Surgical findings. (A) Preoperative photograph showed the root of appendix was inflated and pulled into the cecum and (B) the appendix was enlarged and hyperaemic.

vomited 2 times. She also complained of frequent diarrhea, without pain relief. She did not have a fever, shivering, radiative and referred pain. She underwent caesarean section in 2007 and had no other previous medical history. Physical examination revealed significant pain and tenderness in the right, lower abdominal quadrant. Blood analysis showed that the leukocyte count was 16.5×10^9/L, the percentage of neutrophils was 82.2%, and a normal hematocrit. Serum C-reactive protein was increased (11 mg/L). Blood biochemistry as well as human chorionic gonadotropin (HCG) levels were normal. Abdominal computed tomography revealed a well-encapsulated cystic mass surrounded by the caecum, an enlarged appendix, intussusception and pelvic effusion (*Figure 1*). The patient was taken to the operating room for laparoscopic exploration. There was no evidence of adhesions. The ovaries appeared normal. The appendix was enlarged and the root of appendix was inflated and pulled into the cecum (*Figure 2*). There were no signs of mucinous implants or pseudomyxoma peritonei in the abdominal cavity. The patient underwent a laparoscopic appendectomy and part caecum resection with a primary stapled anastomosis. The specimen which consisted of appendix and part of caecum was removed from the abdomen using an endobag. The portion of cecum measured 5 cm in length and had a diameter of 1.5–2.5 cm. The cecum wall had edema and bulges were observed at the junction of the appendix and the cecum that blocked the exit of the appendix. The appendix measured 5.5 cm in length and had a diameter of 1.2–1.5 cm. The appendiceal serosa was hyperemia and the root of appendix was enlarged with a cystic mass that pulled into the cecum (*Figure 3A*). The pathologic diagnosis showed ileocecal intussusception and chronic inflammation, as well as appendiceal mucocele and acute suppurative appendicitis (*Figure 3B,C*). The patient was discharged home on postoperative day 8 without complications and

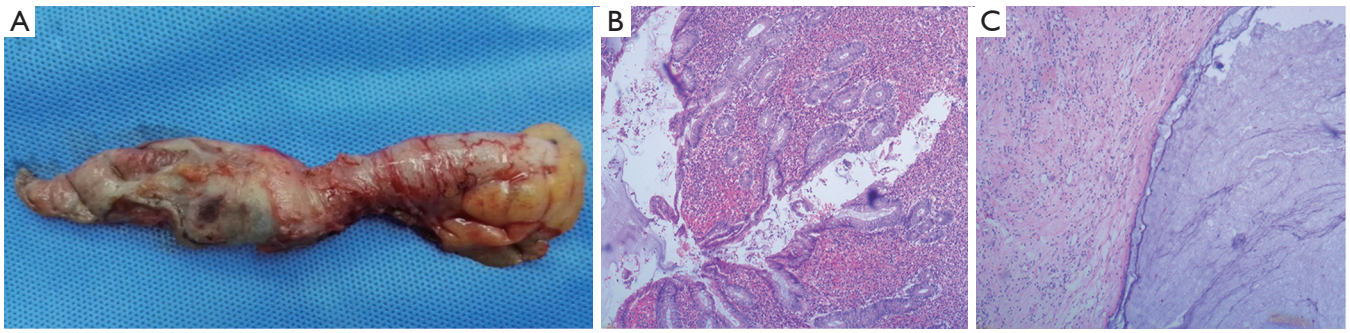

Figure 3 Postoperative histopathological findings. (A) The root of appendix was enlargement with a cystic mass and pulled into the cecum. (B,C) The pathologic diagnosis showed ileocaecal intussusception and chronic inflammation, as well as acute suppurative appendicitis and appendiceal mucocele (H&E stain, ×200).

showed satisfactory recovery that was observed during 15-month follow-ups.

