GASTRIC GLOMIC TUMOR: A CASE REPORT

TUMOR GÁSTRICO GLÔMICO: UM RELATO DE CASO

MARIA CLARA MENEZES DE **MONTEIRO**[1], RAMON SOTTO DE **CASTRO**[2], GABRIEL S. THIAGO **CAVALLEIRO**[3], VINICIUS MOREIRA **PALADINO**[3], ANA CLARA LUZ **PRATES**[3], AMANDA GUIMARÃES DE **SOUZA**[3], DANIEL ALMEIDA DA **COSTA**[4], FERNANDO ANTONIO GALVÃO PEREIRA **JUNIOR**[5]

1. Residente de Oncologia na Santa Casa de Misericórdia de São Paulo/SP; **2.** Médico, pela Universidade de Vassouras/RJ; **3.** Residente de Clínica Médica do Hospital Escola de Valença/RJ; **4.** Professor e Mestre em Reabilitação do Centro Universitário de Valença/RJ; **5.** Professor de Gastroenterologia Centro Universitário de Valença/RJ.

*Sargento Vitor Hugo St, 161, Fatima, Valença, Rio de Janeiro, Brazil. ZIP CODE: 27600-000. daniel.almeida@faa.edu.br

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ABSTRACT

Gastric glomic tumor is a rare type of subepthelial mesenchymal tumor, arising from the neuromyoarterial glomus, an arteriovenous anastomosis that works without the intermediary capillary, usually located in the skin, being responsible for its thermoregulation. It appears in the extremities and the stomach is described as a rare site for these tumors clinically recognized as benign, but some show a biological behavior like those of malignant origin. We report the case of a male patient in a private office with a recent-onset dyspeptic syndrome, with heartburn as a symptom. The main differential diagnosis was made with the GIST, which cannot exclude their malignant potential, an adequate follow-up of these patients was necessary, concluding that case reports are necessary for a better understanding of the gastric glomus tumor, which is a rare entity, difficult to differentiate from other submucosal tumors, being the histopathological and immunohistochemical study essential for its differentiation. Surgical resection is the definitive treatment.

KEYWORDS: Gastric glomus tumor, dyspeptic syndrome, immunohistochemistry.

1. INTRODUCTION

Gastric glomic tumor is a rare type of subepithelial mesenchymal tumor, arising from the neuromyoarterial glomus, an arteriovenous anastomosis that works without the intermediary capillary, usually located in the skin, being responsible for its thermoregulation. It appears in the extremities and the stomach is described as a rare site for these tumors[1].

The glomus of this tumor, whose function is to regulate blood circulation and body temperature, consists of an afferent arteriole, an anastomotic vessel called the Soucquet-Hoyer canal, surrounded by smooth muscle fibers, an efferent vein, nerve fibers and peripheral capsule[2].

This type of tumor has been described as a benign lesion. Its most common location is in the hand, in the subungual region, but it is found in other places, 65% are found under the nails and, sometimes, bilateral[2]. These tumors are more common among women between 50 and 60 years of life[1].

It is usually located in the sublingual region, but can rarely be found in the trachea, mediastinum, lung, bladder, skeletal muscle tissue and soft tissue. Its gastric presence is rare and only isolated cases are reported. The most frequent clinical manifestation is upper digestive bleeding due to tumor ulceration; other symptoms described are dyspepsia, abdominal pain and pyloric obstruction, upper digestive hemorrhage, postprandial fullness, dyspepsia, vomiting and rarely weight loss[3]. Sporadic case**s** of malignant glomus tumor with the presence of distant metastases have been reported[4].

Endoscopically, it is a solitary nodule in the antrum or prepyloric space, commonly in the greater curvature, damaging the submucosal or muscular layer itself. They are clinically profiled as benign, but some indicate biological behavior analogous to those of malignant origin, with the capacity to invade, metastasize and recur, when larger than 5 cm[5], and may affect cervical blood vessels, cranial nerves and the autonomic nervous system, with incidence of 1:30.0006.

In morphology they are like rounded, uniform and hypervascularized cells. In immunohistochemistry, they are positive for α-SMA, calponin, laminin, vimentin, type IV collagen and negative for desmin[4]. The main differential diagnoses of gastric glomus tumor are carcinoid tumors, GIST (gastrointestinal stromal tumor), paraganglioma, leiomyoma and hemangiopericytoma[1,6].

The theme is justified by the need for understanding, for subsequent application of the appropriate diagnostic methods and their therapeutics, aiming at a case report of gastric glomus tumor, with a literature review, exposing the diagnostic methods and therapeutics used in the service in patients with gastric glomic tumor in a particular office.

This article will report a case of Glomic Gastric Tumor that occurred in private care in Valença-RJ, correlating clinical data and conduct with others found in the literature.

