

## Synchronous triple occurrence of MALT lymphoma, schwannoma, and adenocarcinoma of the stomach

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### Abstract

We present a case of a 56-year-old man with 3 synchronous gastric tumors. The patient presented with melena, and 3 gastric abnormalities were detected on gastroduodenoscopic examination, including a small ulcerative lesion in the gastric antrum, a submucosal mass in the gastric body, and severe erosion in the fundus. Histological examination of biopsy samples yielded respective diagnoses of gastric adenocarcinoma, gastritis, and mucosa-associated lymphoid tissue (MALT) lymphoma. The patient first received medication to eradicate any underlying *Helicobacter pylori* infection, which might have been a cause of the MALT lymphoma. Four weeks later, after examination of repeat biopsy samples revealed that the MALT lymphoma had resolved, the patient underwent subtotal gastrectomy. Further histological examination of resected tissue confirmed the antrum lesion as adenocarcinoma and the body lesion as schwannoma. To our knowledge, this is the first reported case of synchronous triple primary gastric adenocarcinoma, MALT lymphoma, and schwannoma.

**Key words:** Gastric cancer; Synchronous; Mucosa-associated lymphoid tissue lymphoma; Schwannoma; Triple

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**Core tip:** Synchronous occurrence of two types of gastric tumor is relatively well described, but this is the first report of synchronous triple gastric adenocarcinoma, mucosa-associated lymphoid tissue

(MALT) lymphoma, and schwannoma. Consideration of the possible underlying involvement of *Helicobacter pylori* in MALT lymphoma and preoperative treatment with appropriate medication allowed performance of subtotal gastrectomy and a successful outcome.

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## INTRODUCTION

The most frequent diagnosis of tumor of the stomach is adenocarcinoma (90%-95%), followed by lymphoma, gastrointestinal stromal tumor (GIST), and carcinoid tumor. Synchronous occurrence of two types of tumor in the stomach is relatively well known. Here, we present an extremely rare case of synchronous gastric adenocarcinoma, mucosa-associated lymphoid tissue (MALT) lymphoma, and schwannoma in a 56-year-old man in Korea.

## CASE REPORT

A 56-year-old male visited to our clinic with melena. Apart from the presence of diabetes, he had no other remarkable symptom or past medical history. There were no specific findings in physical examination except for obesity (body mass index, 32). All laboratory results, including tumor markers, were in normal range. Gastroduodenoscopy showed slightly raised ulcer on the antral anterior wall. Additional findings included a positive rolling sign in the mid-body suggestive of a submucosal lesion and a diffuse erythematous lesion in the fundus area, and these areas were also biopsied (Figure 1). A computed tomography scan of the abdomen showed an ovoid homogeneous 2 cm × 2 cm mass at the greater curvature of the mid-body and mildly enlarged perigastric lymph nodes. Positron emission tomography - computed tomography showed no evidence of any other lymphoid involvement. Histological examination of the antral and fundic biopsy samples yielded respective diagnoses of moderately differentiated adenocarcinoma and low-grade MALT lymphoma with plasmacytic differentiation (Figure 2).

The patient was first prescribed medication to eradicate any underlying *Helicobacter pylori* (*H. pylori*) infection, which might have been a causative factor in the MALT lymphoma, and examination of repeat biopsy obtained at the fundus after 4 wk medication confirmed that the MALT lymphoma had resolved. The patient was then referred for surgical intervention. At laparotomy, the patient underwent radical subtotal gastrectomy with gastrojejunostomy. During

gastrectomy, a well-defined nodular lesion measuring 2 cm × 2 cm was palpated in the greater curvature 5 cm proximal to the adenocarcinoma.

Histological examination of the partial stomach showed the presence of an early gastric cancer consisting of a moderately differentiated intestinal-type adenocarcinoma located in the antrum and measuring 3.2 cm × 2.5 cm. The tumor was infiltrating into the deep submucosa of the antrum. There was no lymph node metastasis in 17 retrieved nodes. The final pathologic stage for gastric adenocarcinoma was pT1bN0M0, p-Stage IA (American Joint Committee on Cancer 7<sup>th</sup> edition). Histological examination of hematoxylin and eosin-stained sections of the other submucosal tumor revealed that it consisted of spindle cells covered by a smooth muscle layer, lymphoid cuffs, and fundic-type glands. On immunohistochemical analysis, this submucosal tumor showed strong positivity for S-100 and negative expression for c-Kit. This immunostaining pattern differentiates gastrointestinal schwannoma from GIST (Figure 3).

Electron microscopic examination of ultrathin sections of the submucosal tumor revealed sheets of elongated cells with numerous complex cytoplasmic processes. The cytoplasmic membrane was completely covered with external lamina. The nucleus had irregular margins and a heterochromatic chromatin pattern, and the perikaryal cytoplasm contained several mitochondria, rough endoplasmic reticulum, ribosomes, and many lysosomes. Hence, a final diagnosis of schwannoma was made (Figure 4).

