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An Infant with Dieulafoy's Lesion: A Case Report from the Philippines

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Abstract

This is a case of a 5-month-old infant who experienced repeated episodes of hematemesis and no known underlying health conditions. It was subsequently diagnosed as Dieulafoy's lesion localized in the lesser curvature of the stomach. Endoscopic diagnosis and treatment were done by angiographic embolization. Dieulafoy's lesion is considered rare even for adult cases, much more for pediatric patients and usually underdiagnosed. Hence, patients presenting with gastrointestinal bleeding should be managed in a multidisciplinary approach. Spreading awareness about this lesion by including it in the considerations, may help improve early detection and treatment.

Keywords

Dieulafoy's Lesion, Gastrointestinal Bleeding, Upper Gastrointestinal Bleeding, Hematemesis, Bleeding

1. Introduction

Dieulafoy's lesion is a relatively rare, but potentially life-threatening condition. It is a congenital arteriovenous malformation characterized by an unusually large tortuous submucosal artery. Worldwide, it only accounts for 1% - 2% as the primary source of upper gastrointestinal bleeding [1]. In the Philippines, there is insufficient data in the pediatric population. The exact pathogenesis is still unknown. It is either inherited or acquired [2]. Its true incidence in the general population is difficult to determine accurately as the symptoms are silent until presentation. Increasing the awareness of this malformation will improve early diagnosis and treatment.

2. Case Description

A 5-month-old, female infant from Davao City, Philippines was brought to the

emergency department due to hematemesis of seven days duration with at least two episodes per day. This was associated with pallor. There were no other symptoms like fever, diarrhea or bloody stools. The patient was managed as a case of anemia secondary to upper gastrointestinal bleeding probably Mallory Weis. She was confined for four days. Complete Blood Count (CBC) showed anemia with hemoglobin of 71 g/dl and hematocrit of 0.22. Bleeding parameters were within normal limits. Patient was advised for endoscopy but the parents initially refused. Transfusion of packed red blood cells was done. Patient was discharged, well. However, the recurrence of hematemesis few days post discharge prompted re-admission of the patient.

The patient was born full term to a 29-year-old G3P3 mother in a local hospital. She was delivered via normal spontaneous delivery with a Ballard score of 39 weeks. Weight is appropriate for gestational age. She was admitted at the Neonatal ICU (NICU) for 19 days. Umbilical vein catheterization was done. Routine newborn care which includes vitamin K was also done. She was managed as a case of meconium aspiration syndrome, hypoxic ischemic encephalopathy stage II. Expanded newborn screening and hearing screening tests were all normal.

The patient had previous admissions due to severe pneumonia and acute gastroenteritis. She had no allergies, non-asthmatic, and no known exposure to tuberculosis. Growth and development are at par with age. She is bottle fed with formula milk since discharge. She received the required vaccination for age. The family has no atopy and no history of bleeding.

The patient was examined awake, comfortable, and not in respiratory distress. Vital signs revealed that she was tachycardic at 162 bpm, eupneic at 30 cpm, afebrile at 37.1˚C, normotensive at 90/60mmHg with oxygen saturation of 99% at room air. Her weight, length and head circumference are all normal for age. Pertinent physical examination showed absence of rashes or purpura on the skin with pale palpebral conjunctiva. There was no palpable cervical lymphadenopathy. Heart sounds were distinct and no murmurs. She had bronchovesicular breath sounds with no crackles. Abdomen is globular with normoactive bowel sounds and no hepatosplenomegaly.

According to Nelson's Textbook of Pediatrics, bleeding that originates in the esophagus, stomach, or duodenum can cause hematemesis. It is the cardinal sign of upper gastrointestinal bleeding (UGIB) which comprises 73% of the presenting symptoms of UGIB [3] [4]. The history should include any previous GI problems (e.g., jaundice, liver disease, ulcers, GER, other GI hemorrhages), blood transfusions, coagulopathies, and iron deficiency. A neonatal history of total parenteral nutrition, omphalitis, or umbilical vein catheterization is a risk factor for portal vein thrombosis (**Figure 1**). Examination of vital signs is important to ensure that bleeding does not require emergent treatment for circulatory compromise.

Confirming the presence of blood is important to avoid unnecessary evaluation.

