REPORTE DE CASO

Glomangioma of the gastric antrum: case report and literature review

Glomangioma del antro gástrico: reporte de caso y revisión de la literatura

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Recibido: 07/02/2024 Arbitrado por pares Aprobado: 19/06/2024

Citar como: Yan-Quiroz EF, Vásquez-Jaico E, Agreda-Castro FM, Tenazoa Villalobos JR, Pérez-Moran ELS, Fernández-Rodríguez LJ. Glomangioma of the gastric antrum: case report and literature review. Acta Med Peru. 2024;41(3):219-24. doi: 10.35663/amp.2024.413.2815.

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ABSTRACT

Gastric glomangiomas are rare vascular tumors originating from glomic cells. We report the case of a 52-year-old woman who presented with early satiety and discomfort in the upper abdomen. Initial biopsy and imaging studies suggested a gastrointestinal stromal tumor (GIST). The tumor was surgically removed through antrectomy and lymphadenectomy, ensuring clear surgical margins. However, immunohistochemistry and histopathological examination of the removed tissue revealed that the initial diagnosis was incorrect; the patient was found to have a gastric glomangioma. This case highlights the challenges in accurately diagnosing gastric glomus tumors before surgery and reviews the prognosis and available treatment options.

Keywords: Glomus tumor; Neoplasms, vascular tissue; Surgery (Source: MeSH-NLM).

RESUMEN

Los glomangiomas gástricos son tumores vasculares raros que se originan en las células glómicas. Presentamos el caso de una mujer de 52 años que acudió con saciedad precoz y molestias en la parte superior del abdomen. La biopsia inicial y los estudios de imagen sugirieron la presencia de un tumor estromal gastrointestinal (GIST). El tumor fue extirpado quirúrgicamente mediante antrectomía y linfadenectomía, asegurando márgenes quirúrgicos libres. Sin embargo, la inmunohistoquímica y el examen histopatológico del tejido extirpado revelaron que el diagnóstico inicial era incorrecto; se determinó que la paciente tenía un glomangioma gástrico. Este caso destaca los desafíos en el diagnóstico preciso de los glomangiomas gástricos.

Palabras clave: Tumor glómico; Neoplasias de tejido vascular; Cirugía (Fuente: DeCS-BIREME).

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INTRODUCTION

Glomus tumors are benign mesenchymal neoplasms that originate in modified smooth muscle cells of the glomus body. They are composed of varying proportions of glomic cells, blood vessels, and smooth muscle. Therefore, depending on the predominant component, they can be classified as a solid glomus tumor (few vascular structures and few muscle cells), glomangioma (with a prominent vascular component), or glomangiomyoma (with predominant vascular and smooth muscle cells). The most common variant is solid tumor (75%), followed by glomangioma (20%) and glomangiomyoma (5%). [1] Diagnosis of glomus tumors is difficult without a careful histopathological analysis of biopsies, as glomus tumors resemble other neoplasms such as hemangiomas, adnexal tumors, myopericytoma and myofibromatosis. [1] Glomus tumors usually occur in areas where there is a high concentration of glomus bodies, such as subungual regions and the deep dermis of the distal extremities. In addition to the deep dermis, glomus tumors have been described in almost every other part of the body, rarely including the stomach.[1]

Vascular tumors account for less than 2% of all benign tumors of the gastrointestinal tract. Gastric glomus tumors account for approximately 1% of these. Gastrointestinal glomus tumors occur almost always in the stomach, arising in the intramuscular layer. These tumors are similar to peripheral glomus tumors but are different from other stomach neoplasms, such as epithelioid gastrointestinal stromal tumors (GIST) and adenocarcinoma, which are more common. [2-4] The frequency of gastric glomus tumors is estimated to be 100 times lower than that of GIST. [5] Treatment of glomus tumors usually involves excision and they have a good postoperative prognosis; they rarely become malignant. [3] Nonetheless, gastric glomus tumors are usually symptomatic when they become large enough and are likely to cause epigastric pain, nausea, vomiting, hematemesis, and/or melaena. [6]

Correct diagnosis of gastric glomus tumors is challenging because these tumors are like more common neoplasms of the stomach, cause symptoms associated with ulcers and *Helicobacter pylori* infection and cannot be differentiated through imaging or observation. Therefore, histopathology and a high index of clinical radiological suspicion are necessary to suspect and diagnose this unusual cancer.

