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A Collision Tumor Composed of a Granulocytic Sarcoma and an Adenocarcinoma of the Stomach
– A Case Report –

Kyu Yeoun Won · Juhie Lee
Yong Ho Kim[1] · Youn Wha Kim

Departments of Pathology and [1]Surgery, Kyung Hee Medical Center, Kyung Hee University College of Medicine, Seoul, Korea

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Corresponding Author
Youn Wha Kim, M.D.
Department of Pathology, Kyung Hee University College of Medicine, 1 Hoegi-dong, Dongdaemun-gu, Seoul 130-702, Korea
Tel: +82-2-958-8743
Fax: +82-2-957-0489
E-mail: kimyw@khu.ac.kr

Granulocytic sarcoma, also called chloroma or myeloblastoma, is an extramedullary invasive tumor composed of neoplastic myeloid cells. In this report, we describe a 43-year-old male patient with a collision tumor composed of an adenocarcinoma and a granulocytic sarcoma in the stomach. The coexistence of a granulocytic sarcoma and adenocarcinoma in the stomach has, to the best of our knowledge, not been reported in the literature. The diagnosis of granulocytic sarcoma is very difficult; especially in the absence of concurrent hematologic disease or in the uncommon setting of coexistence with another tumor. Cautious observation is needed when a finding of unusual atypical cells admixed with an adenocarcinoma in the stomach is confronted.

Key Words: Collision; Sarcoma, myeloid; Adenocarcinoma; Stomach

Granulocytic sarcoma, also called chloroma or myeloblastoma, is an extramedullary invasive tumor composed of neoplastic myeloid cells.[1] This tumor may be associated with acute myelogenous leukemia or chronic myeloproliferative disorders including chronic myelogenous leukemia, myelofibrosis with myeloid metaplasia, polycythemia vera, and myelodysplastic syndrome.[2,3] Common sites of involvement include the skin, gums, lymph nodes, soft tissue, periosteum, and bone.[2,4] Granulocytic sarcomas in the gastrointestinal tract are uncommon[4] and represented 6.5% of cases in one large series.[2] A gastric granulocytic sarcoma may occur as a gastric ulcer or polyp, and patients often present with acute gastrointestinal bleeding.[5,6] In this report, we describe a rare case of a collision tumor composed of an adenocarcinoma and a granulocytic sarcoma in the stomach.

CASE REPORT

A 43-year-old male was admitted to our hospital complaining of postprandial abdominal discomfort for two months. The patient had no specific past medical history. Initial blood studies showed a white blood cell (WBC) count of 47,270/mm^3, hematocrit of 34.6%, hemoglobin level of 11.7 g/dL, and platelet count of 217,000/mm^3. A differential WBC count showed 10% neutrophils, 8% lymphocytes, 32% monocytes, 0% eosinophils, 38% atypical lymphocytes, 10% myelocytes, and 2% immature cells. Moreover, a gastroscopy showed the presence of a 4×4 cm sized huge ulceroinfiltrative mass occupying the entire antrum and involving the angle. A computed tomography scan showed annular and diffuse wall thickening at the antrum, that extended to the high body along the lesser curvature. Also, multiple perigastric enlarged lymph nodes were noted. An endoscopic biopsy was performed, which showed the adenocarcinoma with atypical hematopoietic/lymphoid cells aggregation (Fig. 1A). The atypical cells were found to be positive for myeloperoxidase (Fig. 1B).

A laparoscopic subtotal gastrectomy was performed, and on gross examination, a huge ulcerative lesion measuring 6.0×5.5 cm was observed in the antrum. The surrounding mucosa showed diffuse erythematous change and edematous rugae (Fig. 2). The cut surface of the stomach antral lesion showed diffuse irregular thickening of entire wall. Microscopy demonstrated the pres-