In patients presenting with gastrointestinal bleed, it is fundamental to distinguish an upper from a lower GI bleed. Upper gastrointestinal bleeding refers to bleeding from a site proximal to the ligament of Treitz at the level of duodeno-jejunal flexure. Symptoms of Upper GI Bleeding include hematemesis (73%), melena (21%), and coffee-ground emesis (6%). While hematochezia or passage of bright red blood in stools is usually a feature of lower GI bleeding, but some infants with UGIB can occasionally present with passage of bright red blood from the rectum because of rapid GI transit in a briskly bleeding child. The possible etiologies in Upper GI Bleeding usually vary with age but considerable overlap exists between the different age groups. The rate and extent of bleeding depend on these etiologies and the presence of associated conditions such as coagulation defects [4] (**Table 1**).

Gastrointestinal bleed is rare in neonates however it may be present in newborns with Vitamin K deficiency as well as maternal idiopathic thrombocytopenia

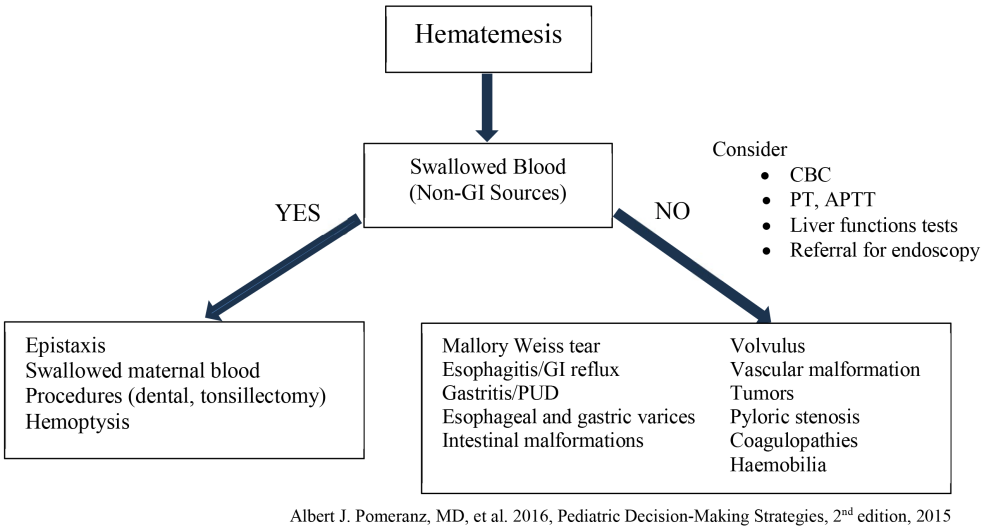


Figure 1. Diagram for approach to diagnosis for patient presenting with hematemesis

Table 1. Upper gastrointestinal bleeding (UGIB) vs Lower gastrointestinal bleeding (LGIB).

	Upper GI Bleed	Lower GI bleed
Location	**Proximal to Ligament of Treitz** (esophagus, stomach, or duodenum)	**Distal to Ligament of Treitz** (jejunum, ileum, colon, rectum)
Symptoms	Hematemesis Coffee-ground emesis Melena Hematochezia (very brisk bleeding)	Hematochezia Melena (SI, ascending colon)
Causes	Age-specific, associated conditions (PUD bleeding, varices, erosive esophagitis, and vomiting-induced hematemesis)	Crohn's disease, ulcerative colitis, hemorrhoids, anal fissures, and Meckel diverticulum
Diagnostics	Esophagogastroduodenoscopy	Colonoscopy

Nelsons Textbook of Pediatrics, 19th Ed.

and maternal NSAID use. Other causes include stress gastritis or ulcers, vascular anomalies, coagulopathy caused by infection, liver failure, or a congenital coagulation factor deficiency. For infants and toddlers, stress ulcers and gastritis are common especially in sick patients. Variceal bleeding with portal hypertension and vascular malformations may also be noted. Other causes include reflux esophagitis, esophageal or gastrointestinal foreign body, communicating duplication cysts, NSAIDS use and corrosive injury. Foreign body ingestion should be also considered especially in patients with a history of choking episodes. Causes of UGIB in older children and adolescents are like those seen in adults. Of this, varices, peptic ulcers, Dieulafoy's lesions and vascular malformations cause major bleeds [3]. For this case esophageal varices, Mallory Weis tears, Cow's milk protein allergy and coagulation problems were all considered as the cause of hematemesis.

