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Synchronous Duodenal and Jejunal Gastrointestinal Stromal Tumors

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ABSTRACT

BACKGROUND

Gastrointestinal stromal tumor (GIST) constitutes up to 1% of all malignant neoplasms of the gastrointestinal tract, with the stomach and small intestine being the most frequently affected organs. The purpose of this report is to present the first published case of synchronous duodenal and jejunal GISTs.

CASE REPORT

A 57-years old male patient with type II diabetes complained of asthenia and vertigo, resulting from anemia. On upper digestive endoscopy, a friable lesion was found in the second portion of the duodenum. The biopsy diagnosed GIST and its removal was indicated. During the surgical procedure, a second tumor was found in the jejunum, which was also removed. Anatomopathological examination of this second lesion also revealed GIST with the same cytological features. Digestive reconstruction was performed in Roux-en-Y of the jejunum sectioned distally to the tumor with the open part of the duodenum. The patient evolved without complications, was discharged on the third postoperative day and has been followed up for eleven years without any other treatment and without neoplastic recurrence. Food transit occurs almost exclusively through the duodenum.

CONCLUSION

This is the first report of two synchronous GISTs of duodenum and jejunum. After removal of both tumors, the Roux-en-Y reconstruction adequately closed the opened duodenum, but food transit persisted physiological through the duodenum.

KEYWORDS

Gastrointestinal stromal tumor; Synchronous; Duodenum; Jejunum; Diagnosis; Roux-en-Y

INTRODUCTION

Although gastrointestinal stromal tumors (GIST) are rare, they are the most frequent mesenchymal tumors of the gastrointestinal tract, with an incidence of approximately ten per million people, which is 1% of all malignant neoplasms of the gastrointestinal tract [1-3]. The most common localization of GISTs is the stomach (50% - 60%), followed by the small intestine (30% - 40%), the colorectal (5% - 10%) and esophagus (<5%) [1,4,5]. Its nonspecific clinical picture, ranging from asymptomatic to

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severe abdominal pain and hemorrhage, may retard the diagnosis [1,6,7]. The best tests to detect this neoplasm are upper digestive endoscopy and computed tomography with oral and venous contrasts [1,7-9].

GISTs can range from small and benign nodules that are incidentally detected, to large and frankly malignant tumors. This tumor arises from the interstitial cells of Cajal and depends on the transcription factor ETV1. Several driver mutations have been identified in the development of GIST, with the most frequent being found in the tyrosine kinase receptor c-kit (c-KIT), present in 75% of GISTs, while 10% reveal mutations in the platelets, derived from growth factor receptor alpha (PDGFR-α). The diagnosis is based on the expression of CD117 (90% - 95%), a membrane-bound receptor of tyrosine kinase, and CD34 (70% - 80%), The remaining "wild-type" GISTs have a variety of other mutations and epimutations that may affect the SDH pathway [1,3,4,9,10].

The majority of GISTs occur as a single lesion in patients between 50 years and 60 years of age, with no association established with tumors of other histologies or other diseases except in patients with neurofibromatosis, Carney triad, and familial GIST, Carney-Stratakis syndrome [2,7,9,10]. The coincidence of synchronous occurrence of GISTs with other non-GIST malignancies has been described with the frequency of 5% to 38% of cases [2,3,5,6,8,9]. The most frequent tumors are in the stomach, colon and rectum (adenocarcinomas), breast (ductal carcinoma), kidney (clear cell carcinoma), prostate (adenocarcinoma), endometrium and ovary (adenocarcinomas), and adrenal (neuroblastoma). With the advancement of diagnostic techniques, the detection of multiple GISTs have also been described in 2% of gastric GIST [2,5,8,10].

No report of two synchronous GISTs in the intestine was found in the literature, therefore, this seems to be the first reported case of synchronous GIST of duodenum and jejunum.

CASE REPORT

A 57-years old black male with clinically controlled type II diabetes was referred to the surgery department due to asthenia, dizziness and anemia (hemoglobin concentration of 8.9 mmol/L) associated with the detection of occult blood in stool. Upper digestive endoscopy revealed a friable ulcer in the anterior mucosa of the second portion of the duodenum with bleeding to the touch. The biopsy indicated an active, ulcerated inflammatory process with fusocellular proliferation. Computed tomography of the abdomen revealed an accentuated anterior thickening in the second part of the duodenum, non-uniform and with contrast enhancement, narrowing the duodenal lumen and measuring 3.9 cm × 2.8 cm. There was no evidence of lymphadenopathy or other abdominal disorder. Surgical removal of this ulcerated tumor was indicated.

During the surgical procedure, the previously diagnosed duodenal tumor did not invade or present adhesions to other structures. Another exophytic jejunal tumor was found 18 cm from the duodenojejunal flexure, without invasion or adhesions to adjacent structures. Upon inventory of the abdominal cavity, no enlarged lymph node or other abnormality was found. The duodenal tumor was removed with three centimeters of free margins, preserving the posterior duodenal wall, including the two duodenal papillae. Four periduodenal lymph nodes were also removed. Then, the entire segment of jejunum that contained the tumor was removed, with seven centimeters of free margins proximally and distally, in monobloc with the mesojejunum, which contained five lymph nodes.

The distal stump of the jejunum was sutured terminolaterally to the duodenal opening wall, resulting from the removal of the first tumor. The proximal stump of the jejunum was sutured in a Roux-en-Y end to side 20 cm

distal to the first anastomosis. The patient progressed uneventfully, received food on the third day, and was then discharged from the hospital.

