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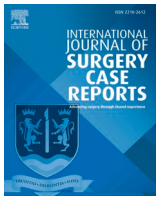

Case report

Bleeding ileal schwannoma resulting in severe anemia requiring massive blood transfusion: A rare case report

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ABSTRACT

Introduction: Melena is a common symptom of schwannoma of the small intestine, a rare type of tumor. Even more rare is schwannoma of the small intestine that requires a massive blood transfusion due to hemorrhage. Herein, we report such a case successfully treated with surgical resection.

Presentation of case: A 72-year-old woman presented to the previous hospital with melena. The patient was taking antiplatelet drugs for a previous cerebral infarction. The patient had progressive anemia due to continuous melena. Thus, she needed a massive blood transfusion with 12 units of packed red blood cells within 1 week of admission. A diagnosis was not possible based on the esophagogastroduodenoscopy and colonoscopy findings. Therefore, the patient was referred to our hospital for further examination and treatment. Computed tomography (CT) showed a well-circumscribed tumor with hyperattenuation in the small intestine, and double-balloon endoscopy (DBE) revealed a submucosal tumor (SMT) in the ileum. The patient was diagnosed with a bleeding gastrointestinal stromal tumor (GIST) and underwent laparoscopic partial resection of the ileum. The histopathological findings revealed spindle-shaped cell growth and a peritumoral lymphoid cuff. Furthermore, immunohistochemistry demonstrated that the tumor cells were negative for c-kit and CD34 but positive for S100 staining. Finally, the patient was diagnosed with ileal schwannoma. The postoperative course was uneventful, and the patient was discharged on postoperative day 10.

Conclusion: This report describes an extremely rare case of ileal schwannoma requiring massive blood transfusion. Furthermore, it highlights that schwannomas of the small intestine can cause severe anemia, especially in patients receiving antiplatelet drugs.

1. Introduction

Schwannoma of the small intestine is very rare, even among intra-abdominal mesenchymal tumors [1]. Melena is a relatively common symptom [2,3]. Most cases of intestinal schwannoma are identified by accident during imaging studies [4]. However, a few reports have described cases of intestinal schwannoma requiring massive blood transfusions due to acute bleeding from the tumor [5–7].

Herein, we report an extremely rare and serious case of ileal schwannoma in a 72-year-old woman which required a massive blood transfusion, and was treated successfully with surgical resection. This case report was prepared according to the SCARE Criteria [8].

2. Presentation of case

A 72-year-old woman presented to the previous hospital with melena. The patient had a history of hypertension and asymptomatic cerebral infarction. She also had a surgical history of open cholecystectomy for gallstones and open total hysterectomy for uterine fibroids. Furthermore, the patient was taking clopidogrel for a previous cerebral infarction. There was no family history of neurofibromatosis, and the patient's physical findings were unremarkable.

On admission, the laboratory findings revealed low hemoglobin (11.5 g/dL) and high blood urea nitrogen (34.5 mg/dL) levels. However, the coagulation parameters were normal. The serum levels of tumor markers, including carcinoembryonic antigen and carbohydrate antigen

Abbreviations: CT, computed tomography; GIST, gastrointestinal stromal tumor; DBE, double-balloon endoscopy; SMT, submucosal tumor; FDG, fluorodeoxyglucose.

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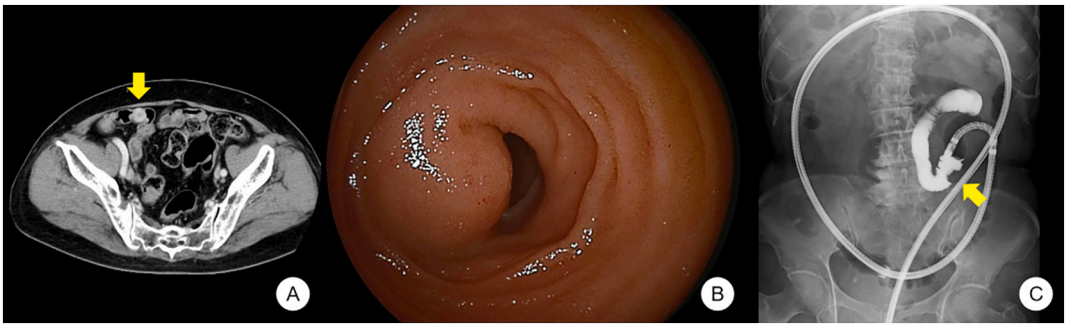

Fig. 1. Imaging findings. (A) Enhanced computed tomography shows a well-defined tumor with heterogeneous hyperattenuation in the small intestine (yellow arrow). (B) Double-balloon endoscopy shows a submucosal tumor in the ileum. (C) An ileal series on double-balloon endoscopy shows a filling defect corresponding to the tumor site (yellow arrow). (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