The patient was admitted under Gastroenterology Service. She was given tranexamic acid for bleeding. Baseline laboratory tests were requested such as: Complete Blood Count, OPS/NPS, Serum electrolytes (Na, K, Ca, Cl, Mg), BUN, Creatinine, SGPT, SGOT, CRP and PCT. Imaging studies such as CXR-APL and Upper GI Endoscopy were also requested. Results showed anemia of 91 g/L (N 96 - 12 g/L), normal hematocrit of 0.30 (N 0.29 - 0.37), normal MCH, increased MCV, decreased MCHC revealing a hypochromic, macrocytic anemia, severe. Coagulation studies showed only a mild elevation of the APTT (35.3 sec, N 27 - 34 sec). The rest of the results were unremarkable.

Upper GI endoscopy findings revealed presence of a protruding vessel with multiple blood clots at the lesser curvature of the stomach, suggesting Dieulafoy's lesion, thus patient was referred to pediatric surgery and vascular and interventional radiology for evaluation and planning of the contemplated procedure (**Figure 2**).

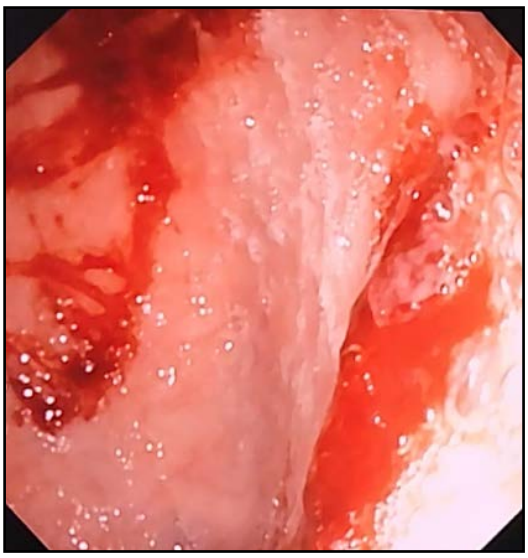


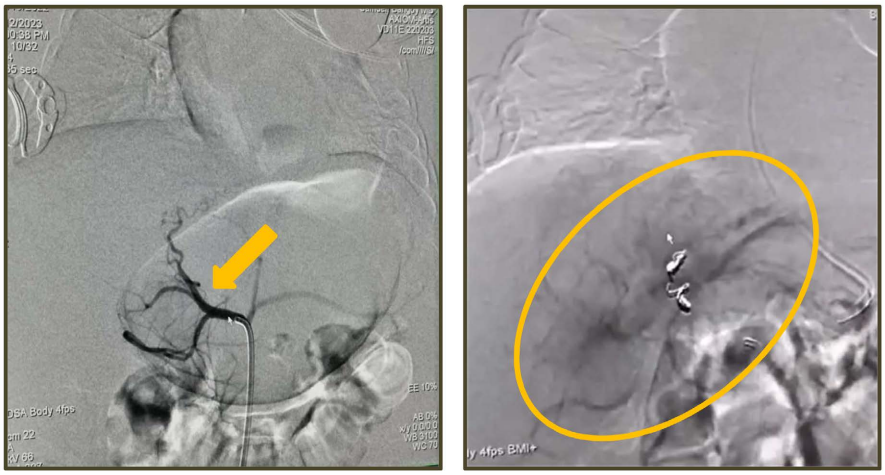
Figure 2. Endoscopic finding of Dieulafoy's lesion. Presence of a protruding vessel at the lesser curvature of the stomach surrounded by clots.

Several approaches were considered such as exploratory laparotomy, gastrostomy, oversewing of the Dieulafoy's lesion vs. wedge resection, and upper GI endoscopy with possible clipping of the lesion. The team decided to do a transarterial embolization. Angiographic embolization was done. The right common femoral artery was accessed and a vascular sheath was inserted. The celiac trunk was selected using a catheter. Upon selective angiography, a tortuous right gastric artery was noted and confirmed at the lesser curvature of the stomach. Embolization was done using 2 3 × 6 mm detachable coils. Post embolization angiography showed a successful embolization of the said artery (Figure 3). Postoperatively, the patient tolerated the procedure well with no recurrence of bleeding and other post-op complications. The patient was discharged well. With good follow-up and monitoring, patients with this malformation have a good prognosis.

