Scientific Research Publishing

Case Reports in Clinical Medicine, 2023, 12, 306-311
https://www.scirp.org/journal/crcm
ISSN Online: 2325-7083
ISSN Print: 2325-7075

Primary Gastric Synovial Sarcoma Diagnosed by Endoscopic Surveillance of a Gastric Ulcer

Spencer Goble[1], Alan Ayoub[2], Bradley Linzie[1], James Fink[1], Ahmad Malli[1]

[1]Hennepin Healthcare, Minneapolis, USA
[2]Medical School, University of Zagreb, Zagreb, Croatia
Email: Spencer.Goble@hcmed.org

How to cite this paper: Goble, S., Ayoub, A., Linzie, B., Fink, J. and Malli, A. (2023) Primary Gastric Synovial Sarcoma Diagnosed by Endoscopic Surveillance of a Gastric Ulcer. *Case Reports in Clinical Medicine*, **12**, 306-311.
https://doi.org/10.4236/crcm.2023.128043

Received: July 18, 2023
Accepted: August 18, 2023
Published: August 21, 2023

Abstract

Primary gastric synovial sarcoma is rare and challenging to diagnose. We present a case of a gastric ulcer that was diagnosed as primary gastric synovial sarcoma only after surveillance endoscopy with repeat biopsies. The diagnosis was established with the identification of the pathognomonic chromosomal translocation t(X;18)(p11;q11). The patient was treated with wedge resection and has remained disease-free on surveillance imaging and endoscopy. This case demonstrates the difficulty in diagnosing primary gastric synovial sarcoma and the benefits of early disease detection.

Keywords

Primary Gastric Synovial Sarcoma, Gastric Ulcer, Surveillance Endoscopy

1. Introduction

Synovial sarcoma is a mesenchymal neoplasm that most frequently occurs near articular structures (80%) but has also been described in the heart, lungs, kidneys, prostate and gastrointestinal tract [1] [2]. Synovial sarcoma is rare, representing only 5% - 10% of soft tissue sarcomas, and primary gastric synovial sarcoma is exceedingly rare with one recent review identifying only 40 cases [1] [2]. It is challenging to diagnose as immunohistochemistry and molecular analysis is critical to establishing the diagnosis [3]. A chromosomal translocation involving the SYT gene, t(X;18)(p11;q11), is considered pathognomonic for synovial sarcoma, with both a high sensitivity and specificity, but testing for the translocation is not routinely performed on gastric biopsies and an index of suspicion for the diagnosis is important for guiding the appropriate workup [2]. Gastric synovial sarcomas can present as gastric ulcers without overt malignant features on the endoscopic exam and the diagnosis can be missed if repeat endoscopy is not performed to assess for

ulcer resolution with a repeat biopsies and further pathologic assessment of persistent ulcers [1] [4]. We present a case of a gastric ulcer that was diagnosed as primary gastric synovial sarcoma only after surveillance endoscopy with repeat biopsies.

2. Case Report

A 51-year-old man with chronic epigastric pain was referred for esophagogastroduodenoscopy (EGD) due to the worsening of his pain and weight loss. EGD revealed a 10 millimeter (mm) non-bleeding, cratered gastric ulcer in the cardia towards the lesser curvature with a clean base. Biopsies were negative for *Helicobacter pylori* (*H. pylori*) and malignancy and showed stromal fibrosis and chronic inflammation. He was prescribed pantoprazole twice daily and counseled to avoid non-steroidal anti-inflammatory drugs (NSAIDs). On repeat EGD after six weeks, the ulcer was unchanged. Biopsies were again negative for *H. pylori* and malignancy. Acute inflammation and underlying fibrous scar were present with spindle cell tissue that was negative for CD117, CD34, pankeratin, desmin, ALK1 and S100 on immunohistochemical staining.

Repeat EGD after eight weeks again showed an unchanged ulcer. Biopsies showed a dense spindle-cell process with admixed epithelioid and some fasciculations that appeared too organized and fascicular to be a scar and more likely to be malignant (**Figure 1**). Gastrointestinal stromal tumor (GIST) stains were again negative. An SYT gene X;18 translocation was present, consistent with synovial sarcoma (**Figure 2**). Localized wall thickening in the area of the ulcer on the lesser curvature of the stomach was appreciated on subsequent endoscopic ultrasound (EUS) (**Figure 3**). Inflammation from the ulcer prevented the determination of the precise wall layer involved, but the serosa was intact and not involved. Contrast-enhanced computed tomography (CT) was negative for evidence of metastatic disease (**Figure 4**).

