Laparoscopic resection of an incidental appendiceal mucocele: is it correct



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Case report

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

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 takenintraoperatively.

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 mucoceles 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 surgery³.

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

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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 mesoappen-

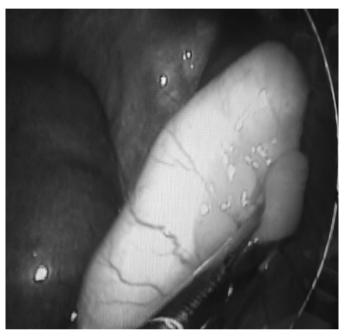


Fig. 1: Appendiceal mucocele visible on laparoscopy.



Fig. 2: A long segment of normal appendix was found distal to its cystic swelling,



Fig. 3: Histological diagnosis was mucinous cystadenoma.

dix 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 usally 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 ². 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 hemicolectomy 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 hemicolectomy 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 7,12,14,16. 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 18. 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 caecetomy 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 possilbile 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 7. 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 3,20. We think that a mass freely involving the appendix body could be treated by a safer standard open approach. Although our followup 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.

Riassunto

Il mucocele dell'appendice è un non comune disordine, caratterizzato da una dilatazione cistica del lume; è principalmente dovuto ad un cistoadenoma mucinoso. La diagnosi definitiva è difficile preoperatoriamente. Può essere scoperto accidentalmente in corso di una laparotomia o laparoscopia eseguita per altre ragioni. Il trattamento consiste nella resezione completa evitando la rottura della cisti in cavità peritoneale. Infatti, la rottura della lesione, spontanea o accidentale, durante la chirurgia può risultare in un pseudomixoma-peritonei. Quindi, l'approccio open è raccomandato per il trattamento chirurgico di queste lesioni. Il ruolo della chirurgia laparoscopica nel trattamento dei mucoceli appendicolari rimane controverso. Noi riportiamo un caso di cistoadenoma mucinoso dell' appendice, rimosso durante una laparoscopia per ulcera peptica perforata, che stava bene ad un follow-up di 12 mesi. L'appendicectomia laparoscopica, non è controindicata nel mucocele dell'appendice, se opportune precauzioni possono essere prese intraoperatoriamente.

References

- 1. Khan MR, Ahmed R, Saleem T: Intricacies in the surgical management of appendiceal mucinous cystadenoma: A case report and review of the literature. J Med Case Reports, 2010; (5)4:129.
- 2. Dhage-Ivatury S, Sugarbaker PH: *Update on the Surgical Approach to Mucocele of the Appendix.* J Am Coll Surg, 2006; 202:680-84.
- 3. Behranwala KA, Agarwal T, El-Sharkawi D, Shorvon D, Chang A: Laparoscopic resection of mucinous cystadenoma of appendix. A Careful Decision. Surg Laparosc Endosc Percutan Tech, 2006; 16:347-48.

- 4. Liberale G, Lemaitre P, Noterman D, Moerman C, de Neubourg E, Sirtaine N, El Nakadi I: *How should we treat mucinous appendiceal neoplasm? By Laparoscopy or Laparotomy? A case report.* Acta Chir Belg, 2010; 110:203-07.
- 5. Gentile M, Guarino V, Mosella F, Carbone G, Beneduce L, Mosella G: *Giant mucocele of the appendix. Case report and review of the literature.* Ann Ital Chir, 2008; 79:293-97.
- 6. Lau H, Yuen WK, Loong F, Lee F: *Laparoscopic resection of an appendiceal mucocele*. Surg Laparosc Endosc Percutan Tech, 2002; 12:367-70.
- 7. Karakaya K, Barut F, Emre AU, Ucan HB, Cakmak GK, Irkorucu O, Tascilar O, Ustundag Y, Comert M: *Appendiceal mucocele: Case reports and review of current literature.* World J Gastroenterol, 2008; 14:2280-83.
- 8. Higa E, Rosai J, Pizzimbono CA, Wise L: Mucosal hyperplasia, mucinous cystadenoma, and mucinous cystadenocarcinoma of the appendix: A re-evaluation of appendiceal mucocele. Cancer, 1973; 32:1525-41.
- 9. Sato H, Fujisaki M, Takahashi T, Maruta M, Maeda K, Kuroda M: *Mucinous cystadenocarcinoma in the appendix in a patient with nonrotation: Report of a case.* Surg Today, 2001; 31:1012-15.
- 10. Ruiz-Tovar J, Teruel DG, Castiñeiras VM, Dehesa AS, Quindós PL, Molina EM: *Mucocele of the Appendix.* World J Surg, 2007; 31:542-48.
- 11. Stocchi L, Wolff BG, Larson DR, Harrington JR: Surgical treatment of appendiceal mucocele. Arch Surg, 2003; 138:585-89.
- 12. Pitiakoudis M, Tsaroucha AK, Mimidis K, Polychronidis A, Minopoulos G, Simopoulos C: *Mucocele of the appendix: A report of five cases.* Tech Coloproctol, 2004; 8:109-12.
- 13. Caspi B, Cassif E, Auslender R, Herman A, Hagay Z, Appelman Z: *The onion skin sign: A specific sonographic marker of appendiceal mucocele.* J Ultrasound Med, 2004; 23:117-21.
- 14. Gonzalez Moreno S, Shmookler BM, Sugarbaker PH: *Appendiceal mucocele: Contraindication to laparoscopic appendectomy.* Surg Endosc, 1998; 12:1177-79.
- 15. Miraliakbari R, Chapman WH III: Laparoscopic treatment of an appendiceal mucocele. J Laparoendosc Adv Surg Tech A, 1999; 9:159-63.
- 16. Rampone B, Roviello F, Marrelli D, Pinto E: *Giant appendiceal mucocele: Report of a case and brief review.* World J Gastroenterol, 2005; 11:4761-763.
- 17. Zagrodnik DF 2nd, Rose DM: Mucinous cystadenoma of the Appendix: Diagnosis, surgical management, and follow-up. Curr Surg, 2003; 60:341-43.
- 18. Rangarajan M, Palanivelu C, Kavalakat A, Parthasarathi R: Laparoscopic appendectomy for mucocele of the appendix: Report of 8 cases. Indian J Gastroenterol, 2006; 25:256-57.
- 19. Sfairi A, Detchev R, Patel JC: Mucocele of the appendix: value of excision by celioscopy. J Chir, 1995; 132:403-05.
- 20. Navarra G, Asopa V, Basaglia E, Jones M, Jiao LR, Habib NA: *Mucous cystadenoma of the appendix: is it safe to remove it by a laparoscopic approach?* Surg Endosc, 2003; 17:833.
- 21. Ng SS, Lee JF: *Mucinous cystadenoma of the appendix*. Can J Surg, 2009; 52:158-59.