

Case Report

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A rare case of post endoscopic retrograde cholangiopancreatography bleeding: Visceral arterial aneurysms

Dinesh Koirala*; Rahul Pathak; Brindeswari Kafle Bhandari; Anurag Jha; Rabin Hamal; Rajesh Pandey; Arun Gnawali

Department of Gastroenterology and Hepatology, Maharajgunj Medical Campus, TUTH, Institute of Medicine, Kathmandu, Nepal.

*Corresponding Author: Dinesh Koirala

Department of Gastroenterology and Hepatology,
Maharajgunj Medical Campus, TUTH, Institute of
Medicine, Maharajgunj, Nepal.
Email: dkoirala045@gmail.com

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Abstract

Endoscopic Retrograde Cholangiopancreatography (ERCP) leading to arterial vascular complication is exceedingly rare. This case report describes a life threatening hemobilia, from a pseudoaneurysm of the pancreaticoduodenal artery, which occurred post ERCP. The pseudoaneurysm was diagnosed by selective angiography of the superior mesenteric artery, and successfully treated with polyvinyl alcohol particle embolization.

Keywords: ERCP; Complication; Hemobilia; Angiography; Pseudoaneurysm; Embolization.

Introduction

The most frequent complications related to endoscopic retrograde cholangiopancreatography (ERCP) and endoscopic biliary sphincterotomy are pancreatitis, cholangitis, hemorrhage, and duodenal perforation in descending order. Post-ERCP pancreatitis occurs in the ranges from 1% to 6% in general population with rates approaching almost 30% in certain groups of high risk population [1]. Other less reported adverse events include cardiopulmonary complications, contrast allergy, impaction of retrieval basket, and formation of pseudoaneurysm [1]. There are many case reports of visceral artery pseudoaneurysm. Overall, it mostly develops from pancreaticoduodenal artery esp. after sphincterotomy.

Visceral artery aneurysms (VAAs) may occur with a wide range of manifestations and prognostic significance. There are two types of VAAs: true and pseudo aneurysm. True aneurysms result from intrinsic vessel wall defects, whereas pseudoaneurysms are often precipitated by underlying traumatic or inflammatory etiologies such as pancreatitis, vascular or laparoscopic interventions, and surgery [2]. We present a case of a pseudoa-

neurysm of the pancreaticoduodenal artery developed after sphincterotomy.

Case presentation

A 53-year-old woman presented with a 2-week history of jaundice and intermittent pain abdomen. Biochemical findings indicated obstructive jaundice. Her physical examination was normal. Ultrasound examination demonstrated cholelithiasis with dilated common bile duct (CBD). Magnetic Resonance Cholangiopancreatography (MRCP) was done which showed approximately 11 mm diameter calculus in the CBD with dilated CBD of 12 mm at the widest segment with dilated intrahepatic bile ducts. Following this, ERCP was done. CBD was cannulated and cholangiogram was done which showed dilated CBD of approximately 15 mm with a filling defect of 10 mm in distal CBD. Stone extraction was tried but could not be done despite adequate sphincterotomy. Double pig tail (DPT) stent of 10 Fr X 8 cm was kept in situ with plan of reattempting ERCP for stone extraction after a month. During ERCP pancreatic duct was not cannulated. Patient was stable post procedure with plan of discharge the next day. But patient developed hemateme-

sis at night. There was an abrupt drop in hemoglobin. Patient was stabilized with blood transfusion and upper gastrointestinal (UGI) endoscopy was planned. UGI endoscopy revealed large blood clot at sphincterotomy site but no active bleeding at that moment. Adrenaline injection with sclerotherapy needle was done around the clot but the clot was not dislodged. Patient was kept for observation. But the following night patient developed further hematemesis with drop in hemoglobin as well as hypotension and tachycardia. Blood transfusion was done and patient resuscitated adequately. Given the severe nature of hemorrhage, computerized tomography (CT) angiography was done with plan to do angioembolisation if bleeding source could be identified. CT angiogram revealed focal contrast filled outpouching measuring 4 X 4 mm adjacent to duodenal papilla which appeared to be continuous with the vessel in this region but the exact course of vessels could not be traced, thus likely giving the diagnosis of pseudoaneurysm of branch of pancreaticoduodenal artery without extravasation of contrast. With the help of interventional radiologist embolization of aneurysm was done with polyvinyl alcohol (PVA) particle. Patient was kept in observation for 72 hours but she had no further episodes of GI bleeding. Her hemoglobin was normal. Patient was observed for 48 hours in the ward and was discharged.

