

Case Report

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First report of an appendiceal perforation caused by a migrated biliary stent after orthotopic liver transplantation

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Introduction

The overall incidence of biliary strictures in liver transplanted patients is around 13% [1], mostly located at the biliary anastomosis, occurring from 6 to 12% of cases [2]. The first treatment option of anastomotic biliary strictures is the insertion of a plastic or metallic biliary prosthesis with endoscopic retrograde cholangiopancreatography (ERCP). Distal migration of biliary stent is an unusual complication related to this procedure, occurring in about 6% of patients [3], but in literature cases of abdominal visceral perforation due to this complication are described, mainly involving small intestine and colon [4-6]. An isolated case of appendiceal perforation related to stent dislocation is found in literature [6].

Case report

A 63-year-old male patient with a history of orthotopic liver transplantation in June 2021 underwent an ERCP procedure in October 2021 for the treatment of anastomotic biliary stricture. Three months later, he came to the emergency department for acute onset abdominal pain and localized peritonitis in the right lower quadrant, emesis, anorexia, and acute kidney failure. Blood tests showed neutrophile leukocytosis, elevated C-reac-



Figure 1: CT scan showing the dislocated biliary stent in the right iliac fossa with the tip extruding from an oedematous and inflamed appendix within a collection (black arrow).

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tive protein (400 mg/ml) and preserved liver function. A plain abdominal X-ray detected a foreign body in the right iliac fossa, more likely the dislocated biliary prosthesis. An abdominal CT scan with contrast enhancement confirmed the dislocation of the biliary prosthesis in the right iliac fossa and the presence of free abdominal air and multiple fluid-filled collection [Figure 1]. Suspicion of cecal or appendiceal perforation was posed. Therefore, the patient underwent an urgent exploratory laparotomy that showed appendiceal perforation caused by the dislocated biliary stent (Figures 2 and 3); the stent was extracted, appendicectomy performed and the peritoneal cavity washed to evacuate the abscesses. Post-operative course was featured with ileus and the need of a second-look laparotomy for wash out of residual fluid collection. Two years after surgery the patient is alive and fully recovered.

Discussion

Dislocation of biliary stent is an uncommon complication related to ERCP with an incidence of 3-6%. It can migrate proximally or distally in the biliary duct. Distal migration is more likely in case of benign disease, papillary stenosis, use of straight stents (compared to pigtail), sphincterotomy, common bile duct diameter >10 mm and use of larger stents [7]. In 43% of cases, migrated biliary prosthesis are spontaneously eliminated with evacuation [8]. When endoscopic access is available, endoscopic treatment of migrated biliary stent becomes the first choice [9,10]. Intestinal perforation is a rare consequence of biliary prosthesis distal migration [11]. Extrinsic fixation of the bowel secondary to postoperative bowel adhesion, diverticulosis, hernia, or stricture is a risk factor for stuck biliary stent in the intestinal wall, increasing the chance of visceral perforation [12,13]. Intestinal perforation requires prompt investigation and urgent surgical treatment for stent removal and drainage of intra-peritoneal or retroperitoneal abscesses [14]. Duodenum is the most frequent site of perforation reported in literature [15]. Perforations in the liver, sigmoid colon, caecum, and pancreas are also described in different studies [4,16]. As far as we are concerned, only one case report of an appendiceal perforation occurred after ERCP in a patient affected by chemotherapy-induced neutropenic enterocolitis is described in literature. A conservative treatment with broad-spectrum antibiotics, fluid resuscitation, correction of hematological parameters was the first choice due to the high surgical and anesthesiologic risk of the patient; after an unsuccessful endoscopic attempt, it was surgically removed and appendicectomy was performed with local lavage and drain placement [6]. So far, in English literature, there is no report of appendiceal perforation due to biliary stent migration in a liver transplant recipient. We believe that in the setting of iatrogenic immunosuppression after liver transplantation, the possible consequences of appendiceal perforation could be catastrophic and the management of these patients require high suspicion index to diagnose the condition, prompt definition of therapeutic plan and fine post-operative management, including modulation of immunosuppression in order to balance the risk of overwhelming sepsis and rejection, especially if the perforation occurs in the very first months after transplantation.



Figure 2: The biliary stent perforating the appendix (black arrow).



Figure 3: The appendix with the site of perforation at mid-body level (black arrow).

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