

Dieulafoy Gastric Lesion: An Unusual Cause of Pediatric Hematemesis

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Abstract

Dieulafoy lesion is a vascular anomaly predominantly found in the stomach, which represents a rare cause of gastrointestinal bleeding and life-threatening haemorrhages in pediatric age with few cases reported in the literature. We experienced a 7-year-old previously healthy boy with hematemesis endoscopically diagnosed and successfully treated for a Dieulafoy lesion in the stomach. This case report illustrates the initial diagnostic workup and the possible differential diagnosis in presence of an acute episode of hematemesis in children. It focuses on an uncommon cause of gastrointestinal bleeding that is probably underestimated because of missing diagnosis. Any unusual and acute case of upper gastrointestinal bleeding should raise the suspicion of Dieulafoy lesion also in children, especially in those who have a past medical history negative for peptic disease and varices due to portal hypertension: a promptly endoscopy can provide visual diagnostic criteria and ensure an adequate hemostasis that is generally the definitive treatment of the lesion.

Keywords

Dieulafoy Lesion, Hematemesis, Pediatric Endoscopy

1. Introduction

Dieulafoy lesion (DL) is characterized by an abnormally large submucosal caliber-persistent arterial that protrudes through a small (2 - 5 mm) mucosal defect in the gastrointestinal tract. No surrounding mucosal ulcerations are generally found. Although a congenital origin of the lesion is supposed, it is found more often in adult

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patients and it is rare to find it in pediatric ones. About three quarters of all DLs are in the stomach and isolated lesions are mainly reported. DL is also described in other gastrointestinal sites such as the esophagus, duodenum, ileum, jejunum, colon, anal canal and rectum. Clinical presentation varies with the location of the lesion and the type of bleeding (massive or intermittent). It includes hematemesis, hematochezia, melena, a combination of them or occult anemia [1]. Endoscopy is the standard approach for the diagnosis and treatment of gastric DL although angiography and capsule endoscopy can be reserved for the diagnosis of occult lesions. The three main endoscopic treatment options include coagulation by several means, local injection of vasoactive or sclerosing agents and mechanical binding. Surgery is reserved for the 4% - 8% of cases in which endoscopic haemostasis failed or for rebleeding lesions. We report a pediatric case of hematemesis caused by gastric DL, promptly treated by endoscopy.

2. Case Presentation

A 7-year-old healthy boy was admitted to the emergency department of our hospital because of five episodes of hematemesis within the same morning. His past medical history was unremarkable and he was on no regular medication. The family history was negative for peptic ulcer disease or bleeding disorders. On physical examination the patient was alert but pale and complained fatigue. Pulse was 110 beats/min and blood pressure was 106/56 mmHg. Abdominal examination revealed a moderate epigastric tenderness without any other remarkable findings. Nasogastric tube aspirate contained 400 ml of fresh blood. The hemoglobin level was 10.5 g/dl and the initial hematocrit was 22.6% with otherwise normal blood chemistry and clotting profile. After moderate volume crystalloid resuscitation, the child received an intravenous dose of H₂ receptor antagonist therapy. An urgent esophagogastroduodenoscopy was performed under general anesthesia and revealed moderate clotted and fresh blood in the gastric fundus and in the second-third portion of the duodenum respectively. A visible vessel with pulsatile bleeding and protruding from a normal mucosal surface was identified on the lesser curvature of the stomach at the angular portion (**Figure 1**). The origin of bleeding endoscopically resembled a DL. Hemostasis was successfully and quickly achieved by local injection of epinephrine (1 ml, 1:10,000 dilution). Finally, an endoscopic clip was positioned (**Figure 2**). The child's hemodynamic status stabilized with progressively im-



Figure 1. Endoscopy image of Dieulafoy lesion. A visible vessel with pulsatile bleeding and protruding from a normal mucosal surface on the lesser curvature of the stomach at the angular portion.



Figure 2. Hemoclipping of Dieulafoy lesion. Endoscopic clip was positioned after local injection of epinephrine.

provement of hemoglobin and hematocrit levels and no blood transfusion was necessary. After the procedure, an omeprazole therapy was temporarily administered for 1 week. An ultrasound and MRI angiographic studies of the abdomen revealed no other vascular anomalies. Refeeding was uncomplicated and, 11 days after endoscopy, the child was discharged from the hospital receiving iron and folic acid therapy. After an 8-month follow-up, no further evidence of gastrointestinal rebleeding has been reported yet.

3. Discussion

Life threatening gastrointestinal hemorrhage in children is uncommon and it is usually related to mucosal erosive disease or variceal bleeding secondary to portal hypertension [2]. Therefore, like the previous case, a past medical history negative for gastrointestinal bleeding disorders must give rise to the suspicion of an uncommon diagnosis of hematemesis.

Vascular anomalies are rare causes of gastrointestinal bleeding and often represent a diagnostic and therapeutic challenge for the clinician, especially in children. DL accounts for 0.5% to 14% of upper GI bleeding in adults and is extremely rare in children [3], as only 26 cases are currently reported worldwide [1] [4] [5]. Therefore the peculiarity of this report consists in the immediately diagnosis of a rare condition. The proximal stomach, especially the lesser curve, is the most common localization (75% - 95%) of these lesions, similarly to the reported case. This may be explained by the fact that submucosal arterial branches in that region arise directly from the left gastric artery [3]. However, DL has also been described in duodenum, colon, small bowel and in nongastrointestinal sites such as the bronchus [1].

Endoscopy is the method of choice for diagnosis of gastro-duodenal and colon lesions. The endoscopic visual criteria for the diagnosis include a) active arterial spurting or oozing from a small (<3 mm) defect in the mucosa, b) visualization of a vessel protruding from a slight defect or normal mucosa, and/or c) a fresh blood clot adherent to a defect of normal mucosa as described by Dieulafoy [1]. A prompt diagnosis of DL performed by a well-experienced endoscopist is fundamental for an adequate management of a possible life-treatment haemorrhage like in the present case. Multiple endoscopies may be required for diagnosis in case of small size lesions, inaccessible location, presence of large amount of blood in the stomach/bowel and intermittent bleeding. For these reasons the prevalence of DL can be underestimated also in pediatric age and the diagnosis can be a challenge even for the more experienced endoscopists. In difficult cases other diagnostic tools can be indicated. Capsule endoscopy can be helpful for small bowel lesions. However, it is not suitable in case of emergency and pediatric data are limited. On the contrary, angiography is a useful diagnostic and therapeutic technique, especially for extra gastric DL [6].

Endoscopy is also the first-line method of treatment of DL, while surgery or embolization is nowadays reserved for endoscopic failures or inaccessible sites. In order to achieve an adequate hemostasis, different endoscopic techniques can be used, such as sclerotherapy, argon laser, elastic band ligation and injection of acrylic resins and hemoclipping. Moreover, the injection of a millesimal solution of epinephrine determines a reduction in bleeding and facilitates definite therapy [7]. Endoscopic treatment is successful in 95% of cases [8] with an excellent prognosis, as demonstrated in this case.

Nevertheless, the published literature on treated DL in children is scarce and there is no general consensus in treatment and follow-up protocols.

In conclusion, although DL is a rare cause of hematemesis in children, pediatricians and pediatric endoscopists have to be aware of this challenging diagnosis because timing of endoscopy and adequate hemostasis are crucial for a favorable outcome and definitive treatment of the lesion.

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