

Xanthogranulomatous gastritis of the remnant stomach mimicking a malignant tumor: A case report

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Abstract. Xanthogranulomas are known to develop in the gallbladder and kidney. Xanthogranuloma of the stomach is a rare disease, and to the best of our knowledge, only a few cases have been reported to date. The present patient was a 64-year-old man who underwent a wide resection of the stomach following a Billroth-I reconstruction for a gastric ulcer ~40 years prior to the current presentation. Due to tarry stools, a gastrointestinal endoscopy was performed, leading to identification of an ulcerated gastric lesion located at the previous suture line at the lesser curvature of the remnant stomach. This lesion was elevated, appearing to indicate a submucosal tumor. Positron emission tomography revealed uptake of fluorodeoxyglucose radiotracer by the tumor. Although not indicated by the biopsy specimens, a malignant tumor of the remnant stomach was suspected, in the form of a malignant gastrointestinal tumor or remnant gastric cancer. Curative resection of the tumor was successfully performed. Histological examination of the resected specimens revealed xanthogranulomatous inflammation consisting of foamy histiocytes and plasma cells, however, no cancer cells were observed. The tumor was diagnosed as xanthogranulomatous gastritis that mimicked a malignant tumor of the remnant stomach. The present study therefore indicates that inflammatory tumors should be considered in the differential diagnosis of malignant tumors.

Introduction

Xanthogranulomas are known to develop in the gallbladder and kidney (1,2). However, rare cases of xanthogranulomatous inflammation have additionally been reported in other organs, including the stomach, colon, pancreas and uterus (3-6). Despite being a benign disease, this uncommon inflammation can progressively invade adjacent organs, mimicking malignant tumor, which often leads to unnecessary resection. Preoperative diagnosis of xanthogranulomatous inflammation is difficult and its features are unknown (1-6). A xanthogranuloma occurring in the stomach is rare, and to the best of our knowledge, only a few cases have been reported to date (6-16). In such cases, the disease was preoperatively misdiagnosed as a submucosal tumor or advanced gastric cancer, and gastrectomy was performed (6-16). Due to the rarity of xanthogranulomatous gastritis, incidence and mortality rates remain unclear. In addition, no optimal treatments have been identified for this condition. The present study reports a rare case of xanthogranulomatous gastritis of the remnant stomach following partial gastrectomy, mimicking a malignant tumor. Written informed consent was obtained from the patient

Case report

A 64-year-old man previously underwent a wide resection of the stomach, following a Billroth-I reconstruction for a gastric ulcer (details unknown). Approximately 40 years after this, in July 2014, the patient presented to Kawagoe Gastrointestinal Hospital (Kawagoe, Japan) due to tarry stools. A gastrointestinal endoscopy was performed, leading to identification of a gastric lesion at the previous suture line of the lesser curvature of the remnant stomach, which was elevated and appeared to indicate a submucosal tumor (SMT) with an ulcerated lesion (Fig. 1). The patient was then referred to Keio University Hospital (Tokyo, Japan) in August 2014. There was no increase in the serum levels of carcinoembryonic antigen or carbohydrate antigen 19-9. Computed tomography revealed wall thickening in the lesser curvature of the remnant stomach and swollen regional lymph nodes (Fig. 2A). Positron emission tomography (PET) revealed uptake of the fluorodeoxyglucose (FDG) radiotracer by the tumor (Fig. 2B), with a maximum standard uptake value (SUV) of 8.41 at the early phase and

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Abbreviations: SUV, standard uptake value; GIST, gastrointestinal stromal tumor; SMT, submucosal tumor; PET, positron emission tomography; FDG, fluorodeoxyglucose

Key words: xanthogranulomatous gastritis, xanthogranuloma, stomach, remnant stomach, inflammatory tumor

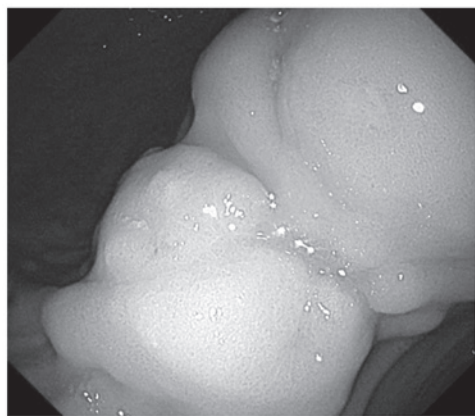


Figure 1. Endoscopic image of the tumor. The lesion was located at the previous suture line of the lesser curvature of the remnant stomach and was elevated, appearing to indicate a submucosal tumor with an ulcerated lesion.

