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Incidental gastric schwannoma identified during open Roux-en-Y gastric bypass

Schwannoma gástrico incidental identificado durante bypass gástrico em Y de Roux

Victor K. Ivano[1]; Cassiano Machado[1]; Lucas Thá Nassif[1-3]; André Thá Nassif[1, 4]; Larissa Miyashiro[5]; Letícia Miyashiro[4]; Larissa L. G. Silva[6]

1. Hospital Santa Casa de Misericórdia de Curitiba, Curitiba, Paraná, Brazil. 2. Hospital Nossa Senhora das Graças, Curitiba, Paraná, Brazil.
3. Faculdades Pequeno Príncipe, Curitiba, Paraná, Brazil. 4. Universidade Positivo, Curitiba, Paraná, Brazil.
5. Pontifícia Universidade Católica do Paraná, Curitiba, Paraná, Brazil. 6. Laboratório Citopar, Centro de Citologia e Patologia do Paraná, Curitiba, Paraná, Brazil.

ABSTRACT

When a gastric mass is encountered, the surgeon must make the decision to resect the lesion or stop the surgery (for less morbidity) and discuss the best treatment. it's rare to find mesenchymal gastric tumors during bariatric procedures, specially gastric schwannomas (GS). 57-year-old woman with hypertension and diabetes under treatment, body mass index (BMI) of 36 kg/m², referring difficulties to lose weight with diet and physical activity. The patient underwent a Roux-en-Y gastric bypass (RYBG) with a wedge resection of a gastric nodule in the anterior wall of the incisura angularis. The immunohistochemistry (IHC) staining was strongly positive for S100, whereas c-kit (CD117), CD45, smooth muscle action (SMA), AE1/AE3 were negative. Hence, a final diagnosis of gastric schwannoma was made. GS should be included in the differential diagnosis of gastric nodules. We also present a surgical option to treat obese patients with gastric nodules during the bariatric procedure.

Key words: incidental findings; gastric bypass; stomach; bariatric surgery; gastrointestinal neoplasias; gastrointestinal stromal tumors.

RESUMO

Durante um procedimento cirúrgico, ao se deparar com uma massa gástrica, o cirurgião deve tomar a decisão de ressecá-la ou adiar a cirurgia (menor morbidade), bem como discutir com a equipe médica qual o melhor tratamento. Tumores gástricos mesenquimais encontrados durante a cirurgia bariátrica são raros, especialmente o schwannoma gástrico (SG). Relatamos o caso de uma mulher, 57 anos, hipertensa e diabética em tratamento, com índice de massa corporal (IMC) de 36 kg/m², referindo dificuldade em perder peso com dieta e atividade física. A paciente foi submetida ao bypass gástrico em Y de Roux com ressecção em cunha de nódulo localizado na parede anterior da incisura angular. A análise imuno-histoquímica foi fortemente positiva para proteína S100, enquanto c-kit (CD117), CD45, actina de músculo liso (SMA) e AE1/AE3 foram negativos. O SG deve ser considerado como diagnóstico diferencial de tumores gástricos. Apresentamos uma opção de tratamento que permite tratar pacientes obesos com nódulos gástricos em qualquer localização no mesmo procedimento da cirurgia bariátrica.

Unitermos: *achados incidentais; derivação gástrica; estômago; cirurgia bariátrica; neoplasias gastrointestinais; tumores do estroma gastrointestinal.*

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RESUMEN

Durante un procedimiento quirúrgico, ante una masa gástrica, el cirujano debe tomar la decisión de resecarla o posponer la cirugía (menor morbilidad), así como discutir con el equipo médico el mejor tratamiento. Los tumores mesenquimales gástricos que se encuentran durante la cirugía bariátrica son raros, especialmente el schwannoma gástrico (GS). Presentamos el caso de una mujer de 57 años, hipertensa y diabética en tratamiento, con un índice de masa corporal (IMC) de 36 kg/m², refiriendo dificultad para adelgazar con dieta y actividad física. La paciente fue sometida a un bypass gástrico Y de Roux con resección en cuña de un nódulo ubicado en la pared anterior de la escotadura angular. El análisis inmunohistoquímico fue fuertemente positivo para la proteína S100, mientras que c-kit (CD117), CD45, actina de músculo liso (SMA) y AE1/AE3 fueron negativos. El GS debe considerarse como un diagnóstico diferencial de los tumores gástricos. Presentamos una opción de tratamiento que le permite tratar a pacientes obesos con nódulos gástricos en cualquier lugar en el mismo procedimiento que la cirugía bariátrica.

