

Rectal resection for Schnitzler's metastasis in a patient presenting with severe rectal stenosis: case report and review of the literature

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ABSTRACT

Rectal metastasis resulting from the hematogenous spread of gastric cancer (Schnitzler's disease) is rarely seen. We report a case of 53-year-old female with rectal metastasis from gastric cancer who presented with abdominal pain, tenesmus, and severe constipation. Digital rectal examination revealed a severe stenosis 5-6 cm from the dentate line. Sigmoidoscopy showed a narrowed lumen at 5 cm up from the dentate line. The evident radiologic finding was circumferential stenosis of the rectum located at the same level. Very low anterior resection and total mesorectal excision with coloanal anastomosis in addition to protective ileostomy were performed. Pathological examination revealed metastatic adenocarcinoma (Schnitzler's metastasis). The patient was discharged on the 9th postoperative day with no adverse events. It should be kept in mind that rectal stenosis could be metastasis from gastric carcinoma if consistent with the patient's history.

Keywords: Schnitzler's disease, gastric cancer, rectum metastasis

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The known routes of distant metastasis after curative gastrectomy include lymphatic spreading (Virchow's node), peritoneal seeding (Krukenberg's tumor or Plummer's rectal shelf) or hematogenous spread (Schnitzler's metastasis) [1-3]. We present a patient with rare Schnitzler's metastasis who lived 42 months after curative treatment of gastric cancer.

CASE PRESENTATION

A 53-year-old woman presented with constipation, tenesmus, abdominal pain, and weight loss of 8-9 kg over a period of 2 months. Her past medical history

was significant for signet ring cell gastric adenocarcinoma (T2N0M0), for which she underwent total gastrectomy with D2 lymph node dissection 42 months earlier. Pathological examination at that time showed the surgical resection margins to be clear. Twenty-one lymph nodes were harvested, and were found to be tumor-free. In the follow-up period of 42 months after total gastrectomy, she was admitted to our General Surgery Clinic with constipation, tenesmus, and abdominal pain.

Digital rectal examination revealed severe stenosis 5-6 cm from the dentate line without any mucosal lesion. Blood test results and serum tumor markers were normal. Sigmoidoscopy showed a narrowed



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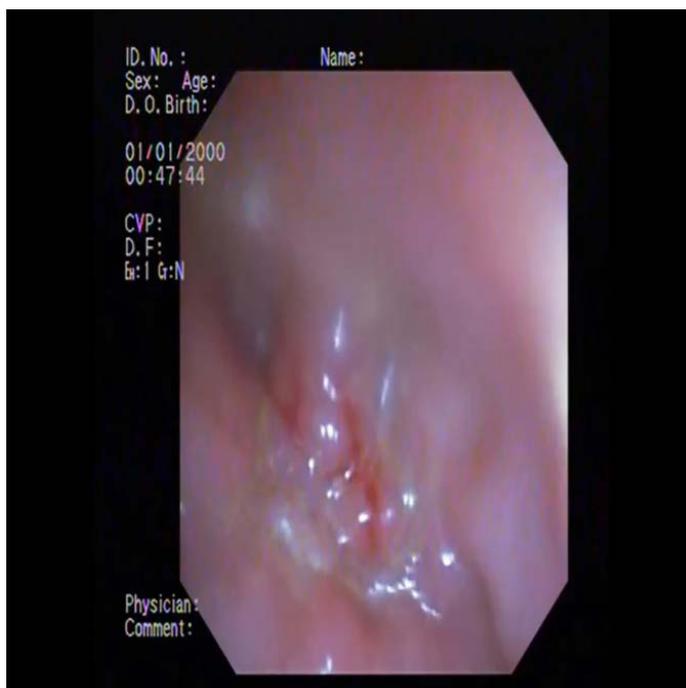


Figure 1. Sigmoidoscopy showing the narrowed lumen at 5th cm from the dentate line.

lumen at the 5th cm from the dentate line (Figure 1). Histopathological examination of the rectal mucosal biopsy did not find any signs of tumoral involvement.

