



Case Report

Granular cell tumor coexisting with adenocarcinoma in the stomach: Report of a rare case

Toshikatsu Nitta^{a,*}, Masato Ohta^{a,b}, Jun Kataoka^a, Masatsugu Ishii^a, Yasuhiko Ueda^a,
Sadakatsu Senpuku^a, Atsushi Takeshita^c, Takashi Ishibashi^a

^a Division of Surgery Gastroenterological Center, Medico Shunju Shiroyama Hospital, Osaka, Japan

^b Department of Gastroenterological Surgery Osaka Medical College, Osaka, Japan

^c Department of Pathology Osaka Medical College, Osaka, Japan



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ABSTRACT

Introduction: and importance: Granular cell tumor (GCT) is a benign, mesenchymal tumor that originates from Schwann cells.

Case presentation: A 75-year-old Japanese woman was referred to our hospital due to epigastric discomfort. Upper gastrointestinal endoscopy revealed an ulcerated cancer lesion, approximately 1.2 cm in diameter, at the angle of the stomach in the posterior gastric wall and a yellowish submucosal tumor, 5 mm in diameter, near an anal ulcerated lesion. Based on these findings, the patient was scheduled for laparoscopic distal gastrectomy and lymph node dissection. The pathological diagnosis was a moderately differentiated tubular adenocarcinoma invading mucosal stroma without lymph node metastasis and GCT in the stomach. Tumor cells were positive for S-100 and were consistent with the characteristics of GCT.

Clinical discussion: To the best of our knowledge, this is an extremely rare case with reports of only 6 cases of such a combination, including our case.

Conclusions: We believe that the coexistence of a GCT and gastric cancer in our patient was accidental and that there was no relation between them.

1. Introduction

Granular cell tumor (GCT), also called Abrikosoff tumor, was first described by Abrikosoff in 1926 [1]. GCTs are benign, mesenchymal tumors that originate from Schwann cells and are thought to occur at many sites, the most frequent being the skin and subcutaneous tissue [2, 3]. However, its occurrence in the gastrointestinal tract is rare and accounts for only approximately 4%–6% of all GCTs [3]. The esophagus is the most common location of GCT in the gastrointestinal tract, followed by the colon [4], and GCT in the stomach is very uncommon.

Only 54 cases of GCT in the stomach have been reported in the English literature, according to PubMed (key words: granular cell tumor and stomach). The coexistence of GCT and cancer in the gastrointestinal tract is rare [5,6], especially in the stomach. We report a case of GCT occurring simultaneously with a well-differentiated adenocarcinoma in the stomach. To the best of our knowledge, this is an extremely rare case report, with only 6 reported cases of such a combination. Here, we

describe the coexistence of GCT and cancer in the stomach.

The work has been reported in line with the SCARE creiteira [7].

2. Case presentation

A 75-year-old Japanese woman was referred to our hospital due to the epigastric discomfort. There was no history of altered bowel habits or blood in the stool. On examination, physical findings included a soft, non-tender abdomen. Upper gastrointestinal endoscopy revealed an ulcerated lesion, approximately 1.2 cm in diameter, at the angle of the stomach in the posterior gastric wall (Fig. 1a) and a 5 mm diameter yellowish submucosal tumor near an anal ulcerated lesion (Fig. 1b). Biopsy from the ulcerated lesion revealed moderately differentiated tubular adenocarcinoma. Abdominal computed tomography did not demonstrate early gastric cancer and lymph node or distant metastasis (Fig. 2). The laboratory findings on admission were within normal limits, including tumor markers. Based on these findings, the patient was

Abbreviations: GCT, Granular cell tumor.

* Corresponding author. 2-8-1 Habikino, Habikino-city, Osaka, 583-0872, Japan.

E-mail address: nitta@shiroyama-hsp.or.jp (T. Nitta).