Palabras clave: *hallazgos incidentales; derivación gástrica; estómago; cirugía bariátrica; neoplasias gastrointestinales; tumores del estroma gastrointestinal.*

INTRODUCTION

The main techniques to treat obesity are the sleeve gastrectomy (SG) and the roux-en-Y gastric bypass (RYGB). Each surgical technique has its advantages and the choice must be individualized. Sometimes during the surgery, even with normal preoperative exams, the bariatric surgeon encounters some unexpected events such as bleeding, or pathologies such as adhesions, hernias, and even tumors in different locations[1, 2].

Specifically, in the stomach, when a gastric mass is encountered, the surgeon must make the decision to resect the lesion or stop the surgery (for less morbidity) and discuss the best treatment. It's rare to find mesenchymal gastric tumors during bariatric procedures, with a 0,6%-0,8% gastrointestinal stromal tumors (GISTs) incidence in bariatric patients[3-5]. Surgeons are usually familiar with GIST, but not with the differential diagnosis [leiomyomas and gastric schwannomas (GS)] and their different prognosis. While GS usually have excellent prognosis after surgical resection, GIST can recur and have malignant potential[6-9].

Considering the rarity of GS (0,2% of gastric tumors and 6,3% of gastric mesenchymal tumors)[9-13], we present the first GS resected during a gastric bypass surgery. It is a case report where we incidentally found during an open bariatric surgery a gastric nodule in the anterior wall of the incisura angularis, that was treated with RYGB with partial gastrectomy of the excluded stomach.

CASE REPORT

A 57-year-old woman, non-smoker, with hypertension and diabetes under treatment, body mass index (BMI) of 36 kg/m², with surgical history of hysterectomy in 2009 for uterine cancer and cholecystectomy in 2018.

In November of 2019, the patient presented to the medical appointment referring difficulties to lose weight. The patient tried diet and physical activity without success. She also complaint of mild symptoms suggesting gastroesophageal reflux disease (GERD) and eventual epigastric pain with normal physical exam. The abnormal preoperative laboratory tests were serum B12 vitamin (inferior limit: 307 pg/ml), fasting glucose (superior limit: 99 ng/dl), triglycerides (319 mg/dl). The nutritionist, psychologist and endocrinologist cleared the patient for surgery. The cardiologic evaluation concluded that the patient had low risk for a bariatric procedure. Pharmacologic stress echocardiogram had no evidence of myocardial ischemia or disfunction. The electrocardiogram had sinusal rhythm with isolated ventricular extrasystole.

The only image done was an abdominal ultrasound that reported only moderate hepatic steatosis and supraumbilical hernia.

The upper gastrointestinal endoscopy (UGE) reported "ectopic gastric mucosa in the distal part esophagus; elevated lesion covered with normal mucosa with 20 mm in the anterior wall of the incisura angularis suggesting subepithelial lesion; negative

H. pylori biopsy; mild antral gastritis". The mucosa biopsy of the lesion's region showed antral gastric mucosa with foveolar hyperplasia and moderate inflammatory infiltrate, without intestinal metaplasia or dysplasia.

After discussion with the general and gastrointestinal surgery teams, a presumptive diagnosis of GIST was made and the patient underwent a surgical gastric bypass due to her comorbidities and GERD complaints, with partial gastrectomy of the excluded stomach. The operation began by retracting the omentum and the transverse colon cephalad and located the Treitz's ligament on the left corner of the base of the transverse mesocolon. From the Treitz's ligament, 100 cm of jejunum was measured and divided using a GIA-80 linear stapler with invaginant 1-layer oversuture of the staple line with 3-0 polydioxanone (PDS) to prevent postoperative bleeding. Then, 100 cm of alimentary limb (Roux Limb) was measured and a 1-layer handsewn side-to-side jejuno-jejunal anastomosis was performed 200 cm distal from the Treitz's ligament with 3-0 PDS. The intermesenteric space was then closed with 3-0 PDS. For the 30-ml gastric pouch, the stomach was divided using 2 loads of GIA-80 linear stapler with invaginant oversuture of the staple line with 3-0 PDS. After the gastric stapling, a wedge extramuscular resection of the gastric nodule in the incisura angularis of the excluded stomach was performed using eletrocautery and oversuture with 3-0 PDS. The antecolic gastrojejunal anastomosis was calibrated using a 36 French orogastric tube Fouchet (20 mm) in a two layers hand-sweing sutures with 3-0 PDS, and negative intraoperative leak test using methylene blue. The postoperative period was uneventful with intravenous hydration for 36 hours. The patient was dismissed from the hospital on the second postoperative day, after good acceptance of 50 milliliters liquid diet. The 18-days, 3 and 4-month medical appointments were with progressive weight loss, glucose levels and arterial blood pressure improvements, and no clinical complaints.