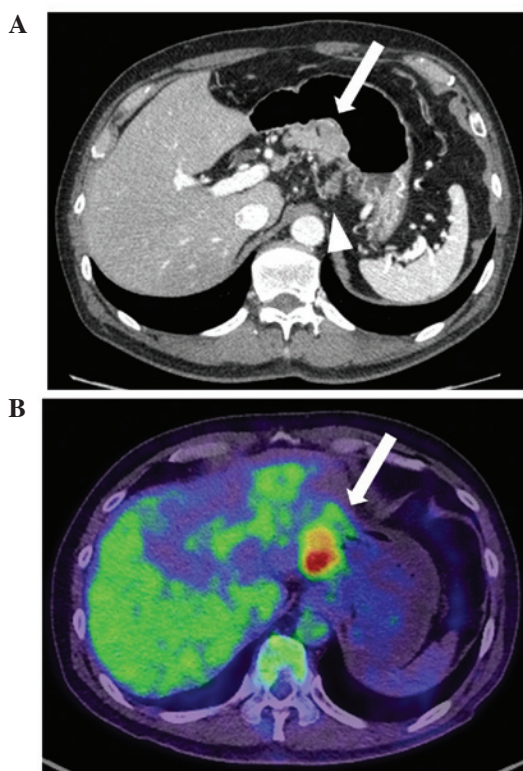


Figure 2. Abdominal computed tomography and positron emission tomography images of the tumor. (A) Computed tomography image revealing wall thickening in the lesser curvature of the remnant stomach (white arrow) and swollen regional lymph nodes (white arrowhead). (B) Positron emission tomography image revealing fluorodeoxyglucose radiotracer uptake by the tumor (white arrow).

7.94 at the late phase. Biopsy specimens from the lesion indicated chronic gastritis with regenerative changes and intestinal metaplasia.

Although not indicated by the pathological findings, it was suspected that the tumor was a highly malignant entity, and was potentially a malignant gastrointestinal stromal tumor (GIST) or remnant gastric cancer with extended submucosal invasion due to the marked FDG uptake. As a curative resection appeared possible, complete resection of the remnant stomach

was performed, with a lymphadenectomy and splenectomy. According to the Japanese Gastric Cancer Treatment Guidelines 2010 (ver. 3), these procedures are required for the performance of curative resection for advanced remnant gastric cancer (17).

Open surgery was performed, which revealed severe adhesion of the left hepatic lobe and gastric wall. The lesion was located at the lesser curvature of the remnant stomach and was relatively similar to a lipoma. However, the regional lymph nodes were swollen; thus, the intended procedures were performed as planned. The specimens were removed, and a Roux-en-Y reconstruction was performed. The post-operative course was positive, and the patient was discharged from hospital 13 days after surgery.

Macroscopically, the tumor was soft, measuring 65x40 mm, and appeared to be a combination of a slightly depressed lesion and SMT (Fig. 3). Histological analysis of the resected remnant stomach revealed xanthogranulomatous inflammation with foamy histiocytes and plasma cells from the submucosal to subserosal layers (Fig. 4). Chronic gastritis with regenerative changes and intestinal metaplasia was observed on the surface of the type 0-IIc lesion. A small number of multinucleated giant cells were observed in the marginal sinuses of the resected lymph nodes. However, no cancer cells were observed in the resected specimens. Therefore, the tumor was diagnosed as xanthogranulomatous gastritis.