We present a case of a 52-year-old Peruvian woman who was initially diagnosed with a gastrointestinal stromal tumor that was later shown to have an antral glomangioma.

CASE REPORT

In June 2023, a 52-year-old Peruvian woman from Piaján, Ascope, La Libertad, Peru was seen in a private clinic. She complained of gastric symptoms of upset stomach, early satiety, and belching lasting for 10 months. Her medical history included hypertension

and diabetes, which were treated with irbesartan and metformin. She was also diagnosed with *H. pylori* infection by antral biopsy and was also taking magaldrate, simethicone, mosapride, and pantoprazole for her gastric symptoms. Her father and paternal aunt were diagnosed with prostate and breast cancer.

In June 2023, an upper gastrointestinal endoscopy revealed a crescent-shaped subepithelial lesion located between the major and minor curves of the anterior face of the antrum. It obstructed approximately 70% of the pylorus. Histology revealed diffuse moderate chronic gastritis with intestinal metaplasia.

Because the lesion was subepithelial, an endoscopic ultrasound (echo-endoscopy) was performed approximately one month later to further characterize the lesion. A subepithelial lesion measuring 51x38 mm was observed in some sections from the 4th layer. It was heterogeneous with hyperechogenic sections, which suggested calcifications. A biopsy of the lesion was performed using an echography-guided needle. Cytological analysis revealed spindle cells with no apparent atypia of neoplastic or mesenchymal origin. Immunocytochemical analysis of the biopsied cells gave positive results for antibody to CD117 and were 5% positive to antibody for Ki 67.^[7] They were not reactive to the active alpha oestrogen receptor. This result suggests a stromal gastrointestinal tumor, as approximately 90% of cases are positive for CD117.^[8]

With these results, the patient was referred to the Oncologic Surgery department of the Hospital EsSalud de Alta Complejidad Virgen de la Puerta, where an abdominal and pelvic tomography with contrast and blood analysis were completed. Tomography largely confirmed the endoscopic observations (Figure 1). A slightly distended stomach, diffuse liver disease, and mural thickening at the level of the pyloric canal were observed. The thickening shows enhancement with contrast and some partial calcification. The results of the blood analysis were mostly unremarkable, except for a slightly decreased prothrombin



Figure 1. Tomography of abdomen and pelvis. The stomach is slightly distended; there is a mural thickening at the level of the pyloric canal, which shows enhancement with the contrast substance, as well as some parietal calcifications.

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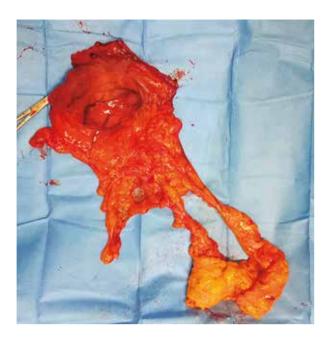


Figure 2. Surgical Specimen. Anterior view of the surgically removed antrum and omentum, showing a tumor on the anterior side of the antrum, with a rounded, regular, exophytic appearance.