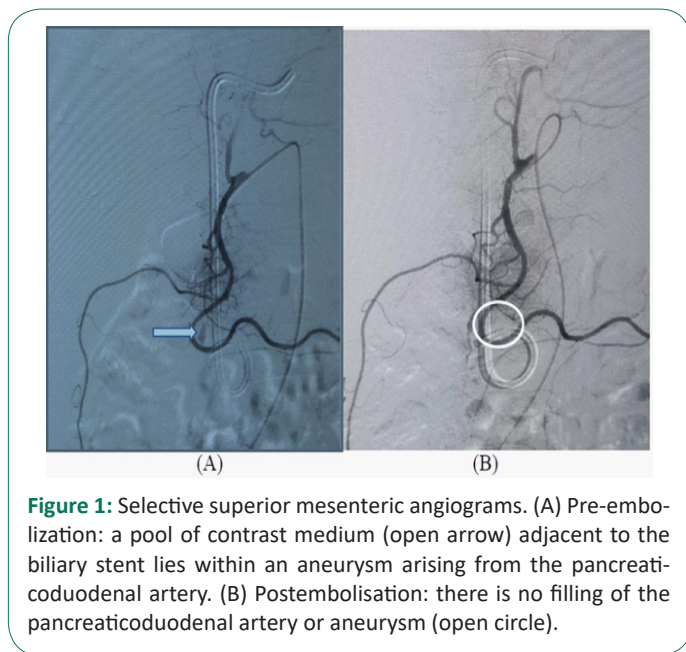


Figure 1: Selective superior mesenteric angiograms. (A) Pre-embolization: a pool of contrast medium (open arrow) adjacent to the biliary stent lies within an aneurysm arising from the pancreaticoduodenal artery. (B) Postembolisation: there is no filling of the pancreaticoduodenal artery or aneurysm (open circle).

Discussion

ERCP procedure has been associated with many complications among which pancreatitis is the most common occurring in almost 2% of ERCPs [3]. Other common complications are cholangitis, perforation and hemorrhage. Post-ERCP bleeding can be defined as clinically significant or nonclinically significant, and can be graded as mild, moderate, or severe based upon a consensus

Definition [4]:

- Mild: clinical evidence of bleeding (i.e. not just endoscopic), hemoglobin drop <3 g/dL, and no need for transfusion
- Moderate: need for transfusion (4 units or less), no angiographic intervention or surgery

- Severe: transfusion of 5 units or more, or intervention (angiographic or surgical)

Approximately one-half of bleeding complications occur immediately after sphincterotomy while others have a delayed presentation ranging from 24 hours up to several days. Delayed bleeding can occur up to 2 weeks post procedure [4]. Bleeding is mostly due to intraluminal cause and rarely ductal and vessel related. In a meta-analysis of 21 prospective trials, the rate of hemorrhage as a complication of ERCP was 1.3% (95% CI, 1.2%-1.5%) with mild bleeding occurring in 70% episodes [5]. Severe bleeding following ERCP has been reported in 0.1-0.5% of cases [1]. Hemorrhage is supposed to be the complication of sphincterotomy rather than the procedure itself. The risk of severe hemorrhage is estimated to occur in fewer than 1 per 1000 sphincterotomies [4]. The presence of melena, hematochezia, or hematemesis associated with a hemoglobin level decrease of at least 2 g/dL and/or the need for a blood transfusion signals a major bleeding episode and requires intervention [6]. Other factors believed to be associated with bleeding include coagulopathy, the use of anticoagulants within 72 hours of sphincterotomy, the presence of acute cholangitis, papillary stenosis, the use of precut sphincterotomy, and low case volume of the endoscopist (i.e. 1 sphincterotomy per week or less) [6]. Factors such as aspirin and nonsteroidal anti-inflammatory drug (NSAID) use, ampullary tumors, long sphincterotomy, and extension of prior sphincterotomy were not associated with a greater risk of postsphincterotomy bleeding, whereas liver cirrhosis, dilated common bile ducts, periampullary diverticulum, precut sphincterotomy, and common bile duct stones appear to increase the risk of postsphincterotomy bleeding [7].