The patient's postoperative period was uneventful and he was discharged in good health. Follow-up visits, including endoscopy every 6 mo, for up to 2 yr after operation were unremarkable.

## DISCUSSION

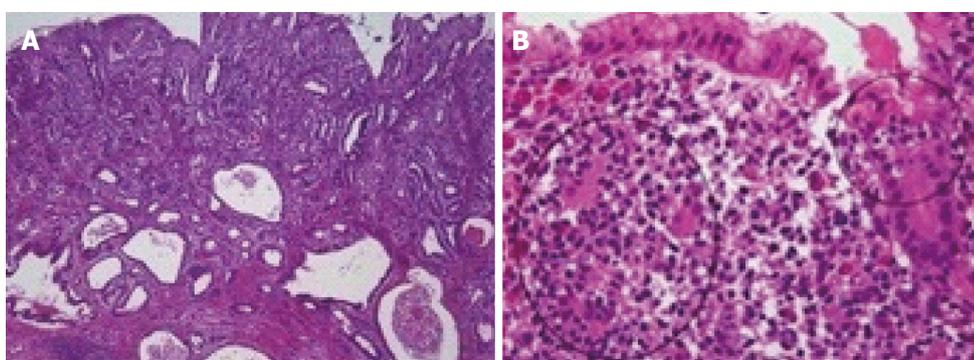
Occurrence of two types of primary gastric neoplasm is relatively well known, but no reports have been published regarding the simultaneous presence of gastric adenocarcinoma, MALT lymphoma, and schwannoma.

Gastric adenocarcinoma accounts for more than 90% of all malignant gastric tumors and may co-exist with synchronous tumors of a different histologic type, most commonly lymphoma.

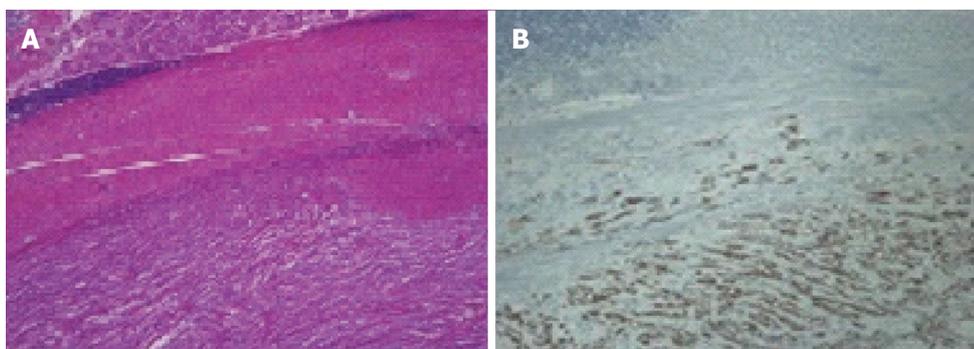
Primary gastric lymphoma occurs in 5% of gastric malignancies and their worldwide incidence is increasing. Primary gastric lymphoma is mostly non-Hodgkin lymphoma. Diffuse large B cell and marginal zone B cell type are the most common subtypes. The pathogenesis is often related to *H. pylori* infection<sup>[1]</sup>. Since Rabinovitch *et al*<sup>[2]</sup> published the first case in 1952, 56 cases of simultaneous occurrence of gastric adenocarcinoma and gastric lymphoma have been reported. In that series, gastric lymphoma was mainly MALT type (69.6%) or low grade (87.2%), the



**Figure 1** Images obtained during endoscopic examination. A: There is a slightly raised ulcerated area on the anterior wall of the antrum; B: A positive rolling sign is present in the mid-body, suggestive of a submucosal lesion; C: A diffuse erythematous lesion is seen in the fundic area. These three lesions underwent biopsy sampling.



**Figure 2** Histologic images. A: The tumor in the antrum is a moderately differentiated adenocarcinoma [ $\times 40$ , hematoxylin and eosin (HE)]; B: In the fundus section, lymphoepithelial lesions (circled in black), typical for mucosa-associated lymphoid tissue (MALT) lymphoma, are seen, which are formed by infiltration of centrocyte-like cells into the gastric glandular epithelium ( $\times 400$ , HE).



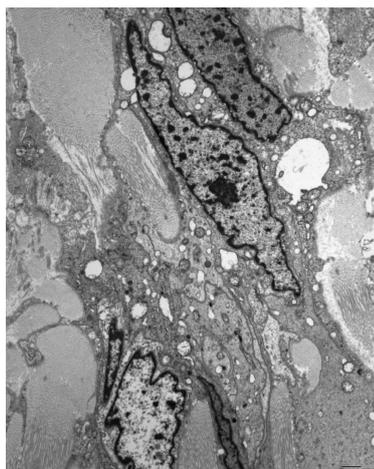
**Figure 3** Histologic images. A: The tumor in the mid-body consists of spindle cells. Above the spindle cell tumor, a smooth muscle layer, lymphoid cuffs, and fundic-type glands are found ( $\times 40$ , HE); B: On immunohistochemical analysis, the spindle tumor cells are positive for S-100, in contrast to the overlying smooth muscle fibers and lymphoid cells ( $\times 100$ ).

correlation between *H. pylori* and MALT lymphoma was 86%/72% in the Eastern and Western cases<sup>[3]</sup>.