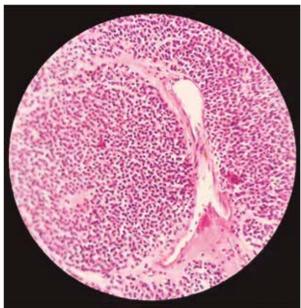


Figure 3. Haematoxylin and eosin (40x) micrograph of the tumor. The tumor consists mainly of small, rounded, well-defined cells, arranged around slightly dilated vessels and low mitotic activity.

time and slightly elevated alanine aminotransferase activity. Physical examination revealed ECOG level 1 and no peripheral lymphadenopathy. The abdomen was globulous, soft, and depressible with superficial tenderness. No palpable masses were perceived. Together, these results lead to an initial diagnosis of GIST, and the patient was recommended for surgery to remove the tumor.

Surgery consisted of an exploratory laparotomy, D0 subtotal gastrectomy, and a Hofmeister-Finsterer ansioperistaltic transmesocolic anastomosis with placement of a Blake drain and nasojejunal tube. The surgical findings included an open cavity, a soft, rounded, and mobile lesion approximately 6 x 5 cm located at the gastric curvature that does not retract into the serous membrane upon pressure (Figure 2). One lymph node in group 4 had a palpable enlargement. The liver had smooth edges, no peritoneal implants were observed, and the rest of the abdominal organs were normal without secondary lesions.

Histopathology of the tumor revealed that it had a size of 4 cm, and mild nuclear grade (Figure 3). Neither necrosis nor perineural invasion was evident, but lymphovascular invasion was. Surgical edges were free, and no lymph nodes (0 of 19) were positive for neoplasm. Immunohistochemistry results were: cKIT (CD117) negative, CD34 negative, alpha actin positive, S-100 negative, calponin positive, desmin negative, EMA negative, vimentin positive, focal synaptophysin (-/+), chromogranin negative, CD31 negative, PK negative, LCA (CD45) negative, Ki67 approximately 1%, caldesmon positive, and the glomus cells shows as rounded shaped, pale cytosol and round to oval-shaped nuclei (Figures 4 and 5). Histomorphology and immunohistochemistry of

the excised piece favour diagnosis of glomangioma over gastrointestinal stromal tumor. [3,9-12]

In the postoperative period, nutritional therapy with polymeric isotonic nutrients was initiated through the nasojejunal tube placed during surgery. Subsequently, there was a progression to oral feeding, but this was discontinued due to stenosis caused by inflammation at the site of the gastrojejunal anastomosis, which was resolved with a one-week course of corticosteroids. The patient was discharged from the hospital in October 2023 with good tolerance to oral feeding. As of November 2023, she is receiving treatment for her *H. pylori* infection under the care of a gastroenterologist, which consists of esomeprazole, amoxicillin, and clarithromycin. Additionally, she continues to have follow-ups in the oncology department.

DISCUSSION

Glomus tumors are benign tumors of glomic cells, which are modified smooth muscle cells located in the walls of the Sucquet-Hoyer canals (arteriovenous anastomoses or shunts), whose function is to regulate skin temperature by regulating blood flow. Glomus tumors are usually found in the peripheral soft tissues, but can arise elsewhere, including the stomach or intestines, where they can cause acute or chronic bleeding, which results more frequently in melena than haematemesis. Other ulcerlike symptoms are frequent. The tumor may also cause gastric perforation or be symptom-free. [1,10,11]

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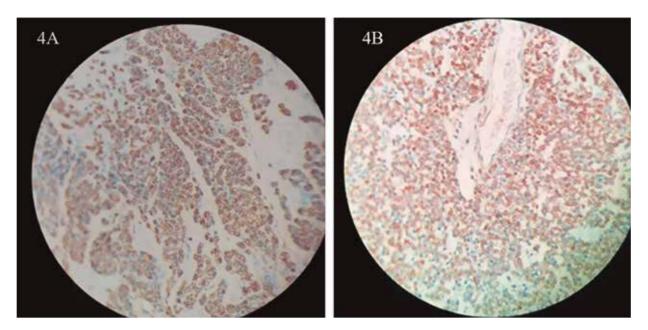


Figure 4. Immunohistochemistry. **A.** Smooth muscle actin stain of the surgical specimen (40x) revealing a positive result. **B.** Calponin stain of the surgical specimen (40x) revealing a positive result.

