Case Report

Adenocarcinoma of the Small Intestine in a Young Adult Diagnosed by Double-balloon Enteroscopy

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A 29-year-old man presented with a 3-month history of abdominal pain and appetite loss. Superficial lymph node adenopathy was noted. Systemic computed tomography showed multiple liver and lung metastases, as well as ascites. No abnormalities were found on upper gastrointestinal endoscopy and colonoscopy; therefore, double-balloon enteroscopy was performed. A stenosis with reddish and edematous mucosal changes from the third part of the duodenum to the upper jejunum was noted; on histopathology of the biopsy specimens, adenocarcinoma was diagnosed. Thus, the patient had advanced small intestinal cancer with carcinomatous peritonitis and liver metastases. Although the patient was given chemotherapy with cisplatin and 5-fluorouracil, he died 2 months after commencing treatment. Primary small intestinal carcinoma is a rare malignancy; most cases cannot be detected on routine gastrointestinal endoscopy due to their location. Our experience suggests that double-balloon enteroscopy is useful for diagnosing small intestinal adenocarcinoma.

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Introduction

Primary adenocarcinoma of the small intestine is rare. Although the small intestine accounts for about three-quarters of the gastrointestinal tract's length, malignant tumors of the small intestine only account for 1-5% of all gastrointestinal tract tumors.¹ The rarity of these tumors, combined with the presence of non-specific signs and symptoms, can delay prompt diagnosis and treatment.¹² As well, the early detection of these lesions has been hindered by their inaccessibility to endoscopy and the limitations of radiographic techniques.¹²

Recently, double-balloon enteroscopy (DBE) has been developed in Japan.³ DBE allows high-resolution visualization, as well as diagnostic and therapeutic interventions, for all segments of the small intestine.³ In our patient, DBE was useful for identifying the origin of the patient's systemic lymph node and distal metastases, which were the result of a primary adenocarcinoma of the small intestine.

Case report

A 29-year-old man presented with abdominal pain, appetite loss, and general fatigue of 3 months' duration. His past history was noncontributory. His mother had died of pancreatic cancer. On physical examination, lymph node adenopathy of the neck and axilla was noted. On histopathological examination of the excisional biopsy of a neck lymph node, a moderately differentiated adenocarcinoma was found. Admission hematological data showed an iron deficiency anemia (Hb 8.3 g/dL, Fe 15 mg/dL, and UIBC 306 mg/dL) and an elevation of CRP (4.03 mg/dl). CA19-9 was elevated to 1580 U/ml, while the CEA was 1.2 ng/mL (within normal). Computed tomography of the abdomen and chest showed multiple liver and lung metastases, peri-aortic lymph node swelling, and ascites. No tumors were not identified in the biliary tract and pancreas on the imaging. No abnormalities were found on upper gastrointestinal endoscopy and colonoscopy. An upper gastrointestinal barium examination with small bowel follow-through showed irregular narrowing from the

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ascending part of the duodenum to the upper jejunum (Figure 1), suggesting neoplasia of the small intestine. Thus, DBE was performed to confirm the x-ray findings and to obtain a tissue diagnosis. On DBE, an annular constricting lesion with reddish and edematous mucosal changes was noted from the ascending part of the duodenum to the upper jejunum (Figure 2). On histopathological examination of the biopsy specimens, moderately to poorly differentiated adenocarcinoma was noted. Based on these findings, the patient was diagnosed as having very advanced small intestinal cancer with distant metastases. Given that the patient presented with stage IV disease, 4 courses of monthly chemotherapy consisting of cisplatin (5 mg/body/day) on the first day and 5-fluorouracil (750 mg/body/day) for 5 consecutive days were given. Nevertheless, the patient's condition deteriorated, and he died 2 months after com-



Figure 1. Barium radiography of the small bowel shows irregular narrowing from the ascending part of the duodenum to the upper jejunum.



Figure 2. Double-balloon enteroscopy shows an annular constricting lesion with reddish and edematous mucosal changes from the third portion of the duodenum to the upper jejunum.