Discussion

Appendiceal mucocele, an obstructive dilatation of the appendix caused by intraluminal accumulation of mucoid material, is very rare. The incidence is less than 1% of all appendectomied specimens (6). Appendiceal mucocele does not exhibit unique clinical symptoms. The asymptomatic mass in the lower right quadrant of the abdomen is a common clinical feature. Sometimes a patient has abnormal pain due to acute appendicitis. In addition, patients may present an intussusception, appendiceal torsion, gastrointestinal bleeding (7-10). The intussusception caused by appendiceal mucocele is extremely rare.

Appendiceal intussusception occurs when an appendix segment is pulled into itself or the cecum. Since the first report by M'Kidd in 1859 (8), literature on this disease has been confined to only a few case reports. Appendiceal intussusception may mimic various chronic and acute abdominal conditions and some cases may be completely asymptomatic (9-13). Patients with appendiceal intussusception and their symptoms can be divided into four groups including asymptomatic patients, patients with symptoms similar to acute appendicitis, patients with symptoms present as intestinal intussusception or patients with a prolonged history of right, lower quadrant abdominal pain. As in the described case, the presenting symptoms presented as acute appendicitis.

Diagnosis is difficult before operation due to the variable presentation and unspecific symptoms of appendiceal intussusception. Some patient cases were accidentally identified during surgery (11). However, the preoperative diagnosis is very important for the selection of an adequate surgical method and could prevent intraoperative and postoperative complications. Imaging examinations might be useful for the preoperative diagnosis of appendiceal intussusceptions. In most cases, X-rays of the abdomen are normal. Dilated small bowel loops with air-fluid levels may be found when there is associated small bowel obstruction. Ultrasound is the method of choice, especially in children. The cecum might appear as concentric loop pattern or the appendix invaginated into the cecum and the classic image of intussusception being of "onion skin-like lesion" (14,15). Abdomen-pelvis computed tomography is the most commonly used clinical diagnostic method. This method show a well-encapsulated cystic mass surrounded by the caecum, or the "cup-and ball" pattern, which is the appearance of the appendiceal mucocele surrounded by the caecum (16,17). Colonoscopy is another method that shows a cystic mass occupying most of the lumen of the caecum (18). Our patient showed the typical "cup and ball" pattern in the CT scan. However, the report of imaging examination just includes an enlarged appendix, intussusception and pelvic effusion, due to the limited knowledge of this disease.

Since appendiceal mucocele could lead to malignant transformation and spontaneous or iatrogenic ruptures, surgical resection is recommended (19). One of the cardinal principles of surgical treatment is avoiding rupture of the mucocele and potential spillage of mucin into the peritoneal cavity during resection, which could result in implantation of mucin-producing cells inside the peritoneal cavity and subsequent pseudomyxoma peritonei (20). An algorithm for the selection of a surgery type has been formulated

Table 1 The factors of the extent of surgery as treatment for appendiceal mucocele

The extent of surgery	The factors
Appendectomy	Non-perforated; Simple mucoceles/cystadenomas with an intact appendiceal base; Negative cytology; Negative margin of appendiceal stump; Negative appendiceal lymph nodes
Partial cecectomy	Non-perforated; The mucocele is broad and protrudes into the cecal wall; Negative/positive cytology; Negative/positive margin of appendiceal stump; Negative appendiceal lymph nodes
Right hemicolectomy	Non-perforated; The mucocele invades the cecal wall or ileum; An adequate resection margin cannot be secured; Positive cytology; Positive margin of resection; Positive margin of appendiceal stump; Positive appendiceal lymph nodes; If malignancy is strongly suspected

(I) If the mucocele is perforated and/or epithelial cells are found in the mucoid fluid of abdominal and/or pelvic, the cytoreductive surgery (CRS), early postoperative intraperitoneal chemotherapy (EPIC) or heated intraoperative intraperitoneal chemotherapy (HIIC) is needed; (II) the frozen pathology during surgery is necessary and important.