Abdominal and pelvic computed tomography (CT) showed circumferential stenosis of the middle rectum (Figures 2 and 3). There were no signs of metastatic involvement in her pelvic area. At positron emission tomography-computed tomography (PET-CT), a high standardized uptake value (SUV) of 5.8

was detected at the upper-middle rectum (Figure 4). The rectal stenosis was defined as rectal fibrosis on the PET-CT scan. The patient’s past medical history and imaging features made us consider the lesion as metastasis of the rectum from gastric malignancy rather than fibrosis; this possibility was more likely than that of primary malignancy.

Informed consent has been taken from the patient. Very low anterior resection (VLAR) and total

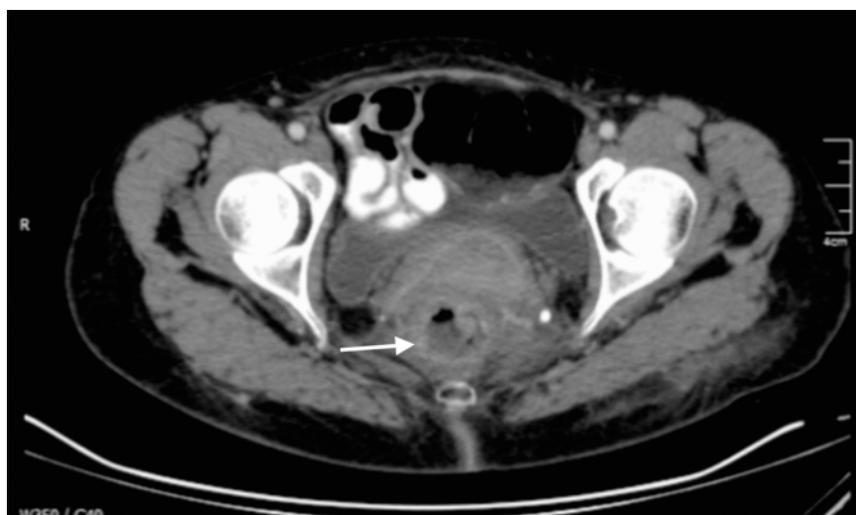


Figure 2. Axial CT image showing normal lumen of rectum from the proximal part of rectal stenosis.

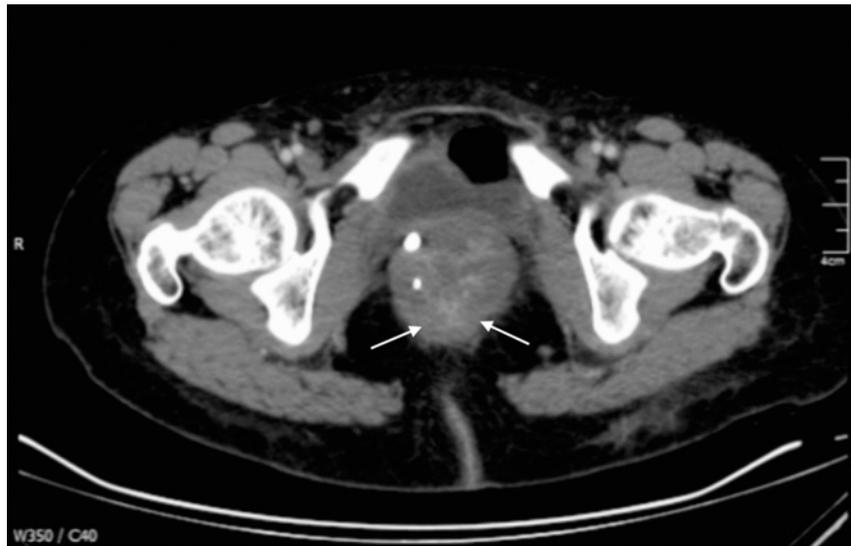


Figure 3. Axial CT image showing circumferential stenosis of the distal rectum.

mesorectal excision (TME) with pull-through coloanal anastomosis were performed (Figure 5). In addition, protective ileostomy was done. During the operation, frozen section revealed an unclear malignancy with glandular structure. Nevertheless, the entire abdominal exploration gave no evidence of malignant nodular involvement, either in the abdominal spaces or on the pelvic peritoneal surfaces.

Histopathological examination revealed metastatic adenocarcinoma with the lesion originating with histopathology similar to the gastric cancer, which occurred most likely before spreading via the hematologic route.

