

A Case of Cecal Cancer With Heterotopic Ossification

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Abstract

Heterotopic bone formation in the colon is rare, and its mechanism is still unclear. A 45-year-old man with severe anemia was referred to our hospital and was diagnosed as with cecal cancer on exploratory colonoscopy. On right hemicolectomy with lymphadenectomy, the cut surface of the resected specimen showed a type 2 tumor with a white solid lesion in the cecum. The histopathological study revealed a moderately differentiated adenocarcinoma with ossification. Herein we report and discuss this rare case.

Keywords: Colon cancer; Heterotopic bone formation; Ossification

Introduction

Heterotopic bone formation in malignant neoplasms is common in renal, liver, breast, and skin carcinomas; however, it is rare in the colon. In 1923, there was a first report on a colon cancer with ossification. In 1939, Dukes was the first investigator in the English literature to describe rectal cancer with ossification. The etiology and mechanism of heterotopic bone formation in the colon are still unclear. Therefore, we present a case of cecal cancer with ossification.

Case Report

A healthy 45-year-old man was referred to our hospital for severe anemia (hemoglobin level, 4.8 g/dL). A colonoscopy revealed an irregular protruding tumor in the cecum (Fig. 1), which was infiltrating the Bauhin valve (Fig. 2). Irrigoscopy showed the typical apple-core sign; the patient was diagnosed as having cecal cancer.

The computed tomography scan showed an irregular cecal wall thickening without any signs of calcification or ossification. We observed several lymph node metastases around the

cecum and the ileocolic arteries (Fig. 3); therefore, we performed a right hemicolectomy with a regional lymphadenectomy.

Pathological findings

The resected right hemicolon specimen contained a 70 × 65 mm mass with a white, solid region (Fig. 4). Cross-sections cut from the fixed tissue revealed a moderately differentiated adenocarcinoma with mucus collection. The tumor extended into the subserosal tissue, with absence of necrotic tissue. The white solid lesion comprised an ossification tissue with normal osteoblasts completely surrounded by the benign fibrous tissue. No histological evidence of venous or lymphatic invasion was observed. A total of 27 lymph nodes were recovered from the mesentery; no lymph node metastasis was observed (Fig. 5).

Discussion

Bone formation in the colon is a rare phenomenon. Colon cancer with ossification has been reported as a slowly progressive, moderately to highly differentiated tumor that is not invasive to vessels or lymph nodes. Mucosal and necrotic tissues often coexist [1-4].

Heterotopic bone formation has been observed within

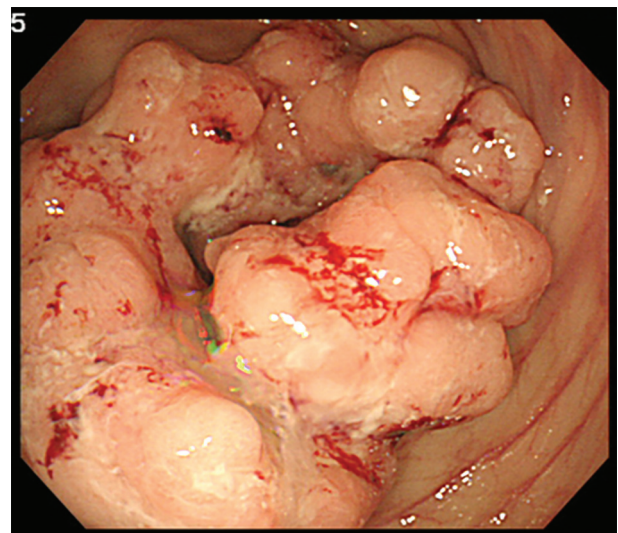


Figure 1. Colonoscopy revealing a cecal cancer with bleeding.

