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CASE REPORT

A case of a rectal adenocarcinoma mimicking a neuroendocrine tumor in the background mucosa of amoebic colitis

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Abstract
A 54-year-old man with diarrhea and hematochezia for 2 months was referred to our department. A total colonoscopy revealed amoebic colitis caused by *Entamoeba histolytica.* Concurrently, a submucosal tumor-like yellowish hemispheric polypoid lesion was incidentally detected in the rectum. We speculated that the lesion was a NET, which could be cured by endoscopic treatment. However, histopathological assessment of a biopsy specimen unexpectedly revealed a well- or moderately differentiated adenocarcinoma. After treatment of the amoebic colitis, endoscopic ultrasound revealed a low, hetero-echoic, 6-mm-diameter lesion mainly in the submucosal layer. We performed surgical resection because the invasion was estimated to be to the deeper submucosal layer. Histopathological assessment of the surgically resected specimen revealed a focal lesion of a well-differentiated adenocarcinoma in the granulation tissue of the submucosal layer. In cases accompanied by amoebic colitis, a tumor's initial gross type might change. Diagnostic endoscopic resection could be acceptable in such cases.

Keywords Adenocarcinoma · Amoebic colitis · Neuroendocrine tumor

Introduction

Submucosal tumor (SMT) is a well-known type of lesion that can be seen during surveillance colonoscopy in clinical daily practice. Most small SMTs do not require treatment, although some may require further investigations and treatment. Among them, rectal neuroendocrine tumors (NETs) are one of the common rectal SMTs encountered [1]. Because NETs < 10 mm in diameter can be cured by endoscopic treatment, they are sometimes treated without confirmation of the histopathological diagnosis by endoscopic biopsy [2, 3]. Here, we present a rare case of a rectal adenocarcinoma mimicking a NET that drastically changed in the clinical time course of amoebic colitis.

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Case report

A 54-year-old man with symptoms of diarrhea and hematochezia for 2 months was referred to our department. A total colonoscopy revealed scattered erosions in the mucosa of the full-length colon. On the basis of the histopathological assessment using periodic acid–Schiff staining of an endoscopic biopsy specimen, he was diagnosed as having amoebic colitis caused by *Entamoeba histolytica* (Fig. 1a). Concurrently, an SMT-like yellowish hemispheric polypoid lesion < 10 mm in diameter was incidentally detected in the rectum during the colonoscopy (Fig. 1b, c). Magnifying endoscopy using blue-laser imaging revealed a regular, round, enlarged-pit pattern on the surface of the lesion. Considering the endoscopic findings, we speculated that the lesion was a NET, which could be cured with endoscopic treatment. However, histopathological assessment of a biopsy specimen unexpectedly revealed a well- or moderately differentiated adenocarcinoma surrounded with the granulation tissue (Fig. 1d).

After 14 days of metronidazole administration, the patient showed improvement in the symptoms caused by amoebic colitis. A colonoscopy during the 4th week after his first admission showed complete disappearance of mucosal

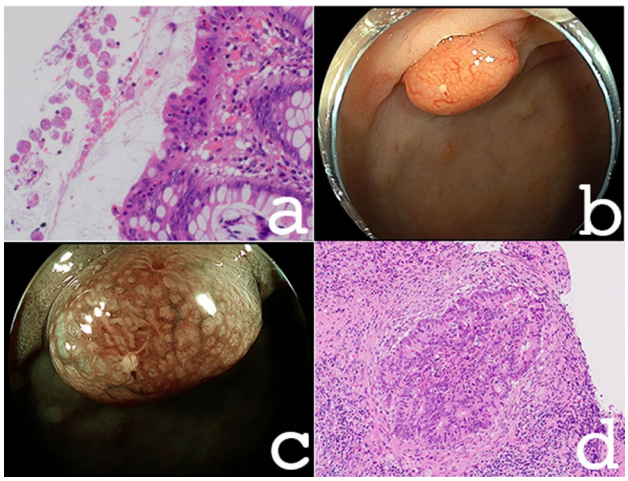

Fig. 1 Endoscopic and histopathological images before the treatment for amoebic colitis. **a** Histopathological image of amoebic trophozoites on the surface of the mucosa, **b** white-light endoscopic image, **c** enhanced endoscopic image, and **d** histopathological image of adenocarcinoma in the granulation tissue

erosions. Simultaneously, the gross tumor type of the polypoid lesion had drastically changed to a flat lesion covered with almost normal mucosa showing regular pits (Fig. 2a, b, c). To assure the location of the lesion, we performed endoscopic ultrasonography (EUS). EUS revealed a low, 6-mm-diameter, hetero-echoic lesion mainly located in the submucosal layer (Fig. 2d). Considering these findings, we speculated that the adenocarcinoma proven in the previous endoscopy was located mainly in the deeper submucosal layer. To rule out the possibility of metastatic lesions from other organs, esophagogastroduodenoscopy, contrast-enhanced computed tomography, and positron emission tomography were performed but showed no other primary neoplasm. Therefore, we concluded that this lesion was a primary adenocarcinoma that originated from the rectum. Finally, we decided to perform surgical resection after obtaining informed consent because the depth of invasion was estimated to be the deeper submucosal layer.

Histopathological assessment of the surgically resected specimen revealed a focal lesion of a well-differentiated adenocarcinoma in the granulation tissue of the submucosal layer (Fig. 3a, b, c). The residual size of the adenocarcinoma was 2 mm in diameter. The depth of invasion was 1.5 mm from the muscularis mucosae, and no vessel invasion was observed. No lymph node metastasis was observed.