The specimen was sent to pathology. On the gross examination, a grey brown nodular, solid, smooth lesion partially covered with serosa, measuring 2 × 1.5 × 0.9 cm. A cut section revealed a whirling trabeculation, whitish, opaque and firm tissue. Microscopic examination showed fusocellular stromal tumor with mild atypias associated with lymphoid component. No mitotic activity and no necrosis were identified. The surgical margin was negative for tumor cells. Immunohistochemistry (IHC) staining was strongly positive for S100, whereas c-kit (CD117), CD45, smooth muscle actin (SMA), AE1/AE3 were negative. Hence, a final diagnosis of GS was made.

Figure 1 shows the results from histopatological and immunohistochemical pattern analysis.

Figure 2 shows the endoscopic aspect.

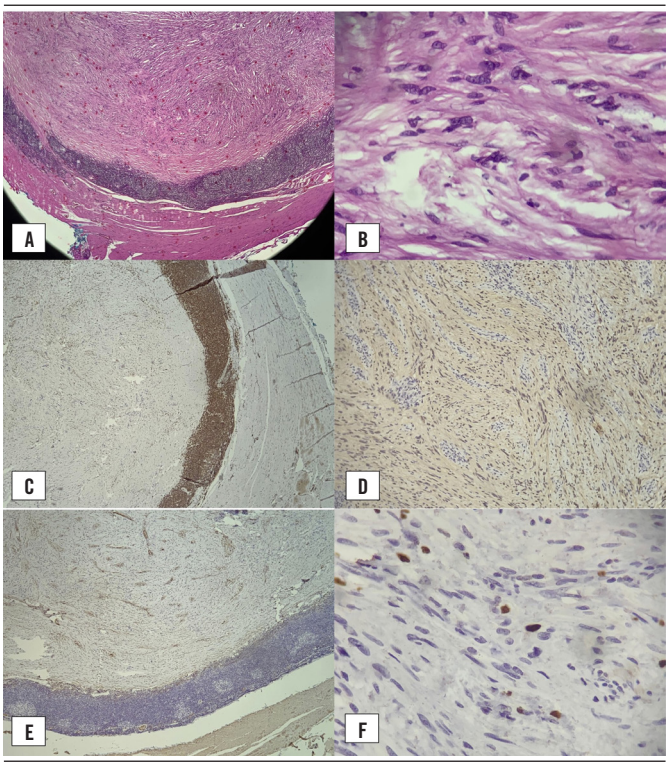

FIGURE 1 – *Histopathological and immunohistochemical pattern analysis*

A and B) shows microscopic examination with fusocellular stromal tumor with mild atypias associated with lymphoid component. No mitotic activity and no necrosis were identified; C) IHC staining and CD45 negative; D) strongly positive for S100; E) SMA negative; F) Ki-67 of 5%.

IHC: immunohistochemistry; SMA: smooth muscle actin.

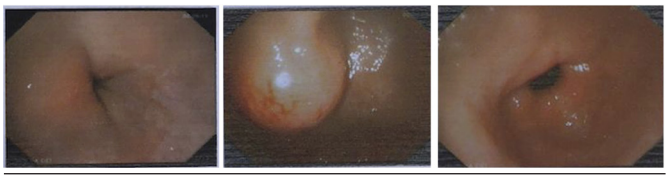

FIGURE 2 – *Endoscopic aspect*

A) ectopic gastric mucosa in the distal part esophagus; B and C) elevated and covered with normal mucosa with 20 mm in the anterior wall of the incisura angularis suggesting subepithelial lesion; mild antral gastritis.

Different researches were performed in PubMed and LILACS using terms related to "incidental findings; gastric bypass; stomach; bariatric surgery; gastrointestinal neoplasms; gastrointestinal stromal tumors; gastric schwannomas; mesenchymal tumors; GIST; leiomyomas; sleeve gastrectomy". The results were then carefully analyzed by the authors aiming mesenchymal gastric tumors and its incidence during a bariatric procedure. There was only one study that found GS during a bariatric procedure. This study was conducted according to the ethical guidelines for clinical studies of the Ministry of Health. The protocol was approved by the local ethics committee of Instituto Federal do Paraná (IFPR), number 30332020.0.0000.8156.