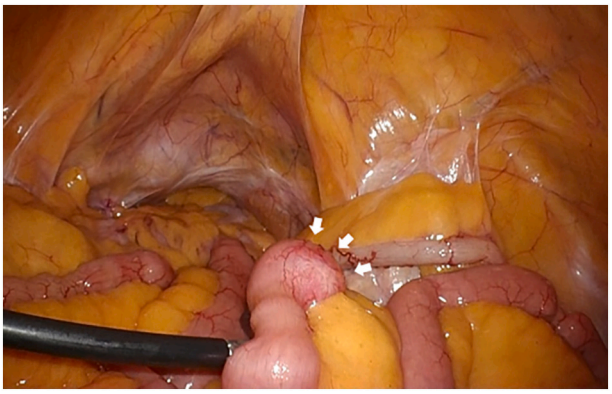

Fig. 2. Intraoperative findings reveal redness on the serosa of the ileum 90 cm from the ileocecal valve (white arrow), consistent with the tumor.

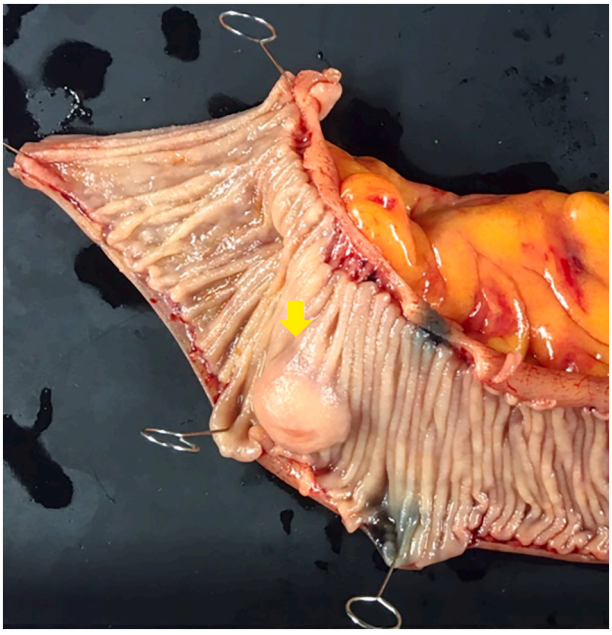

Fig. 3. Macroscopic findings identified a round 15-mm submucosal tumor (yellow arrow). (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

19–9, were within reference ranges. It was not possible to make a diagnosis based on the unremarkable esophagogastroduodenoscopy and colonoscopy findings. However, the patient had progressive anemia due to bleeding, suspected to be from the small intestine. On the third day of hospitalization, the patient's hemoglobin level decreased to 7.3 g/dL. The intestinal bleeding had continued for 1 week; therefore, she received a massive blood transfusion with 12 units of packed red blood cells. Following this, the patient was referred to our hospital for further examination and treatment. Computed tomography (CT) revealed a well-circumscribed tumor with heterogeneous hyperattenuation in the small intestine (Fig. 1A). Double-balloon endoscopy (DBE) revealed a submucosal tumor (SMT) in the ileum (Fig. 1B), and the ileal series showed filling defects corresponding to the tumor site (Fig. 1C). Therefore, the patient was diagnosed with a gastrointestinal stromal tumor (GIST) with bleeding and underwent laparoscopic partial resection of the ileum.

Intraoperative findings revealed erythema of the serosa of the ileum 90 cm from the ileocecal valve (Fig. 2). The macroscopic results showed a round 15-mm SMT (Fig. 3), and the histopathological examination revealed spindle-shaped cell growth and peritumoral lymphoid cuffs (Fig. 4A–C). Immunohistochemical analysis demonstrated that the tumor cells were negative for c-kit and CD34 but positive for S100 staining (Fig. 4D–F); therefore, the patient was diagnosed with ileal schwannoma. The postoperative course was uneventful, and the patient was discharged on postoperative day 10.

3. Discussion

Herein, we report a rare case of ileal schwannoma. Schwannomas are the most common type of peripheral nerve tumor, usually occurring in the peripheral nerves of the extremities or spinal cord [9,10]. Conversely, gastrointestinal schwannomas are rare, and mostly occur in the stomach [1,11]. Schwannomas of the small intestine are extremely rare, comprising only 0.5 % of intra-abdominal mesenchymal tumors [1]. Therefore, reports on schwannomas of the small intestine are limited.

