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Clinical Case **Gastroenterology C**

Rare Cause of Upper Gastrointestinal Hemorrhage: Ectopic Duodenal Varices

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Abstract

Ectopic varices represent 1–5% of variceal bleeding and carry a high mortality rate. Duodenal varices are more frequent in extrahepatic portal hypertension, often related to prior abdominal surgery. Diagnosis requires high suspicion and use of imaging when endoscopy is inconclusive. Treatment involves endoscopy, interventional radiology, or surgery, depending on the case. We report the case of a 37-year-old woman who presented with upper gastrointestinal bleeding due to ectopic duodenal varices, revealing extrahepatic portal hypertension with a portal cavernoma. Diagnosis was made through endoscopy and imaging. Despite endoscopic and surgical treatment, the outcome was fatal. This case highlights the severity and management challenges of ectopic varices. This case illustrates the severity and diagnostic difficulty of ectopic varices and the need for a multidisciplinary, tailored approach to improve outcomes. Early diagnosis and timely treatment are critical to reduce mortality.

Keywords: Gastrointestinal hemorrhage; Portal hypertension, Ectopic varices.

INTRODUCTION

Portal hypertension leads to the development of esophageal and gastric varices, but rarely to the appearance of varices in other sites, known as ectopic varices. These varices are often responsible for Gastrointestinal (GI) hemorrhagic complications. Ectopic varices are defined as large portosystemic venous collaterals arising anywhere in the gastrointestinal tract outside the eso-cardial region. We reported a rare case of upper GI hemorrhage from ectopic duodenal varices.

CLINICAL CASE

A 37-year-old female, with a medical history of repeated transfusions, no personal or family history of thrombosis, and a history of laparoscopic cholecystectomy in September 2020 for gallbladder stones.

The patient's symptomatology was started 2 months after the cholecystectomy with the onset of clinical anemia with a laboratory test showing hemoglobin (HB) at 4g/dl that was treated with iron infusion without improvement. Two months later, the patient was admitted for several haematemesis with melena and the Hb was at 5g/dl. An upper endoscopy revealed significant bleeding from the duodenum and the cause could not be determined. The patient was admitted to intensive care for hemodynamic instability, and she was transfused with five Red blood cells (RBC) units. After hemodynamic stabilization, abdominal CT scan angiography revealed a venous process between the duodenum and the hepatic hilum, which was probably responsible for the bleeding. The patient was urgently admitted to the operating room, and exploration showed the presence of enormous veinous circulations on the anterior face of the hepatic pedicle and the upper edge of the pancreas. A duodenotomy from genu superius to the genu inferius revealed significant bleeding, that was initially controlled by using a balloon to tamponade the bleeding, then a ligation of the venous collaterals.

The patient remained asymptomatic for 08 months before being readmitted to emergency for anemia, and GI hemorrhage. On clinical examination, the patient was hemodynamically stable. The abdominal examination was unremarkable and the rectal exam revealed fresh red blood.

Biological tests showed anemia at 5.2 g/dl, and Liver function tests were normal. Abdominal ultrasound

with Doppler showed a normal liver with a normal porta vein with the presence of several periportal, periduodenal, peri-jejunal, and peripancreatic collaterals vessels. Abdominal -CT- scan angiography didn't show the portal trunk that was replaced by hilar serpiginous structures associated with a portal cavernoma, thrombosis of splenic vein, and superior mesenteric vein with development of varices at the level of the duodenal wall D3, D4.

The Upper endoscopy showed the absence of esophageal varices and gastric varices, normal gastric and bulbar mucosa, but showed the presence of two varices at the D2/D3 junction with red signs. We injected 02 cc of biological glue, and 01 cc for each varix with good induration at the varices. During the aetiological work-up, we first ruled out malignant hematological causes; myeloproliferative syndromes by osteomedullary biopsy which came back normal; JAK2 mutation was negative; Thrombophilia work-up was normal, no anti-phospholipid syndrome, homocysteinemia normal, no signs of systemic illness or Inflammatory bowel disease, no coeliac disease, no active neoplasia; HIV test was negative.

The clinical outcomes were good, Hb was stable at 7.7 g/dl. The upper endoscopy of follow-up at 15 days showed two varices at the D2/D3 junction with no red signs. Two weeks later, the patient was readmitted to the emergency department for upper GI hemorrhage with hemodynamic instability for which was admitted to the emergency department and unfortunately, the patient died of hemorrhagic shock

