Laparoscopic resection of an incidental appendiceal mucocele: is it correct
Case report

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Mucocele of the appendix is an uncommon disorder, characterized by a cystic dilatation of the lumen; it's mainly due to mucinous cystadenoma. Definite diagnosis is difficult preoperatively. It can be discovered incidentally at laparotomy or laparoscopy performed for other reason. Treatment consist in complete resection avoiding rupture of the cyst in the peritoneal cavity. Indeed, rupture of the lesion either spontaneous or accidental, during surgery may result in the clinical condition of pseudomyxoma peritonei. Therefore, open approach is recommended for the surgical treatment of these lesions. The role of laparoscopic surgery in the management of appendiceal mucocele remains controversial. We report a case of mucous cystadenoma of the appendix, successfully removed during a laparoscopy for perforated peptic ulcer, which was well at a 12-month follow-up. Laparoscopic appendectomy is not contraindicated in mucocele of appendix, if appropriate precautions can be taken intraoperatively.

KEY WORDS: Appendiceal mucocele, Appendectomy, Laparoscopy.

Introduction

Appendiceal mucocele (AM) is a rare entity that can present with a variety of clinical symptoms or occur as an incidental surgical finding. A mucocele of the appendix is an obstructive dilation of the appendix by intraluminal accumulation of mucoid material. It may be a benign or malignant process. Mucinous cystadenomas being the most commonly encountered appendiceal mucocles 1. Proper treatment of mucocele is critical because, if managed incorrectly, it may progress to pseudomyxoma peritonei 2. Even if laparoscopy has been successfully used to perform appendectomy, some concerns exist regarding its use in dealing with mucinous secreting lesions because of possible spillage of mucin during surgery 3.

The role of laparoscopic surgery in the management of AM therefore remains contentious 4-6. We present a successful laparoscopic resection of an AM and review pertinent literature.

Case report

A 42-year-old man presented with severe epigastric pain and nausea that began 4 hours before emergency room admission. On physical examination he was afebrile, with a heart rate of 77 and blood pressure of 135/80. His abdomen was diffusely tender with guarding and rebound. Bowel sounds were present. The plain radiograph of the abdomen demonstrated free intraperitoneal air. Our patient underwent an emergency exploratory laparoscopy. The laparoscopic procedure was performed with the patient and the team set up in the “French” position. Trocars were placed at the umbilicus (video scope) and on the left and right midclavicular line above the level of the umbilicus (instruments). Laparoscopy revealed peritonitis due to a perforated ulcer on the anterior wall of the duodenum, which was sutured in combination with an omental patch. Free fluid and purulent...
material were noted in the subdiaphragmatic region. After a thorough lavage of the peritoneal cavity, further exploration of the intra-abdominal organs revealed a mucocele of the appendix with an estimated diameter of 50 x 30 mm (Fig. 1). The patient was then placed in a Trendelenburg position. A long segment of normal appendix was found distal to its cystic swelling (Fig. 2). There was neither mucinous ascites nor lymphadenopathy. The normal segment of appendix was grasped using bowel-holding graspers (non-traumatic) and mesoappendix was mobilized using Harmonic scalpel; two vicryl endoloops were applied on both sides before division. The cyst was retrieved with an abdominal bag.

Pathological examination revealed a mucinous cystadenoma without cecal involvement (Fig 3). Postoperative course was unremarkable and he was discharged home on the 7th postoperative day in good conditions. During follow-up, the patient was asymptomatic and well. A CT scan 12 months later showed no signs of recurrence.

Discussion

Mucocele of the appendix is an uncommon tumor, with an incidence of 0.29%-0.4% of all appendectomied specimens 7. It may be caused by one of four processes: retention cyst, mucosal hyperplasia, mucinous cystadenoma, or mucinous cystadenocarcinoma 2. The last form is the rarest 8,9. Mucinous cystadenoma represents 50% of mucoceles 10. Furthemor, mucocels from cystadenoma or adenocarcinoma are usually larger, measuring up to 6 cm 6. Acute or chronic pain in right iliac fossa is the most frequent symptom, appearing sometimes as a mass at physical examination 10. The patients with symptoms, 27% had abdominal pain, 14% had an abdominal mass, 13% lost weight, 9% had nausea, vomiting, or both, and 8% had acute appendicitis. Symptomatic patients were more likely to have a malignant AM 2. However, AM may be asymptomatic in 50% of the cases 11. AM was associated with colon adenocarcinomas with a frequency around 20% 12. The preoperative diagnosis of mucocele can be difficult because of its rarity. USG, CT and colonoscopic examinations can facilitate preoperative diagnosis of AM 7. Ultrasound shows cysts with variable echogenicity, depending of the composition of the mucus. Multiple echogenic layers along a dilated appendix produce the appearance of “onion-skin” circles and may be pathognomonic for mucocele 13. Diagnosis
of a mucocele is often confirmed by a CT examination of the abdomen, which is characterized by a well-encapsulated cystic mass 2 to 20 cm in diameter that occurs usually in the right lower quadrant. Curvilinear mural calcification is seen about 50% of the time 7. USG and CT findings are non-specific and the differential diagnosis includes periappendiceal abscess, cystic ovarian neoplasms, mesenteric cysts and hydrosalpinx 12.

Colonoscopy is a useful tool for determination of mucocele. Generally, an elevation of the orifice of the appendix is seen. A yellowish mucous discharge would be visible from appendiceal orifice during colonoscopy 7. However, definitive diagnosis of AM is only achieved in 19% of the cases 11. Stocchi et al. found that the 60% of the patients who underwent surgery had incidental removal of their AM during an operation performed for concurrent conditions 11. All mucoceles should probably be removed to eliminate the chance of progression to malignancy.