The pancreaticoduodenal artery crosses the bile duct on average 3.75 cm from the tip of the papilla, which is well out of the range of a sphincterotomy. However, in 4% of autopsy specimens examined this artery was within range of the sphincterotomy [8]. It is probably this small group of patients who are at risk of hemorrhage. Special attention should be paid to the anatomy of arterial supply to the major papilla when performing a sphincterotomy to reduce the risk of bleeding. The 11-1 o'clock arc above the duodenal papilla reportedly has the least concentration of arteries, which is the currently the most common practiced site of cutting for performing biliary sphincterotomy to minimize the risk of bleeding [3]. Special attention should be given to identify coagulopathy (international normalized ratio >1.5) or a low platelet count (<50,000/cu mm) which are risk factors for bleeding [4]. Accurate positioning, proper electrocautery technique, and prevention of long, erratic cuts during sphincterotomy (referred to as "zipper") are also equally important.

Treatment options for postsphincterotomy bleeding consist of injection of dilute epinephrine solution through a sclerotherapy needle, balloon tamponade, heater probe or bipolar coagulation, placement of endoscopic clips and self-expandable metal stents (SEMS) [4,6]. Most episodes of bleeding stop spontaneously; thus, treatment should be reserved for patients who have clinically significant and severe bleeding. Injection of epinephrine solution through a sclerotherapy needle around the bleeding site or at the apex of the sphincterotomy incision is the most commonly utilized method of hemostasis and is quite effective in majority of cases. When endoscopic treatment mo-

dalities fail in a clinically significant bleeding (i.e. hemodynamically unstable), the patient may require to undergo selective mesenteric angiography together with embolization or even open surgery if angiography and embolization not possible or unsuccessful.

Visceral artery aneurysm (VAA) formation is one of the very rare complications of ERCP, resulting directly from traumatic injury to the visceral arteries during pancreaticobiliary manipulations or indirectly from post ERCP pancreatitis [9]. Pseudoaneurysms occurs due to injury to one or multiple layers of vessel wall. There are only few case reports of ERCP leading to formation of VAA. Sphincterotomy and stent placement are the most common maneuvers resulting in the formation of VAA. Visceral angiography is the gold-standard for the diagnosis of VAA and their rupture [2].

Pancreaticoduodenal artery aneurysms (PDAs) are rare, accounting for 1-2% of all visceral aneurysms [10]. True PDAs are usually associated with stenosis of the coeliac axis, whereas pseudo/false PDAs commonly occur as a complication of pancreatitis [10]. Pseudoaneurysms occur as a result of damage to the wall of an artery due to multiple causes. In our case this probably occurred during sphincterotomy. It has been observed that pseudo PDA as a complication of pancreatitis usually arises from the superior pancreaticoduodenal artery, whereas it arises from the inferior pancreaticoduodenal artery as a complication of ERCP. Furthermore, if the aneurysm is rapidly arising without a significant history of pancreatitis, it is most likely due to complication of ERCP [10]. Our patient rapidly developed a 4 X 4 mm aneurysm and urgent treatment was indicated to stop further bleeding. Mortality may occur in up to 49% with rupture and bleeding. Mortality may occur in 19% of surgical repair patients [11]. Surgery has largely been replaced by angiography and embolization, which has a lower procedure mortality and high success rate. This approach had a successful outcome in 15 of 19 patients (79%) with peripancreatic aneurysms [12].

Conclusion

This report documents that pseudoaneurysm formation is a potential complication of endoscopic sphincterotomy during ERCP. Pseudoaneurysm rupture is one of the life-threatening but rare cause of post ERCP bleeding which should always be borne in mind in a case of post ERCP bleeding. In a case of significant and severe bleeding, angiography should be done at a low threshold. Urgent referral for angiographic embolization is advised in such situation to avoid aneurysmal rupture and bleeding which could be lifesaving to the patient.

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