

Case Report

A Rare Case of Appendiceal Stump Adenocarcinoma and Review of Literature

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ABSTRACT

Adenocarcinoma of appendiceal stump is extremely rare, with only four such patients reported to date. It has no specific clinical signs, symptoms, or radiologic features, making preoperative diagnosis difficult. Secondary right hemicolectomy is recommended and is difficult to perform, with peritoneal dissemination and lymph node metastases sometimes found at the second operation. We report on a case of a 72-year-old patient who underwent

an appendectomy in 2005 and was admitted because of a 3-month history of repeated constipation with vomiting. He was not relieved with the use of gastrokinetic drugs. Initial diagnosis was chronic adhesive intestinal obstruction due to previous lower abdominal surgery. He recovered well postoperatively. A histological examination showed a well-differentiated adenocarcinoma of the appendiceal stump.

KEY WORDS: adenocarcinoma, appendectomy, appendiceal stump

INTRODUCTION

Since appendectomy is usually performed for patients with appendicitis, patients with an appendiceal stump are not uncommon. Malignant tumours of the appendix, however, are rare and adenocarcinoma involving the post-appendectomy appendiceal stump is extremely rare, with only four such patients reported to date^[1]. This diagnosis cannot be determined until laparotomy or pathologic evaluation of the appendectomy specimen. Reoperation, consisting of right hemicolectomy, is recommended in patients diagnosed with adenocarcinoma of the appendiceal stump after pathologic evaluation of an appendectomy specimen^[2].

CASE REPORT

In September 2010, a 72-year-old man was admitted to our institution with a 3-month history of repeated constipation along with vomiting. He had undergone an appendectomy in 2005 due to acute appendicitis. Since then, he had remained well. The pathological results of the lesion and the sections were not available. On admission, no obvious abnormality was found, except for a decrease in bowel sounds. There was no family history of cancer or colon polyps.

His hemoglobin concentration was 105 g/l (normal, 110 - 165 g/l), his white blood cell count was 9.5×10^9 cells/l (normal, $3.5 - 10.0 \times 10^9$ cells/l), and neutrophil were 77.10% (normal, 50 - 70%). Biochemical analyses were within normal range. During the last three months, he was managed conservatively, including fasting with fluid support and tube decompression when intermittently admitted to a local hospital on three occasions. Since his symptoms were partially alleviated by administration of gastrokinetic drugs with no obvious positive findings on abdominal X-ray, he was initially diagnosed as a case of intermittent incomplete adhesive intestinal obstruction due to lower abdominal surgery. As the diagnosis remained obscure, we carried out an endoscopy examination for the old man to rule out malignancy. Colonoscopy only showed an obscure view of the ileocecal valve opening and inflammatory changes in the mucous membrane of the colon.

During this hospitalization, he experienced abdominal discomfort. At the urging of the patient and his family, an exploratory laparotomy was performed six days after admission due to unalleviated abdominal pain. At exploratory laparotomy, we observed no obvious adhesions in the abdominal cavity. We also

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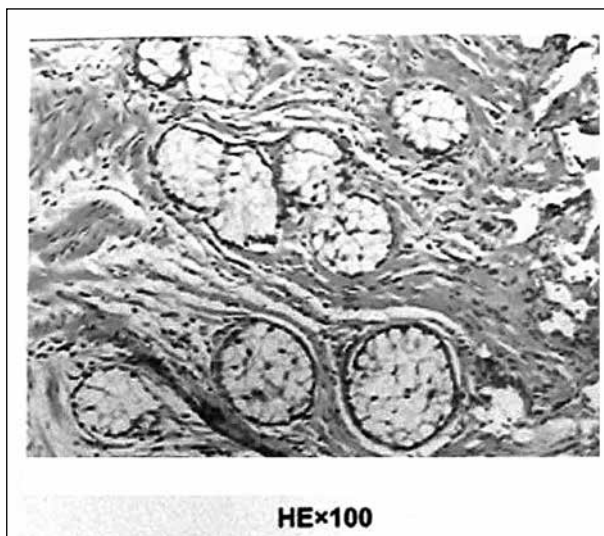


Fig. 1: Pathologic examination showing a well-differentiated adenocarcinoma of the appendiceal stump

found thickening of 50 cm of the terminal ileum, indicative of chronic obstructive changes, as well as inversion of the preileal appendiceal stump around the ileocecal valve. The stump mass had a diameter of 2.5 - 3.0 cm and felt somewhat hard. The stump mass was resected because of its potential for incomplete constriction of the ileocecal valve, leading to obstruction. Although he recovered well, histological examination of the resected mass showed that it was a well-differentiated adenocarcinoma of the appendiceal stump (Fig.1). The patient refused to undergo a right hemicolectomy, and he was discharged 10 days after the exploratory laparotomy.