The worst complication is pseudomyxoma peritonei, characterized by peritoneal dissemination caused by iatrogenic or spontaneous rupture of the mucocele 14. Cystadenomas and cystadenocarcinomas present an incidence of perforation around 20% 10. Mucoceles, if perforated, may be associated with mucoid material in the peritoneal cavity. This mucoid material may be acellular or can contain cells with low-grade dysplasia or cells with high-grade dysplasia 2. Mucinous ascites in the pelvis and in the right upper quadrant between liver and right hemidiaphragm indicates rupture of the mucocele 2. Even a benign disease such as cystadenoma, can cause pseudomyxoma peritonei 2,10. So it is important to keep a mucocele intact during operations 2.

The best surgical management of a patient with an appendiceal mucocele remains a subject of controversy. There is little consensus on the optimal choice of procedure (right hemicolecotomy versus appendectomy) as well as the approach (laparoscopic versus laparotomy) 1. Minimal surgery should be performed when a dysplastic mucocele has ruptured and mucinous carcinomatosis or pseudomyxoma peritonei syndrome is diagnosed. This means that an appendectomy with a clear margin at the base of the appendix, appendiceal lymphadenectomy, and a generous sampling of the IP mucinous fluid are indicated 2. If epithelial cells are found in the mucoid fluid, the patient must be referred to an established peritoneal carcinomatosis treatment center for cytoreductive surgery and intraperitoneal chemotherapy 10. A right hemicolecotomy must be performed if the gross appearance and then cryostat section showing malignancy within the appendiceal or the ileocolic lymph nodes 2,10. If there is any doubt about the extension of the tumor mass longitudinally through the appendix, cryostat sectioning of the surgical margin is indicated. Successful removal of appendiceal mucoceles laparoscopically, even those requiring ileocecal resection, has been reported 15. However, the problem is not technical feasibility, but surgical indications, considering the possible adverse events related to a type of surgical procedure. The laparoscopic approach has an increased risk of rupture and subsequent pseudomyxoma peritonei formation 2,12,14,16. Dhage-Ivatury et al. recommend that, when a mucocele is visualized during a laparoscopic procedure, the safer maneuver is to convert to an open laparotomy for mucocele excision 2. Moreno et al. suggest conversion to an open appendectomy in case of mucocele when laparoscopic appendectomy is intended 14. Open laparotomy is necessary to prevent rupture of the mucocele, seeding of trocar sites and allows better visualization of the abdominal cavity. Reports of missed lesions and widespread peritoneal implants after laparoscopic removal would tend to support open conversion in most circumstances 1,3,14,17. Thus, conventional surgery is preferred rather than laparoscopic approaches for the treatment 2,6,14,16. Few authors still recommend a minimally invasive approach in selected patients for this rare entity 4,15,18-21. The article by Rangarajan et al is perhaps the largest case series (8 cases) on laparoscopic appendectomy for mucocele of appendix 19. Navarra et al reported successful, removal of an appendiceal mucinous cystadenoma contained within the appendix using a laparoscopic approach with the patient remaining free of disease at 12-month follow-up 20. If laparoscopy is to be used and performed safely, grasping the mucocele should be avoided and an endo-bag must be used 2. Liberale et al.suggest to manipulate the normal appendix (when possible) without grasping the cyst and to use an endostapler for the appendiceal section, allowing also the realisation of a partial cecectomy if the AM is proximal 4. Manipulation of the lesion can be facilitated by gravity after appropriate adjustment of the position of operation table 2. In our patient was possibile take appropriate precautions. The laparoscopic approach avoided a large incision for exploration of the peritoneal cavity and conferred the benefits of minimal access surgery. Surgeons experienced in minimally invasive surgery, can minimize the unnecessary complication of peritoneal spillage of mucin, which may occur during open or laparoscopic appendectomy 21. The outcome of simple mucocele, mucosal hyperplasia, and mucinous cystadenoma after appendectomy is excellent, reaching 91% 10-year survival 10. However follow-up is recommended in all case, even those with benign histology, because of the demonstrated possibility of pseudomyxoma peritonei and reported association with other cancers of the large bowel 1,10.

J. Ruiz-Tovar et al reported the case of a patient with a diagnosis of mucocele (apparently not perforated), which 6 years later developed a recurrence in the form of pseudomyxoma peritonei 10. There is no consensus about the frequency of the follow-up; but it seems reasonable to propose it every six months for the first two years after surgery, and then yearly 4.
**Conclusion**

In literature the laparoscopic approach in the treatment of mucocele has been adopted for a small number of patients. We report a case of laparoscopic appendectomy for a mucinous cystadenoma of the appendix. At a 12-month follow-up assessment, the patient was free of disease. During a laparoscopy performed for other reasons, an AM must be recognized, so that precautions to avoid the rupture of the cyst can be taken; need to manipulate the normal appendix without grasping the cyst and to use endopouch retriever. Conversion to laparotomy should be considered if the lesion must be grasped, or if the tumor clearly extends beyond the appendix. We think that a mass freely involving the appendix body could be treated by a safer standard open approach. Although our follow-up is limited, we believe that laparoscopy is not contraindicated, if appropriate precautions can be taken. However, we need a large series and longer follow-up to substantiate recommendations of laparoscopic approach.

**References**