At follow-up 1 year after surgery, there was no evidence of recurrence of inflammation. The patient continues to undergo postoperative gastrectomy follow-up.

Discussion

Xanthogranulomatous inflammation is a rare inflammatory lesion characterized by marked proliferative fibrosis, with infiltration of foamy histiocytes and other acute and chronic inflammatory cells (13). These lesions are common in the gallbladder as xanthogranulomatous cholecystitis and in the kidney as xanthogranulomatous pyelonephritis (1,2). However, rare cases of xanthogranulomatous inflammation have additionally been reported in other organs, including the stomach, colon, pancreas and uterus (3-6). The occurrence of xanthogranuloma of the stomach is rare, and only a few cases have been reported to date (6-16). To the best of our knowledge, this is the first reported case of xanthogranulomatous gastritis in the remnant stomach.

The pathogenesis of xanthogranuloma remains to be elucidated, although it is proposed to be a chronic lesion associated with infection, immunological disorders, lymphatic obstruction and lipid transport (14). It is additionally speculated that xanthogranulomatous cholecystitis results from sterile chronic inflammation due to extravasation of bile into the gallbladder wall with involvement of the Rokitansky-Aschoff sinuses or via a small mucosal ulceration (18). Although no studies have described a correlation between xanthogranulomatous gastritis and previous gastric surgery, the pathogenesis in the present case may be associated with the response following the previous surgery, as the tumor was located at the previous suture line. In addition, Guarino *et al* (11) suggested the potential correlation of bile

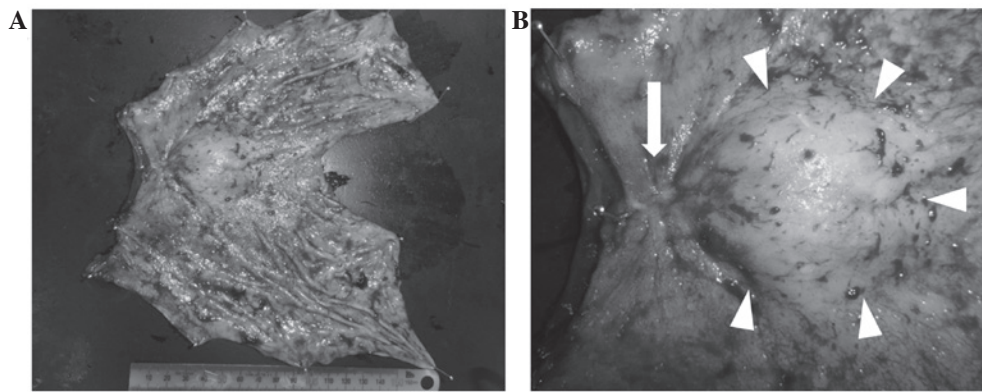


Figure 3. Macroscopic imaging of the resected remnant stomach. (A) Resected remnant stomach. The tumor was relatively soft similar to a lipoma, and measured 65x40 mm. (B) The tumor appeared to be a combination of a type 0-IIc lesion (white arrow) and a submucosal tumor (white arrowheads).