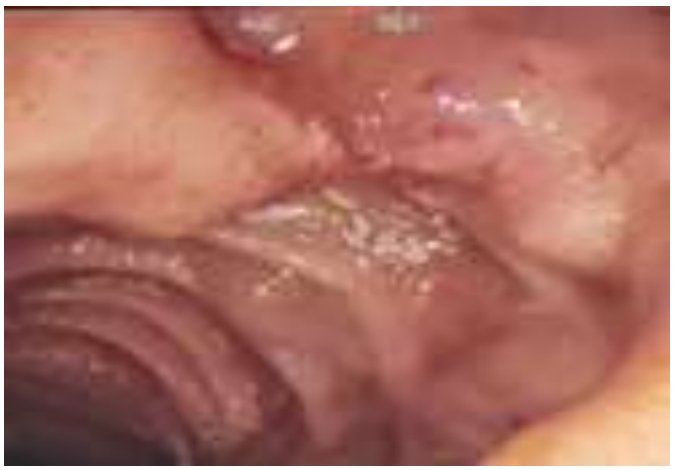

Figure 1: Ectopic duodenal varices with red signs

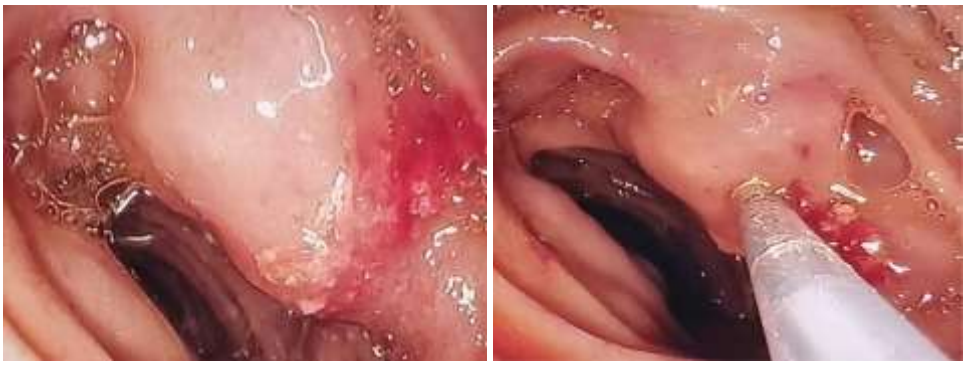

Figure 2: Injection of biological glue into ectopic duodenal varices

DISCUSSION

Ectopic varices account for 1 to 5% of all hemorrhages due to variceal rupture [1]. They are mainly found in the digestive tract (small intestine, colon, rectum, and enterostomies) but have also been described in the peritoneum, biliary tract, vagina, and bladder. Their prevalence does not exceed 1 to 3% in patients with cirrhosis but may be as high as 20 to 30% in patients with portal hypertension of extra-hepatic origin [2]. A history of abdominal or pelvic surgery predisposes to the risk of ectopic varicose veins due to the large venous communications that develop between the systemic and portal venous circuits via post-operative adhesions [1,3-5]. Duodenal varices are preferentially associated with extrahepatic portal hypertension and occur in 40% of cases in patients with a history of abdominal surgery [2]. In a review of 169 cases of hemorrhage from ectopic varices, 17% were of duodenal origin, 17% were of jejunal or ileal origin, 14% were colonic, 9% peritoneal and 8% rectal. Hemorrhage was peristomal in 26% of cases, and very rarely came from other sites (ovaries, vagina) [1]. Our patient underwent surgery for cholecystectomy that showed she already had portal

hypertension and the upper endoscopy confirmed the ectopic duodenal varices.

Hemorrhage from ruptured ectopic varices is often life-threatening, with a mortality rate of up to 35% [3]. The clinical picture of ectopic variceal rupture generally combines signs of hypovolemic shock with melena or profuse rectal bleeding. Haematemesis may also occur in cases of duodenal variceal rupture. Ectopic variceal rupture should be suspected in patients presenting with gastrointestinal bleeding in the context of portal hypertension, in whom upper and lower gastrointestinal investigations have not revealed the origin of the bleeding [1].

Diagnosing and managing ectopic varices are complex, requiring a multidisciplinary approach. The investigation of digestive hemorrhage relies on endoscopy. A negative upper endoscopy and colonoscopy should lead to the investigation of the small intestine using an endoscopic video capsule. Abdominal CT scan angiography is a very good alternative for investigating GI hemorrhage, especially in an emergency and when endoscopy is inconclusive or unavailable. It enables a positive diagnosis, localizes the site of bleeding, makes an aetiological diagnosis, and carries out even prior vascular mapping if an endovascular procedure is indicated [6-8]. Our endoscopic and imagery investigations allowed the diagnosis and treatment of the duodenal varices.

Therapeutic options are various and depend on the expertise of the center. Endoscopic treatment is indicated in cases of active hemorrhage and may be combined with TIPS (transjugular intrahepatic portosystemic shunt). Retrograde transvenous obliteration of varices has also been reported [10]. Surgery consists of resection of the collaterals with ligation and/or surgical portosystemic bypass. Surgery is associated with high morbidity and mortality, especially as it is generally performed on patients with advanced cirrhosis. In our case, the patient (03 weeks after glue injection) presented with two attacks of haematemesis in less than 24 hours, one of which was fulminant with hemorrhagic shock before she benefited from a scheduled surgical portosystemic shunt.

CONCLUSION

Ectopic varices are a rare cause of GI hemorrhage in patients with portal hypertension. They predominate in the digestive tract. Portal cavernoma and adhesions resulting from abdominal or pelvic surgery are the main cause of ectopic varices. Their management is difficult and requires the skills of endoscopists, interventional radiologists, and surgeons.

Author Contribution Statement:

1. Made a substantial contribution to the concept or design of the work; or acquisition, analysis or interpretation of data.
2. Drafted the article or revised it critically for important intellectual content.
3. Approved the version to be published.

Declarations:

Ethical Approval: Informed consent to participate and consent to publish was obtained for this case report.

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