by Dhage-Ivatury *et al.* (21) and Kim *et al.* (22) (*Table 1*). There are several important factors to consider including whether the mucocele is perforated, whether the base of the appendix is involved and whether there are positive lymph nodes of mesoappendix and ileocolic. Simple appendectomy is the choice for patients in cases when the mucocele is not too large and is distant from the appendicular base, there is a presence of a normal caecum and appendicular base and there is no evidence of perforation. Partial cecectomy is required when the base of the mucocele is broad and protrudes into the cecal wall, or if the margin of appendiceal stump is positive and the lymph nodes are negative. Right hemicolectomy is recommended when malignant mucocele is suspected, and when there is an enlarged mesenteric lymph node or a positive cytology. So sometimes the frozen pathology during surgery is necessary and important. Comprehensive exploration of the abdomen is advised, especially for female patients, due to the association between the appendiceal mucocele and mucin-secreting cells cancers such as colon and ovarian cancers. In our case, the mucocele located at the appendicular base, partial cecum was involved and exploration of the abdomen was negative. As there was no evidence of malignancies, if the patient was treated with right hemicolectomy, the injury of operation was high. Appendectomy and partial colectomy were undertaken at the condition of avoiding mucocele rupture.

Historically, laparoscopic technology has been considered a contraindication for the surgical treatment of appendiceal mucoceles since patients undergoing laparoscopic appendectomy for intact mucoceles developed peritoneal implants after surgery (23). These results may indicate limitations of laparoscopic surgery for maintaining the integrity of the mucocele walls. Some researchers emphasize if appendiceal mucocele is found in laparoscopic surgery, the surgery must then be converted into open surgery. As open surgery could be performed carefully and proper exploration of the abdomen cavity can be done completely to avoid mucocele rupture and subsequent pseudomyxoma peritonei in comparison to the laparoscopic surgery (21,24,25). With developing laparoscopic technology, there were several reported cases that laparoscopic surgery can be conducted for appendiceal mucoceles without intraoperative rupture of the mucocele (26-28). Especially, Tae Kyu Kim reported a multicenter study of the safety and feasibility of laparoscopic surgery for appendiceal mucocele which showed that laparoscopic technology is safe and feasible for the surgical treatment of appendiceal mucocele (22). However, the safety and outcome of laparoscopic surgery for appendiceal intussusception caused by mucocele of the appendix is unclearly and the report is few (4,5,26). In our

case, the patient underwent laparoscopic appendectomy and partial cecectomy and showed satisfactory recovery that was observed during 15 months of follow-ups. Our experiences indicate: (I) the surgeon should have enough experience with cases of laparoscopic surgery, including appendectomy, right hemicolectomy, and so on, could accurately locate the relation of mucocele, appendix and caecum; (II) to prevent mucocele rupture and implantation metastasis, the operation should be performed carefully, avoiding touching the mucocele during the procedure; (III) the endobag must be used when removing the mucocele from the abdomen for preventing port-site seeding; (IV) the limitation of this study is we should take frozen pathology during surgery, in order to make definitive diagnosis.

In summary, our case report provides evidence for the safety of laparoscopic surgery used in appendiceal intussusception caused by mucocele of the appendix. The local excision using laparoscopic surgery is safe and feasible and the tissue was handled with minimal manipulation avoiding larger injury in the patient. This may be a potential operative method for similar specified appendiceal intussusception cases.

Acknowledgments

We thank our coworkers of the departments of emergency, radiology, laboratory, electrocardiogram, and operating room for their invaluable technical help.
Funding: This work was supported by the Fund of PLA Strategic Support Force Characteristic Medical Center (09ZX25).

Footnote

Conflicts of Interest: The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. Written informed consent was obtained from the patient for publication of this manuscript and any accompanying images.

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Cite this article as: Sun P, Jiang F, Sun H, Zhao X, Ma J, Li C, Yang H, Cui Y. Minimally invasive surgery for appendiceal intussusception caused by mucocele of the appendix: case report and review of the literature. J Gastrointest Oncol 2020;11(1):102-107. doi: 10.21037/jgo.2019.12.01