3. Discussion

Dieulafoy's lesion is also known as gastric aneurysm. It is a congenital arteriovenous malformation characterized by an unusually large tortuous submucosal artery. If this artery is eroded, it could lead to pulsatile bleeding causing recurrent gastric bleed [4]. Our case had two visits to the emergency department due to episodes of hematemesis. However, it is only on the present admission that the etiology was determined.

The incidence of acute gastrointestinal bleeding ranges from 50 - 150 per 100,000 of the population each year, in all ages [1]. To account, most journals would say it only comprises 1% - 2% of acute Upper GI Bleeding in adults and is



(a) (b)

Figure 3. (a) Pre-embolization of the Dieulafoy's lesion. (b) Post-embolization of the lesion. Right common femoral artery was accessed and vascular sheath was inserted. Celiac trunk was selected using a catheter. Upon selective angiography, a tortuous right gastric artery was noted and confirmed to be seen at the lesser curvature of the stomach. Embolization was then done using 2 3 × 6 mm detachable coils. Post angioembolization shows successful embolization of the said artery.

extremely rare in children. There are no accurate statistics on the incidence of this disease in children [5]. From 1995 to 2021, only sporadic cases were reported in children worldwide. In the Philippines, there is only 1 case reported to date, however, this is an adult case presenting with melena. There is insufficient data recording its occurrence in the pediatric population [5].

According to a journal by Dulic-Lakovic, *et al*, upper GI tract is the predominant location for dieulafoy's lesion [6]. Approximately, 80% - 90% is located within 6 cm from the gastroesophageal junction within the lesser curvature of the stomach, with a direct correlation with the blood supply that directly arise from arterial chain, compared to the other part of the stomach in which blood supply is derived from submucosal plexus of a larger vessel [7] [8]. However, this could also be found elsewhere along the gastrointestinal tract—duodenum (15%), esophagus (8%), rectum (2%), colon (2%), and jejunum (1%) [5]. Dieulafoy's lesion involving the colon and rectum is extremely uncommon, diagnosis was by rigid sigmoidoscopy, with fewer than 30 cases [9]. One study reported its occurrence in the tracheobronchial branch presenting as hemoptysis [10]. The patient's endoscopic findings showed protruding vessel with multiple blood clots at the lesser curvature of the stomach which is the most common site for the lesion to arise.

The exact pathogenesis is still unknown, either it is inherited or acquired. Older age patients with the said lesion may suggest it is acquired, while pediatric literature suggests it may represent a congenital anomaly [11]. One literature proposed an underlying mechanism: unlike the normal arterial tree that usually tapers off when approaching the distal branches, the lesion maintains dilated approximately 1 - 3 mm all throughout. This caliber is ten-fold larger than the normal maximal caliber of submucosal vessels. Dieulafoy's lesion can protrude through a small mucosal defect making it susceptible to minor mechanical trauma and eventually erode into the lumen to cause acute gastrointestinal bleeding [1].

Patients usually present with painless recurrent intermittent hematemesis associated with melena, hematochezia, and hypotension. The clinical manifestations of the patient depend on the location and the diameter of the bleeding vessel involved [5]. Gastric and duodenal lesions present with massive upper gastrointestinal bleeding, small intestinal lesions present with UGIB bleeding and/or hematochezia, while colonic lesions present with fresh blood per rectum [12]. In our case, our patient presented with recurrent hematemesis eventually during the course in the wards was associated with melena.

For patients who present with acute upper gastrointestinal bleeding, initial management for these patients would include fluid resuscitation and as well as risk stratification, whether low or high risk. Patients are classified under high-risk when: Patient presents with hypotension, tachycardia, oliguria, pallor, altered sensorium, hematemesis, decreased hematocrit which are signs of large blood loss. Patients with chronic conditions or comorbidities such as any heart, liver or kidney problems which may affect/compromise the body's physiologic reserve,

immunosuppressed patients or those taking anticoagulants. With esophagogastroduodenoscopy EGD done, those who will present with active bleeding, deep ulcer, visible vessels. Patients who are considered low-risk are usually started with supportive management and given IV proton pump inhibitor while patients with higher risk may need transfusion of blood products and admission to ICU for close monitoring. Once patients are stabilized, diagnostics to identify the source of bleeding such as EGD may be commenced [5].

