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Case Report

Duodenal Dieulafoy Lesion in an 8-Year-Old Boy: A Case Report

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Abstract

Dieulafoy lesion is a rare often under diagnosed cause of gastrointestinal bleeding in children. Its presentation is varied from occult blood loss in stools to frank hematemesis and melena. It is a large torturous sub mucosal blood vessel that protrudes through mucosa and can rupture with minimal trauma and cause massive fatal hemorrhage. Its etiology is enigmatic. Endoscopy is considered gold standard for its diagnosis as well as management. We present here a case of dieulafoy lesion in first part of duodenum of an 8 year old boy presenting with massive melena and was successfully managed with endoscopic hemoclipping.

Key words*: dieulafoy lesion, upper gastrointestinal bleeding, angiodysplasia, melena, children.*

Introduction

Dieulafoy lesion also known as dieulafoy disease is a rare often underdiagnosed cause of upper gastrointestinal bleeding in children.**[**1**]** The incidence of dieulafoy lesions is under documented in children and only 28 cases have been documented so far in literature.**[**2**]** It is a fatal yet preventable cause of gastrointestinal bleeding and early diagnosis and prompt management is life saving.**[**3**]** We present here a case of an 8 years old boy with dieulafoy lesion in first part of duodenum, presented to us with melena and pain abdomen and was successfully managed endoscopically with hemostatic clipping.

Case Presentation

An 8 year old boy presented to the emergency department of Children Hospital Faisalabad with abdominal pain and black stools for last 4-5 days. He had blunt trauma to the back when during playing his siblings jumped over his back repeatedly after which he developed melena. He was initially managed by a paediatric surgeon who after baseline workup and consultation with radiologist did CT abdomen with contrast and found clots of blood in stomach and bilateral gluteal hematomas (Fig.1) he put drains in anterolateral aspects of his both thighs.

He also did exploratory laparotomy which was inconclusive and he did colostomy after that. The child is loosing blood massively in stools and needs 3-4 blood transfusions per day. He was then referred to paediatric gastroenterologist because of continuous loss of blood in stools and transfusion dependency due to persistently low hemoglobin despite 3-4 blood transfusions in a day. There is no history of loose motions, hematochezia, jaundice, petechie, bruises, ascites , edema, hematemesis, heart burn, malabsorption or rashes on the body, joint pains, any medication use.

There is no family history of any liver diseases, bleeding disorders, malabsorption syndromes or autoimmune diseases. On examination he is markedly pale with drains on anterolateral aspect of both thighs and colostomy bag attached containing melena stools. He is having tachycardia with pulse rate of 110 beats/minute. His Respiratory rate is 26 breaths /minute and temperature is 98.6°F. His weight is 22kg. There is no jaundice, petechie and bruises. His abdomen is tense and distended with bluish discoloration of right upper quadrant with subcutaneous edema. There is no hepatosplenomegaly or signs of chronic liver disease. His liver function tests and coagulation profile was normal. Complete blood counts showed hemoglobin 9.3g/dl with microcytic hypochromic anemia. His viral serology for hepatitis is negative.

After stabilization his endoscopy was performed which showed a bleeding dieulafoy lesion in the 1st part of the duodenum, clotted blood in the stomach and attenuated blood in the 2^{nd} and 3^{rd} part of duodenum. Hemoclips were applied at the site of lesion and hemostasis secured. (Fig. 2) After hemoclipping his melena stopped and his hemoglobin level was maintained. He was on our follow up in paediatric gastroenterology clinic and his repeat endoscopy after 2 months showed hemoclips in place and no active bleeding. (Fig. 3) His Hemoglobin was 12.5g/dl on follow up and he is thriving well.