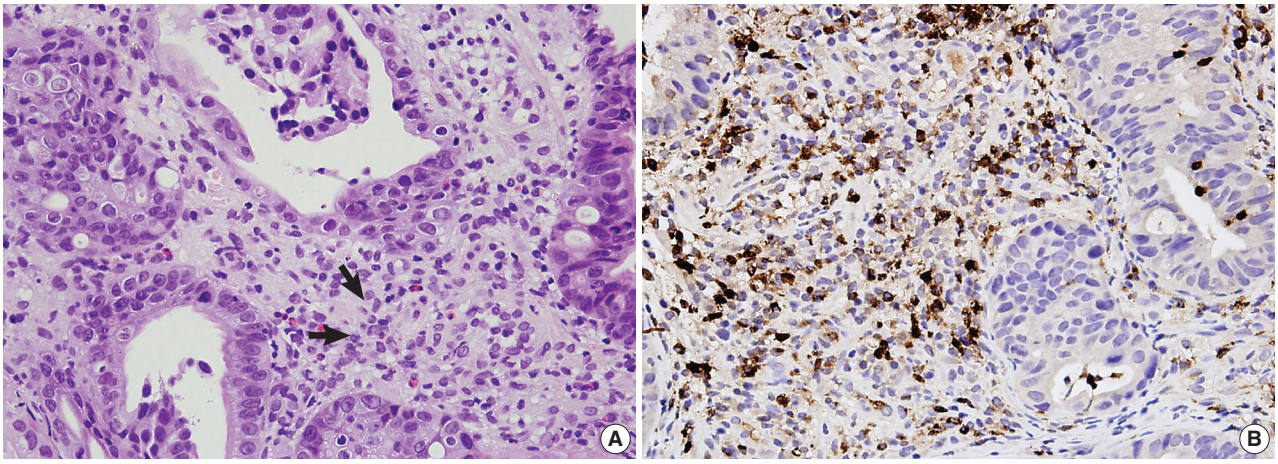

Fig. 1. (A) The biopsied specimen shows a well differentiated adenocarcinoma with atypical hematopoietic/lymphoid cells aggregation (arrows). (B) The atypical hematopoietic/lymphoid cells are positive for myeloperoxidase.

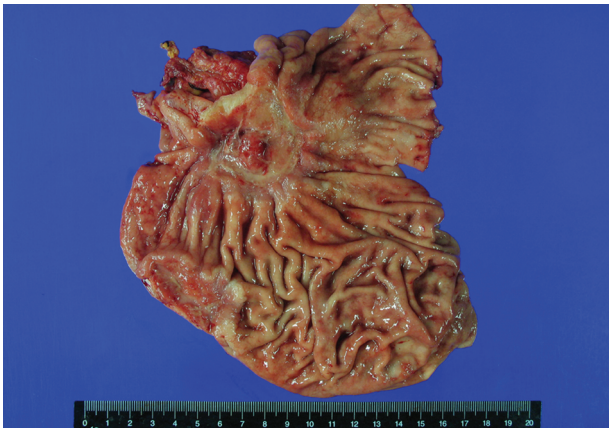

Fig. 2. A huge ulcerative lesion is seen in the antrum of the stomach. The remainder of the stomach shows diffuse erythematous change and edematous rugae.

ence of well to poorly differentiated adenocarcinoma including signet ring cells that extended to the subserosa without serosal penetration. Unexpectedly, diffuse infiltration of medium-sized atypical cells resembling a lymphoid malignancy was noticed throughout the entire stomach wall. The atypical cells were admixed with adenocarcinoma cells (Fig. 3A). The atypical cells were positive for myeloperoxidase (Fig. 3B), leukocyte common antigen, and CD68. In addition, the atypical cells were negative for cytokeratin, CD3, CD20, CD10, MUM1, kappa, lambda, Bcl2, and Bcl6. On the third day post surgery, a bone marrow aspirate demonstrated the presence of acute myelomonocytic leukemia (FAB M4) (Fig. 4). Cytogenetic studies showed no abnormality. The final diagnosis of the stomach was a collision tumor composed of a well to poorly differentiated adenocarcinoma and granulocytic sarcoma.

The perigastric lymph nodes showed the presence of a metastatic adenocarcinoma in three of the 57 lymph nodes and a metastatic granulocytic sarcoma in 54 of 57 lymph nodes. Interestingly, the three lymph nodes showed a mixed metastatic adenocarcinoma and granulocytic sarcoma. Just after surgery, the patient received induction and consolidation chemotherapy (cytarabine and daunorubicin) for treatment of the leukemia. Unfortunately, about four months later, the patient died from septic shock.

DISCUSSION

A gastric collision tumor of the stomach is uncommon,[2] with reported cases including gastric adenocarcinoma admixed with gastric lymphoma,[7] carcinoid,[8] leiomyosarcoma,[9] or rhabdomyosarcoma.[10,11] Most common gastric collision tumors are composed of an adenocarcinoma intermixed with a gastric lymphoma which occurs in 0.08% of all adenocarcinoma cases.[7,11,12] However, a collision tumor composed of a granulocytic sarcoma and an adenocarcinoma in the stomach has, to the best of our knowledge, not been reported in the English literature.

The diagnosis of granulocytic sarcoma is very difficult, especially in the absence of concurrent hematologic disease or in the uncommon setting of coexistence with another tumor.[13] Misdiagnosis of a granulocytic sarcoma may be as high as 75% of cases.[14] The most frequent misdiagnosis is large cell lymphoma.[15]

The gastrointestinal involvement of a granulocytic sarcoma is rare, and still rarer is the isolated gastro-duodenal localization.[1,6,16] Only a few cases of gastrointestinal granulocytic sarcoma have been reported in the literature with less than 20 cases having been described.[2] The prognosis of a patient with a gastroin-