Gastric adenocarcinoma and gastric MALT lymphoma are considered to be one of the results of *H. pylori* infection. Gastric cancer can occur in about 1%-2% of *H. pylori* infection cases, and the risk is nine times higher than in patients without *H. pylori* infection. In addition, *H. pylori* infection is highly correlated with gastric MALT lymphoma and is found as a low-grade type MALT lymphoma in over 90% of

cases<sup>[4]</sup>. At present, the most widely accepted initial therapy for localized low-grade MALT lymphoma is aimed at the eradication of *H. pylori* infection, with regimens combining antibiotics and proton-pump inhibitors. Many reports have confirmed the efficacy of antibiotic therapy and showed long-term remission in 60%-100% of patients with localized *H. pylori*-positive MALT lymphoma<sup>[5]</sup>.

In our patient, we decided to treat the MALT lymphoma first by prescribing medication for the



**Figure 4** Electron microscopic findings of the submucosal tumor. Elongated neoplastic cells show with numerous complex interdigitating cytoplasmic processes. The cytoplasmic membrane was completely covered with external lamina. The nuclei reveal irregular margins and a heterochromatic chromatin pattern, and the perikaryal cytoplasm contained several mitochondria, rough endoplasmic reticulum, ribosomes, and many lysosomes.

eradication of *H. pylori* organisms to avoid total gastrectomy. After 4 wk, repeat biopsy was conducted at the fundic site and we confirmed that the MALT lymphoma had resolved. Subsequently, the patient underwent radical subtotal gastrectomy, which included removal of the submucosal tumor in the mid-body. The final pathologic findings were moderately differentiated intestinal-type adenocarcinoma (submucosal invasion without regional lymph node metastasis) and schwannoma, and follow-up visits up to 2 y after operation were unremarkable.

According to Nakamura's study<sup>[6]</sup>, the survival rate of patients with synchronous gastric adenocarcinoma and gastric lymphoma was similar to survival rate of gastric adenocarcinoma alone and was significantly lower than survival rate of gastric lymphoma alone.

Schwannoma is a rare gastrointestinal mesenchymal tumor. The most common site for schwannoma in the gastrointestinal tract is the stomach, followed by the colon. There are some histological differences between gastric schwannoma and soft-tissue schwannoma. Unlike soft tissue schwannoma, encapsulation, nuclear palisading and vascular hyalinization are rare in gastric schwannoma<sup>[7]</sup>. Gastric schwannoma seems to have a good prognosis without recurrence and metastasis. As for simultaneous occurrences of schwannoma with other types of gastric cancer, only three cases have been reported prior to 2015<sup>[8]</sup>.

In conclusion, synchronous triple gastric adenocarcinoma, MALT lymphoma, and schwannoma has not been reported previously in the literature. *H. pylori* eradication and surgery are the mainstay treatment. Further biologic and genetic studies will be required to explain the simultaneous development of tumors of different histotypes.

## COMMENTS

### Case characteristics

A 56-year-old man presented to our hospital with melena and three gastric abnormalities were detected on gastroduodenoscopic examination.

### Clinical diagnosis

Mucosa-associated lymphoid tissue (MALT) lymphoma, adenocarcinoma, and submucosal tumor in stomach.

### Differential diagnosis

Carcinoid tumor, primary lymphoma, gastrointestinal stromal tumor.

### Laboratory diagnosis

All laboratory results were within normal limits.

### Imaging diagnosis

Gastroduodenoscopic examination showed a small ulcerative lesion in the gastric antrum, a submucosal mass in the gastric body, and severe erosion in the fundus. CT showed an ovoid homogeneous 2 cm × 2 cm mass at the greater curvature of the mid-body and mildly enlarged perigastric lymph nodes.

### Pathological diagnosis

MALT lymphoma, adenocarcinoma, schwannoma.

### Treatment

Antibiotics treatment and radical subtotal gastrectomy.

### Related reports

Synchronous occurrence of two types of tumor in the stomach is relatively well known. This is rare case of synchronous triple tumors in stomach.

### Term explanation

MALT lymphoma is a form of lymphoma involving the MALT, frequently of the stomach. Gastric schwannoma is a benign nerve sheath tumor composed of Schwann cells.

### Experiences and lessons

Further biologic and genetic studies will be required to explain the simultaneous development of tumors of different histotypes.

### Peer-review

It is a well written article, a case report about synchronous triple primary gastric adenocarcinoma, MALT lymphoma, and schwannoma.

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