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Accidental discovery of appendiceal carcinoma during gynecological surgery: A case report

Lin Wang, Yan Dong, Ya-Hui Chen, Ya-Nan Wang, Lin Sun

Abstract

BACKGROUND
Malignant tumors of the appendix are extremely rare, constituting about 1% of all gastrointestinal tumors. Generally, pathology identifies these tumors during or after appendectomy because they are difficult to detect at the preoperative stage. This case report aims to introduce the definitive diagnosis and treatment of mucinous adenocarcinoma of the appendix.

CASE SUMMARY
A 49-year-old female patient came to our hospital with right lower abdominal pain, nausea, and vomiting for three days. There was no change in the menstrual cycle. Gynecological ultrasound showed a cystic, solid mass in the right adnexa. Abdominal enhanced computed tomography showed a thick appendix. Cancer was found on exploration of the appendix during gynecological surgery. The right colon was removed. After surgery, the patient received chemotherapy and is recovering well.

CONCLUSION
Appendiceal carcinoma is frequently found during or after surgery, and both preoperative examination and early evaluation of clinical manifestations are extremely important.

Key Words: Abdominal pain; Pelvic mass; Appendix carcinoma; Mucinous adenocarcinoma; Case report

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Core Tip: Mucinous adenocarcinoma of the appendix has a low incidence rate and is relatively rare. Increased tumor markers in patients has certain guiding significance. Imaging examination can indicate that the appendix is thickened, and diagnosis depends on histopathology.

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INTRODUCTION
Malignant appendix epithelial tumors can be divided into three categories: mucinous adenocarcinoma of the appendix (MAA)\textsuperscript{[1]}, intestinal type of adenocarcinoma, and signet ring cell carcinoma. Of these, MAA is the most prevalent histological form. Its occurrence may be linked to chronic inflammatory infiltration of the appendix. Here, we report a case of appendiceal mucinous adenocarcinoma, accidentally found during surgery for right lower abdominal pain and a pelvic mass. We also reviewed available relevant literature.

CASE PRESENTATION

Chief complaints
A 49-year-old female patient came to our hospital with right lower abdominal pain, nausea, and vomiting.

History of present illness
The patient’s symptoms started three days ago with right lower abdominal pain, nausea, and vomiting.

History of past illness
There was no change in the menstrual cycle. The patient had no previous medical history.

Personal and family history
The patient is in good health and has no family genetic diseases.

Physical examination
Gynecologic examination suggested normal vulvar development, a smooth vagina, little vaginal discharge, a soft cervix, an average size uterus, and no tenderness. There was no abnormality in the left accessories, and a cystic solid tumor approximately 6 cm in size was palpated in the right accessories.

Laboratory examinations
Human epididymal protein 4 (HE4), carcinoembryonic antigen (CEA), and alpha-fetoprotein (AFP) levels were normal on May 14, 2021. The patient’s CA125 level was 392.9 U/mL and CA199 level was 88.27 U/mL on May 14, 2021.

Imaging examinations
Gynecological ultrasound on May 15, 2021, showed a cystic, solid mass in the right adnexa area approximately 6.2 cm × 5.6 cm × 5.8 cm in size, with an unclear right ovary. On May 18, 2021, a complete abdominal computed tomography (CT) scan showed a hypocystic shadow in the right adnexa area with a visible compartment inside and appendiceal thickening with a maximal thickness of around 12 mm (Figure 1). The mass in the pelvic cavity was unidentified.

FINAL DIAGNOSIS
The patient underwent an exploratory laparotomy on May 21, 2021, during which the right accessory and cystic mass were removed, thickening of the appendix (~6 cm in length and ~1 cm in diameter) was noted, with a hard texture and edema, attached to the posterior wall of the ascending colon. Appendiceal malignancy could not be excluded, and was diagnosed following rapid intraoperative pathology. The final diagnosis of the presented case was MAA.
TREATMENT

Gastrointestinal surgery consisting of a right hemicolectomy and peripheral lymph node dissection was performed. The residual intestine, stomach, liver, greater omentum, and peritoneum surface were examined at the end of surgery, and no abnormalities were found.