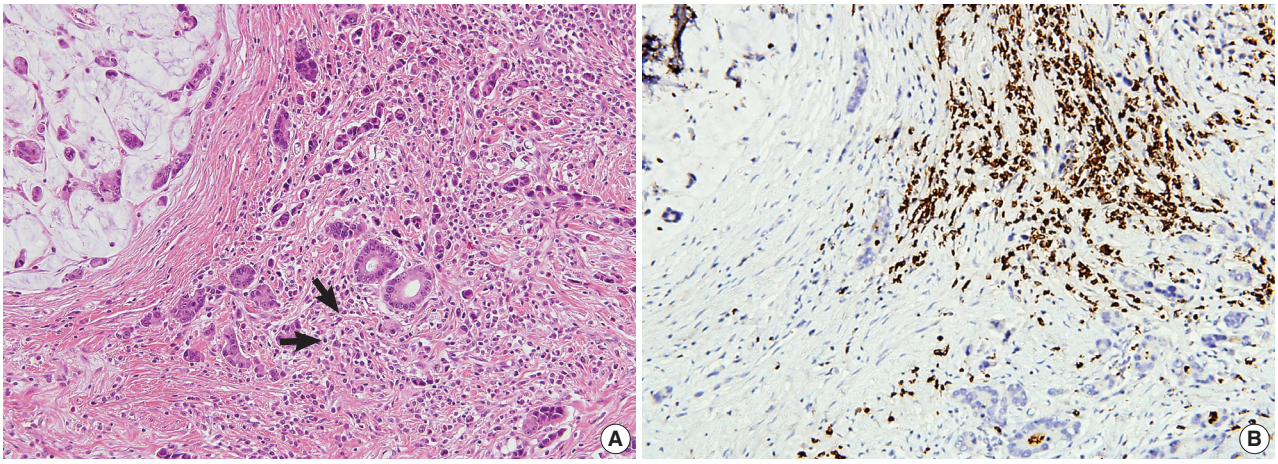

Fig. 3. (A) Granulocytic sarcoma cells (arrows) are admixed with well to poorly differentiated adenocarcinoma cells. (B) The granulocytic sarcoma cells are positive for myeloperoxidase.

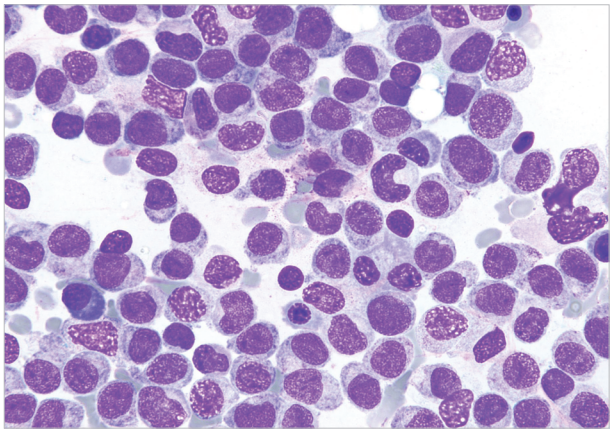

Fig. 4. A bone marrow aspirate demonstrates the presence of acute myelomonocytic leukemia (FAB M4).

testinal granulocytic sarcoma is poor, particularly because of the high rate of early complications such as bleeding and perforation.[17]

We overlooked the granulocytic sarcoma component on the biopsied specimen because of the definite adenocarcinoma component. Lazaris *et al.*[13] reported the coexistence of a granulocytic sarcoma and an adenocarcinoma of the rectum. These investigators observed that a well-differentiated adenocarcinoma extended into the muscle layer. Additionally, a diffuse monotonous infiltrate of medium-sized cells was noted throughout the entire intestinal wall and into the surrounding fat. Adjacent lymph nodes demonstrated granulocytic sarcoma cell involvement of the subcapsular sinuses. In comparison to the case by Lazaris *et al.*,[13] our case was composed of a poorly differentiated adenocarcinoma including signet ring cells admixed with a granulocytic sarcoma. Interestingly, some lymph nodes showed a combination of a metastatic adenocarcinoma and a granulocytic sarcoma.

In general, the growth pattern and cell morphology between a granulocytic sarcoma and adenocarcinoma are very different.[13] Granulocytic sarcoma cells invade tissue with relatively good preservation of the tissue architecture[3,6,18] and can have round to oval, reniform or multilobed nuclei, and occasional prominent nucleoli or mitosis.[13] Taking into account the different degrees of cell maturation in the tumor, a correct diagnosis depends on the use of immunohistochemical staining.[19]

The mechanism of formation in a collision tumor consisting of a granulocytic sarcoma and an adenocarcinoma is questionable. Some investigators have suggested that the mere coincidence or specific genetic aberrations occurring at the gene loci form the basis of the coexistence.[13] Analysis of previously reported granulocytic sarcomas have shown abnormalities involving chromosome 17, in which the *p53* tumor suppressor gene is located.[20] However, our case showed no such cytogenetic abnormality.

In conclusion, the coexistence of a granulocytic sarcoma and an adenocarcinoma is very rare. A correct and prompt diagnosis is very important for appropriate treatment. Therefore, cautious observation is needed when a finding of unusual atypical cells admixed with an adenocarcinoma in the stomach is confronted.

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