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Case Report

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Small Intestinal Glomus Tumor as an Uncommon Cause of Gastrointestinal Hemorrhage: A Case Report and Review of the Literature

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Abstract

Gastrointestinal glomus tumors are rare and almost always occur in the stomach. Up to now, only a very few cases have been reported in small intestine. Here, we present the ninth case of small intestinal glomus tumor in English literature. The 30-year-old female was referred to our hospital with chief complaints of melena and fatigue. Oral balloon-assisted enteroscopy revealed a 2.0*2.0 cm mass in the jejunum. Partial enterectomy was then performed and postoperative pathology reported a benign glomus tumor. No further melena was observed and also no malignant transformation or recurrence was detected after surgery. In conclusion, together with the literature review, small intestinal glomus tumor is an extremely rare cause of gastrointestinal hemorrhage. Early diagnosis and treatment are important to improve the prognosis since the potential risk of malignancy.

Keywords: Gastrointestinal hemorrhage • Benign • Glomus tumor • Small intestine

Introduction

Glomus Tumors (GTs) within the gastrointestinal tract have been reported to usually occur in the stomach, especially at the gastric antrum. The vast majority of gastric GTs follow a benign clinical course, without histologic or clinical evidence of malignancy [1,2]. GTs rarely involve in the small intestine, with only 8 cases reported in English literature, among which 4 cases present malignant features [3-10]. Here, we report the ninth case of small intestinal GT in a female adult. Additionally, we reviewed all the reported cases to summarize its clinicopathologic features, diagnosis and treatment.

Case Presentation

A 30-year-old female referred to our hospital with chief complains of melena and fatigue for 3 weeks. There were no significant positive signs other than anemic appearance (hemoglobin 6.7 g/dL). Routine endoscopy (gastroscopy and colonoscopy) and abdominal Computed Tomographic (CT) in fundamental hospital, revealed no significant findings. After admission, capsule endoscopy was immediately applied and indicated a possible mucosal eminence in the jejunum. Oral balloon-assisted enteroscopy was subsequently applied because the images captured by capsule endoscopy were limited. A 2.0 × 2.0 cm tumor without epithelial lining was discovered after insertion through pylorus about 250 cm (Figures 1A and 1B). Then abdominal enhanced CT was performed to further confirm the nature of the tumor. As shown in Figure 2, a solid tumor was observed in the upper-middle abdomen, which displayed homogeneous enhancement in the arterial phase and continuous enhancement in the delayed phase. No apparent invasion to the adjacent organs and lymphatic metastasis were observed from the CT scan (Figures 1A and 1B)(Figure 2).

The patient was then transferred to the Department of Gastrointestinal Surgery and underwent a partial enterectomy. A 3.5 cm long segment of jejunum was removed by surgical operation. Gross pathology showed a 2.0 × 1.3 × 1.3 cm gray-red polypoid protuberance in the surface of small intestinal mucosa. HE stain illustrated that the tumor was well limited by a thin fibrous capsule, interspersed with congestive capillaries of various sizes, and admixed with smooth muscle bundles. Muscularis propria and serosa didn't show any damage. Immunohistochemical study showed that the tumor cells were strongly positive for SMA and collagen type . And negative for CD31, CD34, CD117, Desmin, DOG-1, CgA, Syn, S100, EMA, STAT6, -catenin, Caldesmon and Calponin. Ki-67 proliferation index was less than 1%. And mitotic activity was about 1/50 HPF. The lesion was finally diagnosed as benign GT (Figures 3A-3F).

After surgery, the hemoglobin of this female patient recovered to a normal level soon and no further melena was observed. Due to half of small intestinal GT presenting malignant features, abdominal and pelvic CT reexaminations were performed every 6 months. No malignant transformation or recurrence was detected during a follow-up period of 12 months.

Results and Discussion

GTs arising from the small intestine is extremely rare. So far, only 8 cases have been described in English literatures [3-10]. Among them, two cases located at duodenum, one case at jejunum, and five cases at ileum. The clinicopathologic features of small intestinal GTs are summarized in Table 1. Similar to other small intestinal tumors, gastrointestinal hemorrhage is the most common symptom of small intestinal GTs. Also, half of the reported 8 cases presented abdominal pain. The mean age of onset was 55.1 ± 17.9 years (range 29-82 years). Together with the present case, no sex trend was revealed (5 males and 4 females) (Table 1).

