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Case Report

Duodenal Dieulafoy lesion: a rare pathogeny of gastrointestinal bleeding in children

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ABSTRACT

Background. Dieulafoy lesion is a calibre persistent submucosal artery associated with a minuter mucosal defect. Dieulafoy lesion has been reported to account for 1-5.8% of acute nonvariceal upper gastrointestinal bleeding in adults, but it is rarely reported in children.

Case. Here we report a case of duodenal Dieulafoy lesion in a 13-year-old boy. After endoscopy and laparotomy, he still had no definite diagnosis and effective treatment. The duodenal Dieulafoy lesion was finally identified by selective angiography and was effectively treated by intravascular embolization.

Conclusions. For unexplained upper gastrointestinal bleeding, the possibility of duodenal Dieulafoy lesion should be considered. A combination of multiple diagnosis and treatment methods can improve the success rate of diagnosis and treatment when a single test or treatment method cannot provide definitive diagnosis or effective treatment.

Key words: duodenal, Dieulafoy lesion, gastrointestinal bleeding, children.

Dieulafoy lesion (DL) is a calibre persistent submucosal artery associated with a miniature mucosal defect, which was first described by Paul Georges Dieulafoy in 1897.[1] Most of DL is located in the stomach, within 6 cm of the gastro-esophageal junction, and rarely occur further along the gastrointestinal tract including duodenum, jejunum, ileum, cecum, appendix, colon and anal canal.[2] DL in children is rarely reported which is difficult to timely diagnose and can be fatal without appropriate treatment. Here we report a case of DL in a 13-year-old boy. Through the diagnosis and treatment of this case, we hope to provide experience for gastrointestinal bleeding of unknown pathogeny in children.

Case Report

A 13-year-old boy was admitted to pediatric surgery clinic for hematochezia and dizziness. The patient had been hospitalized in another hospital with the same symptoms 3 months prior. No definite bleeding was found on abdominal contrast-enhanced computerized tomography (CT), and no abnormal lesions in the esophagus, stomach and duodenum were found by gastroscopy. The patient denied any surgical history and was not taking any medication. The family history was negative for colorectal cancer, *Helicobacter pylori* infection and intestinal polyps.

Physical examination showed hypotension (blood pressure 92/50 mmHg), pallor and normally capillary filling test. The initial laboratory testing showed severe anemia (white blood cell count 7.6×10^9/L, hemoglobin 5.6 g/dL, hematocrit 18.5%, platelet count 322×10^9/L). Coagulation tests showed normal liver function, kidney function, cardiac enzymes and electrolytes were normal.

On the first day of hospitalization, the treatment regimen of blood transfusion (10 units of packed red blood cells) and application of hemostatic

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drugs were adopted, but the laboratory testing still showed severe anemia (white blood cell count 8.9×10^9/L, hemoglobin 6.5 g/dL, hematocrit 21.0%, platelet count 255×10^9/L). The first endoscopy revealed no abnormalities in the esophagus and stomach, but showed suspicious duodenal polyp without active bleeding (Fig. 1). Because the patient had always presented with black tarry stool, we considered that it might be small intestinal bleeding, so colonoscopy was not given priority. The exploratory laparotomy was taken immediately to identify the reason of hematochezia. No malformations of intestines and blood vessels were found during the operation. No jejunum abnormality was detected by enteroscopy. However, there are many convex nodules and fresh blood on the wall of the distal ileum. The distal ileum was resected, but histopathological analysis showed no abnormalities such as granuloma. On the fifth day after surgery, the patient had hematochezia again.

During the second endoscopy, we found a suspected actively bleeding protruding vessel at the suspicious duodenal polyp found in the first endoscopy and fresh blood in the stomach and duodenum (Fig. 1). Endoscopic ligation failed due to rapid bleeding and poor visual field exposure.

We urgently performed selective angiography of the celiac trunk and mesenteric artery. The angiography of each branch of celiac trunk