2. CASE REPORT

A 48-year-old male case is presented, weighing 88 KG with a personal pathological history of dyspeptic syndrome of recent onset, who was seen in an office on 10/23/2019. At the consultation, he indicates that he obtained relief with the use of omeprazole, reporting other previous episodes of lesser intensity.

The patient has a previous pathological history to vasectomy, is unaware of drug allergy and drug use. Denies family comorbidities

Her family history indicates that the father died from carcinomatosis and the mother from cancer with an unknown focus. He denies a history of peptic ulcers and his social history denies alcoholism and smoking. The examination indicated good general condition. Colored, hydrated, anicteric, acyanotic and afebrile. Her blood pressure was 120/80 mmg, with a heart rate: 80 bpm and a respiratory rate of 20 bpm. Her abdomen was saggy, peristaltic, and painless. Do not palpate masses or visceromegaly. The pulmonary and cardiac apparatus showed no changes

On 11/06/2019, he returned to the office with the result of the Upper Digestive Endoscopy, which presented a subepithelial lesion of the antrum, the patient was asymptomatic, for this reason the doctor requested an ultrasound endoscopy.

On 02/28/2020, the patient came to the office with the result of the Ecoendoscopy, indicating a 20.9 mm subepithelial lesion restricted to the muscle layer, the doctor considered the hypothesis of leiomyoma or GST, but required immunohistochemistry as a differential diagnosis.

On 03/11/2020, he presented himself again with the laboratory results that indicated Leukocytes: 4500, Platelets: 225000, Creatinine: 1.0, Glucose: 96, K: 3.4, Cholesterol: 223, Glucose: 132, Uroculture: NEGATIVE

Based on the results of the immunohistochemistry, the physician diagnosed a glomus tumor and considered continuing the treatment in 6 months, a period given for monitoring the growth or not of the glomus tumor.

The patient in question addressed in this study after being guided by the assistant physician about the surgical treatment that consisted of partial gastrectomy to remove the lesion, this being a definitive and curative treatment. Decides not to undergo surgical treatment at first. The patient is then under outpatient follow-up in a way that has been maintained with clinical consultations and serial imaging tests to date.

Complying with the ethical precepts of Resolution 196/96 for research with human beings, the project was submitted and approved by the Ethics and Research Committee of the Faculdade de Medicina de Valença.

3. DISCUSSION

In this article, the gastric glomic tumor is discussed, which is a rare benign tumor originating from modified smooth muscle cells of the glomus body, which is a neuromyoarterial structure that intervenes in the regulation of peripheral body temperature and in the control of arteriolar blood flow[7,8].

In turn, authors[9,10] define it as a benign tumoral lesion, located in the digital pulp and subungual region, and the literature mentions rarer locations, such as the central nervous system, stomach, liver, mediastinum, trachea, lungs, bones, joints, and organs genitals.

Due to the diversity of glomus bodies in our body, tumors of these structures can be found in various locations, being reported in the subungual region and more rarely in other regions such as liver, stomach, lungs, bones, joints, genitals, central nervous system, and blood vessels according to teaching[11,12].

According to others[13,14], its formation encompasses nerve fibers, muscle cells and vascular components and for this fact it was called glomangioma, which is a rare tumor, representing less than 2% of soft tissue tumors.

Studies[8,15] indicate that this tumor is located preferentially in the skin and in the subcutaneous cellular tissue, with primacy in subungual beds. Cases in bones and joints, skeletal muscle, mediastinum, trachea, kidney, uterus, vagina, and gastrointestinal tract are described.

The gastric presentation of glomus tumor is rare, representing 1% of gastrointestinal stromal tumors. Most are benign tumors, even though malignant tumors have been reported in only 1% of the diagnosed glomus tumors, with metastases in the liver, brain, kidney, and skin, according to articles[16,17] and according to the case presented.

In a review of surgical specimens[16] resected for gastric mesenchymal tumors, they indicated that only 0.58% of cases are of glomus tumor. The glomus tumors in the stomach are usually located in the antrum, are usually single and measure between 2 and 2.5 cm, with an allusion that the size for suspected malignancy is greater than 5 and 10 cm, as shown by the figures presented in the case report.

Miettinen *et al.* (2002)[18] found a difference in the malignant behavior of deep glomus tumors of the extremities in relation to gastric tumors. In the case of soft tissue tumors, the size above 2 cm has malignant potential, while in the gastric tumors most have a size above 2 cm, but the behavior is benign.

Therefore, other studies[14,18] indicate a size above 5cm for those of gastric origin as a potential for malignancy, and the number of mitoses does not influence the malignant potential in large tumors. The main differential diagnosis is made with GISTs, with glomus tumors being responsible for 1% of this entity, as demonstrated by the examination requests in the case report.