Discussion

We presented a case of a rectal adenocarcinoma mimicking a NET that drastically changed shape after treatment of amoebic colitis. Considering the very small volume of adenocarcinoma in the submucosal layer, the polypoid lesion observed during the first colonoscopy mainly showed an inflammatory change caused by amoebic colitis. We speculated that this small adenocarcinoma was not associated with amoebic colitis, although it was incidentally involved in the inflammatory change, which is called an "ameboma."

A persistent *E. histolytica* infection in the colon has been reported to cause formation of the mass-like granuloma known as an ameboma [4]. *E. histolytica* infection typically begins with ingestion of mature quadrinucleated cysts. Excystation occurs in the small intestine with the release of motile trophozoites, which migrate to the large intestine.

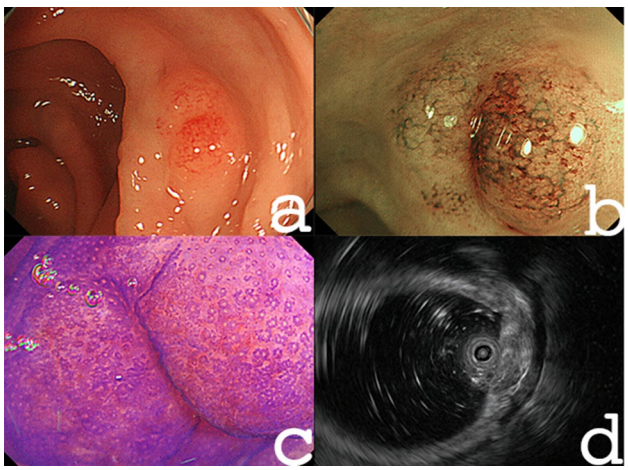

Fig. 2 Endoscopic images after the treatment for amoebic colitis. **a** White-light endoscopic image, **b** enhanced endoscopic image, **c** crystal violet-stained image after treatment of amoebic colitis, and **d** endoscopic ultrasonography image

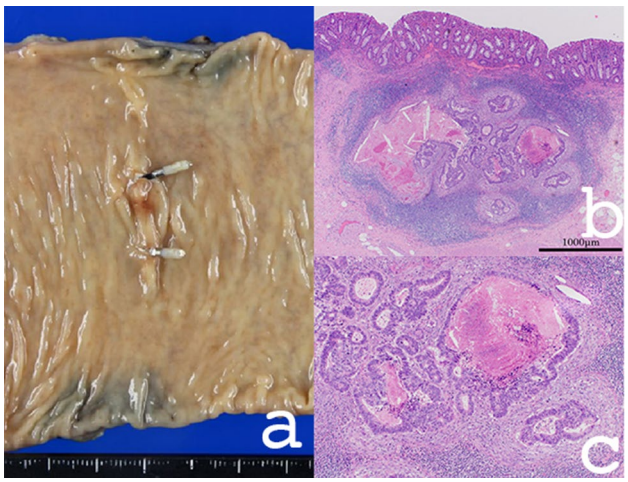

Fig. 3 Histopathological images of the surgically resected specimen. **a** Macroscopic view and **b**, **c** microscopic images showing a well-differentiated adenocarcinoma in the granulation tissue of the submucosal layer

Through binary fission, trophozoites form new cysts, and both stages are shed in feces. Only trophozoites have the capacity to adhere and lyse colonic epithelium and subsequently spread hematogenously to distant sites, such as the peritoneum, liver, lung, or brain [5]. After penetration of the mucus barrier, trophozoites reach the luminal surface, lyse surrounding cells, and degrade the extracellular matrix components of the colonic mucosa. As a result of lysed cells and necrosis, a thick exudate containing acellular proteinaceous material, red blood cells, and strands of fibrin is formed under the viable submucosa. Finally, infiltration of abundant neutrophils, lymphocytes, and macrophages is observed, and granuloma is formed around the necrotic tissue [6].

In the present case, considering that most of the tumor in the surgical specimen was composed of granuloma without trophozoites and that the surgical operation was performed after the treatment of amoebic colitis, it might be natural that a SMT-like polypoid lesion would be a small ameboma and drastically change its shape in the healing process of amoebic colitis.

Undoubtedly, we must consider the possibility of a rare type of SMT-like adenocarcinoma. Colon cancers resemble SMTs and are called dome-type carcinomas or gastrointestinal-associated lymphoid tissue carcinomas, which are thought to arise from the M-cells of the lymphoglandular complex of the intestine [7]. Histologically, they are characterized by submucosal localization, a prominent lymphoid infiltrate with germinal center formation, and dilated glands lined by bland columnar epithelial cells with eosinophilic cytoplasm. However, no such findings were observed in the present case. Considering all histopathological findings and the clinical course, we speculated that the adenocarcinoma on the rectal mucosa was incidentally involved in the submucosal layer in the process of ameboma formation caused by amoebic colitis.

In conclusion, the estimated massive invasion observed on EUS might have overestimated granulation around the adenocarcinoma. In cases accompanied by amoebic colitis, a tumor's initial gross type might change. Radical resection was chosen for the patient in our case, although diagnostic endoscopic resection could be acceptable in similar cases.

Compliance with ethical standards

Conflict of interest Shun Ito, Satoshi Ono, Akihiro Kobayashi, Kyohei Maejima, Shosuke Hosaka, Kiyotaka Umeki, Shin-ichiro Sato, Satoshi Wakasugi, and Kenji Ogata declare that they have no conflict of interest.

Human and animal rights All procedures followed have been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki and its later amendments.

Informed consent Informed consent was obtained from all patients before the treatment.

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