The patient was not directly asked because all the information was obtained from medical report, protecting the patient's anonymity and ensuring the confidentiality of the data collected.

DISCUSSION

Obesity

Obesity is defined as BMI ≥ 30 kg/m^2 and is a risk factor for some of the leading causes of death in the world. Besides clinical treatment, there are surgical methods to reduce weight with better long-term results. The main techniques are SG and RYGB. Each surgical technique has its advantages and the choice must be individualized. For example, patients with GERD have better results with RYGB, whereas obese patients benefit from the SG method,with lower incidences of severe malnutrition and dumping syndrome[2, 14-18].

In Brazil, 77.4% of bariatric surgeries are performed with health insurance assistance, 17.8% with the Public Health System [Sistema Único de Saúde (SUS)] and 4.8% with unknown resources. According to the Brazilian Society for Metabolic and Bariatric Surgery [Sociedade Brasileira de Cirurgia Bariátrica e Metabólica (SBCBM)], most bariatric surgeries provided by SUS are open surgery, mainly because of the cost and the learning curve[14-16, 18].

Schwann cells and schwannomas

Schwann cells are the central neuroglial cells of the peripheral nervous system and are responsible for the regeneration of the peripheral nerves after injury and sustaining the remyelination of central nervous system axons[19].

Schwannomas are benign encapsulated tumors that may occur in soft tissues, internal organs, or spinal nerve roots. They are characteristic lesions of familiar neurofibromatosis type 2 (NF2), when bilateral vestibular schwannomas occur. Tumors arising in the nerve root or the vestibular nerve (the most affected cranial nerve) may be associated with symptoms related to nerve root compression[20].

Gastrointestinal mesenchymal tumors

Mesenchymal tumors of the gastrointestinal tract have spindle cells, and include three types: GIST, SMA and nerve sheath tumor (mostly schwannomas)[11, 21].

GISTs and the interstitial cells of Cajal express CD117 (*c-kit* gene product), which is typically coexpressed with DOG1. These are used to differentiate GISTs from other spindle-cell neoplasms such as leiomyomas and schwannomas[22, 23]. Schwannomas preferentially express S100 but not CD117, DOG1, desmin, or SMA[23, 24]. Leiomyomas conversely express desmin and SMA but do not express CD117, DOG1, or S100[4].

GISTs represent most of the gastrointestinal mesenchymal tumors (1%-2% of gastrointestinal neoplasms)[23]. It is estimated that for every 45 cases of gastric GISTs there is one GS[7]. Complete resection of the tumors is necessary to determine the diagnosis. Surgical margins do not need to be clear, and in most cases is curative[25]. The importance of the correct diagnosis lies on GS having an excellent prognosis after surgical resection, while GIST can recur and potentially have a malignant outcome[6].

For GIST, it is important to monitor the possibility of metastasis in patients with high-risk categories [tumor size > 5 cm with > 5/50 high-power fields (HPF); tumor size > 10 cm with any mitotic count] with serial computed tomography (CT) scans every three months for five years[23]. The guidelines of the National Comprehensive and Cancer Network (NCCN) recommend an abdominal and pelvic CT scan every 3-6 months for 3-5 years, and annual postoperative follow-up[25].

GS

Gastrointestinal neurogenic tumors derive from different components of nerve fibers. Their known subtypes are schwannoma (91%), neurofibroma, and granular cell tumors[26, 27]. GS are defined as benign mesenchymal tumors that originate from schwann cells of the nerve sheath of Auerbach's or Meissner's plexus[6, 10, 22]. They are mostly found in the stomach typically involving submucosa/muscularis propria[11, 22], and represent 0.2% of all gastric tumors; 6.3% of gastric mesenchymal tumors; 4% of all benign gastric tumors[10-13, 28, 29] and were first described by Daimaru *et al*. in 1988[30].

It is suggested that the non-expressing monosomy on chromosome 22 and rare mutations in *NF2* can be present in the physiopathology of GS[7]. According to some authors, GS can rarely transform into malignant tumors, especially when they have a mitotic index higher than 10/50 HPF, presence of necrosis and nuclear atypia[6, 7, 28, 29, 31]. However, this idea of malignant transformation has been questioned by Voltaggio *et al*. (2012)[7], since most of the malignant transformation were reported prior to modern immunohistochemistry, which they probably corresponded to GISTs instead of schwannomas.