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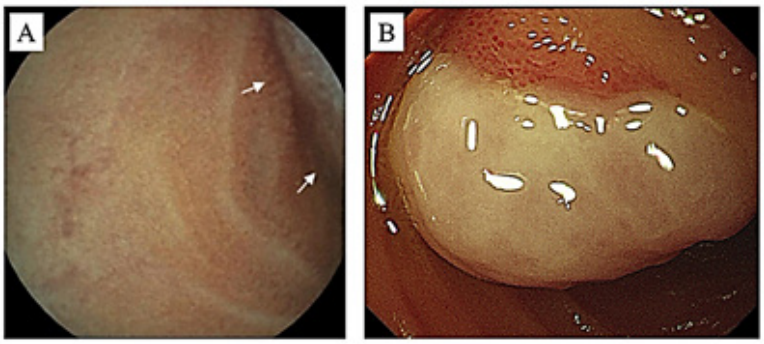

Figure 1. Endoscopic features of the GT. **Note:** A. Image of capsule endoscopy; B. Image of oral balloon-assisted enteroscopy

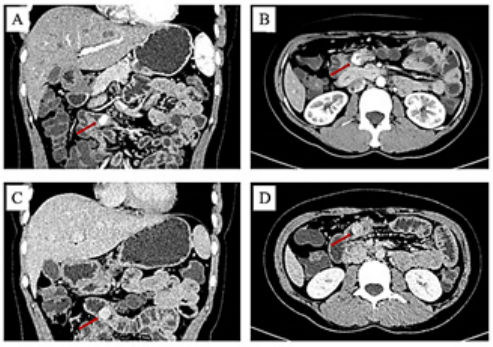

Figure 2. Features of the GT on abdominal enhanced CT. A and B. CT images during arterial phase (coronal and horizontal planes) showed an obvious enhanced nodule in the small intestine (arrow head); C and D. CT images during delayed phase (coronal and horizontal planes)

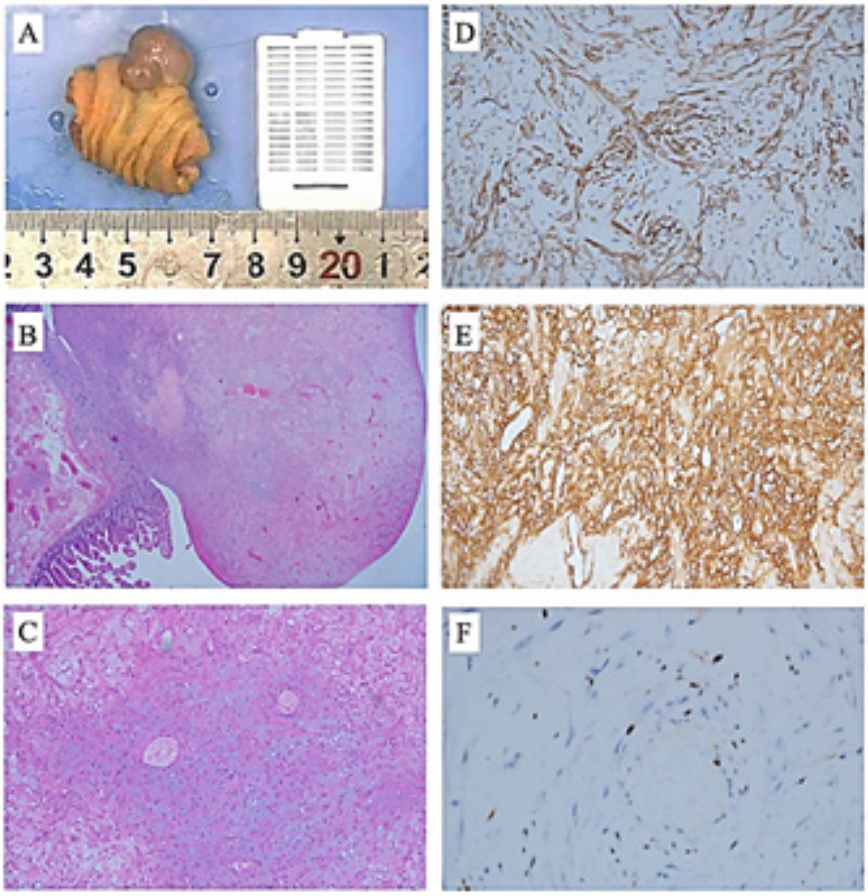

Figure 3. Gross findings, histological and immunohistochemical characteristics of the GT. A. Gross image of resected mass; B. H&E, X40; C. H&E, X 100; D. SMA immunostaining, X 200; E. Collagen type IV immunostaining, X 200; F. Ki-67 immunostaining, X 400