OUTCOME AND FOLLOW-UP

Postoperative pathology confirmed mucinous adenocarcinoma of the appendix with partial signet-ring cell carcinoma. Immunohistochemistry was performed using the following markers: CDX-2 (+), CK7 (-), CK20 (+), CA125 (+), CD56 (-), WT-1 (-), and SATB2 (+) (Figure 2). The patient received chemotherapy 45 days after the operation. Six courses of XELOX chemotherapy (Oxaliplatin + Capecitabine) were completed, and there was no evidence of recurrence. MAA with signet-ring cell features is considered more invasive and has a worse prognosis. The patient requires regular follow-up every four months for three years, every six months for the next two years, and then every year for the next 15 years, following initial therapy[2].

DISCUSSION

MAA is a rare disease characterized by elevated CA199 and CEA levels. C. PABLO et al showed that tumor markers CEA and CA199 have high clinical value in diagnosing MAA[3,4]. Moreover, the increasing level of CA125 while maintaining normal HE4 helps discriminate between benign and malignant ovarian tumors. Our patient showed elevated CA199 and CA125 levels, which can help diagnose appendiceal lesions[3]. Moh M used immunohistochemistry to identify SATB (-) in ovarian mucinous tumors. The presence of SATB2 (+) and CDX-2 (+) highly suggests that the tumor originates from the colon or appendix[4]. Imaging is a useful diagnostic tool for MAA. Ultrasound observations revealed a cystic mass in the appendix, heterogeneous echogenicity, hypoechogenic or tubular lesions in the appendix, and irregular thickening. CT scans can rule out appendiceal inflammation and abscess, and all the above findings help diagnose mucinous cystadenoma[1,6]. MAA is difficult to diagnose due to the non-specific nature of early symptoms, including lower abdominal pain, weight loss, nausea, vomiting, a palpable mass, and acute appendicitis, and is frequently misdiagnosed as a gynecological condition such as right adnexal mass[7]. The bladder may also be affected, with symptoms of bladder irritation or the formation of hematuria, leading to a misdiagnosis of urinary tract infection or bladder cancer[8-10]. In most cases, appendiceal malignancy is detected accidentally by abdominal CT or surgery for appendicitis due to other reasons. Appendiceal cancer is difficult to identify even by preoperative colonoscopy[11]. In the case of submucosal lesions of the cecum near the mouth of the appendix, mucus flows out of the mouth of the appendix; thus, the treating physician should be highly vigilant against appendiceal lesions. Mucinous adenocarcinoma of the appendix is associated with a high risk of peritoneal seeding along with hematogenous and lymph node metastasis. As a surgical treatment for mucinous adenocarcinoma, simultaneous surgical removal of the appendix and right hemicolectomy with peripheral lymph node dissection is preferred[12]. Laparotomy is superior to laparoscopic surgery as it facilitates identification of the involvement of other organs. It is better to protect against incision, thereby avoiding mass rupture, leading to intra-peritoneal dissemination and affecting prognosis.
CONCLUSION

MAA is remarkably rare, difficult to diagnose and distinguish from other tumors. Preoperative laboratory and imaging examinations, and a well-planned diagnostic and treatment strategy are essential. In mucinous neoplasms, we believe that a right hemicolectomy should definitely be performed if required for tumor clearance because a complete cytoreduction of mucinous tumors of the appendix is associated with improved survival[12,13]. The patient described here presented with common clinical symptoms of MAA. For patients with the appearance of an abnormal appendix during preoperative examination, it is necessary to examine the appendix at the same time to avoid missed diagnosis and misdiagnosis.

FOOTNOTES

Author contributions: Wang L drafted, reviewed, and revised the manuscript; Sun L was the primary physician during the patient’s inpatient stay; Dong Y, Wang YN and Chen YH provided the images; all authors have read and approved the final manuscript.

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