According to Chong *et al.* (2009)[19] and Fujioka *et al.* (2009)[20], the clinical history of its appearance includes some type of trauma. Pain is shared by all sufferers. Initially it is tame, but over time it becomes intense and unbearable. There is a periodicity between pain episodes, but as the disease progresses, the frequency of pain increases. The time of appearance is

usually at night, preventing the patient from sleeping. One of the theories to explain the intense pain is based on the expansion of the tumor, as it is restricted to a pseudocapsule, its growth is hampered, with necrosis of central cells and replacement by connective tissue. Another theory is the location of this tumor in the myoneural plate.

Another author[16] states that the clinical manifestations are changeable, ranging from asymptomatic cases to dyspeptic symptoms, chronic anemia, epigastric pain, nausea and/or vomiting or acute digestive bleeding. It occurs in men and women with a ratio of 1:1.6 and a median age of 45 years, which was corroborated by Castro *et al*. (2016)[11.] The clinical case presented indicates that the patient has dyspeptic syndrome of recent onset, corroborating what was mentioned by the authors.

Diagnosis is usually early due to pain, but the literature indicates cases with 40 years of evolution. As for the histopathological diagnosis, some authors suggest that cellular atypia comes from the accumulation of heterochromatin linked to DNA inactivation. Capillarity is increased, its local architecture in the arrangement of tumor cells is performed with uniform size and shape, and a cellular uniformity of the base indicates its diagnosis. It should be noted that, when diagnosing a glomangioma, another one never goes unnoticed by the examiner, due to its characteristics. Glomus tumors are rare, and their diagnosis can be difficult, since the presentation does not include the 3 classic symptoms of intense and disproportionate pain, paresthesias and cold intolerance, as understood by Fujioka *et al*. (2009)[20]. In the case report, it was shown that the diagnosis was early, as the patient presents heartburn as a symptom.

Kato *et al*. (2015)[7] indicate that its endoscopic diagnosis is problematic, being generally observed as submucosal lesions, indistinguishable from gastrointestinal stromal tumors (GITS), leiomyomas, lipomas, ectopic pancreas, and carcinoid tumor, due to its submucosal origin, the biopsy obtained by the endoscopic method conventional is not favorable for the diagnosis, which was confirmed[11]. This understanding is corroborated by what was presented in the case report, by the requests made by the physician.

Kato et al.[7] further explains that imaging techniques are not useful for the differential diagnosis of these tumors. Most reported cases are diagnosed by histological study of the surgical sample. But EUS-guided fine needle aspiration biopsy (EUS-FNA) is beneficial in the preoperative diagnosis of gastric glomus tumor.

The histological study of this type of tumor, according to teachings[3,] indicates uniform, small, round, or polygonal cells, with highly visible cell membranes. The definitive diagnosis is based on an immunohistochemical study, with positivity for smooth muscle actin, calponin, h-Caldesmon and negative desmin. The definitive treatment is surgical, which was ratified[21.]

In 1980, Schick *et al*. (1980)[22] reported the first successful embolization of a glomus tumor followed by surgical removal. Embolization of the occipital and posterior auricular arteries and the thyrocervical trunk was performed; post-embolization arteriography showed a 30% decrease in tumor size, corroborated by the literature[23.]

As an option to arterial embolization[24,25], they indicate that direct puncture of hypervascularized tumors is safe. Even though embolization is not a definitive treatment, the literature indicates that tumor removal is more accurate after embolization, due to the devascularization of the neoplasm, reducing intraoperative bleeding and allowing for better visualization of adjacent structures, avoiding damage to them, and confirming the plan of cleavage. The cooling of intraoperative bleeding and greater ease of dissection were noted by the authors.

Only pathological anatomy gives the definitive diagnosis. The indicated treatment is complete surgical excision, which is usually possible due to its superficial and well-defined location, with cure rates higher than 95% of cases. The relapse rate of the glomus tumor varies from 1% to 33%, depending on the series analyzed. The treatment can also be by laser technique, being applied in cases of multiple glomangiomas, but there can be implantation of tumor cells due to rupture of the capsule, with implantation of a new tumor, presenting an average recurrence of 13.3%. Tumor removal leads to immediate symptom relief and complete pain resolution according to other authors' understanding[1,4].

When recurrence occurs in the early postoperative period, it is believed to be the result of incomplete excision. When it occurs late, it is understood that it is due to the development of a new glomic tumor or to the non-recognition of other lesser tumors in the first operative period. After excision, and if no complications are observed, a complete remission of symptoms is expected from the patient, as understood by the above authors[1,4].

4. CONCLUSION

Case reports such as this one is necessary for a better understanding of the gastric glomus tumor, which is a rare entity, difficult to differentiate from other submucosal tumors, and the histopathological and immunohistochemical study is essential for its differentiation. Surgical resection is the definitive treatment. EUS-FNA is useful for preoperative diagnosis.

Even though most gastric glomus tumors are benign, their malignant potential cannot be excluded, always bearing in mind the differential diagnosis of gastric submucosal lesions. Its treatment is surgical, and a routine follow-up of these patients is necessary.

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