CASE REPORT

Mesenteric arteriovenous fistula causing portal hypertension and bleeding duodenal varices

João Baranda, José M. Pontes, Francisco Portela, Luís Silveira*, Pedro Amaro, Paula Ministro, Albano Rosa, Isidoro Pimenta, Paulo Andrade, António Bernardes*, João Pereira*, Maximino C. Leitão, António Donato and Diniz de Freitas

We report a case of portal hypertension associated with a non-traumatic arteriovenous fistula, presenting with bleeding duodenal varices. The patient was admitted for melaena. Emergency endoscopy showed oesophageal varices with no signs of recent bleeding and with no blood in the upper gastrointestinal tract. Arteriography of the coeliac axis and superior mesenteric artery failed to detect any bleeding source. Endoscopy was repeated because of persistent bleeding and revealed active bleeding from varices in the distal duodenum. The patient underwent surgery and a large paraduodenal varicose vein associated with an arteriovenous fistula was found. Resection of the paramural varix and surgical occlusion of the arteriovenous fistula were effective in the control of bleeding. Liver biopsy revealed mild portal fibrosis without cirrhosis. Three years after surgery the patient still has oesophageal varices but has not had recurrent bleeding. There was regression of intraduodenal varices.

European Journal of Gastroenterology & Hepatology 1996, 8:1223–1225

Keywords: portal hypertension, splanchnic arteriovenous fistula, duodenal varices

Introduction

Prehepatic portal hypertension caused by splanchnic arteriovenous fistulas (AVF) are relatively rare, with around 200 cases reported in the literature [1].

We report a case of portal hypertension due to an AVF between the left colic artery and a branch of the inferior mesenteric vein, associated with bleeding duodenal varices.

Bleeding from duodenal varices is uncommon in itself [2], but when occurring in association with portal hypertension of a rare aetiology such as in our patient, it represents an exceedingly rare case. Indeed, we did not find any similar case reported in the literature.

Case report

A 69-year-old white man, retired from the metallurgical industry, was admitted for melaena. He had a history of heavy alcohol consumption (around 100 g/day), was a non-smoker and there was no recent intake of non-steroidal anti-inflammatory drugs. There was no history of previous gastrointestinal bleeding, surgery, abdominal trauma or foreign travel.

On admission, he presented with signs of acute anaemia with hypotension, cold extremities and profuse sweating. Physical examination showed abdominal collateral veins and splenomegaly, with no stigmata of chronic liver disease and no hepatomegaly. Laboratory evaluation showed haemoglobin 56 g/l, prothrombin time 14 s (control 12), creatinine 212 µmol/l, total bilirubin 22 µmol/l; alanine aminotransferase (ALT) 28 UI/l (normal < 20), aspartate aminotransferase (AST) 80 UI/l (normal < 20), gamma glutamyl transferase (γGT) 46 UI/l (normal < 38), alkaline phosphatase 43 UI/l (normal 20–90).

Emergency upper endoscopy showed grade II oesophageal varices with no signs of recent bleeding, erosive gastritis and bulbitis, and no blood in the lumen. Persistence of melaena and haematochezia prompted endoscopy to be repeated, showing the same findings. A total colonoscopy revealed angiodysplasia in the sigmoid colon, without signs of bleeding.

Arteriography of the coeliac axis and superior mesenteric artery during venous stages showed varices in the gastric fundus, without any bleeding source.

From the Departments of Gastroenterology and *Surgery I, Coimbra University Hospital, Coimbra, Portugal.

Requests for reprints to João Baranda MD, Department of Gastroenterology, Coimbra University Hospital, Praceta Prof. Mota Pinto, 3049 Coimbra Codex, Portugal.

Date received: 21 May 1996; revised: 1 August 1996; accepted: 30 August 1996.

A third endoscopy was performed with further progression of the endoscope beyond the second portion of the duodenum, showing actively bleeding duodenal varices (Fig. 1). Efficiency of endoscopic haemostasis was considered doubtful and the patient underwent surgery.

Laparotomy demonstrated a large varicose vein near Treitz's arch, supplied by a branch of the inferior mesenteric vein showing fistulas with the left colic artery. The paramural component of the varix was resected (Fig. 2). AVF and communicating vessels with intraluminal varices were ligated. A per-operative liver biopsy showed moderate macrovesicular steatosis with portal fibrosis, without septa or cirrhotic nodules.

Recovery after surgery was uneventful and there was no recurrent bleeding.

Serum markers of hepatitis B and C, auto-antibodies and stool examination for parasites were negative. Iron kinetics was normal. Follow-up endoscopy was performed every 6 months and showed persistence of oesophageal varices and regression of intraduodenal varices. This finding, associated with the superficial per-operative biopsy, prompted the execution of a percutaneous liver biopsy 6 months later. Histopathology confirmed portal fibrosis without septa or nodules, and showed no steatosis.