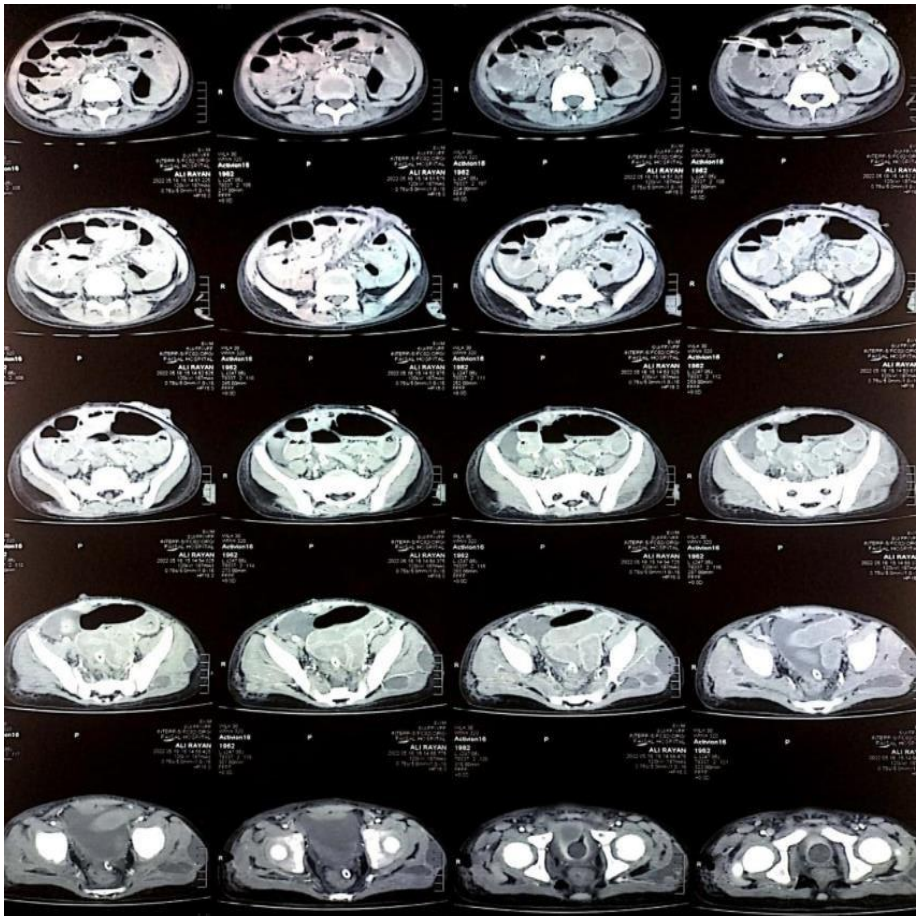

Figure 1

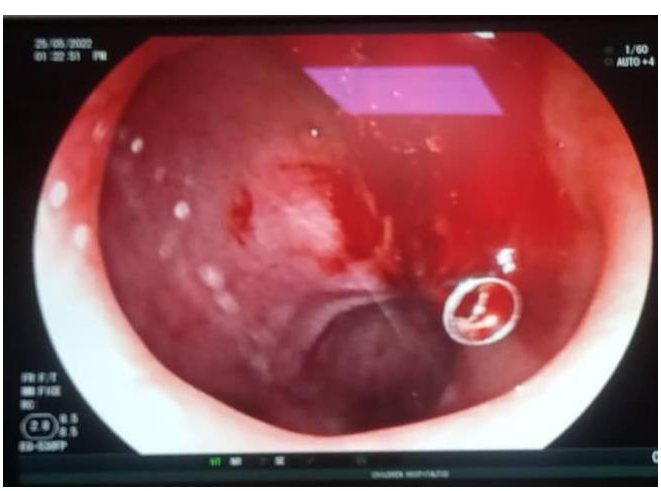

Figure 2

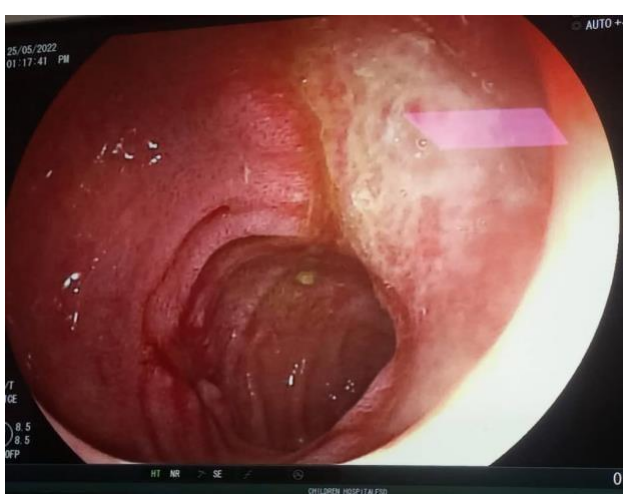

Figure 3

Discussion

Diuelafoy lesion is an Arteriovenous malformation of submucosal arterioles proturding through a small defect in mucosa or even underneath a normal looking mucosa.**[**4**]** It was first noticed by Gallard in 1884 but later on described in detail by Paul George Diuelafoy a French surgeon in 1898 and he named them as "exulaceratio simplex".Its incidence is 0.3% - 6.7% in adults but data in children is lacking.**[**5**]**

It is twice as common in males than females.**[**6**]** 75% of lesions are along the lesser curvature of stomach about 6cm from the oesophagogastric junction due to direct supply of this region from left gastric artery, less commonly involved sites are duodenum(14%), jejunum(10%),colon(5%), surgical anastamoses(5%), esophagus(1%) and even outside gastrointestinal tract in the traecheobronchial tree. **[**7**]** The exact pathogenesis of dieulafoy lesion is unknown, normally the arterioles in sub mucosa they taper as they traverse through the muscularis mucosae layer beneath mucosa but the dieulafoy lesion do not tapers and this torturous large calibre dysplastic submucosal arterioles of 1-5mm diameter (more than 10 times the diameter of capillaries in this region) puts pressure due to pulsatile nature, causing ischemia and thinning of overlying mucosa leading to exposure of these arterioles and subsequent bleeding with slight insult without any ulceration of mucosa.**[**8**]**

Dieulafoy lesion lack aneurysmal, arterioseclerotic and vasculitic changes.**[**9**]** The risk factors associated with dieulafoy lesions in adults are chronic use of NASIDS and anticoagulants, autoimmune disorders, alcohol consumption, chronic kidney disease, peptic ulcer disease, hypertension, hyperlipidemia and chronic liver disease but in children it is usually congenital.