Five months after his initial endoscopy, the patient underwent a 4 × 3 × 1 cm wedge resection with gastroscopy with curative intent. He tolerated the procedure well without complication. The ulceration was 0.8 × 0.5 × 0.3 cm in area and involved the mucosa, muscularis mucosa and submucosa with no invasion of the muscularis propria. All lateral resection margins were negative by at least 10 mm and the radial margin was negative by 2 mm. The patient followed up with two different oncologists who both recommended against adjuvant chemotherapy as there was not felt to be an indication. Instead, oncology recommended periodic surveillance with upper endoscopy and CT scans. A genetics referral was also offered, but the patient declined this. Contrast-enhanced CTs were performed at three, six, 12 and 18 months post-operatively and repeat endoscopy was done at three, six and 18 months post-operatively. No recurrence or signs of metastasis has been found on surveillance imaging (**Figure 5**) or endoscopy and the patient is doing well 24 months after the resection. He is continuing surveillance with a CT scan every six months with plans to transition to annual scans if his next CT is without disease evidence.

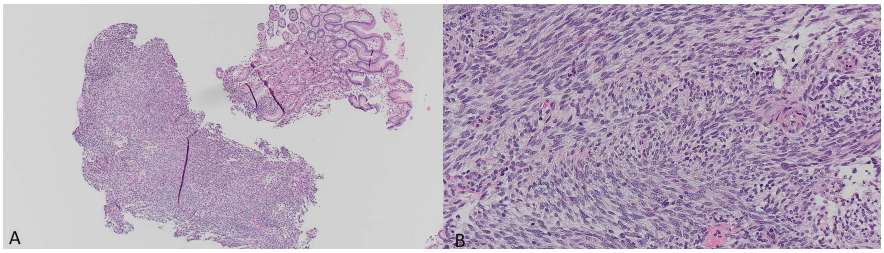

Figure 1. Pathological findings of the tumor. (A) Normal gastric mucosa (right) in comparison to the tumor with atypical spindle cells (left) (hematoxylin and eosin stain, 40× magnification). (B) Proliferation of spindle cells forming the tumor (hematoxylin and eosin stain, 200× magnification).

Figure 2. Gastric cells hybridized with fluorescent probes to the SYT locus on 18q11.2 on two separate slides. Arrows indicate cells with the pathognomonic translocation (fluorescence in situ hybridization).

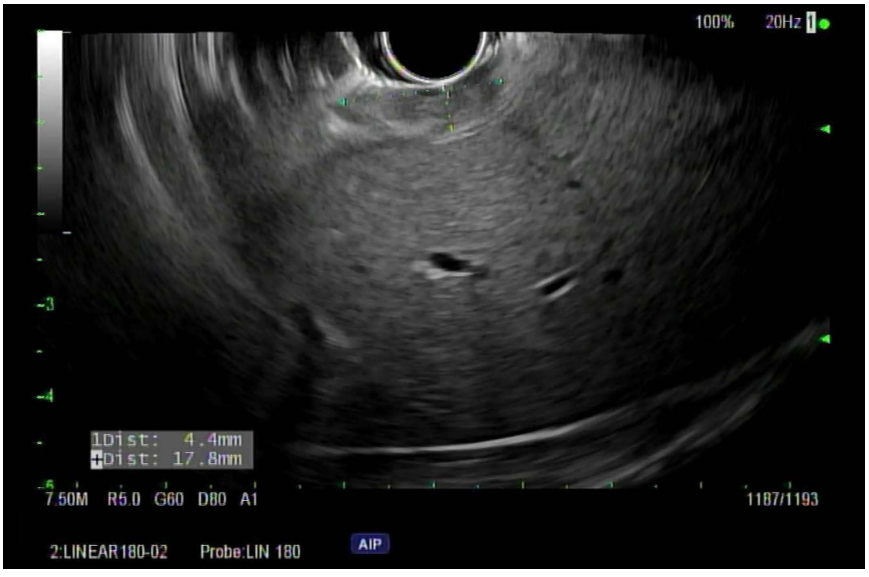

Figure 3. Endoscopic ultrasound demonstrating localized wall thickening in the lesser curvature of the stomach.

