Gastric Dieulafoy lesion as a cause of upper gastrointestinal bleeding in a 4-year-old child: a case report

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ABSTRACT

Dieulafoy lesion, although rare, is a significant gastrointestinal condition that can affect children. These quiescent lesions can easily be overlooked and bleeding lesions are occasionally misidentified on endoscopy. So, accurate diagnosis depends on increased knowledge and improvements in endoscopic procedures. Treatment of Dieulafoy lesion in children usually involves in different modalities of endoscopic interventions to stop the bleeding and promote healing. In more severe cases or when endoscopic treatments are unsuccessful, surgical intervention may be required to remove the lesion. In this case report, we present the case of a boy of 4 years and 4 months old, who presented with hematemesis with melena. He appeared mildly pale, moderately dehydrated, had a moderate pulse volume, prolonged capillary refill time, a narrow pulse pressure and tenderness in the epigastrium when the abdomen was examined. His tests showed a low hemoglobin level (7.5 g/dl), a normal coagulation profile and several bleeding sites in the fundus and upper part of stomach on endoscopic examination, which are consistent with Dieulafoy lesions.

Key words: Dieulafoy lesion, upper gastro-intestinal bleeding, severe anemia.

BIRDEM Med J 2023; 13(3): 163-166 DOI: https://doi.org/10.3329/birdem.v13i3.68829

INTRODUCTION

Dieulafoy lesion, also known as "submucosal arteriole malformation" and "cirsoid aneurysm,¹ is a rare, sometimes fatal illness that can affect both children and adults. It is characterized by the presence of submucosal, abnormally large artery that erodes through the wall of the gastrointestinal tract, leading to spontaneous and sometimes severe bleeding.² Dieulafoy lesions can

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Received: May 28, 2023 Revision received: August 19, 2023 Accepted: August 29, 2023 develop in any part of the gastro-intestinal (GI) tract, as well as extra GI tract such as the bronchus. It is usually detected in the fundus and lesser curvature of the stomach.^{2,3} The precise reason of Dieulafoy lesions is not fully understood⁴ but the neonatal presentation supports the theory of its congenital origin.⁵ Depending on the lesion's location and the level of bleeding, the Dieulafoy lesion's symptoms can vary. The most common symptom is sudden and severe GI bleeding, which can manifest as hematemesis or melaena. In some cases, there may be massive bleeding which could be fatal without prompt initiation of adequate treatment.⁶ Diagnosing Dieulafoy lesion can be challenging because of the intermittent pattern of bleeding. Various diagnostic tests may be performed to identify the source of bleeding, including upper GI endoscopy, colonoscopy, angiography or technetium 99m labelled red blood cell scanning etc. However, endoscopy is the preferred method, as it allows direct visualization and potential treatment of the lesion during the procedure.¹ Treatment of Dieulafoy lesion in children focuses on stopping the

bleeding and preventing further complications. With advances in endoscopic techniques, endoscopic therapy has gradually supplanted surgery as the primary method for identifying and treating Dieulafoy lesions.⁶ Here, we describe a case of a gastric Dieulafoy lesion that caused circulatory compromise due to hematemesis, melena three to four times. To improve the quality of life of such cases, more awareness must be raised about the necessity of early identification and treatment of this rare condition.

CASE REPORT

A 4-year-4-month old male child was admitted to our hospital with intermittent fever for 7 days along with vomiting 4-5 times a day which initially contained food material but day before admission he develops hematemesis and melena 3-4 times with poor feeding and oliguria. He had no past history of any bleeding manifestations before this illness and did not have any history of taking non-steroidal anti-inflammatory drugs (NSAIDs).

On physical examination, he was afebrile initially, moderately pale, not icteric, moderately dehydrated, pulse volume was moderate, CRT > 2 sec, mild tachycardia (HR 130 b/min), BP 100/80 mmHg, RR 22 br/ min, SPO2 98% in room air. Abdominal examination revealed tenderness in epigastric region without any organomegaly; other systemic examination was unremarkable. His laboratory results showed, Hb 9.8 gm/dl initially which was dropped to 7.5 after 12 hours (Hct was 29.3 and 23.4 respectively), normal white cell and platelet count; CRP was 2.16 (normal value <0.3 mg/dl), procalcitonin was 2.37 (normal value <0.05 ng/ml), dengue IgM, IgG was negative, liver and renal function were within normal limits, DIC profile was also came out normal. His blood and urine cultures revealed no growth.

