Cavernous haemangioma of the duodenum with acute massive bleeding in the ascending portion: a case report

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Abstract
Duodenal cavernous haemangiomas are rare, benign disorders, and massive gastrointestinal (GI) bleeding is a rare clinical condition. The present case report describes a 50-year-old male patient who presented with severe, ongoing haematochezia. A peripheral blood smear at the time of admission showed significant anaemia, and haemoglobin level was 52 g/l (normal range, 120–175 g/l). Albumin level was also low at 28 g/l (normal range, 40–55 g/l). Standard computed tomography (CT) showed mural thickening and relative lumen stenosis in the ascending (fourth) portion of the duodenum. Contrast-enhanced CT using hypotonic solution revealed the lesions to be hypervascular haemangiomas. Laparotomy and segmental duodenum resection were performed, and the first jejunal limb was anastomosed using a side-to-end technique. Histopathological examination confirmed the diagnosis of cavernous haemangioma. The patient showed marked improvement during follow-up. The present case findings emphasize that duodenal haemangioma is possible without a history of chronic anaemia, and should remain a consideration in differential diagnosis for patients presenting with massive GI bleeding. CT is useful for preoperative diagnosis of massive bleeding, and surgery with segmental resection is usually curative.

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Introduction
Haemangiomas, characterized by disorganized angiogenesis, are an infrequent occurrence in the gastrointestinal (GI) tract.1,2 The most common clinical manifestation is chronic GI bleeding, leading to anaemia, but massive or life-threatening bleeding occurs in rare cases.3 Patients may also present with icterus if localization of the haemangioma is exceptional, such as near the ampulla of Vater.3 Based on the scope of affected vessels, haemangiomas are histologically divided into cavernous, capillary or mixed-type, with the cavernous type being the most common.4

Cavernous haemangiomas of the duodenum originate from the submucosal vascular plexus, and generally, the submucosa and muscular layer of the intestinal wall are involved. Endoscopy combined with computed tomography (CT) allows for better evaluation of lesions. Endoscopically, it is possible to visualize the mucosal oedema, nodules, and vascular congestion. CT can be used to differentiate duodenal subepithelial lesions.5,6 Here, a rare case of a cavernous haemangioma in the ascending (fourth) portion of the duodenum with massive bleeding is reported. CT and endoscopy were used to define the preoperative diagnosis. Duodenal resection with duodenoejejunostomy was performed, and histological examination confirmed cavernous haemangioma.

Case report
The study was approved by the ethics committee of the First Affiliated Hospital of Nanchang University. Written informed consent was obtained from the patient for publication of this case report and accompanying images.

A 50-year-old male patient was admitted to the First Affiliated Hospital of Nanchang University in August 2019, due to sudden onset of severe, ongoing haematochezia. He reported no haematemesis, no history of anaemia or surgery, and no family history of anaemia. Physical examination upon admission showed a body temperature of 36.4°C, heart rate of 92 beats per min, respiratory rate of 19 breaths per min, and blood pressure of 108/67 mmHg. The patient’s height was 171 cm, and weight was 60 kg. His conjunctiva was pale and his abdomen was flat and soft, without tenderness and rebound pain, but with active bowel sounds. Laboratory examination revealed significant anaemia with a haemoglobin level of 52 g/l (normal range, 120–175 g/l). In addition, his albumin level was decreased at 28 g/l (normal range, 40–55 g/l).

Standard abdominal CT was performed with a SOMATOM Definition Flash dual-source scanner (Siemens Healthcare, Forchheim, Germany) to investigate the causes of GI bleeding. The findings showed thickening of the mural wall and relatively narrow lumen in the fourth portion of the duodenum (Figure 1). After resuscitation, upper GI endoscopy revealed a dark red submucosal lesion occupying the fourth portion of the duodenal cavity, with mucous oedema in the raised edges and a variceal appearance in the centre, and with active spontaneous bleeding (Figure 2). Biopsy was not performed due to the high risk of bleeding. The lesion was further
investigated by contrast-enhanced CT using hypotonic contrast agent. The lesions were found to be hypervascular with a peripheral, progressive, and discontinuous contrast uptake in the arterial phase (Figure 3A). The uptake tended to be homogeneous in the portal phase (Figure 3B), and low-fissure or low-cobble density in the central area was observed as contrast faded in the delayed phase (Figure 3C). Therefore, the lesion was suspected to be a haemangioma. No other lesions or extramural extension of the tumour were observed, and no inflammatory exudation was observed in the mesenteric fat. There were no enlarged abdominal lymph nodes to suggest malignancy, and there was no involvement of adjacent organs (Figure 3). CT angiography did not detect the presence of thick blood vessels or a large vessel supplying the lesion (Figure 4).

Based on these comprehensive evaluations, duodenal haemangioma was preoperatively diagnosed as the most likely source of the bleeding. With conservative therapy, comprising 4 units of leukoreduced red blood cells and 1000 ml lactated Ringer’s solution by intravenous infusion, 1 unit of

Figure 1. Representative abdominal computed tomography images of a lesion in the fourth portion of the duodenum: (a) transverse plane, showing a mass-forming lesion (yellow arrow) in the fourth portion of the duodenum and thickening of the mural wall; (b) sagittal reconstruction showing similar findings (yellow arrow); and (c) coronal reconstruction showing the duodenal cavity stricture (yellow arrow).