Table 1. Clinicopathological characteristics of documented small intestinal GTs

Reference	Age/Sex	Symptoms	Location	Size in cm	Invasion	Treatment	Immunohistochemistry	Nature	Outcome
Chen, et al [3]. 2020	73/F	Weakness	Ileum	2.0*2.8*1.2	Muscularis propria	Laparoscopic resection	SMA(+), vimentin(+), caldesmon(+), CD34(+), Ki-67(80%+), CD117(-), desmin(-), Dog-1(-), S100(-), CD45(-), cytokeratin(-)	Malignant	Metastases, Died
Ma, et al [4]. 2020	58/M	Abdominal pain, Hematocheza	Ileum	6.0*5.0	Serosa	Laparoscopic resection	SMA(+),Ki-67(70%+), CD34(+), Nestin(+), EMA(-), CD117(-), Dog-1(-), S100(-), desmin(-)	Malignant	Liver metastases
Campana, et al [5]. 2014	51/M	Melena	Ileum	3.7	Serosa	Laparoscopic resection	Ki-67(<5%+)	Benign	Follow-up
Abu-Zaid, et al [6]. 2013	29/F	Constipation, Vomiting, Melena	Ileum	12.8*10.2*13.1	Whole layer	Surgical resection	SMA(+), collagen type IV(+), caldesmon(+), calponin(+), CD117(-), CD34(-), S100(-), cytokeratin(-), desmin(-), HMB-45(-), chromogranin(-), synaptophysin(-)	Malignant	Follow-up
Knackstedt, et al [7]. 2007	65/M	Vomiting	Duodenum	Not available	Submucosa	ESD	SMA(-), CD117(-), CD56(-), CD34(-)	Benign	Follow-up
Shelton, et al [8]. 2007	48/F	Melena, Abdominal pain	Ampulla	3	Minimal invasion	Whipple	Not available	Malignant	Unknown
Geraghty, et al [9]. 1991	60/M	Abdominal pain, Diarrhea	Ileum	0.6	Serosa	Surgical resection	SMA(+), desmin(-), chromogranin(-), NSE(-)	Unknown	Die of unrelated causes
Hamilton, et al [10]. 1982	82/M	Abdominal pain, Anorexia, Nausea	Jejunum	1.0*1.5	Not available	Surgical resection	Not available	Unknown	Unknown

Due to the deep location and non-specific symptoms, small intestinal tumors are usually difficult to differentiate and diagnose. Abdominal enhanced CT or CT enterography is usually used to evaluate small intestinal lesions on imaging diagnosis. In the present case, a mass with homogeneous enhancement in the arterial phase and continuous enhancement in the delayed phase was found in the upper middle abdomen, which is similar to previous descriptions in gastric GTs [11]. GTs in stomach usually displays a submucosal pattern of growth. In the case reported by Knackstedt, et al. the GT located in the duodenal bulb was polyp-like [7]. Shelton, et al. reported a GT of the ampulla, in which a protruding mass was seen [8]. Although 6 cases had been reported, there is still no enteroscopic image presented for GTs located in jejunum or ileum. In this study, for the first time, we acquired the endoscopic imaging of small intestinal GT by balloon-assisted enteroscopy. The most significant feature of the small intestinal GT observed in enteroscopy is that the tumor is covered by a thin fibrous capsule, without epithelial lining.

Most GTs depends on the postoperative pathological diagnosis. Under the microscope, the tumor consists of a large number of smooth muscle bundles and dilated capillaries. Immunohistochemically, tumor cells were stained positive for SMA and collagen type . Negative CD117 and DOG-1 expressions may help in excluding the diagnosis of gastrointestinal stromal tumors, while negative CgA and Syn expressions help in excluding neuroendocrine neoplasms [12]. Criteria for malignant GTs proposed by Folpe, include tumors with a deep location and a size of more than 2 cm, or atypical mitotic figures, or moderate to high nuclear grade and ≥ 5 mitotic figures/50 HPF [13]. According to the criteria, the present case was considered as low possibility of malignancy. And 12 months follow-up showed no indication of recurrence or metastasis after a partial enterectomy, which supports the benign diagnosis.

All the small intestinal GTs recorded in the literature has received surgical resection, but no patients received regional lymphadenectomy. Also, no patients received adjuvant radiotherapy or chemotherapy after surgery. However, 2 patients developed metastases and one died soon after the metastasis [3,4]. Therefore, standardized systemic treatment needs to be further evaluated. For gastric GTs, endoscopic or laparoscopic resection is recommended. Also due to the fact of extremely rare cases of malignant GTs in the stomach, no standardized neoadjuvant or adjuvant treatment was suggested [14]. Radiotherapy or chemotherapy has been utilized for the treatment of malignant GTs of head and neck. However, in the reported cases, tumor progression was not reliably altered.

Conclusion

In conclusion, we have reported a case of small intestinal GT in a 30-year-old female. The final diagnosis is made by histological and immunohistochemical examination. Due to the exceedingly rare occurrence, there is no standardized management pathway for these patients. Since a potential for malignancy, early diagnosis and treatment are important for a better prognosis.

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