In this case, the tumor caused early satiety and gastric discomfort due to its antral location. The patient did not have anaemia, gastric perforation, or bleeding. Therefore, her symptoms were nonspecific, making additional analysis necessary for diagnosis.

Diagnosis of gastric glomus tumors is difficult due to nonspecific symptoms, similarity to other neoplasms, and subepithelial nature. Endoscopic biopsy is not particularly useful in these cases because the tumor resides beyond the reach of the biopsy and other imaging technologies reveal solitary hypervascularized

lesions with an average side between 1 and 4 cm, which resemble more common neoplasms like GIST or lymphoma. [13]

Standard treatment for gastric glomus tumors is surgical excision consisting of complete resection with free margins of the tumor without lymphonodectomy, according to several case reports. $^{[4,5,14-16]}$ In our case, the patient was treated by surgery. As the lesion was circumferential, antral and stenosing, the patient underwent a partial gastrectomy with a proximal surgical margin measuring $4.5 \times 3.5 \text{ cm}$ and a distal surgical border measuring

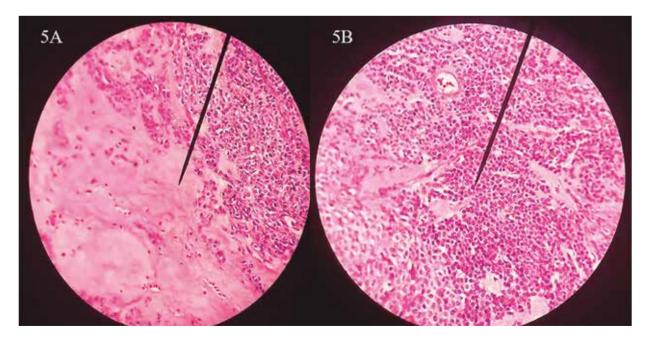


Figure 5. Haematoxylin & Eosin (40x). **A.** Glomus cells located at the right side and hyalinized stroma at the left side. **B.** The glomus cells show as rounded shaped, uniforms, round to oval-shaped nuclei and pale cytosol.

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2.5x2 cm. The affected lymph nodes only corresponded to the antral area, specifically to groups 3b, 5, 6 and 4d, which were resected together to keep the specimen intact and prevent its rupture. The lymphandectomy was not part of the pre-surgery plan but was required as one lymph node was larger than normal.

Immunohistochemistry using appropriate markers is essential in the correct diagnosis of glomus tumors. As seen in this case, neither imaging nor routine endoscopic biopsy was sufficient to rule out GIST. The appropriate immunology tests were required to make the diagnosis. Typically, GIST is positive for cKIT (CD117), carcinoid tumor is positive for chromogranin, and gastric lymphoma is positive for common leukocyte antigen, unlike glomus tumors, which are negative for all of these markers and characteristically positive for smooth muscle actin.[3,10-12 17] One misleading aspect of this case was that the biopsy yielded a positive result for cKIT (CD117), but subsequent immunohistochemical analysis yielded a negative result. Possible explanations include poor sensitivity and specificity of the test, that rare tumors are not expected among pathologists, and that the size of the biopsy was small and did not represent the entire tumor in the way a surgical specimen could.

Adverse histopathological prognostic factors for gastric glomus tumors include tumor size and lymphovascular invasion. Although rare, malignant gastric glomus tumors have been reported in the literature. [3,10,18] Tumor size has been proposed as an indicator for malignancy [3,10], with sizes larger than 5 cm more likely to become malignant or metastasize. Vascular invasion is a second sign of poor prognosis. [3] In this case, the tumor was 4 cm in size with lymphovascular invasion, but the Ki67 nuclear proliferation index was approximately 1% and the mitosis index was 1 in 50 with hematoxylin phloxine saffron staining, so its biological behaviour was likely not aggressive. Given this information, a favourable clinical outcome can be expected.

