CASE REPORT
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Gastric carcinosarcoma with rhabdomyosarcomatous differentiation: a case report and literature review

Hsing-Yu Shih[1], Che-Pin Lin [2], Feng-Chuan Tai[3]*

[1]Department of Surgery, Cathay General Hospital, Taipei, Taiwan.
[2]Division of hematology and oncology, Cathay General Hospital, Taipei, Taiwan.
[3]Division of General Surgery, Cathay General Hospital, Taipei, Taiwan.

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***Correspondence to**
Feng-Chuan Tai, MD
Division of General Surgery
Cathay General Hospital,
280 Renai Rd. Sec.4, Taipei, Taiwan
cghsurgery@gmail.com
Telephone: +886-0931099299
Fax: +886-27540222

ABSTRACT
Gastric carcinosarcoma with rhabdomyosarcomatous differentiation is a rare tumor. Herein, we report the case of a 34-year-old man with a history of dysphagia, upper abdominal fullness, and poor appetite. Endoscopic findings showed a large friable mass that originated from the gastric cardia and lesser curvature of the high body. Consequently, radical total gastrectomy with Roux-en-Y esophagojejunostomy was performed. Histopathological analysis of the resected specimen revealed that the mass had invaded the serosa without regional lymph node metastasis; moreover, the tumor was positive for desmin and myogenin. Finally, we conclude this report with literature review and discussion.

Key Words:
Gastric tumor, gastric carcinosarcoma, rhabdomyosarcomatous.

Background

Carcinosarcoma is an uncommon biphasic malignant tumor composed of carcinoma and sarcoma components. In the upper gastrointestinal tract, carcinosarcoma is more frequently found in the esophagus; however, its gastric localization has been reported rarely.[1-3] The presentation of gastric carcinosarcoma with rhabdomyosarcomatous differentiation is even rarer, with only 12 cases reported thus far.[4-15]

Herein, we report the case of a male patient who developed gastric carcinosarcoma with rhabdomyosarcomatous differentiation and provide the clinical and histopathological features of this tumor. Moreover, we also review and discuss relevant literature.

Case report

A 34-year-old man had been experiencing dysphagia along with upper abdomen fullness and poor appetite for 2 months; he had lost 12 kg of body weight over the previous 6 weeks. Initially, he went visited a local clinic, where panendoscopy was performed; the results revealed a large (size, 4 cm) friable tumor, stretching from the cardia to the lesser curvature side of the high body (*Figure 1*). The tumor arose from the submucosal layer with infiltration to the mucosal layer. A biopsy sample was obtained and sent for pathological examination. The histopathological analysis confirmed the presence of malignancy with rhabdomyosarcomatous differentiation.

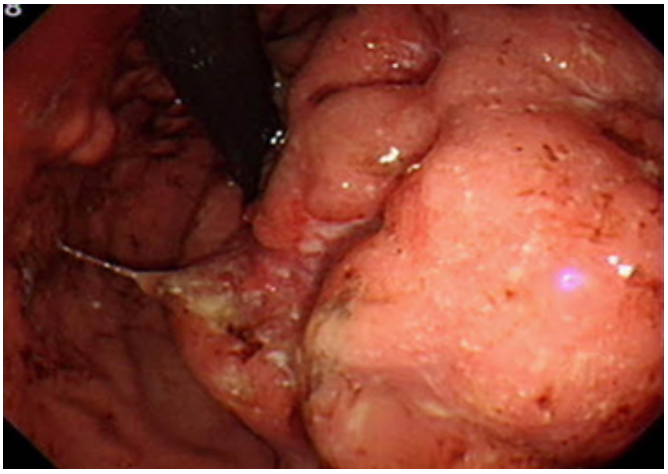

Figure 1: Panendoscopy revealed a tumor stretching from the cardia to the lesser curvature side of the high body.

The patient was referred to Cathay General Hospital for further evaluation. The levels of tumor markers CEA, CA19-9, and SCC were all within the normal range. Abdominal computed tomography (CT) revealed an irregular lobular mass, measuring approximately 7.5 × 6.5 × 8.5 cm3, stretching from the esophagogastric junction (EGJ) to the lesser curvature of the gastric high body, with at least three enlarged regional lymph nodes around the EGJ and lesser curvature of the stomach (*Figure 2A, 2B*). Nevertheless, chest CT revealed absence of pulmonary metastasis.

