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CASE REPORT

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An unusual cause for massive upper gastrointestinal bleeding in children: Dieulafoy's lesion

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Abstract Dieulafoy's lesion is a rare cause of severe upper gastrointestinal hemorrhage in children and predominantly occurs in the proximal stomach. We report a case of massive upper gastrointestinal bleeding in a 3-year-old boy that originated from a Dieulafoy's lesion and was treated by epinephrine injection.

Keywords Child · Dieulafoy's lesion · Upper gastrointestinal hemorrhage · Treatment

Introduction

Massive upper gastrointestinal hemorrhage in children is uncommon and is usually caused by esophageal varices secondary to sclerotic liver disease. Dieulafoy's lesion has been recognized as an important cause of obscure upper gastrointestinal hemorrhage. It is especially rare in children. This lesion is characterized by a minute mucosal defect with a large, tortuous artery at the base of the defect, causing rupture of the artery with potentially life-threatening bleeding [1]. In this study, we report a rare case of massive upper gastrointestinal hemorrhage due to Dieulafoy's lesion in a child.

Case report

A 3-year-old boy was admitted to our pediatric emergency department with a 1-day history of bloody vomiting, epigastric pain, and lethargy. There was no history of melena or regular medication, and his past medical history was unremarkable. During examination the boy was noted to be pale, with tachycardia and hypotension. Other systemic examination findings were normal.

His initial hemoglobin level was 8.6 g/dl, hematocrit was 25%, and platelet count was 210,000/mm^3. Serum electrolytes, transaminases, bilirubin, and amylase levels were normal. Clotting studies and serum fibrinogen level were within the normal range. However, serum urea (42 mg/dl) and creatinine (2.5 g/dl) levels were increased at the initial evaluation.

After transfusions of two units of packed red blood cells, esophagogastroduodenoscopy was performed under sedation. The esophagus and gastroesophageal junction were normal. A large amount of clotted and fresh blood was found in the stomach, but a bleeding solitary artery was identified on the corpus of the stomach approximately 4–5 cm from the cardia. The vessel, approximately 2 mm in diameter, was protruding from a base of mucosal ulceration (Fig. 1). Hemostasis was achieved by injecting a total of 5 ml of epinephrine (1:10,000) into the base of the vessel. The hemoglobin level and hematocrit stabilized after the procedure, and the patient was discharged home 3 days after endoscopy with proton pump inhibitor therapy. No further bleeding episodes occurred during 6 months of follow-up.

Discussion

Dieulafoy's lesion accounts for approximately 2% of acute upper gastrointestinal bleeding [2]. This lesion was first described in 1897 by Dieulafoy, a French surgeon, and is characterized by an artery protruding from the base of a shallow mucosal ulceration 2–5 mm in diameter. As in our case, the artery is located in the submucosal area and is usually large and tortuous [1]. It is usually found in adults and is twice as common in men as in women [3]. Dieulafoy's lesions are rarely described in the English literature in children less than 15 years of

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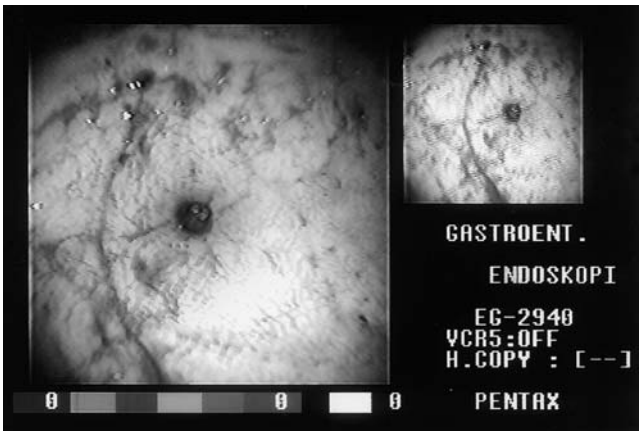

Fig. 1 Endoscopic view of Dieulafoy's lesion in the proximal stomach

age [4, 5]. So far, the youngest reported patient to suffer from Dieulafoy's lesion was 1 year old [6].

These patients present with massive hematemesis but without abdominal pain or other relevant history. During upper endoscopy, a bleeding or clot-bearing artery is seen protruding into the gastric lumen with surrounding normal mucosa [7]. Dieulafoy's lesion is often found in the proximal stomach, within 6 cm of the gastroesophageal junction in more than 80% of cases [3]. However, these lesions have also been noted in the duodenum [8], jejunum [9], colon [10], and esophagus [11]. Histologically, this lesion is a tiny submucosal defect with fibrinoid necrosis at its base, overlying a large, tortuous, thick-walled artery in the muscularis mucosa. It is characterized by subintimal fibrosis of the artery and absence of inflammation at the edge of the mucosal defect [1].

The pathogenesis of Dieulafoy's lesion is unknown. In the past, this lesion has been regarded as an acquired abnormality that may involve aging, leading to elongation and tortuosity of a submucosal artery [12]. However, some believe that Dieulafoy's lesion may represent a congenital vascular malformation due to the vascular architecture of the gastric blood supply [3], as the lesser curve is one area of the stomach that is not perfused by a submucosal plexus, and arteries in the lesser curve arise directly from the arterial chain along the lesser curve outside the stomach [13]. Parro-Blanco et al. have reported that Dieulafoy's lesion usually occurs on the lesser curvature [14].

Endoscopy has proved effective for both diagnosis and treatment. Before the advent of flexible endoscopy, the mortality from a bleeding Dieulafoy's lesion was high, and therapy was predominantly surgical. Recently, endoscopic therapy has been successful in approximately 95% of cases [6, 11]. The techniques used for endoscopic therapy have included, as in our case, injection of epinephrine or sclerosants, electrocoagulation, band ligation, and photocoagulation.

Baettig et al. reported a success rate of 96.4% for endoscopic treatment using injection of a solution containing norepinephrine and polidocanol [7]. Stark et al. also obtained similar results in a group of 19 patients, with the predominant treatment being epinephrine injection and heat probe thermocoagulation [12]. We successfully used epinephrine injection for treating our patient. Additionally, Murray et al. reported using endoscopic band ligation to treat a Dieulafoy's lesion in the small intestine of a child [15].

In summary, Dieulafoy's lesion as a cause of massive upper gastrointestinal bleeding in a child is uncommon. When it does occur, endoscopic injection therapy is effective, well tolerated, and of long-term benefit.

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