Also of interest is that the patient was infected with *H. pylori*, which has been shown to cause gastric symptoms and MALT. Therefore, it is reasonable to question whether *H. pylori* may be associated with gastric glomic tumors as well. A review of the literature [19-25] indicated that there were 6 patients with both gastric glomus tumor and *H. pylori* infection (including this case) and 21 patients with gastric glomus tumor without a diagnosed *H. pylori* infection. Most of these cases come from a single reference, and since *H. pylori* infection is common across the world, it is likely that there is no causal link, but more systematic studies could improve the data quality.

In conclusion, this case underscores the challenge of accurately diagnosing antral glomangioma before surgery. With current practices and technology, a definitive diagnosis will likely still require analysis of the surgical specimen. Nevertheless, this case suggests the importance of immunohistochemical testing for markers such as cKIT (CD117), chromogranin, common leukocyte antigen, and smooth muscle actin to facilitate a rapid and accurate differential diagnosis.

Authors' contributions: EFYQ, FMAC, and JRTV participated in patient care and manuscript drafting. EVJ was responsible for compiling the clinical history, while ELSPM reviewed the pathological data. LJFR contributed to writing and editing the manuscript.

Funding: The research was self-funded by the authors.

Conflicts of interest: The authors declare that they have no economic or non-economic conflicts of interest in the publication of this article.

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REFERENCES

- Gombos Z, Zhang PJ. Glomus tumor. Arch Pathol Lab Med. 2008;132(9):1448-52. doi: 10.5858/2008-132-1448-GT.
- Chao González L, Gil Llanes I, Moret Vara S, Barroso Márquez L, Mesa González M, Pérez González T. Tumor glómico gástrico. Invest Medicoquir. 2019;11(2):1-7.
- Miettinen M, Paal E, Lasota J, Sobin LH. Gastrointestinal glomus tumors: a clinicopathologic, immunohistochemical, and molecular genetic study of 32 cases. Am J Surg Pathol. 2002;26(3):301-11. doi: 10.1097/00000478-200203000-00003.
- Vassiliou I, Tympa A, Theodosopoulos T, Dafnios N, Fragulidis G, Koureas A, et al. Gastric glomus tumor: a case report. World J Surg Oncol. 2010;8:19. doi: 10.1186/1477-7819-8-19.
- Devanathan M, Jamuna Rani S, Chandramohan SM, Anbarasu S. A rare submucosal tumour of stomach-glomus tumour: A case report. Int J Surg Case Rep. 2015;11:64-67. doi: 10.1016/j.ijscr.2015.04.014.
- Wang LM, Chetty R. Selected unusual tumors of the stomach: a review. Int J Surg Pathol. 2012;20(1):5-14. doi: 10.1177/1066896911429300.
- Seidal T, Edvardsson H. Expression of c-kit (CD117) and Ki67 provides information about the possible cell of origin and clinical course of gastrointestinal stromal tumours. Histopathology. 1999;34(5):416-24. doi: 10.1046/j.1365-2559.1999.00643.x.
- Demetri GD, von Mehren M, Antonescu CR, DeMatteo RP, Ganjoo KN, Maki RG, et al. NCCN Task Force report: update on the management of patients with gastrointestinal stromal tumors. J Natl Compr Canc Netw. 2010;8 Suppl 2(0 2):S1-41; quiz S42-4. doi: 10.6004/jnccn.2010.0116.
- Chen KB, Chen L. Glomus tumor in the stomach: A case report and review of the literature. Oncol Lett. 2014;7(6):1790-1792. doi: 10.3892/ol.2014.1986.

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 Lin J, Shen J, Yue H, Li Q, Cheng Y, Zhou M. Gastric Glomus Tumor: A Clinicopathologic and Immunohistochemical Study of 21 Cases. Biomed Res Int. 2020;2020:5637893. doi: 10.1155/2020/5637893.