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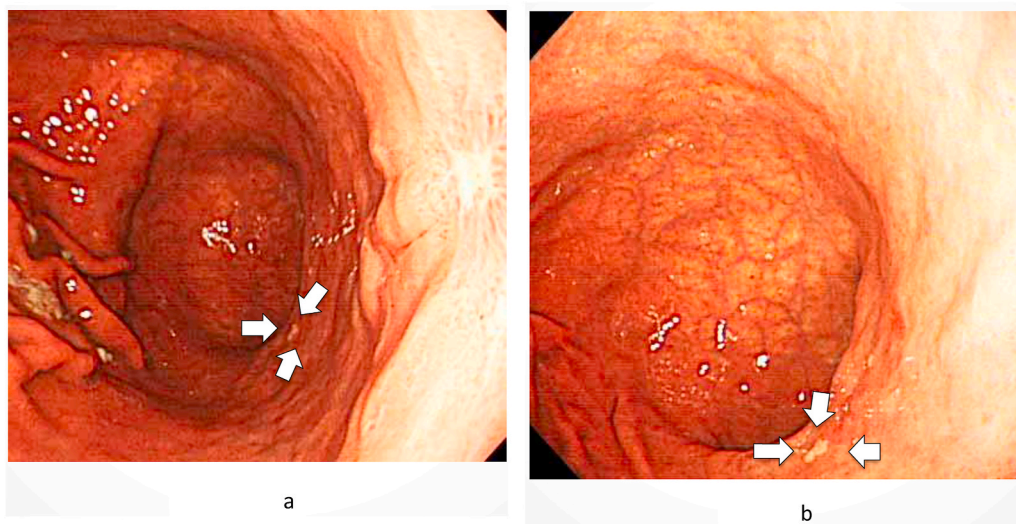


Fig. 1. Upper gastrointestinal endoscopy

1a. Upper gastrointestinal endoscopy revealed an ulcerated lesion, approximately 1.2 cm in diameter, at the angle of the stomach in the posterior gastric wall and a yellowish submucosal tumor (white arrow), 5 mm in diameter, near the anal ulcerated lesion.

1b. Upper gastrointestinal endoscopy revealed a yellowish submucosal tumor (white arrow), 5 mm in diameter, near the anal ulcerated lesion.



Fig. 2. Enhanced computed tomography. Abdominal computed tomography could not demonstrate early gastric cancer and lymph node or distant metastasis.

scheduled for laparoscopic distal gastrectomy and lymph node dissection.

In the resected specimens, gastric cancer lesions were present in the middle of the gastric body, and a yellowish submucosal tumor (Fig. 3a and b) was detected near the cancer lesion, supporting the preoperative diagnosis. The pathological diagnosis of the cancer lesions was moderately differentiated tubular adenocarcinoma invading mucosal stroma without lymph node metastasis and that of the yellowish submucosal tumor was GCT in the stomach. Hematoxylin-eosin staining showed that the submucosal tumor was composed of nests of polygonal cells with small uniform, hyperchromatic nuclei and granular cytoplasm. GCT proliferates mainly in the submucosa (Fig. 4a and b).

We performed immunochemical staining for S-100, c-kit, CD34, p53, and Ki67. Tumor cells were positive for S-100 (Fig. 5a and b) and CD34 in this tumor. These histologic findings were consistent with the characteristics of GCT.

The final diagnosis was benign GCT in the stomach and gastric cancer.

The total operative time was 347 min, and the intraoperative blood loss was 10 mL. The patient demonstrated a good postoperative course and was discharged from our hospital in remission 15 days after the operation because of a grade B pancreatic fistula (grading according to International study group of postoperative pancreatic fistula [8]).

We have observed this case for 2 years so far with no adjuvant chemotherapy, and no recurrence or metastasis has been revealed.

3. Discussion

GCT was originally called granular cell myoblastoma in 1926 [1], since they are generally thought to be of Schwann cell origin and are typically S-100 positive by immunochemical staining.