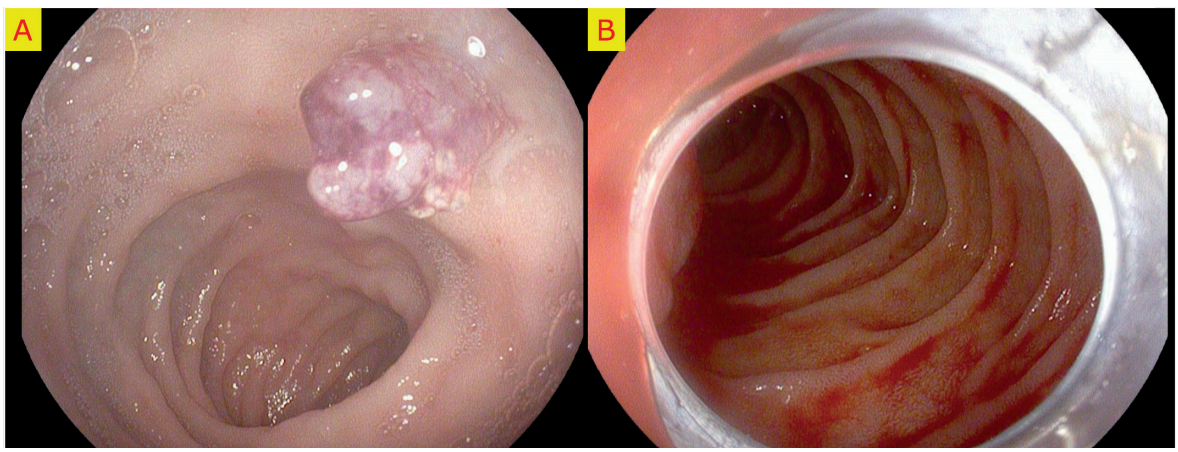

Fig. 1. Endoscopic features. (A) The first endoscopy showed suspicious duodenal polyp without active bleeding. (B) The second endoscopy showed fresh blood in the duodenum.

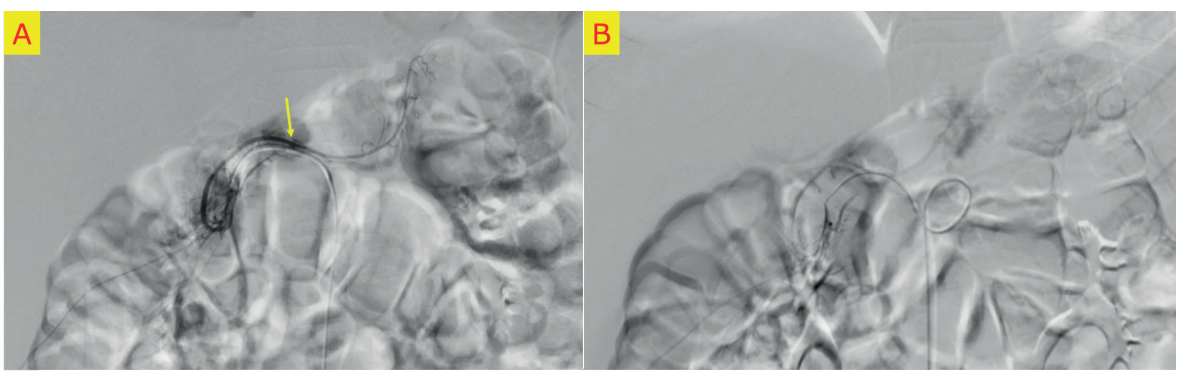

Fig. 2. Image of the selective angiography. (A) The selective angiography showed that the distal branch of gastroduodenal artery had a constant diameter, and there was a leak of contrast agent. (B) Distal embolization of the bleeding branch was performed utilizing gelatin sponge particles, and post-embolization arteriograms showed complete cessation of bleeding.

artery was performed successively. The distal branch of gastroduodenal artery had a constant diameter, and there was a leak of contrast agent (Fig. 2). The diagnosis of duodenal Dieulafoy lesion (DL) was confirmed. Distal embolization of the bleeding branch was performed utilizing gelatin sponge particles (700-1000 μm, 1000-1400 μm, 1400-2000 μm, Hangzhou Alicon Pharmaceutical Co., Ltd), and post-embolization arteriograms showed complete cessation of bleeding (Fig. 2).

The patient recovered steadily and was discharged 10 days after vascular embolization. No further hematochezia occurred after 10 months of follow-up. Informed consent was obtained from his parents for publication and photographs.

Discussion

Pediatric DL is rarely reported.[3] Hematochezia in children is usually caused by intestinal malformation such as Meckel's diverticulum, intestinal duplication. The reason for DL is often overlooked by pediatricians. In this case we report, although suspicious lesions in the duodenal bulb were found under the first endoscopy, the possible presence of duodenal DL was ignored, which was a profound lesson for us.