The histopathological and immunohistochemistry studies of the two tumors confirmed the GIST. The cellular characteristics of the two tumors were similar. Tumor-free resection margins were achieved in both GISTs. No neoplasm or other disorder was found in the lymph nodes sent for examination. The patient did not undergo adjuvant treatment and remained under follow-up for eleven years without complaints and in good general health. Computed tomographic follow-up showed that almost all the gastrointestinal (GI) transit occurred through the duodenum, without stenosis or other abnormality. Very little oral contrast followed through the isolated Y loop, despite the duodenojejunal anastomosis being wide.

DISCUSSION

Most of GISTs are incidentally detected during imaging studies, endoscopy or abdominal surgeries and may co-exist with other primary tumors that can involve either the GI tract or other extra-GI sites [1,5,6,7,11]. Multiple studies showed that patients with GISTs may develop synchronous or metachronous malignancies within the first year before or after the GIST diagnosis [7]. According to the current literature, the most common GIST-associated neoplasms are reported to be adenocarcinomas of the GI tract [2,3,4,9,10]. The highest incidence of GIST is located in the stomach, which is the most frequently associated with co-existing other malignancy of the abdomen [8].

This case report presents two synchronous GIST in the small bowel with similar histopathological and immunochemistry characteristics. The patient was a man with no family history and no clinical manifestations indicating GIST. His age is consistent with a mere coincidence, as most of GIST and non-GIST tumors occur. The genetic or triggering factors of GIST could not explain the location of these two similar tumors in the duodenum and jejunum, considering the prevalence of this neoplasm is in the stomach [1,5,6,8] (Figure 1).

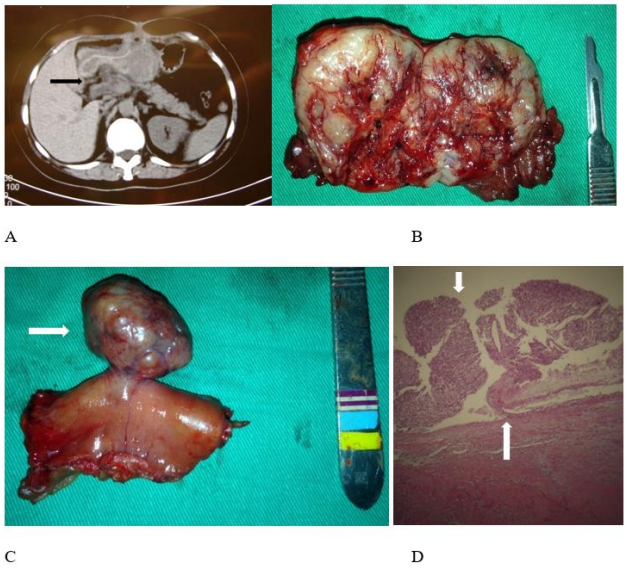

Figure 1: Images of synchronous duodenal and jejunal gastrointestinal stromal tumors (GIST). **A)** Computed tomography image of duodenal GIST (arrow). **B)** Duodenal GIST specimen after being removed and transversally opened. **C)** Exophytic jejunal GIST (arrow) specimen removed with large free margin. **D)** Microscopic view of the exophytic jejunal GIST (arrows), showing fusiform pattern in collagenized stroma, with the muscularis propria and without areas of necrosis (hematoxylin and eosin stain, 20X magnification).

It is worth to emphasize that surgeons must carefully investigate the abdomen during surgical procedures to treat a cancer, not only to stage it, but also to look for synchronous tumors or other disorders. In this case, no preoperative clinical or complementary propedeutics suggested the presence of two neoplasms. This study reinforces that the tomography pitfalls may occur in up to 20% of the exams, and it may not disclose even a large exophytic jejunal tumor. Clinicopathological characteristics and molecular analysis of the KIT/PDGFRA genes did not reveal any positive association between two synchronous GISTs in the small bowel [1,3,4,9,10].

Currently, the most indicated treatment for GIST is the surgical resection with tumor-free margin. GISTs should

be excised when incidentally discovered during surgery and if needed targeted therapy with TKI inhibitors (imatinib) is recommended for advanced or metastatic GISTs [1]. All surgeons must be aware of possible synchronicity, recognize the GI tumor and perform the proper treatment. As GISTs rarely (5%) metastasize to lymph nodes, routine extended regional lymph nodes dissection around the GIST is unnecessary. In this case, no metastasis was found in the resected lymph nodes and no recurrence or metastasis was found during the eleven-year follow-up [6,7].

The normal duodenal transit in this case must be emphasised, since a large duodenojejunal anastomosis was performed at the second part of duodenum. Since the early twentieth century, Cannon showed that even in presence of a large GI anastomosis the digestive transit occurs always through the physiologic way. The transit through an anastomotic shunt occurs only in presence of a GI obstruction [1,4,7]. Therefore, this case confirms the natural tendency to the physiologic GI transit.

CONCLUSION

This is the first report of two synchronous GISTs of duodenum and jejunum. After removal of both tumors, the Roux-en-Y reconstruction adequately closed the opened duodenum and food transit persisted physiological through the duodenum.

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DATA AVAILABILITY

The datasets generated during and analysed during the current study are available from the corresponding author on reasonable request.

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CONFLICT OF INTERESTS

The authors have no relevant financial or non-financial interests to disclose. The authors declare they have no competing or conflicting interests.

ETHICAL APPROVAL

This is an observational study. The Federal University of Minas Gerais, Brazil Research Ethics Committee has confirmed that no ethical approval is required. Ethical approval was not required, considering this paper reports the treatment performed on a patient with no research intention. Informed consent was obtained from the patient included in this report.

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