Immunohistochemical staining for S-100 protein supports the

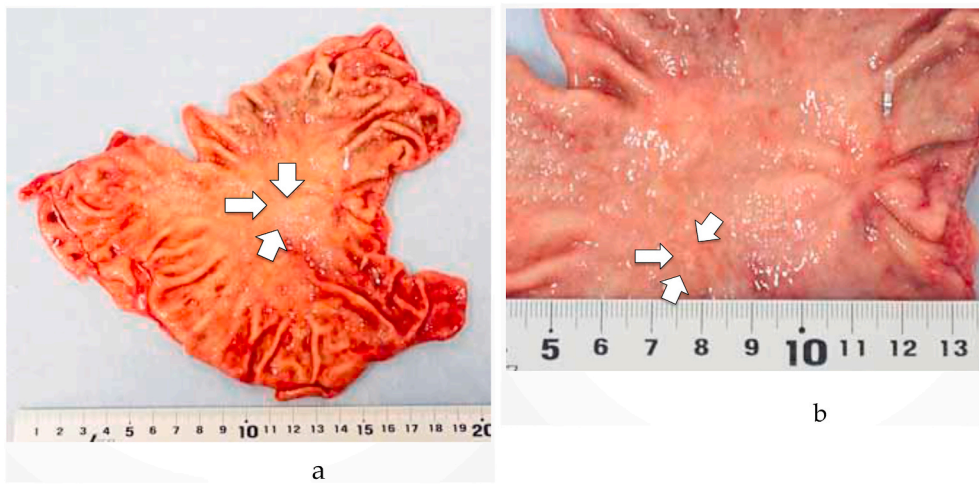


Fig. 3. 3a and 3b. Macroscopy of resected specimen

Gastric cancer lesion was present in the middle of the gastric body and a yellowish submucosal tumor (white arrow) was detected near the cancer lesion.

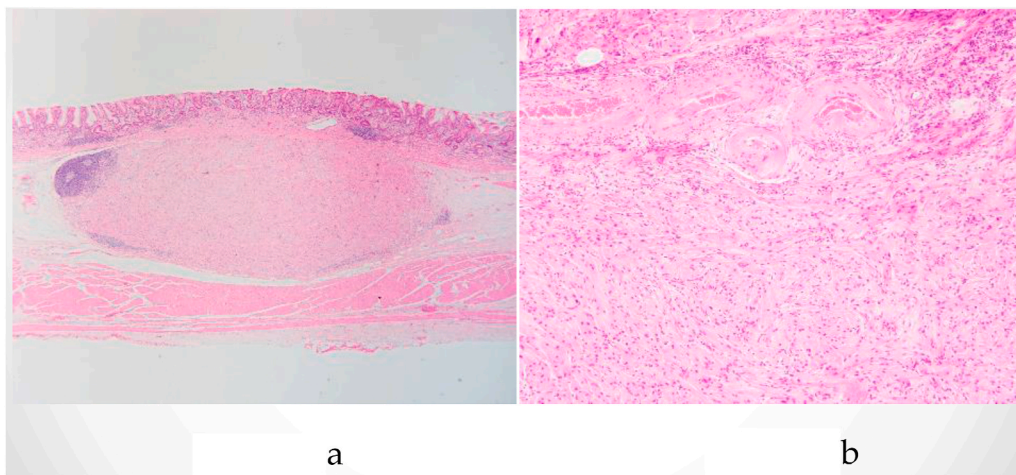


Fig. 4. Pathological findings.

The submucosal tumor was composed of nests of polygonal cells with small uniform, hyperchromatic nuclei and granular cytoplasm.

4a Hematoxylin-eosin staining, $\times 2$

4b Hematoxylin-eosin staining, $\times 20$.