His initial management was focused on correction of dehydration and reassessment of hemodynamic status which became normal after initial resuscitation, other supportive management were antibiotics, proton pump inhibitor, intravenous fluids and nil oral. However, his temperature raised to 101.8° F and melaena was continued though hematemesis was stopped. He was transfused with PRBC as Hb was dropped. Upper GI endoscopy was done after correction of anemia which revealed multiple bleeding points (Figure 1) with intact mucosa at the fundus and upper body along with linear erythema at the upper and mid body of stomach; features were suggestive of Dieulafoy lesions. Hemostasis was done on the bleeding points with argon plasma coagulation (APC) (Figure 2).

Gradually, his melaena subsided after 2 days of endoscopic management of bleeding points. He was discharged 3 days after post-endoscopy in a vitally stable condition without having any hematemesis and melaena with stable Hb level on two occasions done 24 hours apart, oral proton pump inhibitor was advised along with follow up plan.



Figure 1. Upper gastro-intestinal tract endoscopy showed multiple bleeding spots in stomach mucosa



Figure 2. Bleeding spots hemostasis with argon plasma coagulation

DISCUSSION

Dieulafoy lesions, primarily upper gastrointestinal hemorrhage, are a rare cause of GI bleeding in children. Gallard first characterized this illness in 1884 but French surgeon Georges Dieulafoy more precisely labeled it "exulceratio simplex" in 1898.^{1,6}

Most often, there is an abrupt, painless, severe and recurring hemorrhage in the presentation. Depending on where the lesion was, the presentation changed. While colonic lesions showed fresh bleeding per rectum, gastric and duodenal lesions showed substantial upper GI bleeding.⁶ Our patient also had lesions in stomach and presented to us with hematemesis and melaena.

Among the location of the lesion, stomach is the most common site in both children and adults.¹

In 1968, Rossi et al.⁷ reported the first pediatric case of Dieulafoy lesion in a 20-month-old girl who had Christmas illness and required surgery due to significant upper GI bleeding. This lesion can also be a cause of upper GI bleeding in neonate, two case reports have been published by Coit et al and Polonki et al respectively.^{8,9}

The rising prevalence of Dieulafoy lesions in children during the past ten years is reflected in the fact that more than half of all pediatric Dieulafoy lesion cases were documented after the year 2000. The male to female ratio of the lesions was 1.5:1⁵, and the patient in our presenting case is a boy.

The majority of diagnoses for Dielafoy lesions are now made endoscopically and if there is active bleeding occurring at a rate of at least 0.5 ml/min, they are made 70% of the time.¹ The diagnosis of our patient was likewise made via endoscopy. The small size of the lesion, the typical appearance of the mucosa in the area and the intermittent nature of the hemorrhage may make endoscopic diagnosis challenging.⁶ Emerging technologies like capsule endoscopy could be used for tiny intestinal lesions.¹

The mainstay of treatment at the moment is endoscopic hemostasis, which can be carried out by: a. localized injection, such as local epinephrine injection and sclerotherapy; b. thermal coagulation, such as electrocoagulation, heat probe coagulation, and APC and c. hemostatic clips.⁶

Although available data from the literature research showed that >90% of adults with Dieulafoy lesions had effective endoscopic hemostasis, pediatric cases review showed that hemostasis was performed endoscopically in 37% of patients and surgically (either open or laparoscopically) in 48% of patients.¹⁰ Our patient's hemostasis was achieved endoscopically with APC which is being supported by the literature of Owensby S et al.¹¹

The prognosis for children with Dieulafoy lesion depends on the severity of the bleeding, the location of the lesion and the promptness of medical intervention. With timely diagnosis and appropriate treatment, the majority of children can recover successfully. However, if the bleeding is severe and not promptly addressed, it can lead to significant complications, including hemorrhagic shock or death.

Authors' contribution: Drafting and writing the manuscript were done in collaboration with NN, and SS provided guidance and critical revision. All authors were involved in patient management. The submitted manuscript was reviewed and approved by all authors.

Consent: Informed written consent was taken from parents of the patient for publication of this case report and accompanying images.

Conflicts of interest: Nothing to declare.

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