- 11. Wang ZB, Yuan J, Shi HY. Features of gastric glomus tumor: a clinicopathologic, immunohistochemical and molecular retrospective study. Int J Clin Exp Pathol. 2014;7(4):1438-48.
- Rosales-López E, Salceda-Otero JC, Angulo-Molina D, Posada-Torres JA, Canchola-Aguilar MG, Lozoya-Gonzalez D. Gastric glomangioma, differential diagnosis of gastrointestinal stromal tumors. Rev Gastroenterol Mex (Engl Ed). 2018;83(1):72-74. English, Spanish. doi: 10.1016/j.rgmx.2016.08.006.
- 13. Padilla-Herrera C, Suárez LM, Guevara RE, Barriga JC. Tumores gástricos peculiares: tumor glómico, schwannoma, leiomioma y cistoadenoma en el páncreas ectópico. Serie de casos y revisión de la literatura. Rev Colomb Gastroenterol. 2021;36(4):463-472. https://doi.org/10.22516/25007440.716.
- 14. Papadelis A, Brooks CJ, Albaran RG. Gastric glomus tumor. J Surg Case Rep. 2016;2016(11):rjw183. doi: 10.1093/jscr/rjw183.
- Tsagkataki ES, Flamourakis ME, Gkionis IG, Giakoumakis MI, Delimpaltadakis GN, Kazamias GM, et al. Gastric glomus tumor: a case report and review of the literature. J Med Case Rep. 2021;15(1):415. doi: 10.1186/s13256-021-03011-0.
- Pansa A, Samà L, Ruspi L, Sicoli F, Cananzi FCM, Quagliuolo V. Glomus Tumor of the Stomach: A Systematic Review and Illustrative Case Report. Dig Dis. 2023;41(1):17-33. doi: 10.1159/000525513.
- 17. Torío B, Berzal F, Aparicio R. Tumor glómico en antro gástrico. Rev Esp Enferm Dig. 2008;100(10):660-661.

- Zaidi S, Arafah M. Malignant Gastric Glomus Tumor: A Case Report and Literature Review of a Rare Entity. Oman Med J. 2016;31(1):60-4. doi: 10.5001/omj.2016.11.
- Mehmood F, Jamil H, Khalid A. Gastric Glomus Tumor: A Rare Cause of Acute Blood Loss Anemia. Cureus. 2022;14(4):e24511. doi: 10.7759/cureus.24511.
- Nascimento EF, Fonte FP, Mendonça RL, Nonose R, de Souza CA, Martinez CA. Glomus tumor of the stomach: a rare cause of upper gastrointestinal bleeding. Case Rep Surg. 2011;2011:371082. doi: 10.1155/2011/371082.
- 21. Kapiev A, Lavy R, Sandbank J, Halevy A. Glomus tumor of the stomach. Isr Med Assoc J. 2012 Mar;14(3):192.
- Alkhateb O, Daaboul O, Daaboul B, Abouharb R, Alazmeh MA, Hamed G. Glomangiomyoma of the Stomach: Case Report. Case Rep Gastroenterol. 2023;17(1):185-190. doi: 10.1159/000529044.
- 23. Birris T. A Rare Cause of Iron Deficiency Anemia in a Young Woman: Glomus Tumor of the Stomach Associated with *Helicobacter pylori* Infection: 544. Am J Gastroenterol. 2011;106:S210.
- Knackstedt C, Wasmuth H, Donner A, Trautwein C, Winograd R. Diagnosis of an unusual tumor in the duodenum. Endoscopy. 2007;39 Suppl 1:E94. doi: 10.1055/s-2007-966456.
- Xing JJ, Huang WP, Wang F, Chai YR, Gao JB. Computed tomography features and clinicopathological characteristics of gastric glomus tumor. BMC Gastroenterol. 2022;22(1):174. doi: 10.1186/s12876-022-02241-w.