CASE REPORT

A CASE OF ARTERIOVENOUS MALFORMATION IN THE SUBMUCOSAL LAYER OF THE STOMACH

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ABSTRACT: Gastrointestinal bleeding is a commonly encountered emergency. Common causes include bleeding peptic ulcers, gastric erosions and esophageal varices. Rare causes include arteriovenous malformation (AVM) of the gastrointestinal tract. With increasing availability of endoscopy and elective angiography AVM is being more frequently recognized. Literature search shows since 1884 about 42 cases have been reported so far worldwide. Upper GI bleeding caused by AVM usually presents as massive haematemesis or chronic iron deficiency anaemia

KEYWORDS: Arteriovenous malformation (AVM), Upper gastro-intestinal (UGI) Oesophagogastrosocopy (OGD)

CASE REPORT: Arteriovenous malformation (AVM) of the stomach is extremely rare. We report a patient with asymptomatic gastric AVM detected during investigating a patient of severe anaemia [4]. The patient, a 26-year-old male, had no history of melena, presented with severe anaemia, history of repeated blood transfusions. His routine biochemistry and hematology including bone marrow favoured iron deficiency anaemia due to blood loss. Endoscopy revealed multiple vascular lesions, scattered in stomach, duodenum with few gastric erosions. Colonoscopy revealed a small vascular lesion 0.5x0.5 cm in lower rectum (AVM)

Upper gastro-intestinal (UGI) bleeding can be classified into several broad categories based upon anatomic and pathophysiologic factors. Peptic ulcer disease; 55 percent, Oesophagogastric varices; 14 percent, Arterial, venous, and other vascular malformations; 7 percent, Mallory-Weiss tears; 5 percent, Erosions; 4 percent, Tumors; 4 percent and other causes; 11 percent [1]. Gastrointestinal vascular diseases include angiodysplasia, arteriovenous malformation (AVM), cavernous haemangioma, hereditary haemorrhagic telangiectasia (Rendu-Osler-Weber disease), Gastric antral vascular ectasia and Dieulafoy's lesion (DL) [1,2].

Angiodysplasia presents as an irregular shaped clusters of ectatic small arteries, small veins and their capillary connections. These lesions are called by various names such as vascular ectasia or angiectasia. Arteriovenous fistulae, often called "malformations," may be congenital or acquired. AVM remains a relatively rare clinical lesion consisting of abnormal shunts between the arterial and venous vascular systems, the diagnosis of which is problematic because routine barium contrast studies and endoscopy fail to demonstrate the lesion. With increasing use of angiography over the past 30 years in the assessment of gastrointestinal bleeding, AVM has been more frequently recognized [3]. Gastric AVM may clinically be asymptomatic or may present as massive upper gastrointestinal bleeding or chronic iron deficiency anaemia [4]. Gastric antral

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vascular ectasia (GAVE or watermelon stomach) is a rare cause of UGI bleeding. It is often confused with portal hypertensive gastropathy, both of which can occur in patients with cirrhosis [4,5]. The red stripes represent ectatic and sacculated mucosal vessels. Dieulafoy's Lesion (DL) is an uncommon cause of gastric bleeding. It accounts for less than 5% of all gastrointestinal bleeds in adults [2]. However, unlike most other aneurysms these are thought to be developmental malformations rather than degenerative changes. DL lesion has also been given other names: caliber-persistent artery, gastric arteriosclerosis, cirroid aneurysm, and submucosal arterial malformation. Majority of the Dieulafoy's lesions occur in the upper part of the stomach, however they can occur anywhere in the GI tract. Extragastric DLs are uncommon, but have been identified more frequently in recent years because of increased awareness of the condition. Duodenum is the commonest location (18%) followed by colon (10%) and jejunum (2%) and oesophagus (2%) [2]. The pathology of the lesion is essentially the same. The most common presenting symptom is recurrent, often massive haematemesis associated with melena (51%). The lesion may present with haematemesis alone (28%), or melena alone (18%) [5,6]. Clinical symptoms may include perforation or haemoperitoneum. Characteristically, there are no symptoms of dyspepsia, anorexia or abdominal pain. Initial examination may reveal haemodynamic instability, postural hypotension and anaemia. The mean hemoglobin level on admission has been reported to be between 8.4–9.2 g/dl in various studies [7,8]. The average transfusion requirement for the initial resuscitation is usually in excess of three and up to eight units of packed red blood cells [9,10]. Dieulafoy's is inherently a difficult lesion to recognize, especially when bleeding is inactive. In approximately 4–9% of massive upper gastrointestinal haemorrhage, no demonstrable cause can be found [10,11]. Dieulafoy's lesion is thought to be the cause of acute and chronic upper gastrointestinal bleeding in approximately 1–2% of these cases [12,13]. It is thought to be more common in males (M: F = 2:1) [13,14] with a median age of 54 years at presentation [14,15]. Approximately 75% to 95% of Dieulafoy's lesions are found within 6 cm of the gastroesophageal junction, predominantly on the lesser curve [16]. The blood supply to that portion of the stomach is from a large submucosal artery arising directly from the left gastric artery.