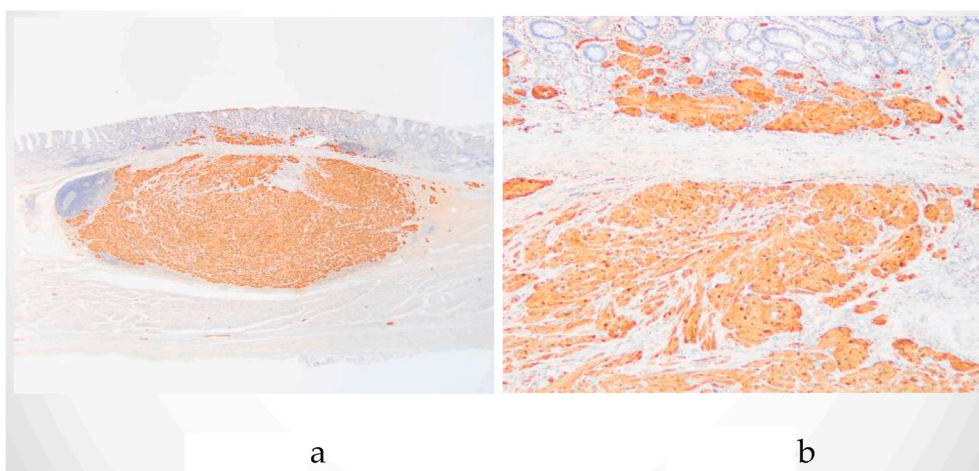


Fig. 5. Immunohistochemical findings

5a Immunohistochemical study of the submucosal tumor Diffusely positive for S100, $\times 2$

5b Immunohistochemical study of the submucosal tumor Diffusely positive for S100, $\times 20$.

proposed origin of the tumor from Schwann cells [9]. Moreover, immunohistochemistry is a useful method to arrive at a correct diagnosis, and it is considered that an immunohistochemical S-100 positivity would support a diagnosis of GCT. In our case, the submucosal tumor was positive for S-100. Recently, it has become easier to diagnose GCT by hematoxylin-eosin staining, because there are many reports of GCT at single or multiple sites [3]. GCT is diagnosed histologically by the following characteristic features: 1. Large polyhedral cells with a markedly granular acidophilic cytoplasm. 2. The cytoplasmic granules and inclusions are PAS-positive and resistant to diastase digestion [10, 11]. The tumor cells are said to be positive for S-100, vimentin, NSE, CD68, and CD57. In our case, S-100, vimentin, and NSE were positive, but we could diagnose GCT in the stomach by hematoxylin-eosin staining, without immunohistochemistry.

However, the epidemiology of GCT is unclear. There is no documented age or gender predilection. GCTs are generally benign tumors with 1–3% malignancy rate [10,12]. GCT occurs at many sites including the gastrointestinal tract [2]. There is no consensus on how to treat GCT. According to Yasuda et al. [13], a minimum surgical margin seems to be adequate if the tumor is completely removed regardless of the type of surgery, such as enucleation, wide resection, or gastrectomy. However, careful follow-up should be maintained even if complete resection is performed, because GCT in the stomach has the possibility of malignancy.

4. Conclusions

As we described in the introduction, GCT in the stomach is rare. Besides, the coexistence of granular cell tumor and cancer in the stomach, as in our case, is extremely rare. To the best of our knowledge, this is an extremely rare case with only 6 reported cases of such a combination, including our case [5,6,9,14,15]. We believe that the coexistence of the GCT and gastric cancer in our patient was accidental and that there was no relation between them [16]. In our case, the distance between the GCT and cancer in the stomach was not much; hence, the GCT and cancer in the stomach were resected simultaneously. We should be careful to resect any lesion found in association with a cancer, since those lesions could be malignant.

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Ethical approval

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Consent

A written informed consent was obtained from the patient for publication of this case report and accompanying images.

Author contribution

Drafting of manuscript, proofreading. All authors read and approved

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Provenance and peer review

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Dr.Narita.

Declaration of competing interest

All authors declared no conflict of interest.

Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.amsu.2021.102271>.