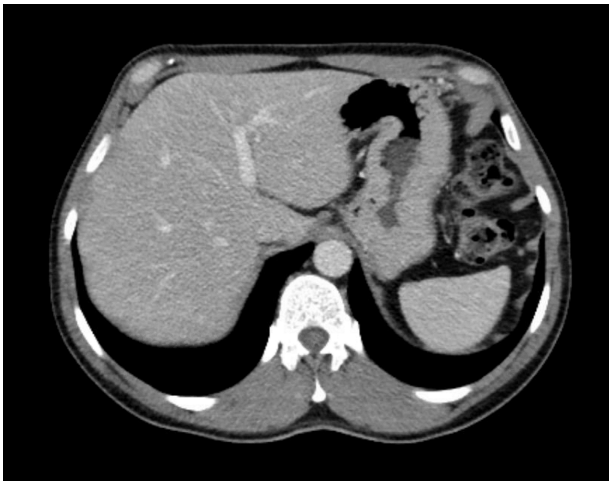

Figure 4. Contrast-enhanced computed tomography with a focal outpouching in the lesser curvature of the stomach consistent with a gastric ulcer without findings concerning for metastatic disease.

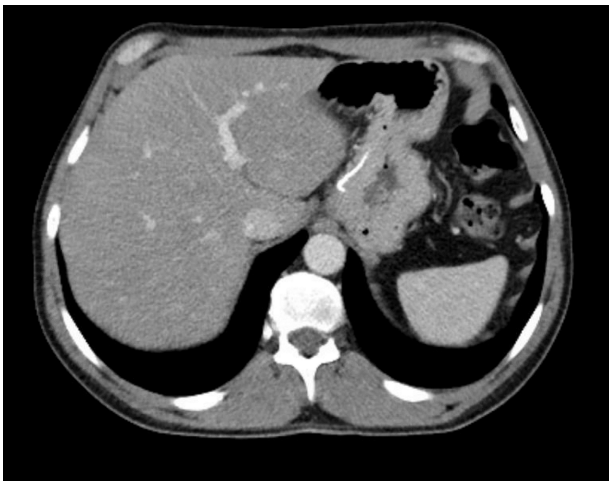

Figure 5. 18-month follow-up contrast-enhanced computed tomography demonstrating post-wedge resection changes without evidence of ulcer recurrence or metastatic disease.

3. Discussion

Clinical suspicion for malignancy, in this case, resulted in surveillance endoscopy with repeat biopsies. Surveillance endoscopy for gastric ulcers with initial pathology negative for malignancy yields a subsequent diagnosis of malignancy in only about 1% of individuals, however, clinical context must be taken into account when considering if surveillance should be pursued [4] [5]. The patient in this case had no clear non-malignant etiology for his ulcer. *H. pylori* testing was negative and despite counseling to avoid NSAIDs, the patient's ulcer persisted on repeat endoscopy. Malignant risk factors were present on endoscopy including location of the ulcer (cardia), elevated borders and lack of resolution with pantoprazole [6] [7]. The presence of malignant risk factors without clear non-malignant etiology warranted continued suspicion of malignancy in this case. Surveillance endoscopy with repeat biopsies and the suspicion of possible syn-

ovial sarcoma by the pathologist ultimately led to the diagnosis and allowed for timely treatment.

The endoscopic and microscopic appearances of gastric synovial sarcoma are similar to that of GISTs [8]. The diagnosis was established in this case by the presence of the pathognomonic SYT gene X;18 translocations. Testing for the presence of the translocation was done based on the pathologist's suspicion after immunohistochemistry testing was inconsistent with GIST on repeat biopsies. While rare, the diagnosis of gastric synovial sarcoma should be considered in scenarios similar to this where immunohistochemistry is negative for GIST despite a persistent lesion with a microscopic appearance similar to GIST.

Tumor resection, wedge resection, partial gastrectomy and total gastrectomy have all been described as surgical treatments for gastric synovial sarcoma and more outcome data is needed to determine the optimal technique for individual presentations. Similarly, further data is required to help guide the need for adjuvant chemotherapy. A review of 40 cases by Marchand *et al.* found that adjuvant chemotherapy was used in 7/29 (24%) cases in which the treatment was specified [1]. Of those individuals, two died as a result of the disease, four remained alive with the disease present, and one was alive and disease-free [1]. In this case, the lack of metastatic disease, the absence of invasion of the muscularis propria and the small size of the lesion argued against the need for adjuvant chemotherapy. Indeed, at 8 mm this is one the smallest in diameter gastric synovial sarcomas to be reported with one previously described 8 mm lesion and one 6 mm in diameter [9] [10]. Including this case, eight cases of lesions 10 mm or less have been described and all have been treated with a wedge or partial resection without adjuvant chemotherapy and no cases of recurrence have been documented with the smallest resected lesion known to have recurred being 20 mm [9]. Based on the available data, we agree that deferring adjuvant therapy is reasonable in cases of lesions 10 mm or smaller.