The patient subsequently received total gastrectomy with Roux-en-Y esophagojejunostomy (*Figure 3*).

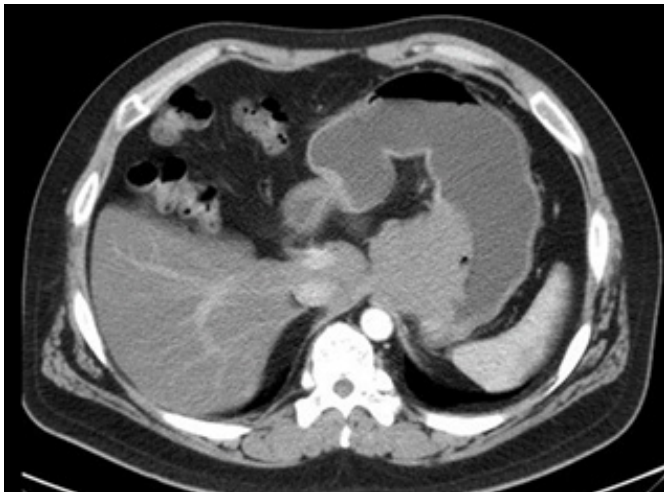

Figure 2A: Irregular lobular mass lesion stretching from the EGJ to the lesser curvature of the gastric high body.

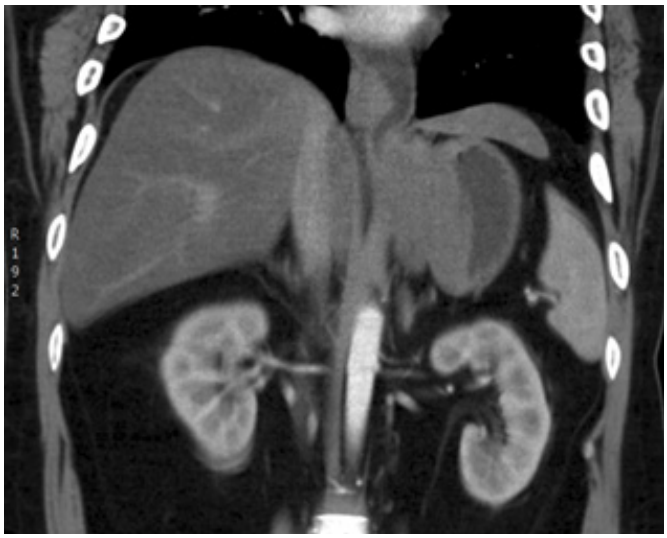

Figure 2B: A 3 cm submucosal mass at distal esophagus, suspect regional lymphadenopathy.