References

- [1] A.I. Abrikossoff, U. Myome, Ausgehend von der quergestreifter willknerlicher Muskulatur, *Virchows Arch Pathol Anat* 260 (1926) 214–233.
- [2] F. Radaelli, G. Minoli, Granular cell tumor of gastrointestinal tract: questions and answers, *Gastroenterol Hepatol N Y* 5 (2009) 798–800.
- [3] S. An, J. Jang, K. Min, M.S. Kim, H. Park, Y.S. Park, et al., Granular cell tumor of the gastrointestinal tract: histologic and immunohistochemical analysis of 98 cases, *Hum. Pathol.* 46 (6) (2015) 813–819.
- [4] A. Nakachi, H. Miyazato, T. Oshiro, H. Shimoji, M. Shiraishi, Y. Muto, Granular cell tumor of the rectum: a case report and review of the literature, *J. Gastroenterol.* 35 (8) (2000) 631–634.
- [5] K. Yamaguchi, S. Maeda, K. Kitamura, Granular cell tumor of the stomach coincident with two early gastric carcinomas, *Am. J. Gastroenterol.* 84 (6) (1989) 656–659.
- [6] Z.X. Xuan, T. Yao, T. Ueyama, M. Tsuneyoshi, CoIncident occurrence of granular cell tumor of the stomach with an early gastric carcinoma, *Fukuoka Acta Med.* 83 (1) (1992) 21–26.
- [7] R.A. Agha, T. Franchi, C. Sohrabi, et al. for the Scare Group, The SCARE 2020 guideline: updating consensus surgical case report (SCARE) guidelines, *Int. J. Surg.* 84 (2020) 226–230.
- [8] C. Bassi, C. Dervenis, G. Butturini, A. Fingerhut, C. Yeo, J. Izbicki, et al., Postoperative pancreatic fistula: an international study group (ISGPF) definition, *Surgery* 138 (1) (2005) 8–13.
- [9] J.L. Sailors, S.W. French, The unique simultaneous occurrence of granular cell tumor, gastrointestinal stromal tumor, and gastric adenocarcinoma, *Arch. Pathol. Lab Med.* 129 (5) (2005) e121–e123.
- [10] H. Matsumoto, Y. Kojima, T. Inoue, S. Takegawa, H. Tsuda, A. Kobayashi, et al., A malignant granular cell tumor of the stomach: report of a case, *Surg. Today* 26 (2) (1996) 119–122.
- [11] M.T. Mazur, J.J. Shultz, J.L. Myers, Granular cell tumor: immuno-histochemical analysis of 21 benign tumors and one malignant tumor, *Arch. Pathol. Lab Med.* 114 (7) (1990) 692–696.
- [12] Franco R, Giorgio M. Granular cell tumors of the gastrointestinal tract: questions and answers. *Gastroenterol Hepatol N Y*;5(11):234–240.
- [13] A. Yasuda, T. Yasuda, H. Imamoto, et al., A case of gastric granular cell tumor preoperatively diagnosed and successfully treated by single-incision laparoscopic surgery, *Surg Case Rep* 6 (1) (2020) 44.
- [14] S. Eguchi, S. Matsuo, M. Hidaka, T. Azuma, S. Yamaguchi, T. Hayashi, et al., Concomitant triple lesions of adenocarcinoma, malignant lymphoma, and granular cell tumor of the stomach, *J. Clin. Gastroenterol.* 35 (1) (2002 Jul) 107–109.
- [15] Y. Watanabe, M. Watanabe, N. Suehara, N. Ishikawa, T. Shinkawa, T. Hosokawa, et al., Early gastric cancer with diffuse heterotopic gastric glands and granular cell tumors mimicking advanced gastric cancer, *Int J Surg Case Rep* 46 (2018) 41–46.
- [16] T. Nitta, Y. Egashira, Y. Kurisu, et al., Granular cell tumor coexisting with adenocarcinoma in the rectum, *Bull. Osaka Med. Coll.* 45 (2) (1999) 43–47.