Oesophagogastroscopy (OGD) can successfully identify the lesions in approximately 82% of patients. Approximately 49% of the lesions are identified during the initial endoscopic examination, while 33% require more than one OGD for confident identification [17-19]. The remainder of the patients with Dieulafoy's lesions is identified intraoperatively or angiographically [20,21]. Endoscopic ultrasound can be a useful tool in confirming the diagnosis of a Dieulafoy's lesion, by showing a tortuous submucosal vessel adjacent to the mucosal defect. Angiography, during active bleeding has been helpful in a small number of cases in which initial endoscopy failed to show the bleeding source. It has been tentatively suggested that, in selected cases where experienced radiological, endoscopic and surgical staff are available, thrombolytic therapy to precipitate bleeding can be used electively as an adjunct to diagnostic angiography to help in localizing Dieulafoy's lesion [22]. Other reported diagnostic methods include CT and enteroclysis [23]. For acute and massive gastrointestinal haemorrhage, immediate embolisation can stop bleeding and maintain vital signs of positive bleeders [24]. Endoscopic techniques used in the treatment include epinephrine injection followed by bipolar electrocoagulation, monopolar electrocoagulation, injection sclerotherapy, heater probe, laser photocoagulation, haemoclipping or banding [2]. Rarely, surgical removal of the lesion may be needed and is recommended only if other treatment options have not been successful. Endoscopic therapy is said to be successful in achieving permanent haemostasis in 85% of cases. Of the remaining 15% in whom re-bleeding

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occurs, 10% can successfully be treated by repeat endoscopic therapy and 5% may ultimately require surgical intervention [19,25].

The endoscopic criteria proposed to define DL are: 1) Active arterial spurting or micropulsatile streaming from a minute mucosal defect or through normal surrounding mucosa, 2) Visualization of a protruding vessel with or without active bleeding within a minute mucosal defect or through normal surrounding mucosa, and 3) Fresh, densely adherent clot with a narrow point of attachment to a minute mucosal defect or to normal appearing mucosa [24,26]. DL is characterized by a single large tortuous arteriole in the submucosa which does not undergo normal branching, or one of the branches retain high caliber of about 1–5 mm which is more than 10 times the normal diameter of mucosal capillaries. The lesion bleeds into the gastrointestinal tract through a minute defect in the mucosa which is not a primary ulcer of the mucosa but erosion probably caused from the submucosal surface by the pulsatile arteriole protruding into the mucosa [2]. It has also been suggested that a congenital or acquired vascular malformation might be the underlying cause [25,26]. Histologically, the eroded artery appears normal. There is no evidence of any mucosal inflammatory process, signs of deep ulcerations, penetration of the muscularis propria, vasculitis, aneurysm formation, or arteriosclerosis [6,27,28]. Patients with lesions in the duodenal bulb and proximal jejunum, present in a similar way to those with gastric lesions. Patients with lesions in the middle or distal jejunum, right colon and rectum present with massive rectal bleeding [29,30]. The risk of re-bleeding after endoscopic therapy remains high (9 to 40 percent in various reports) due to the large size of the underlying artery [31,32]. The mortality rate for Dieulafoy's was much higher before the era of endoscopy, where open surgery was the only treatment option [33,34].

DISCUSSION: vascular diseases of GIT are a known but rare cause of upper or lower GIT bleeds. It may present as a diagnostic challenge because of its diverse manifestations; however, a physician should always consider vascular diseases as a cause of recurrent unexplained GI bleed [35]. Management of AVM may warrant major surgical undertaking both in elective as well as in emergency situation [[4,16], and [35]].

Our patient had a diffuse type of AV malformation involving whole of the stomach as the malformations were multiple therefore surgical procedure was not done. Patient is on repeated blood transfusions and till today he has received more than 130 blood transfusions .

Conclusion :- avm of gastrointestinal tract is an uncommon finding , which can be missed in routine barium studies and endoscopy . Therefore whenever there is a strong suspicion of upper gastrointestinal bleeding which is not seen in routine studies angiography should be considered to rule out aneurysm

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