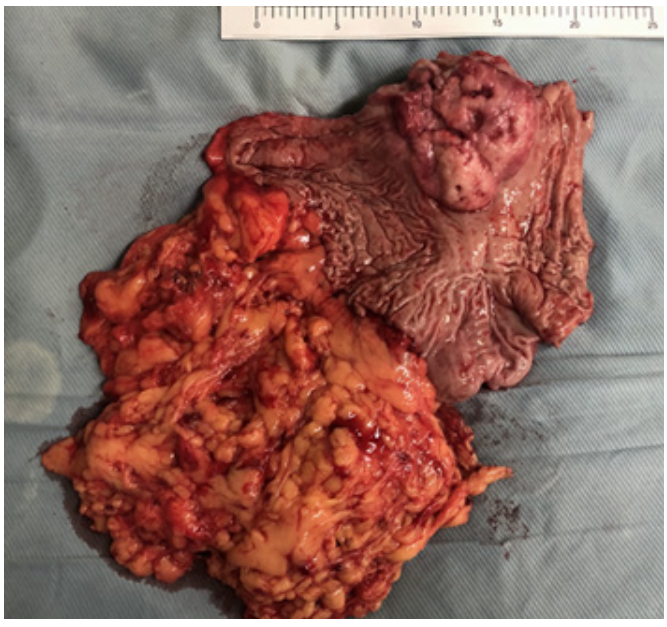

Figure 3: Polypoid tumor with central ulceration at the cardiac region

Macroscopic examination of the excised tissue revealed an 11.2 × 8.9 cm2 tumor mass, invading up until the serosa (*Figure 4*), but no lymph node metastasis was found. Histopathological examination revealed complex glandular formation with marked nuclear atypia and small, loose clusters of atypical cells, with indistinct to little eosinophilic cytoplasm and marked pleomorphic and hyperchromatic nuclei (*Figure 5*). Moreover, immunohistochemical analysis showed that the loose, atypical cells were positive for CK(AE1/3), myogenin, and desmin but negative for S-100, CD117, CD34, and CD45RB. Taken together, these results confirmed the diagnosis of gastric carcinosarcoma with rhabdomyosarcomatous differentiation, at a pathological stage of T4aN0M0 (Stage IIB).

Based on this diagnosis, the patient was administered six cycles of dacarbazine and doxorubicin for chemotherapy. Moreover, because the paraesophageal lymphadenopathy was inaccessible during surgery, the patient was also administered local radiotherapy, as suggested by our radiation oncologist.

Finally, owing to favorable recovery, the patient was discharged on the 14th postoperative day. At the 16-month follow-up, the patient did not show any evidence of recurrence.

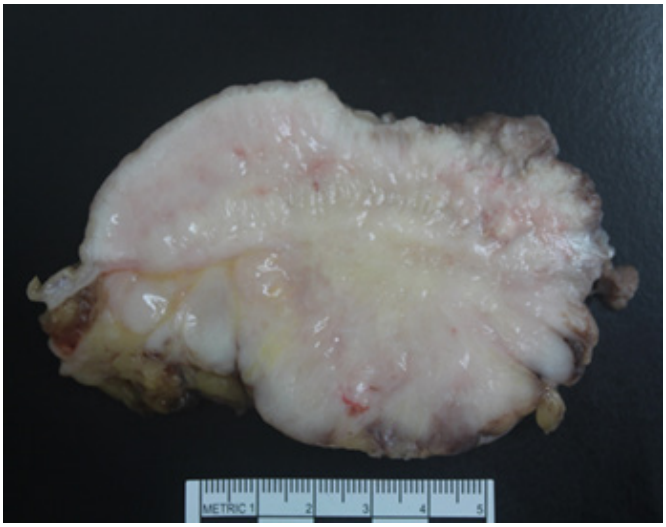

Figure 4: Tan-white and firm tumor invading from the mucosa to the serosa

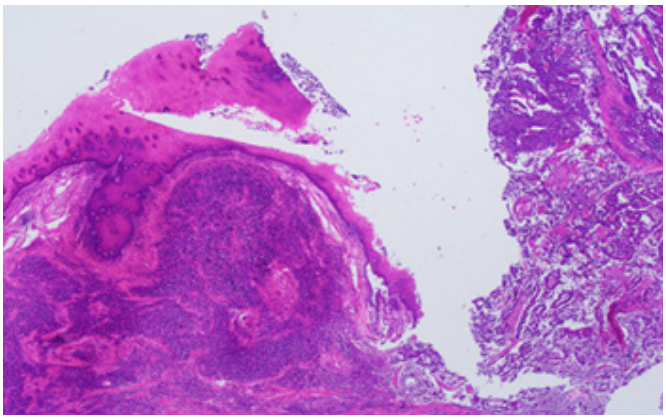

Figure 5A: Glandular structure and solid pattern.

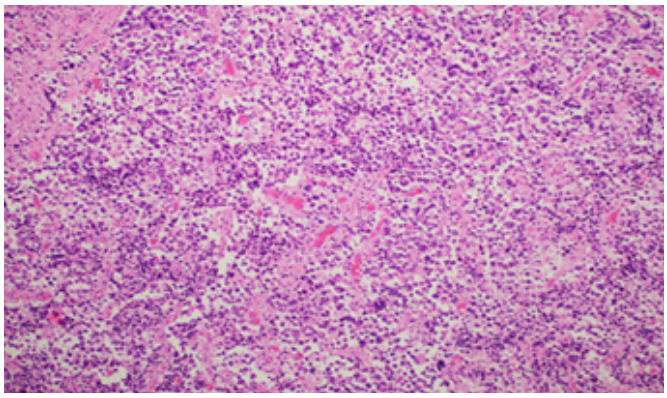

Figure 5B: Single neoplastic cells with marked pleomorphism.