**[**10**]** It presentation is varied from massive upper or lower GI bleeding with shock, recurrent bleeding episodes from upper or lower GI tract, melena, hematochexia, recurrent abdominal pain and severe anemia.**[**11**]** Endoscopy is the gold standard for diagnosis and lesion is seen as an oozing vessel with normal mucosal boundaries differentiating it from gastric ulcers.**[**12**]**

Endoscopic criteria for of a Dieulafoy lesion is; i). Spurting or oozing of blood from a small defect in mucosa <3mm; ii) a visible protruding vessel from a normal or slight defect in the mucosa; iii) a fresh blood clot adherent to the defect or normal looking mucosa.**[**13**]** If lesion is small enough or is at an obscured site that is not visualized endoscopically then mesenteric angiography, Technetium- 99m labelled red blood cell scanning, contrast enhanced CT abdomen and surgical exploration can also be used for diagnosis.**[**14**]**

It is managed endoscopically with hemostatic clipping and band ligation and it has 95% success rate and is

superior to conventional techniques of epinephrine injections, sclerotherapy, argon plasma therapy and thermocoagulation used for managing dieulafoy lesion.[6,15] Other modalities like angioembolization using gel foam and surgical repair are reserved for cases where endoscopy fails.[16] Indications for surgical intervention are hemodynamically unstable patient, more then one lesion or lesion is at site where it is difficult to reach endoscopically.[2,17] The prognosis of dieulafoy lesion is good with risk of rebleeding in <10% patients managed endoscopically.[18] High prothrombin time (>12 seconds), elevated leucocyte counts (>10 ×108/L), old age and use of anticoagulants are associated with rebleeding after repair in few cases.[19] Combination therapy in endoscopy using either band ligation, hemoclips or epinephrine injections is recommended in literature to prevent rebleeding in these lesions.[20]

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