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Glomus tumor of the stomach: a case report

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Summary

Glomus tumors are relatively uncommon benign neoplasms, most common in the skin and subcutaneous tissue, but can also occur in the viscera, most often in the stomach. Preoperative diagnosis, when the tumor occurs in the stomach, seems difficult but is important, since glomus tumors of the stomach are essentially benign and amenable to conservative excision, contrary to aggressive gastric neoplasms. We describe the case of a 75-year-old man presented to the emergency department with gastrointestinal bleeding and epigastric pain. Gastroscopy revealed a submucosal mass in the antrum of the stomach in correspondence to the lesser curvature. Biopsies taken from the lesion showed morphological and immunohistochemical [positivity for alpha-smooth muscle actin (alpha-SMA) and vimentin, negativity for CD34, CD117, chromogranin, synaptophysin, desmin and S-100 protein] characteristics attributable to glomus tumor. The patient underwent a laparotomic antrotomy incision. Histological examination of the tumor confirmed the diagnosis of gastric glomus tumor.

Key words: gastric, glomus tumor, immunohistochemistry

Introduction

Glomus tumors are relatively uncommon benign neoplasms originating from modified smooth muscle cells of the normal glomus body [1,2]. They usually occur in the dermis or subcutis of the extremities. Unusual locations have included the patella, chest wall, bone, stomach, colon, nerve, trachea, mediastinum, mesentery, cervix and vagina [3,4]. Since glomus tumor of the stomach is essentially benign and amenable to conservative excision, preoperative differential diagnosis from other, aggressive gastric neoplasms is of primary importance.

Case presentation

A 75-year-old man presented to the emergency department of our hospital complaining of nausea, vomiting and vague periumbilical pain, accompanied by progressive weakness and weight loss complicated finally by an episode of epigastric pain and gastrointestinal bleeding. Past medical history included essential hypertension and hyperlipidemia. There was no family history for intestinal disease. Physical examination, chest X-ray and ECG revealed no significant features. Laboratory analyses - including complete blood count, biochemical examination, tumor markers (CEA, CA19-9, AFP), microbiological cultures for bacteria and parasites in feces and culture of the urine - were normal. Gastroscopy revealed an ulcer of maximum diameter 1 cm in the antrum of the stomach in correspondence to the lesser curvature. Biopsies were taken from the lesion.

Histological examination of the samples taken from the stomach revealed mild chronic gastritis, without activity or atrophy. In the submucosa a well circumscribed lesion was verified, consisting of tight convolutes of capillary-sized vessels surrounded by collars of small round or cuboidal cells with uniform

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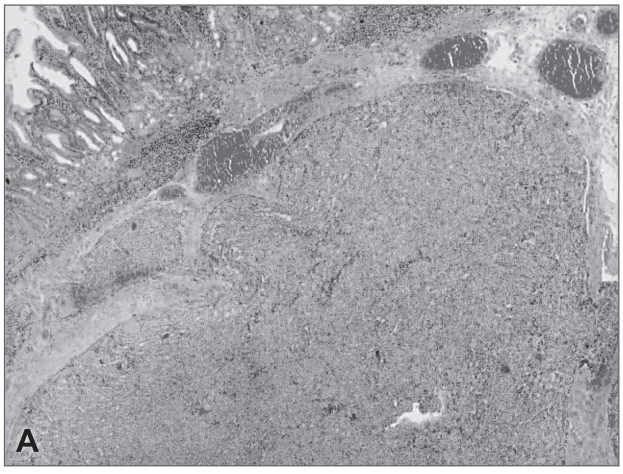
round hyperchromatic nuclei and acidophilic cytoplasm set in a myxoid stroma. Atypia of the neoplastic cells, mitosis or necrosis were absent (Figure 1A, 1B). The immunohistochemical profile of the tumor was positive for alpha-SMA (NovoCastra Newcastle Upon Tyne, UK; Figure 2) and vimentin (NovoCastra) and negative for CD34 (Biogenex, San Ramon, USA), CD117 (DakoCytomation, Glostrum Denmark), CD20 (NovoCastra), CD45RO (DakoCytomation), CD45RA (NovoCastra), Chromogranin (NovoCastra), Synaptophysin (BioGenex), Desmin (NovoCastra) and S-100 protein (DakoCytomation). The expression of the cell proliferation marker Ki67 (DakoCytomation) was low. These characteristics were attributable to glomus tumor of the stomach.

Biopsy samples taken from the duodenum demonstrated edema and increased number of inflammatory cells in the lamina propria.

Colonoscopy showed edema and erythema of the mucosa. Biopsy samples were taken and histological examination showed mild inflammation.



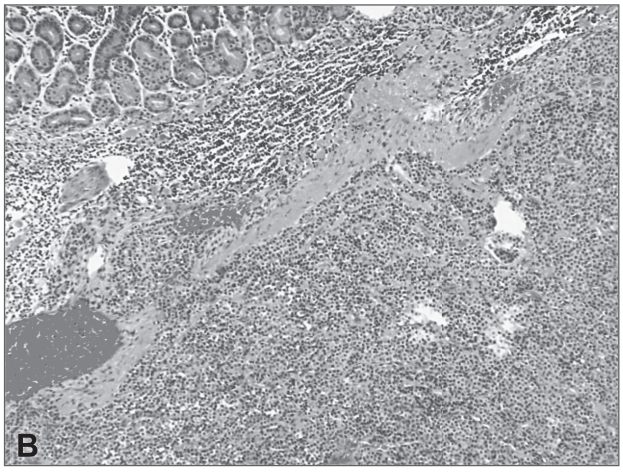


Figure 1. A: Glomus tumor of the stomach (H&E ×40). **B:** Glomus tumor of the stomach (H&E ×100).