In conclusion, we presented a case of gastric synovial sarcoma diagnosed through surveillance endoscopy of a gastric ulcer. Clinical suspicion for malignancy, in this case, resulted in surveillance endoscopy with repeat biopsies which eventually led to the diagnosis. Prompt and accurate diagnosis of gastric synovial sarcoma is important as limited available data suggests good outcomes in those who are diagnosed when tumors are still <20 mm, as was the case here [9]. Further accumulation of documented cases of gastric synovial sarcoma is needed to better characterize presentation, appropriate treatment, and outcomes for this disease.

Author Contributions

S. Goble conducted a chart review and literature review and drafted the initial manuscript. A. Ayoub provided content editing for the final manuscript. B. Linzie provided **Figure 1** and the interpretation of the figure. J. Fink provided the FISH **Figure 2** along with the interpretation of the figures. A. Malli provided care for the patient, reviewed the manuscript, supervised the project and is the article guarantor.

Consent for Publication

Informed consent was obtained from the patient described in this case.

Conflicts of Interest

The contributing authors have no conflicts of interest to declare.

References

[1] Crety, C.M., Bellefqih, S., Amroun, K., Garbar, C. and Felici, F. (2021) Primary Gastric Synovial Sarcoma: A Case Report and Literature Review. *International Journal of Surgery Case Reports*, **78**, 270-273. https://doi.org/10.1016/j.ijscr.2020.12.055

[2] Gazendam, A.M., Popovic, S., Munir, S., Parasu, N., Wilson, D. and Ghert, M. (2021) Synovial Sarcoma: A Clinical Review. *Current Oncology*, **28**, 1909-1920. https://doi.org/10.3390/curroncol28030177

[3] So, I.T., Cho, K.B., Lee, J.Y., Kim, S.J., Jung, H.I., Choi, J.H., *et al.* (2017) A Primary Gastric Synovial Sarcoma: A Case Report And Literature Review. *Medicine* (*Baltimore*), **96**, e8904. https://doi.org/10.1097/MD.0000000000008904

[4] Selinger, C.P., Cochrane, R., Thanaraj, S., Sainsbury, A., Subramanian, V. and Everett, S. (2016) Gastric Ulcers: Malignancy Yield and Risk Stratification for Follow-Up Endoscopy. *Endoscopy International Open*, **4**, E709-E714. https://doi.org/10.1055/s-0042-106959

[5] Yang, L.S., Hartley, I., Thompson, A.J., Desmond, P., Taylor, A.C.F., Moss, A., *et al.* (2022) Evaluation of Endoscopic Practices and Outcomes in Follow-Up of Gastric Ulcers. *Journal of Clinical Gastroenterology*, **56**, 412-418. https://doi.org/10.1097/MCG.0000000000001595

[6] Kocak, E., Kilic, F., Akbal, E., Tas, A., Koklu, S., Filik, L., *et al.* (2013) The Usefulness of Ulcer Size and Location in the Differential Diagnosis of Benign and Malignant Gastric Ulcer. *Wiener klinische Wochenschrift*, **125**, 21-25. https://doi.org/10.1007/s00508-012-0309-8

[7] Gielisse, E.A.R. and Kuyvenhoven, J.P. (2015) Follow-Up Endoscopy for Benign- Appearing Gastric Ulcers Has no Additive Value in Detecting Malignancy: It Is Time to Individualise Surveillance Endoscopy. *Gastric Cancer*, **18**, 803-809. https://doi.org/10.1007/s10120-014-0433-4

[8] Rivelli, M., Fernandes, E., Conti, C., Bernardoni, L., Pecori, S., Cingarlini, S., *et al.* (2021) Laparoscopic Intragastric Resection of Gastric Synovial Sarcoma: Report of the First Ever Case with Video Demonstration. *World Journal of Surgical Oncology*, **19**, Article 65. https://doi.org/10.1186/s12957-021-02172-y

[9] Yoshiyasu, K., Kono, H., Hojo, Y., Ishida, Y., Tamura, A., Nakai, K., *et al.* (2021) A Minute Primary Gastric Synovial Sarcoma with Ulcer: A Case Report. *Diagnostic Pathology*, **16**, Article 115. https://doi.org/10.1186/s13000-021-01175-3

[10] Makhlouf, H.R., Ahrens, W., Agarwal, B., Dow, N., Marshalleck, J.J., Lee, E.L., *et al.* (2008) Synovial Sarcoma of the Stomach: A Clinicopathologic, Immunohistochemical, and Molecular Genetic Study of 10 Cases. *The American Journal of Surgical Pathology*, **32**, 275-281. https://doi.org/10.1097/PAS.0b013e31812e6a58