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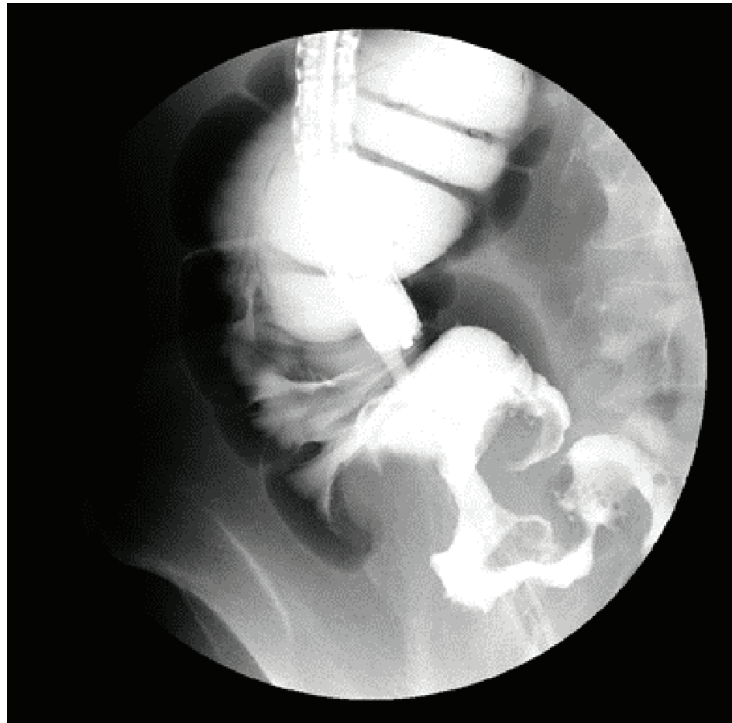


Figure 2. Irrigoscopy showing the typical apple-core sign at the cecum.

the tumor stroma associated with necrosis and within pools of mucus [3, 5-9]. However, most colon cancers with necrotic tissue are not associated with ossification, and not all colon cancers with ossification present with an associated necrotic tissue (similar to the tumor in our case). The association between tissue necrosis and bone formation is unclear; however, it is clear that the presence of necrosis is not a prerequisite for bone formation.

Two hypotheses have been proposed to explain the pathogenesis of heterotopic bone formation: osteoblastic metaplasia of cancer cells and pluripotent mesenchymal cells, both un-

der the influence of factors generated by the cancer cells [5]. Electron microscopic studies in two cases of lung cancers have reported a lack of intermediate stages between tumor cells and osteoblasts [10]. Werner et al have demonstrated the transition from osteoblasts to fully differentiated osteocytes in gastric cancer of rats [11]. These reports suggest the osteoblastic metaplasia of normal mesenchymal cells. In our case, the ossification tissue with osteoblasts was completely surrounded by the benign fibrous tissue, suggesting that the osteoblasts were derived from the pluripotent mesenchymal cells and not the cancer cells.

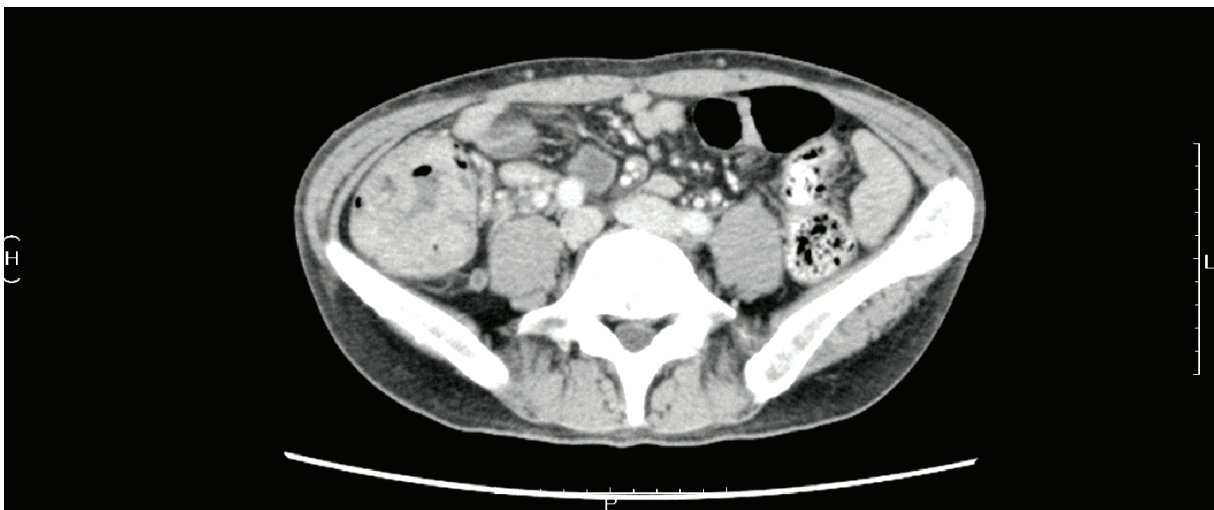


Figure 3. Computed tomography scan showing an irregular cecal wall thickening without any sign of calcification or ossification.