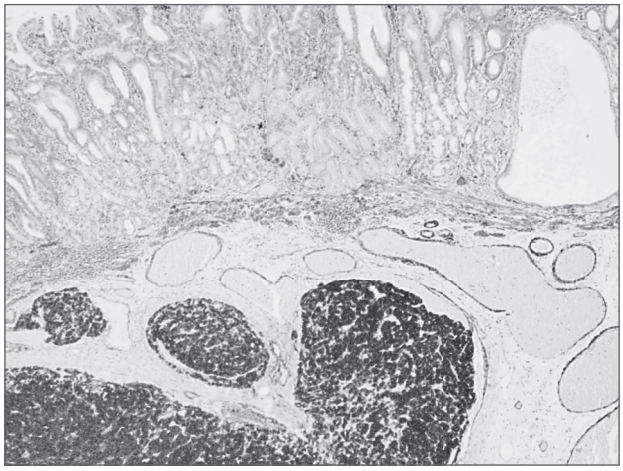


Figure 2. Glomus tumor of the stomach (smooth muscle actin ×100).

The patient underwent a laparotomic antrotomy incision. Grossly, the specimen, after fixation in formalin, consisted of a submucosal fleshy mass 2×1.8×0.9 cm. The mass was fixed to the mucosa which contained an irregular central erosion of maximum diameter 1 cm.

The postoperative course was uncomplicated. The patient remained asymptomatic and during a 6-month follow-up there was no evidence of recurrence. Almost a year later he died of unspecified reasons.

Discussion

Glomus tumor is a distinctive neoplasm originating from modified smooth muscle cells of the normal glomus body [1,2]. The normal glomus body is a specialized form of arteriovenous anastomosis that serves in thermal regulation. Glomus bodies are located in the stratum reticularis of the dermis throughout the body, but they are more numerous in the digits, palms and soles of the feet [5]. Glomus bodies consist of an afferent arteriole, a tortuous arteriovenous anastomosis, a system of collecting veins and a neurovascular reticulum that regulates the flow of blood through the anastomosis.

The classical location is the subungual region, but it can occur elsewhere in the skin, soft tissues, nerves, stomach, nasal cavity and trachea.

Fople et al. [6] studied 52 unusual glomus tumors and proposed the following classification scheme and criteria: malignant glomus tumor (tumor with deep location and maximum diameter of more than 2 cm, or atypical mitotic figures, or moderate to high nuclear grade and 5 mitotic figures/50 HPF); symplastic glomus tumor (tumor with high nuclear grade in the ab-

sence of any other malignant feature); glomus tumor of uncertain malignant potential (tumor that lacks criteria for malignant glomus tumor or symplastic glomus tumor but has high mitotic activity and superficial location only, or large size only, or deep location only); glomangiomatosis (tumors with histologic features of diffuse angiomatosis and excess glomus cells). Using this classification, metastasis was observed in 38% of tumors fulfilling the criteria for malignancy. In contrast, metastatic disease was not seen in any specimen classified as symplastic glomus tumor, glomus tumor of uncertain malignant potential or glomangiomatosis.

Lesions similar to glomus tumors of peripheral soft tissue occur in the gastric antrum as small intramural masses. They are more common in women than in men in a ratio of 2.5:1 and a median age of 55 years [7,8]. One third manifests as ulcer, one third as bleeding and one third is asymptomatic.

In endoscopic ultrasonography gastric glomus tumor appears with homogeneous or heterogeneous hypoechoic pattern mixed with internal high echoic spots [9].

Histologically, the tumors typically have a solid pattern of round glomus cells with low mitotic activity. Vascular invasion and focal atypia have been reported [2]. Immunohistochemically, tumor cells are positive for alpha-SMA, vimentin and calponin, and negative for desmin and S-100 protein. In some cases neoplastic cells in glomus tumor may show a coexpression of alpha-SMA and CD34, an important finding regarding the differential diagnosis of these lesions and other perivascular neoplasms [10,11].

Gastric glomus tumors have a good overall prognosis, but a small unpredictable potential for malignant behavior exists [2] and occurs in larger tumors (>5 cm) showing nuclear atypia. These tumors require strict postoperative follow-up [2,12,13].

A case of malignant gastric glomus tumor with metastasis to the liver, lymph nodes and peritoneum has been reported. The patient, however, survived for more than 15 years after surgical resection [14].

The main differential diagnosis of gastric glomus tumor includes carcinoid and lymphoma. Differentiation from carcinoid and lymphoma may be particularly difficult in frozen section. The histological feature that confirms the diagnosis of glomus tumor is the arrangement of glomus cells in complete and incomplete lobules along the vascular channels, giving an organoid appearance. Recognizing the histological pattern of gastric glomus tumor in frozen section is important in order to prevent unnecessary radical surgery [15]. In addition, glomus tumor, carcinoid and lymphoma have different immunohistochemical profiles: glomus tumor is positive for SMA and negative for neuroendocrine markers and leucocyte common antigen (LCA) [2,10,11].

The lack of CD117 positivity has been described as a feature of glomus tumor of gastrointestinal origin and this helps to distinguish glomus tumor from gastrointestinal stromal tumors [16].

We conclude that most glomus tumors are benign and can be treated adequately by simple excision. Therefore, differential diagnosis from other, aggressive gastric neoplasms is of crucial importance, particularly in frozen sections. However, postoperative follow-up is essential in order to prevent local recurrence.

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