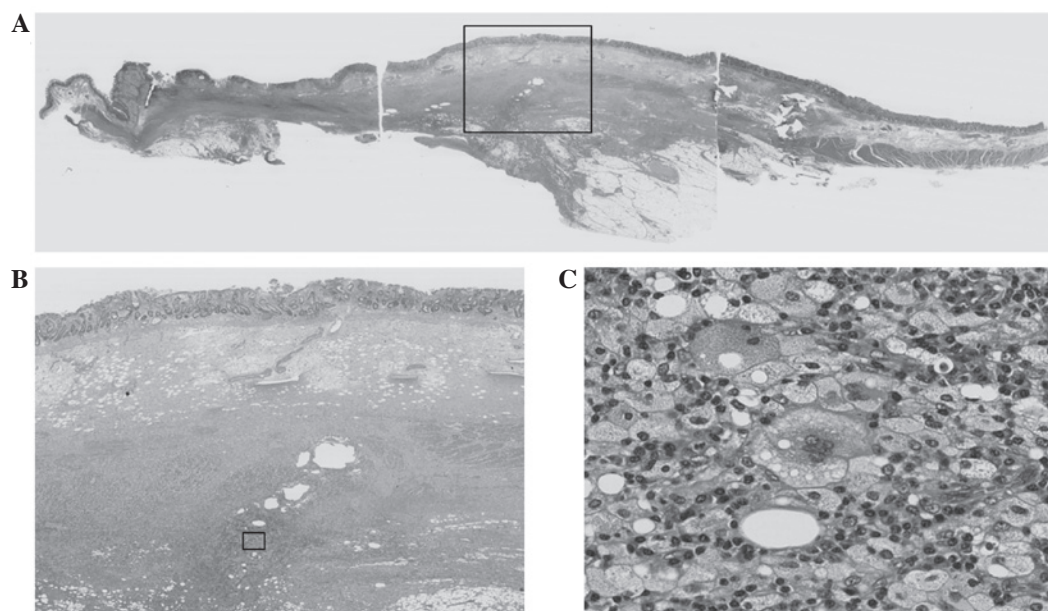


Figure 4. Histopathological results of the tumor. Xanthogranulomatous inflammation, consisting of foamy histiocytes and plasma cells from the submucosal to subserosal layer, was observed by hematoxylin and eosin staining. (A) Whole image (low-power field; objective magnification, x0.5). (B) A magnified view of the inset black box indicated in image A (low-power field; objective magnification, x1.25). (C) A magnified view of the inset black box indicated in image B (high-power field; objective magnification, x40).

reflux into the stomach following a wide gastrectomy subsequent to Billroth-I reconstruction with the development of xanthogranulomatous gastritis.

As in the present case, xanthogranulomatous gastritis has been misdiagnosed as SMT and gastric cancer in previous reports (6-16). Although not indicated by pathological findings, a malignant gastric tumor was initially suspected, for example a malignant GIST or gastric cancer, due to the gross features observed by endoscopy and the marked FDG uptake observed during PET. A diagnosis of xanthogranulomatous gastritis was not considered. Despite a low incidence, it is clear that the possibility of an inflammatory tumor should be included in the differential diagnosis of malignant tumors. However, none of the previously reported cases were definitively diagnosed prior to surgical resection. Therefore, distinguishing xanthogranulomatous gastritis from other malignancies remains difficult. In addition, xanthogranulomatous gastritis combined with gastric cancer should be considered in these cases (14,16).

In the present case, endoscopic evaluation revealed an elevated ulcerated lesion. Biopsy specimens did not indicate malignancy. However, this result did not definitively contraindicate advanced gastric cancer; thus, the tumor was 'overdiagnosed' and a radical resection was performed. Histopathologically, on the surface of the lesion, only chronic gastritis with regenerative changes and intestinal metaplasia was observed. In addition, the layer of muscularis propria was lacking, and the laminar structure of the gastric wall was not retained. It was speculated that destruction of the laminar structure occurred due to the inflammation, or that it had potentially been altered during the previous surgery. The ulcerated section of the lesion may have been associated with the xanthogranulomatous inflammation itself or with the previous suture line.

PET is an imaging method that has a significant role in the evaluation of a wide range of malignancies (19). The SUV is used as a semi-quantitative measure of the degree of metabolic activity in abnormal tissues. In general, inflammation

is indicated by intense FDG uptake, which may additionally indicate malignancy, due to the intense glucose metabolism in inflammatory cells. Previous studies have noted that it is challenging to differentiate between xanthogranulomatous inflammation and malignancy using PET (13,20). Therefore, arriving at a clinical diagnosis of xanthogranulomatous gastritis may be difficult and may only be established via histological examination.

In conclusion, to the best of our knowledge, the present case is the first report of xanthogranulomatous gastritis of the remnant stomach mimicking a malignant tumor. Therefore, inflammatory tumors should be considered in the differential diagnosis of malignant tumors, even though distinguishing them from other tumors remains challenging.

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