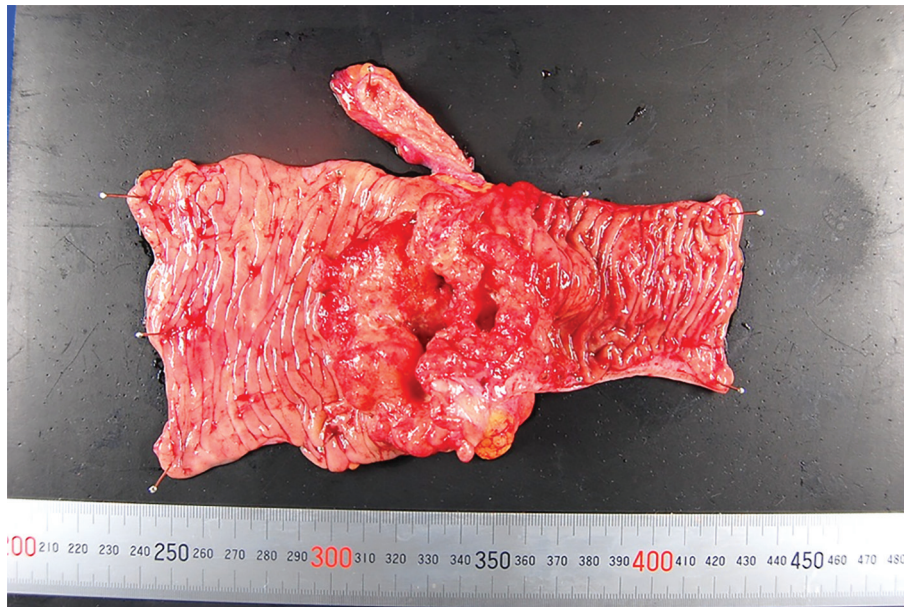


Figure 4. The resected specimen.

There is no difference between colon cancer with and without ossification with respect to the course, management, and prognosis [5, 6]. Apart from colon cancer, osseous metaplasia has also been reported in association with benign polyps of the colon, carcinoids of the stomach, and mucocoeles of the appendix [5, 8, 12].

Osseous metaplasia in colon cancer should not be confused with carcinosarcoma because although the latter is extremely rare, it has a much worse prognosis due to its more invasive nature and high recurrence rate [13]. Colon cancer with ossification can be differentiated from carcinosarcoma because the osseous metaplasia in colon cancer is always associated with benign bone formation, similar to this case [7].

As they have totally different prognoses, colon cancer with ossification needs to be differentiated from other benign lesions and carcinosarcomas. Only few colon cancer cases

with ossification have been reported in the literature, and the pathophysiology is not well known. Further studies are needed to find the etiological, epidemiological, and pathophysiological factors associated with osseous metaplasia in colon cancer.

Conflict of Interest

The authors declare no potential conflict of interest with respect to the research, authorship, and/or publication of this article.

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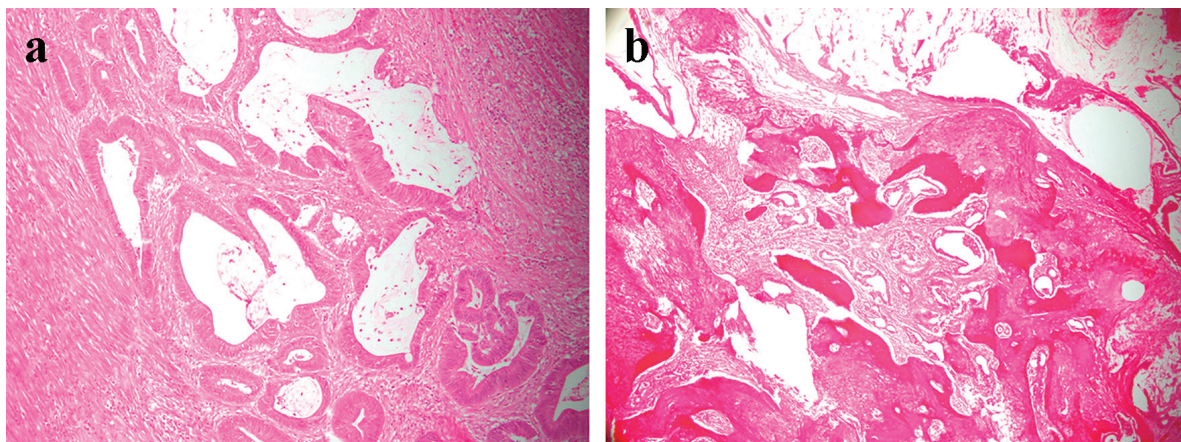


Figure 5. (a) A section of the main tumor comprising moderately differentiated adenocarcinoma tissue. (b) A section of the white solid lesion showing heterotopic bone formation without dysplasia.

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