Case Report


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Abstract
Surgical approaches for an appendiceal mucocoele remain controversial, since caution needs to be exercised during surgery to avoid inadvertent rupture of the lesion, which may cause pseudomyxoma peritonei if the lesion is neoplastic. We report a case of mucinous cystadenoma of the vermiform appendix treated successfully by single-incision laparoscopic surgery (SILS). A 71-year-old woman was admitted for the treatment of a cystic mass in the right iliac fossa. Abdominal computed tomography revealed a well-encapsulated cystic mass measuring 30 mm in diameter, adjacent to the cecum. SILS ileocele resection with lateral-to-medial mobilization and extracorporeal anastomosis was performed via a 3.5-cm-long transumbilical incision. The operative time was 145 minutes and the blood loss was about 10 mL. There were no intra- or postoperative complications. The histological diagnosis was mucinous cystadenoma. SILS seems to be a useful option for minimally invasive treatment of appendiceal mucocoele, however, as mentioned above, care is needed to prevent complications.

Key words: single incision, mucinous cystadenoma of appendix, colectomy

Introduction
Appendiceal mucocoele is usually a unilocular cystic lesion filled with mucin. The term “appendiceal mucocoele” is not used based on the histological diagnosis, but on the basis of the macroscopic appearance of a dilated appendix. In fact, appendiceal mucocoele could be related to both non-neoplastic and neoplastic processes. Since preoperative accurate diagnosis of malignancy is usually difficult to make, the treatment consists of complete surgical removal of a cystic lesion, while carefully avoiding its rupture into the peritoneal cavity. Indeed, rupture of a neoplastic cyst, whether histologically confirmed to be mucinous cystadenoma or mucinous cystadenocarcinoma, can produce pseudomyxoma peritonei, which can lead to decreased survival of the patients.

Currently, the best surgical approach for removing appendiceal mucocoele remains controversial. Some authors recommend conventional open surgical treatment to reduce the potential risk of inadvertent rupture of the lesion, while others have reported the feasibility and safety of laparoscopic treatment. Recently, single-incision laparoscopic surgery (SILS) has been performed for various benign and malignant diseases, including acute appendicitis and colorectal cancer. Surgical treatment for appendiceal mucocoele includes appendectomy or colectomy (ileocele resection or right hemicolectomy), depending on the extent and nature of the lesion. We attempted SILS for the removal of appendiceal mucocoele based on our favorable experience of SILS colectomy (data not shown). We herein report a patient with appendiceal mucocoele, histologically diagnosed as mucinous cystadenoma, who was successfully treated by SILS.

Case Report
A 71-year-old woman complaining of vague abdominal distress and painful constipation for 6 months
was admitted to our hospital for the treatment of a suspected lesion in the right iliac fossa. On physical examination, a cystic mass was palpable in the right iliac fossa, which was not tender to palpation. Colonoscopy demonstrated an extrinsic mass in the cecum. Barium enema demonstrated a filling defect corresponding to the mass detected by colonoscopy. Abdominal computed tomography revealed a well-encapsulated cystic mass adjacent to the cecum. The vermiform appendix could not be detected. Laboratory examination showed normal results, including of the serum levels of carcinoembryonic antigen (CEA) and carbohydrate antigen 19-9 (CA19-9). The patient was clinically diagnosed as having appendiceal mucocele, however, a definitive diagnosis could not be made preoperatively. SILS ileocecal resection was performed with the informed consent of the patient, after learning in detail the surgical technique, the possible complications, and the method of potential conversion to standard multiport laparoscopic surgery or conventional open surgery.

The patient was placed in the supine position with broad base of the thigh. The peritoneum was approached via a 3.5-cm-long transumbilical skin incision and the SILS port® (Covidien, Mansfield, MA, USA) was introduced. After creating pneumoperitoneum at 10 mmHg, two 5-mm trocars for laparoscopic instruments and one 12-mm trocar for a laparoscope were introduced intraperitoneally through the SILS ports. Laparoscopic visualization demonstrated appendiceal mucocele, with no evidence of invasion to any other organs. The entire appendix showed cystic dilatation, especially the root of the appendix was swollen. Further laparoscopic exploration did not reveal any other mucoid lesions. A Ligasure™ device (Covidien, Mansfield, MA, USA) and conventional (straight) laparoscopic forceps and graspers were used (Fig. 1). The lateral-to-medial approach was undertaken to take down the hepatic flexure and mobilize the ascending colon and cecum. After full mobilization of the right-sided colon, the SILS port was removed and a wound retractor (Alexis®, medium size, Applied Medical, Santa Margarita, CA, USA) was placed on the edge of the abdominal incision. The proximal and distal site of the bowel were transected extracorporeally, followed by lymph node dissection, including of the epicoic, paracolic and intermediate lymph nodes, by cutting the ileocolic vessels at their roots using conventional surgical instruments (Fig. 2), since a paracolic lymph node was felt to be hard, metastasis could not be excluded. The operative time was 145 minutes and the estimated blood loss was about 10 mL. There were no intra-operative or postoperative complications. Histological examination revealed an appendicular cyst, its wall lined by a mildly atypical hypersecretory columnar epithelium, containing abundant intraluminal mucin, compatible with the diagnosis of mucinous cystadenoma.

She was discharged on the 7th postoperative day. At present, 15 months after the surgery, the patient remains well, without any wound complications (Fig. 3).