Figure 2. Representative upper gastrointestinal endoscopy images showing a dark red, submucosal lesion filling the duodenal cavity, with oedematous mucous in the raised edges, a variceal appearance in the centre, and spots of active spontaneous bleeding (yellow arrows).
haemocoagulase Atrox by intravenous injection, and 1 unit of haemocoagulase Atrox by intramuscular injection, the haemoglobin level increased to 70 g/l, and there was no obvious ongoing bleeding. A laparotomy and segmental duodenum resection (partial 3rd portion and entire fourth portion of the duodenum) were performed, with side-to-end technique to anastomose the first jejunal limb. No other lesions were detected intraoperatively in the remaining GI tract.

Macroscopically, the specimen was bluish-purple in colour, with cystic nodules of varying sizes fused into a tumour, and the dissected capsule was filled with blood (Figure 5A). Microscopic examination of haematoxylin and eosin-stained resected tumour tissue sections revealed lumens of different sizes, shapes, and thicknesses, and the presence of disintegrated red blood cells. Histopathological examination confirmed the diagnosis of a cavernous haemangioma (Figure 5B and C). The patient’s condition improved during follow-up, and no melena relapse symptoms were reported during the follow-up period. No abnormalities were observed during conventional CT (Figure 6) performed at the last follow-up, at 14 months following treatment.

**Discussion**

Haemangioma is a benign endothelial tumour characterized by an increase in cellular proliferation, with the feature of slow involution and no recurrence. Accounting for approximately 0.3% of all GI tumours,
haemangioma usually occurs in young individuals (aged 5–25 years) and lesions may be present in childhood; however, most reported cases concern adult patients. Infantile haemangioma is usually accompanied by cutaneous haemangiomas and bluish-purple syndrome. Duodenal haemangioma has a very low incidence. Based on the scope of the affected vessels, haemangiomas are divided into three histological types: cavernous, capillary, or mixed, among which, cavernous haemangiomas are the most frequent. Between 1978 and 2008, only 21 cases of duodenal haemangioma were reported in the published literature, and of these, cavernous haemangiomas accounted for just over half (11 of 21 cases). Between 2008 and 2019, two cases of duodenal haemangiomas were reported, including one case of cavernous haemangioma.

Cavernous haemangiomas are soft and compressible, and consist of blood-filled spaces (caverns) between dilated vessels within the mucosa and submucosa. Morphologically, cavernous haemangiomas present as submucosal tumour-like, diffusely infiltrating, or polypoid lesions. The most common clinical manifestation is chronic GI bleeding, leading to anaemia; however, massive or life-threatening bleeding occurs in rare cases. Except for the present case, there have been no published reports of duodenal cavernous haemangioma with massive bleeding during the last decade.

Since there are no specific clinical symptoms, preoperative ancillary inspections are particularly important for differential diagnosis. In the present case, common diseases that cause GI bleeding were considered first, including portal hypertensive gastropathy, oesophageal varices, chronic peptic ulcer, Mallory-Weiss tear, malignant tumour, and haemobilia. These common diseases have some past medical history that can help establish a differential diagnosis.

Figure 4. Representative computed tomography angiography images showing the absence of any large blood vessels supplying the haemangioma lesion: (a) transverse plane; (b) sagittal plane; (c) coronal plane; and (d) three-dimensional reconstruction of the artery. Yellow arrows indicate duodenal lesions.
diagnosis. For example, most oesophageal varices are linked to a history of cirrhosis. Secondly, rare diseases were considered, such as Dieulafoy’s lesion, of which the symptoms, serology, endoscopy, and other auxiliary examinations have been described in detail previously. Endoscopic features of Dieulafoy’s lesion comprise ulcers, small red dots, polyps, dilated vessels, and round mucosal defects, whereas the endoscopic
features of haemangiomas are dark red sub-mucosal lesions, mucosal oedema bulge, and erosion, as observed in the present case. Endoscopy may be very helpful for differential diagnosis, and remains the primary diagnostic technique for duodenal mass-forming lesions, as it allows full visualization of the duodenal tract. It also allows the provision of therapeutic interventions, such as endoscopic haemostatic clipping, thereby halting disease progression while avoiding unnecessary transfusion.\textsuperscript{15} Endoscopy simultaneously combined with CT examination may improve the accuracy of diagnosis.\textsuperscript{16-18} Using contrast-enhanced CT, cavernous haemangiomas present typical imaging features, as described within the present case report.

Endoscopic haemostatic techniques can be used to treat active bleeding sites,\textsuperscript{15} and several approaches have been reported, including endoscopic sclerotherapy, endo-loop snare,\textsuperscript{20} polypectomy, and endoscopic mucosal resection. However, these procedures are usually ineffective in the context of massive haemorrhage, diffused cavernous haemangiomas, or recurrent episodes of bleeding.\textsuperscript{21} In the present case of diffused cavernous haemangioma, the optimal treatment was surgical resection, thus, laparotomy and surgical resection of the segmental duodenum were performed, taking into account the exceptional location and large size of the haemangioma. The perioperative period was safe, and the clinical outcome was extremely favourable after 14 months of follow-up.

Conclusions

Cavernous haemangiomas in the fourth portion of the duodenum are rare causes of massive GI bleeding. Even without a history of chronic anaemia, duodenal haemangioma is a possibility, and should be considered as a differential diagnosis for patients presenting with massive GI bleeding. Endoscopy and CT are useful techniques to define preoperative diagnosis, and surgery with segmental resection usually results in a cure.

Author contributions

Lei X performed the surgery; Lei X and Liang YH structured the article and wrote the manuscript; Tan YM performed a diagnostic CT on the patient; and Huang ZX collected the clinicopathological data. All authors read and approved the final manuscript.

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Declaration of conflicting interest

The authors declare that there is no conflict of interest.

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