There were many deficiencies in the diagnosis of this case. Firstly, we had insufficient understanding of the pathogenies of gastrointestinal bleeding in children, especially for some rare pathogenies. Secondly, for unexplained gastrointestinal bleeding, it was necessary to perform enhanced CT or conventional angiography after endoscopy was negative, even if these tests might be negative during the bleeding interval of DL. Because the enhanced CT examination of the patient was negative 3 months ago, we ignored the necessity of reexamination of enhanced CT or conventional angiography.

The diagnosis and treatment of DL mainly include endoscopy, selective angiography, and surgery. Endoscopy is the preferred treatment for DL, and the success rate can reach 90%.[4] Endoscopic injection, hemostatic clamping, ligation, and electrocoagulation are all common treatments, but there are still risks of hemostatic failure or bleeding recurrence.[5,6] Due to rapid bleeding and poor visual field, hemostasis under endoscopy failed in our case. Selective angiography can be used as a second-line treatment for DL.[7] It is suitable for patients who have failed endoscopic treatment and cannot tolerate the surgery. Interventional vascular embolization can achieve precise treatment of bleeding, but it also has risks of gastrointestinal necrosis and bleeding recurrence, etc. Surgery used to be the preferred treatment for DL, and surgical methods included electrocoagulation, suture hemostasis, and subtotal gastrectomy, etc. Although electrocoagulation and suture hemostasis were simple, the risk of postoperative recurrence was high. With the continuous development of endoscopic and interventional techniques, surgery is mainly used as the choice after endoscopy or interventional treatment fails, while laparoscopic exploration can be used as the preferred surgical method.[8,9] The surgical exploration of this case failed, which was a profound lesson for us. We hope to provide experiences for other researchers with the solution of the appropriate problem and the addition of the lessons learned.

We searched previous literature with "duodenum", "Dieulafoy" and "children" as keywords, and a total of 9 cases related to the diagnosis and treatment of DL were retrieved (Table I).[10-18] 3 cases were successfully diagnosed and effectively treated under endoscopy, 1 case was successfully treated with duodenotomy, 1 case died 4 hours after surgery, 4 cases were successfully treated with other methods after failed diagnosis or treatment under endoscopy (angiographic embolization in 2 cases, exploratory laparotomy in 2 cases). The case we reported was effectively diagnosed and treated after receiving endoscopy, surgical exploration, endoscopy, and selective angiography, and has been followed up for 10 months without recurrence.

Table I. Case reports of duodenum Dieulafoy lesion (DL) in children.

Authors	Age (sex)	Diagnostic tool	Treatment measures	Follow-up	Recurrence
Komissarov I A, et al.[10]	13 m (M)	Endoscopy; Selective angiography	3 coils of Trufill	5 y	No
Alomari A I, et al.[11]	14 y (F)	Endoscopy; Selective angiography	50% NBCA glue	8 m	No
McClave SA, et al.[12]	16 y (M)	Endoscopy; Laparotomy	A "figure eight" suture	3 y	No
Bilal M, et al.[13]	18 y (M)	Esophagogastroduodenoscopy	Band ligation	6 m	No
Rao S, et al.[14]	3 y (M)	Colonoscopy; Endoscopy; Duodenotomy	Duodenotomy.	_	_
Akira Hokama, et al.[15]	10 y (M)	Endoscopy	MD-850 hemoclips	1 y	No
Shi SJ, et al.[16]	9 y (M)	Laparotomy	Gastroduodenectomy	3 y	No
Wang CL, et al.[17]	11 m (F)	Endoscopy; Laparotomy	Ligasure	_	_
Jadhav DV, et al.[18]	1 m (M)	Endoscopy	Epinephrine and sporadic argon plasma coagulation	1 w	No

Because duodenal DL is clinically rare, the possibility of duodenal DL should be considered for unexplained upper gastrointestinal bleeding.[19] Suspicious gastrointestinal lesions found by endoscopy without bleeding should also be actively treated, which may be the bleeding interval of DL or submucosal thrombosis. A combination of multiple diagnosis and treatment methods can improve the success rate of diagnosis and treatment when a single test or treatment method cannot provide definitive diagnosis or effective treatment, and angiography should be tried primarily before laparotomy.

Ethical approval

This research is supported by the Ethics Committee of Shandong University Qilu Hospital (Qingdao) [KYLL-KS-2021030].

Author contribution

The authors confirm contribution to the paper as follows: study conception and design: LC; data collection: LC, LX; analysis and interpretation of results: LC, ZL; draft manuscript preparation: LC. All authors reviewed the results and approved the final version of the manuscript.

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Conflict of interest

The authors declare that there is no conflict of interest.

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