DISCUSSION

Malignant tumours of the appendix are rare, with the rarest type being adenocarcinoma arising from the appendiceal mucosa. Appendiceal malignancies have no specific clinical signs, symptoms, or radiologic features, making preoperative diagnosis difficult. Adenocarcinoma of the appendix has been found in only 0.080% of appendices removed for disease, incidentally or at autopsy^[1-3].

The two specific criteria of carcinoma of the appendix are: 1) continuity of the carcinoma with the appendiceal mucosa; and 2) presence of neoplastic acini containing mucin, thus excluding simple mucocele of the appendix^[4]. Since the muscular layers of the appendix are frequently incomplete or absent, direct extension to adjacent structures may occur early^[3].

An extremely rare subcategory of adenocarcinoma of the appendix is adenocarcinoma involving the post-appendectomy appendiceal stump. The first such

case was described in 1903; at autopsy, a carcinoma was found in the stump of an appendix that had been removed six years previously^[5]. Since then, only four definitive instances of appendiceal stump carcinoma have been reported^[1,3,6,7], including a 54-year-old male with a mucocele of the appendiceal stump 25 years after appendectomy. That tumor was 20 cm in diameter and filled with mucus, which produced a mucocele.

There is no clear evidence of a correlation between removal of the appendix and subsequent development of adenocarcinoma in the appendiceal stump. Radiographically, an inverted appendiceal stump appears as a round, smooth filling defect in the cecal tip at the expected location of the appendix. Some inverted stumps may not be smooth and may have some irregularities, including sharp margins, most likely related to suture granuloma, but the appearance of these stumps may be similar to that of a true neoplastic polypoid lesion.

Radiologic differential diagnosis in post-appendectomy patients with such a finding includes an unusual inverted appendiceal stump, adenomatous polyp, carcinoid of the stump, inflammatory changes, and carcinoma of the cecum or appendiceal stump. Thus, unless previous films are available to document the lack of change in size and configuration, irregular filling defects in the cecal tip must be evaluated by colonoscopy to rule out neoplasm.

Inverted appendiceal stumps may be misdiagnosed as a polyp, granuloma, or lipoma of the cecum. The appendiceal stump can also occasionally cause hemorrhage or ileocolic intussusception. Appendiceal carcinoma is rarely correctly diagnosed preoperatively, with the most common preoperative diagnosis being appendicitis. The rarity of appendiceal carcinoma and its similar presentation to appendicitis make a correct diagnosis difficult.

Among the diagnostic tools available to avoid reoperation are assays for cancer biomarkers in blood. Endoscopically detectable blockage of the ileocecal valve opening is not diagnostic for neoplasms because it may prevent expulsion and cause appendicitis. Endoscopic ultrasonography (EUS) may be useful as also a frozen section examination.

Adenocarcinoma of the appendix may be treated by right hemicolectomy with lymph node dissection, rather than appendectomy. Secondary right hemicolectomy has been recommended, with the risk of recurrence dependent on the degree of histological differentiation and the stage at diagnosis. Thus, surgical method should be determined case by case. In practice, secondary right hemicolectomy following an appendectomy is difficult to perform, with peritoneal dissemination and lymph node metastases sometimes

found at the second operation. Frequently, the only diagnosis possible, even at operation, is an ileocecal mass, for which an ileocecal resection should be performed^[1, 2, 4, 7].

CONCLUSION

Although appendiceal stump adenocarcinoma is rare, surgeons should be aware of possibility of malignancies arising from the stump. Furthermore, they should carefully try to review pathology results of the appendix specimen if available and evaluate patients with chronic obstruction, right lower quadrant pain by CT scan with contrast and colonoscopy. These are the best type of investigation for post-appendectomy abdominal pain. Also, patients should be informed about risks of potential secondary right hemicolectomy.

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