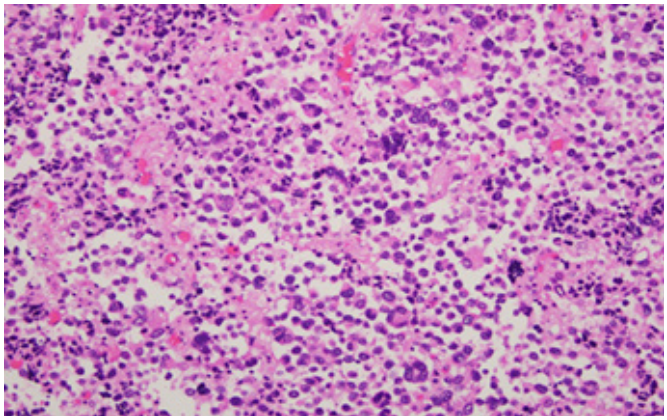

Figure 5C: Single neoplastic cells with marked pleomorphism, eosinophilic cytoplasm, and apoptosis.

Discussion

Based on the conventional histological findings, the World Health Organization defines carcinosarcoma as a malignant tumor composed of intimately mixed epithelial and mesenchymal elements of a type ordinarily found in malignancies of adults.[16] Carcinosarcoma can have a wide variety of localization sites, with the uterus being the most common site. However, as mentioned, localization in the stomach is rare. *Table 1* presents various features of gastric carcinosarcoma with rhabdomyosarcomatous differentiation included in all 12 relevant cases reported thus far.[5-15]

In all 12 cases, no clinical feature was associated with age, sex, or location. However, polypoid lesions were the most commonly reported.[17] The tendency of gastric rhabdomyosarcoma to metastasize to the lymph node and lungs is consistent with the observations of rhabdomyosarcoma originating from other sites.

The current gold standard technique for definitive diagnosis is based on immunohistochemical staining of endoscopic biopsy or on surgical findings. Radical gastrectomy is the treatment of choice when feasible, even when the tumor shows rapid growth and malignant potential. The therapeutic effects of chemotherapy or radiotherapy have not been reported thus far. Moreover, given that this condition is rare and is diagnosed at advanced stage in most cases, no applicable standard chemotherapy regimen is available yet.

Author	Age/sex	Location	Size (cm)	Gross Feature	Depth of invasion	Outcome
Stout (1953) [8]					Submucosa	
Kyogoku (1960) [4]	49/M					Dead, 36 mon
Matsukuma (1977) [9]	74/M	Remnant Stomach	15x11	Polypoid	Serosa	Dead, 5 mon
Machida (1981) [7]	39/F	Cardiac	7x6	Polypoid	Submucosa	Dead, 5 mon
Fox (1990) [10]	68/F	Body	15x10	Polypoid	Mucosa	Dead, 26 mon
Sugai (1991) [11]	78/M	Pylorus	9x7	Polypoid	Serosa	Dead, 5 mon
Melato (1993) [5]	73/M	Remnant Stomach	7x5.5	Polypoid	Serosa	
Nakayama (1997) [6]	69/M	Remnant Stomach	20x18	Polypoid		
Tsuneyama (1999) [12]	63/M	Pylorus	7x6.5	Polypoid	Subserosa	Alive,10 mon
Sato Y(2001) [13]	67/F	Fundus	8x7	Polypoid		Alive, 11 mon
Fujiie (2016) [14]	71/F	Body	2x1.5	Polypoid	Subserosa	Alive, 36 mon
Tokuda (2019) [15]	82/M	Body	8x8	Polypoid	Subserosa	Dead, 3 mon
Our case	34/M	Cardia	11x9	Polypoid	Serosa	Alive, 16 mon

Table 1: Reported cases of gastric carcinosarcoma with rhabdomyosarcomatous differentiation.

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Contributors
HYS, CPL, FCT conceptualized and designed the study, acquired, and analyzed data, interpreted the study results, drafted the manuscript, and critically revised the final version of the manuscript.

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Availability of data and materials
Further information is available from the corresponding author on reasonable request.

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Not applicable.

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