To diagnose Dieulafoy's Lesion, esophagogastroduodenoscopy should be done ideally during the first 12 hours from onset of bleeding. This procedure identifies most of the sources for gastrointestinal bleeding. On EGD, Dieulafoy's lesion may appear as a stream of arterial blood emanating from what seems like a normal mucosa. For those with absence of active bleeding, blood clots may obscure the mucosal defect, and for some a protruding vessel may be seen. However, not all lesions are easily identified by this diagnostic exam due to the intermittent nature of this case which is why for some patients, multiple EGD must be done to confirm the diagnosis. EGD identifies only about 71% of the cases. For our patient, a protruding blood vessel surrounded with blood clots at the lesser curvature was noted suggesting Dieulafoy's Lesion [13] [14]. Other diagnostic modalities used in previous case reports would include mesenteric angiography for patients with massive gastrointestinal bleeding and no history or previous imaging studies which would point out any source of bleeding [14].

Diagnostic modalities for gastrointestinal bleeding may also be used for therapeutic purposes. Treatment options for Dieulafoy's Lesion would include: 1) Endoscopic hemostatic therapy; 2) Angiographic embolization or 3) Surgical plans wherein the lesion would be oversewn and resected [5]. Endoscopic therapy is currently considered as the mainstay therapy for Dieulafoy's lesions as it is able to achieve permanent hemostasis in more than 90% of cases. Endoscopic therapy includes either: the use of vasoconstrictors, such as epinephrine, and sclerosing agents, such as sodium morrhuate, 50% dextrose or absolute alcohol. These agents lead to temporary hemostasis without the risk of any mucosal perforation electrocoagulation/laser photocoagulation. Heater probe, or application of hemoclips was originally opted for our patient, however unavailability of materials led to the decision of angioembolism for our case.

Angiographic embolism is an alternative procedure in managing cases of gastrointestinal bleeding. This procedure enables the physician to locate and identify the site of bleeding and subsequently stops the bleeding by embolizing the bleeding artery with the use of glue or coils. This is usually indicated for patients who 1) failed hemostasis with endoscopic techniques, 2) acute bleeding wherein lesions were beyond the reach of available endoscopes, or 3) poor surgical candidates [13]. Prior case reports opted for this technique since prior EGD had no specific source identified, while some had endoscopic clips applied on the area of involved artery and then subsequent angioembolism done. For our case, the bleeding vessel identified and embolized was the Right gastric artery. The agent

used for embolization was 2 × 3 mm coils with the right femoral artery as the access site. For patients managed with this technique, post operatively, they are monitored for possible re-bleeding episodes due to formation of collateral channels, possible arterial dissection of cannulated artery, or possible occurrence of ischemia or strictures especially when the present collateral channels in the mesenteric area is damaged or when embolic material passes the vascular bed which was not the target vessel. Severity of these complications would then determine if there is a need for anticoagulants or possible surgical management [4] [15].

Surgery was once the first line of management for Dieulafoy's lesion. Currently, this is already reserved for those patients with failure of endoscopic or angiographic interventions. In this technique, the identified vessel is oversewn or resected, given that guided preoperative localization is done. Exploratory laparotomy, gastrotomy with oversewing of Dieulafoy's lesion or possible wedge resectioning was considered for this case if prior options were not possible for our patient.

4. Conclusion

Patients presenting with gastrointestinal bleeding should be managed in a multidisciplinary approach. One must consider other systems aside from gastrointestinal causes. As a rare condition both for adults and children, appropriate diagnostic and treatment modalities should be done. Endoscopic therapy is still the first-line diagnostic and/or treatment option for Dieulafoy's lesion. Other treatment options may be used as adjunct therapy. Despite its good prognosis, treated patients still need to be closely monitored for rebleeding episodes.

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Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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Abbreviations

GIT	Gastrointestinal Tract
LGIB	Lower Gastrointestinal Bleed
UGIB	Upper Gastrointestinal Bleed