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mencing treatment. Autopsy findings included moderately differentiated adenocarcinoma of the duodenum and jejunum (Figure 3 and 4) with multiple metastases (bilateral lungs, liver, bilateral adrenal glands, urinary bladder, peritoneum, omentum, mesentery, and generalized lymph nodes) and direct invasion into the stomach, duodenum, ileum, colon, pancreas, gall bladder, and left kidney.



Figure 3. Macroscopically, circumferential irregular stenosis can be seen from the duodenum to the jejunum.



Figure 4. Histopathology shows moderately differentiated adenocarcinoma of the small bowel.

Discussion

The rarity of adenocarcinoma arising in the small intestine is largely unexplained; however, it is most likely due to a combination of several factors.⁴⁻⁶ The factors that have been suggested as playing an important role in reducing tumor formation include: 1) the dilute nature and rapid transit time of the small bowel contents, which reduce mucosal contact with intraluminal carcinogens; 2) a reduced number of bacteria, which have been associated with the carcinogenic Junyo oh et al.: Double-balloon Enteroscopy for Small Intestinal Cancer

effect that broken down bile salts have in the colon; 3) the alkaline pH of the small intestine, which exerts a protective effect; and 4) a high concentration of secretory IgA antibodies, which may perhaps neutralize oncogenic pathogens.

Several conditions are known to be associated with a higher incidence of primary small bowel carcinoma. Lioe et al. noted that a pre-existing villous adenoma was found adjacent to 6 of 25 (24%) adenocarcinomas (5 jejunal and 1 ileal).¹ T cell lymphomas are known to arise in the jejunal mucosa of celiac disease patients; celiac disease is also known to be associated with adenocarcinoma.⁷ Cunningham et al. documented that, of their 29 patients with small intestinal cancer, all 6 cases with small intestinal cancer in the ileum occurred in Crohn's disease patients.² However, our present case had no conditions that are known to be associated with small intestinal cancer.

The most common symptom of small bowel cancer is pain, followed by gastrointestinal bleeding, weight loss, nausea, and vomiting;12 the current patient had all of these symptoms. The nonspecific nature of the symptoms usually leads to a delay in presentation and diagnostic workup. Adenocarcinoma of the small intestine carries a dismal prognosis. Lioe et al. reported that over half of their 25 patients had nodal disease and distant metastases at the time of diagnosis; the 5-year survival was 15.7%.¹ The median age of the affected patients is 50-60 years old; only 5.0-5.7% of patients are under 30 years of age.⁸⁻¹⁰ Moreover, the younger patients tend to have more advanced disease than the older patients.¹⁰ The present case had an advanced stage with multiple nodal and distal metastases; combined chemotherapy did not appear to prolong his survival. Moreover, due to their location, most lesions cannot be detected by routine gastrointestinal endoscopic procedures. Thus, investigation of the small intestine has been the ultimate challenge for gastroenterologists. DBE is a new endoscopic technique that was developed to allow the entire small intestine to be viewed both in an antegrade and a retrograde manner, and it allows therapeutic procedures to be carried out when appropriate.3 DBE is safe, and complications are uncommon. Several reports have shown that the definite diagnosis of small bowel neoplasms, including adenocarcinoma, can be made using DBE.^{11,12} The arrival of capsule endoscopy early this decade represented a promising, noninvasive, and well-tolerated method of examining the small intestine.¹³ Compared to conventional investigations, such as push enteroscopy and small bowel barium radiography, capsule endoscopy has been reported to have a higher yield.¹³ However, capsule endoscopy is not without its limitations.

Chong et al. described 4 patients in whom small bowel pathology was missed on capsule endoscopy but was detected on DBE;¹¹ two of these patients had gastrointestinal stromal tumors, and one had an adenocarcinoma. May et al. reported that, using DBE, additional and definite bleeding sources were identified in at least 8 of 52 patients, and the diagnosis determined on capsule endoscopy in 5 of the 32 patients who were capsule-positive was changed.¹⁴ These studies suggest that DBE may have a greater yield than capsule endoscopy. Nevertheless, DBE examinations are more labor intensive, require the use of sedation, and may not allow the small intestine to be completely examined from either the antegrade or the retrograde approach alone. Thus, capsule endoscopy and DBE may be considered to be complementary.

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