GS are most common in the fifth and eight decades, with predominance in females[6, 7, 12, 13, 21, 32]. These patients are usually asymptomatic. Symptomatic patients typically present abdominal pain or upper gastrointestinal bleeding[6, 8, 9, 13, 31-34]. Other rare symptoms are palpable abdominal mass (3.05%),

poor appetite (3.05%), dyspepsia (1.82%), weight loss (1.21%) and nausea or vomiting (0.6%)[6, 8, 11, 13, 21, 35, 36]. There is only one case of gastroduodenal intussusception due to GS reported in the literature[37].

In UGE, GS are usually solitary, firm, protruding submucosal masses[6, 13, 35, 36]. Tumors that lie within the muscular propria or are > 3 cm in size have a higher risk of perforation[38]. They can be indistinguishable from a GIST or a leiomyoma[35]. The main location is the body of the stomach (59.3%) with a variable size ranging 0.8 to 15.5 cm, while other locations of GS are the antrum (26.7%), fundus (12%) and cardia (2%)[6, 8, 36]. Endoscopic biopsies have a high rate of false negative results[6, 11, 39-41]. Some methods can increase the diagnostic accuracy of GS, such as fine needle aspiration biopsy (FNA) guided by endoscopic ultrasound (EUS) (50% to 85.2% for mesenchymal gastric tumors), with a hypoechoic appearance in the EUS[13, 39, 40, 42].

In the CT, GS have a homogeneous enhancement, exophytic or mixed growth pattern with cystic degenerations. Leiomyomas often show calcifications and leiomyosarcomas are usually more heterogeneous[43, 44]. The mean doubling time of schwannoma was nearly 5 years in CT images according to Choi *et al*. (2012)[40]. In magnetic resonance imaging (MRI), most GS are low to isointense on T1-weighted images and isointense to high intense on T2-weighted images[41]. However, the radiological findings in both CT and MRI of GS are not specific and can be misinterpreted as other mesenchymal tumors[13, 45]. Therefore, the final diagnosis is the IHC positivity for S100 protein[11, 37-39, 44, 45].

Positive staining for S100 protein and vimentin and negative staining for SMA, c-kit, and CD34 supports the idea that tumor is neurogenic[46].

The treatment can range from endoscopic approach (when the diagnosis of GS is made with endoscopic biopsy) to wedge resection, subtotal or total gastrectomy (when intraoperative diagnosis). Lymphadenectomy is not usually performed unless enlarged lymph nodes are seen, since GS rarely metastasizes to lymph node[6-8, 10].

The recurrence rate of GS is rare and only associated with positive surgical margins. Frequent CT follow-up is not recommended[6, 21] unless malignant GS (CT for at least five years)[8].

Gastric mesenchymal tumors and bariatric surgery

Crouthamel *et al*. (2015)[4] performed a research that correlated gastric mesenchymal tumors and bariatric surgery. In this study, the bariatric procedures were laparoscopic sleeve gastrectomy between 2009 and 2014. Of the 1415 sleeve gastrectomies, 205 cases were performed at a hospital where pathologic examination is routine and identified 17 incidental gastric submucosal tumors (1.2%), including 12 GISTs (0.8%), two schwannomas (0.1%) and three leiomyomas (0.3%). Patients with GIST had a mean age of 55 years ± 9.3 and tended to be older compared with the control group ($p = 0.0069$). There was no significant difference for BMI and gender between GIST and control ($p = 0.38$; $p = 0.72$). The authors affirm that a tumor located at the GE junction or lesser curve would require abandoning the propose SG in favor of an appropriate tumor excision. In this report, due to the GS location in the incisura angularis, a wedge resection was preferred for complete removal of the tumor, and a RYGB was performed considering the benefits of weight loss for the patient.

Sanchez *et al*. (2005)[3] retrospectively review 517 laparoscopic RYGB between 2002 and 2005 and found four cases intraoperatively of GIST (incidence of 0.8%).

This case report corroborates the inclusion of GS in differential diagnosis of mesenchymal gastric tumors for gastroenterologist, surgeons, endoscopists and pathologists. Obese patients with GISTs or carcinoid tumors can benefit from the SG with complete removal of the tumors[2, 14-17], but depending on the tumor's location, the SG cannot be performed. In this report, it is presented a surgical option to treat obese patients with gastric nodules during the bariatric procedure. Besides, the use of IHQ has a crucial role for the mesenchymal tumors differential diagnosis, since some may have malignant potencial.

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CORRESPONDING AUTHOR

Victor Kenzo Ivano ID 0000-0003-0160-2372
e-mail: victor.kenzo11@hotmail.com