Most schwannomas of the small intestine are found by accident during imaging studies [4]. Abdominal pain [12] and melena [2,3] are relatively common symptoms of schwannoma of the small intestine. Some develop into intussusception, although this is rare [13,14]. In our case, the patient had severe anemia associated with acute and continuous bleeding from an ileal schwannoma that required a massive blood transfusion. The patient was on treatment with clopidogrel for a previous cerebral infarction, which may have influenced the persistent bleeding. To our knowledge, few reports have described an intestinal schwannoma requiring a massive blood transfusion for acute bleeding from the tumor. However, interestingly, all patients in the existing reports were on antiplatelet drugs [5–7]. Therefore, we must remember that schwannomas of the small intestine can cause life-threatening

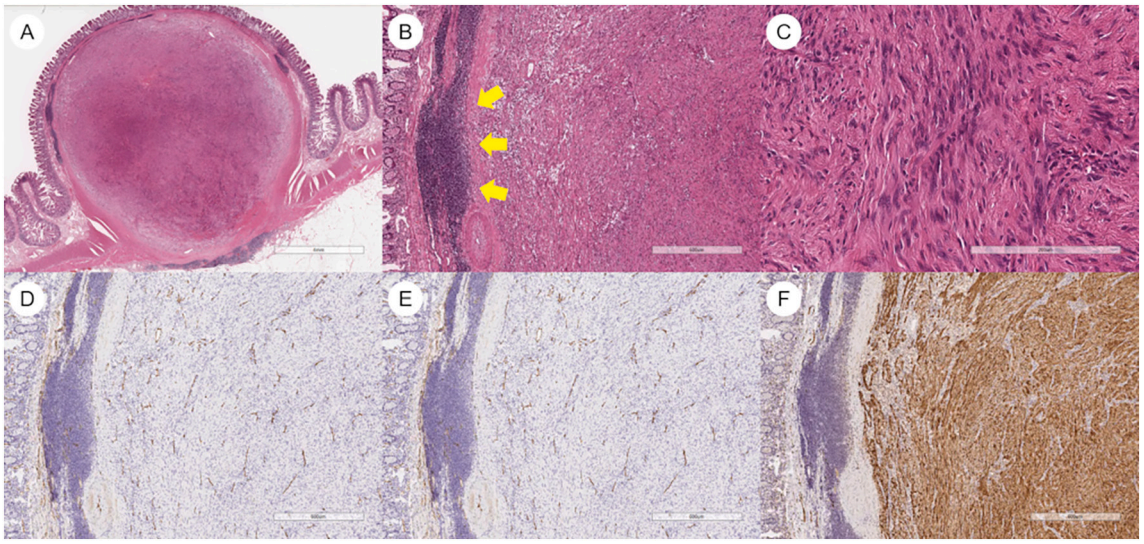

Fig. 4. Histopathological and immunohistochemical findings. (A) The histopathological findings revealed a well-defined submucosal tumor; hematoxylin and eosin staining (6×). (B, C) The tumor had a peritumoral lymphoid cuff, characteristic of intestinal schwannoma (yellow arrow), and spindle-shaped cell growth; hematoxylin and eosin staining: (B) 40× and (C) 200×. Immunohistochemical analysis revealed tumor cells with negative staining for (D) c-kit and (E) CD34, but positive staining for (F) S100 (all 40×). (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

anemia due to bleeding, especially in patients taking antiplatelet or anticoagulant drugs.

Preoperative diagnosis of intestinal schwannoma is challenging [1,4,15]. Goh et al. reported that no patients had been preoperatively diagnosed with schwannoma of the small intestine [1]. Furthermore, diagnosing schwannomas of the small intestine by imaging studies, including CT or magnetic resonance imaging, is not possible because their findings are extremely similar to those of other intestinal tumors, such as GIST or leiomyoma [1,4]. Fluorodeoxyglucose (FDG)-positron emission tomography is a powerful tool for detecting malignant tumors, but FDG accumulation has also been observed in benign schwannomas. Therefore, distinguishing between schwannoma and other malignant tumors, including malignant schwannoma, is almost impossible [9,10]. Diagnosis by endoscopy, including DBE, is also not possible because intestinal schwannoma can appear as a submucosal tumor with a nonspecific morphology [3]. Thus, surgical resection is essential for accurately diagnosing and treating intestinal schwannomas.

The prognosis of schwannoma of the small intestine is excellent [9]. To our knowledge, no cases of benign schwannoma of the small intestine, histopathologically diagnosed based on the surgically resected specimen, have recurred. Therefore, postoperative follow-up of patients who underwent complete resection of benign intestinal schwannomas is not required.

4. Conclusion

In conclusion, we report an extremely rare case of ileal schwannoma. This case highlights the fact that intestinal schwannoma can cause severe anemia, especially among patients taking antiplatelet or anticoagulant drugs. Furthermore, imaging studies are not useful diagnostic tools for schwannoma of the small intestine; therefore, surgical resection is essential for an accurate diagnosis and reliable treatment.

Ethical approval

This study was exempt from ethnical approval in our institution, Iwate Medical University.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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CRediT authorship contribution statement

Hideki Kumagai: Conceptualization, Writing – original draft, Data curation, Writing – review & editing, Visualization. **Mizunori Yaegashi:** Conceptualization, Writing – original draft, Writing – review & editing. **Misato Okutsu:** Writing – original draft, Visualization. **Kanki Otsuka:** Writing – original draft, Data curation. **Tomohiro Iwasa:** Writing – original draft, Data curation. **Akira Sasaki:** Conceptualization, Writing – review & editing, Supervision.

Declaration of competing interest

The authors declare they have no conflicts of interest.

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