**Discussion**

According to the WHO classification**, appendiceal
mucocoele can be divided into three pathological categories: (1) Simple mucocoele, not a neoplasm, resulting from a chronic obstruction of the proximal lumen; (2) mucinous cystadenoma, in which the tumor does not infiltrate the appendiceal wall, but may burst into the abdominal cavity to cause pseudomyxoma peritonei. In cystadenomas, which are the most common cause of appendiceal mucocoele, the luminal dilatation can reach up to 6 cm and is reported to be associated with appendiceal perforation in 20% of cases\(^{18}\); (3) mucinous adenocarcinoma, which may invade the appendiceal wall and other adjacent organs, sometimes associated with regional lymph node metastasis\(^{19-21}\). Mucinous cystadenocarcinomas are less common than mucinous cystadenomas and may present with spontaneous rupture in 6% of those cases\(^{10}\). The latter two categories are often referred to as neoplastic mucocoele. The latter two categories, which account for 62-76%\(^{22-25}\) of all appendiceal mucocoeles, are together referred to as ‘neoplastic mucocoele’.

It is difficult to distinguish among the three categories preoperatively by clinical, laboratory and/or radiographic findings, and also difficult to differentiate between mucinous cystadenoma or mucinous cystadenocarcinoma by intraoperative rapid histologic diagnosis. Therefore, the possibility of both mucinous cystadenoma and mucinous cystadenocarcinoma must be considered in the surgical treatment of appendiceal mucocoele.

Two key factors must be borne in mind while undertaking surgical treatment of appendiceal mucocoele. The first is the operative procedure: which method should be selected for the operation, simple appendectomy or colectomy? Simple appendectomy is reliable and sufficient for uncomplicated mucocoele. There is the opinion that if a cystadenocarcinoma is present without any mesenteric or adjacent organ involvement or lymph node metastasis, then a simple appendectomy with resection of the appendiceal mesentry might be sufficient for this condition as well\(^{26}\). However, if the mucocoele is adherent or suspected to invade the cecum or ileum, then more aggressive intervention, such as right hemicolecotomy or ileocecal resection\(^{27}\), may be indicated\(^{17,22}\). The second factor that needs to be considered is whether or not mesenteric lymph node dissection is needed. Andersson et al.\(^{26}\) reported that lymph node metastasis from mucinous adenocarcinoma of the vermiform appendix was rare and that lymph node dissection could be omitted. González-Morena et al.\(^{21}\) reported that right hemicolecotomy with systematic lymph node dissection did not confer any surgical advantage over simple appendectomy in patients with mucinous cystadenocarcinoma. However, the reported incidence of lymph node metastasis in recently published articles is as high as 48.1%\(^{29}\), and metastasis to intermediate lymph nodes has also been reported\(^{27,29}\). Therefore, we believe that lymph node dissection, including of the pericolic, epiploic and intermediate lymph nodes is necessary in case of suspected neoplastic mucocoele. Based on these considerations, ileocecal resection with regional lymph node dissection around the ileocolic vessels may be the appropriate operative procedure rather than right hemicolecotomy, in terms of the extent of resection. In fact, Japanese surgeons usually perform ileocecal resection for patients with suspected malignant appendiceal mucocoele, according to the guideline for colon cancer\(^{31}\). In the present case, we performed ileocecal resection with regional lymph node dissection, since cystic dilatation of the entire appendix was observed and hard masses were recognized intraoperatively in the regional lymph nodes. As a result, the tumor was pathologically diagnosed as a benign tumor.

Laparoscopic-assisted surgery is associated with fewer postoperative complications, shorter hospital stay, faster recovery of bowel function, and lower incidence of wound infection as compared to conventional open surgery in patients undergoing colorectal resection\(^{38-41}\). Moreover, the oncological safety of laparoscopic-assisted surgery in patients
with colon cancer has been demonstrated in several randomized trials\(^{25-30}\). Even with these advantages, attempts have been made by many surgeons to further decrease the number of ports used during laparoscopic procedures to achieve better cosmesis, less pain, and a lower risk of subsequent incisional hernia. Therefore, SILS has drawn attention and has been used for colon resection\(^{25-30}\) as well as appendectomy\(^2\). SILS poses a number of unique challenges for the laparoscopic surgeon. The potential for triangulation and retraction is significantly limited. The use of a laparoscope and several instruments results in a decreased range of motion and the "clashing" of instruments, rendering meticulous surgical procedures difficult to perform safely. The surgical procedure of choice, including laparoscopic approach and conventional open surgery for appendiceal mucocele, is still under debate. Until now, it has been difficult to arrive at a consensus.

Currently, there are no significant advantages of multiport laparoscopic surgery for appendiceal mucocele as compared to conventional open surgery. Therefore, the feasibility of SILS for appendiceal mucocele would still remain controversial. Under the present situation, however, we believe that SILS would be a useful alternative for potentially malignant appendiceal mucocele, especially if the operating surgeon pays careful attention to the following suggestions; i) full and careful mobilization of the right-sided colon must be ensured, ii) the cystic mass should never be touched by any of the laparoscopic instruments, and iii) resection and anastomosis of the ileocecal region must be conducted extracorporeally, under direct vision. When inflammatory changes around the mucocele are observed in preoperative diagnostic imaging or intraoperative laparoscopic visualization, the decision as to conversion from the multiport laparoscopic approach to conventional open surgery needs to be made quickly. Moreover, it is essential to preoperatively estimate the risk of removal of the cystic mass without damage. In our department, when we realize that the cystic mass cannot be taken out after full mobilization of the right-sided colon due to the small length of the incision, we have no hesitation in extending the incision.

In conclusion, SILS seems to be a useful minimally invasive treatment alternative for appendiceal mucocele. To the best of our knowledge, this is a first reported case of SILS for appendiceal mucocele in the literature.

To validate the usefulness of SILS for appendiceal mucocele, further collection of cases is needed, but it is important for surgeons to plan the resection carefully.

References

SILS for mucinous cystadenoma of appendix