The patient was discharged on the 9th day after surgery without any complications. The patient returned home by flying internationally on



Figure 4. At fused PET-CT image, a high standardized uptake value (SUV) of 5.8 at the upper-middle part of rectum is demonstrated.



Figure 5. Macroscopic appearance of the resected rectal segment.

postoperative day 11. By that time, she had begun to pass her convalescent period. However, at the early stage of that period, she died. The reason has not been clarified to us by her local physician.

DISCUSSION

Gastric cancer is the fourth most commonly diagnosed malignancy and the second most common cause of cancer mortality worldwide [4]. The recurrence patterns after curative surgery for gastric cancer have been described as locoregional recurrence, peritoneal recurrence, and distant metastasis. Hematologic (Schnitzler's metastasis) or lymphatic spread from gastric cancer to the rectum is quite rare. We found the lesion of the present patient located in the middle-lower part of the rectum adjacent to the muco-subserosal layer. During the operation we did not determine the presence of any serosal exposure of tumor, or peritoneal or pelvic dissemination, or ascites. These findings supported the idea that this patient's metastatic rectal cancer was due to hematologic or lymphatic dissemination. Therefore, we are reporting this quite unusual case of rectal Schnitzler's metastasis via a hematogenous route of

gastric adenocarcinoma.

Only ten cases, including the present case, have been reported in the English literature of Schnitzler's metastasis to the rectum [5-13]. The mean age of the reported patients was 58.2 years. Seven of these patients were female. Schnitzler's metastasis to the rectum has been encountered synchronously and/or metachronously. Three of these cases in the literature were synchronous and the others, including our case, were metachronous Schnitzler's metastasis to the rectum. In our patient, metachronous Schnitzler's metastasis occurred 42 months after completion of treatment for gastric adenocarcinoma. In the reported cases, the median disease-free survival after gastrectomy was 38 months (range: 18-72 months) for metachronous metastases [5-12]. The disease-free survival of the present patient was 42 months. The cardinal symptoms of the previously reported cases were constipation, abdominal distension and tenesmus; and in the present case, the cardinal symptoms were severe constipation, abdominal distension and excessive weight loss. In these cases, the most prominent finding was stenosis of the rectum, which did not allow the passage of stool due to the metastatic stenosis, as in our case.

Sigmoidoscopy, colonoscopy and CT with

contrast is the most effective diagnostic strategy for reaching a true diagnosis of rectal metastasis. PET-CT can be used to determine whether the rectal lesion is malignant. In a case presented by Lim *et al.* [8], PET-CT showed hypermetabolic activity in the rectum. In the present case, high SUV 5.8 was detected in the upper-middle rectum, and the rectal stenosis was defined as rectal fibrosis.

The endoscopic appearance of luminal metastasis is variable, but the characteristic features are diffuse circumferential wall thickening and stiffness. Rectal biopsy is generally useful for determining metastatic cancer of the rectum. In our case, rectal biopsy did not give any sign of tumoral involvement.

An expandable metallic stent, lateral decompressive left colostomy, extended Hartmann's procedure, low anterior resection, and chemotherapy were chosen as methods of treatment for the patients of the previous reports. For our patient, we decided to perform VLAR and TME with pull-through coloanal anastomosis to remove the rectum completely in accordance with the oncologic resection on evaluation of the findings of CT, pelvic MRI and PET-CT. Closure of the protective ileostomy was planned to be accomplished quite a long time later to allow a second look in the abdominal cavity. But the patient unfortunately died after several days in her convalescent time period after she returned to her home by an international flight.

CONCLUSION

Rectal metastasis presenting as rectal fibrosis from gastric cancer is very rare. When a patient with a history of gastric cancer presents with symptoms such as change of bowel habits (tenesmus or constipation) and/or rectal stenosis, even if biopsy results are negative for malignancy, it should be kept in mind that rectal stenosis can possibly originate from Schnitzler's metastasis resulting from hematogenous spread of a gastric carcinoma in a patient with a relevant history.

Authorship declaration

All authors listed meet the authorship criteria according to the latest guidelines of the International Committee of Medical Journal Editors, and all authors

are in agreement with the manuscript.

Informed consent

Written informed consent was obtained from the patients for publication of this case report and any accompanying images.

Conflict of interest

The authors declared that there are no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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