One and a half years after the bleeding episode, a primary adenocarcinoma of the lung was diagnosed. It invaded the pleura and the trachea and was found to be unresectable. The patient was treated with chemotherapy.

At present, 3 years after the first admission, the patient is in reasonable general condition, complaining only of cough.

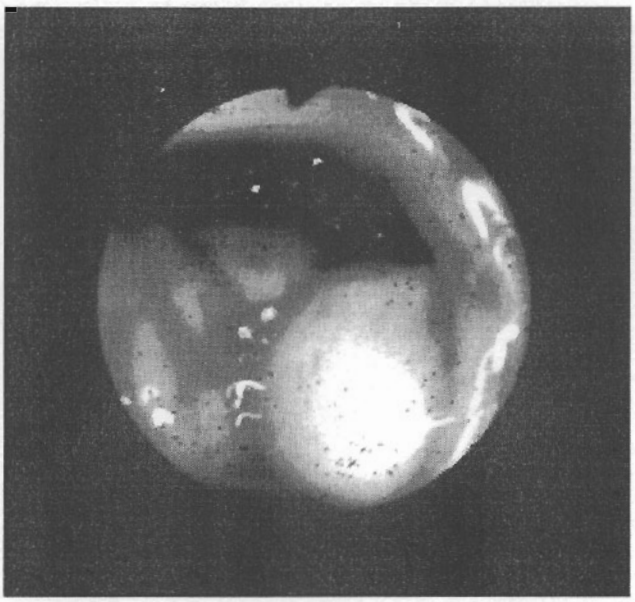

Fig. 1. Endoscopy: duodenal varices with oozing bleeding.

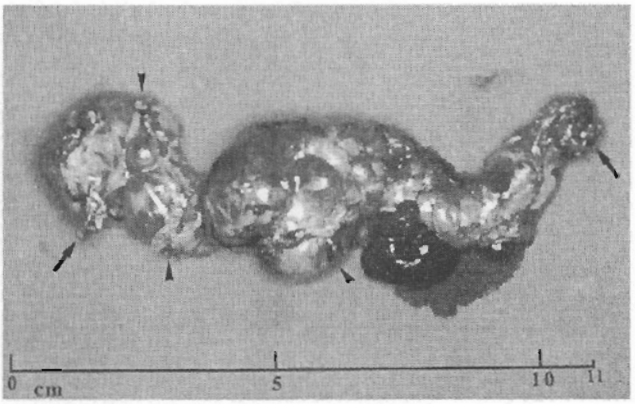

Fig. 2. Resected paraduodenal varix. Communicating vessels with inferior mesenteric vein and left colic artery were ligated (arrows). Supplying veins penetrating the duodenal wall were also ligated (arrowheads). Finally, a large extramural varix was resected.

Discussion

Portal hypertension is the main feature of splanchnic AVF, occurs in more than half of patients with this condition and is known as high-flow hypertension in this setting [1,3]. Pathogenesis is explained by an increased portal blood flow and adaptive portal fibrosis that gradually develops as a result of arterialization of the portal system [4].

These fistulas develop most commonly between the hepatic or splenic arteries and the portal vein or its branches [5]. AVFs involving the inferior mesenteric vessels are rare, only seven cases being reported in the literature [1,6]. In most cases, AVFs are associated with aneurysm rupture or are secondary to traumatic lesions such as surgical procedures or penetrating wounds [5]. Hepatoportal fistulas represent one of the commonest splanchnic AVFs and may result from liver biopsy; or they may be associated with hepatocellular carcinoma [3]. A case of portal hypertension secondary to an AVF in a neuroendocrine tumour of the pancreas has also been reported [7].

AVFs have been considered congenital in very few cases [1,6,8].

The present case has two rare features: an AVF, probably congenital, between branches of the inferior mesenteric vessels, and the development of bleeding duodenal varices

Angiography, although considered to be a first line method for diagnosing AVF, failed to demonstrate the fistula in our patient because a selective catheterism of the inferior mesenteric artery was not performed.

Although endoscopic sclerotherapy has been used in bleeding duodenal varices [9], we decided to send the patient to surgery since experience with endoscopic therapy is very limited. Resection of the paramural varix and surgical ligation of AVF and supplying vessels were effective in controlling bleeding, although regression of oesophageal varices was not achieved. This finding is compatible with long-standing portal hypertension [1,4], secondary to a probably congenital AVF.

To our knowledge, the association of splanchnic AVF and adenocarcinoma of the lung has not been reported in the literature. This association should only reflect a coincidence.

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