ISSN 2689-8268 | Volume 9

American Journal of Surgery and Clinical Case Reports

Case Report Open Access

Multiple Complicated Hepatoduodenal Fistulas Following Previous Hepatic Hydatid Cyst Surgery: A Rare Case Report

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Received: 18 June 2025
Accepted: 30 June 2025
Published: 05 July 2025
J Short Name: AJSCCR

Citation:

Atul Kapoor. Multiple Complicated Hepatoduodenal Fistulas Following Previous Hepatic Hydatid Cyst Surgery: A Rare Case Report. Ame J Surg Clin Case Rep. 2025; 9(1): 1-6

Keywords:

Hepatoduodenal Fistula; Hydatid Cyst; Hepatobiliary Surgery;Endoscopy; MRCP; CECT

1. Abstract

1.1. Background

Hepatoduodenal fistula is an extremely rare complication following hepatobiliary surgery, particularly after hydatid cyst treatment. The condition presents diagnostic challenges due to its nonspecific clinical presentation and delayed onset.

1.2. Case Presentation

We report a case of a 60-year-old female who presented with abdominal pain, fever, and mild jaundice five years after left hepatic lobe hydatid cyst surgery. Diagnostic workup including upper endoscopy, contrast-enhanced computed tomography (CECT), and magnetic resonance cholangiopancreatography (MRCP) confirmed the presence of multiple hepatoduodenal fistulas with associated cholangitis and liver abscesses.

1.3. Conclusion

This case highlights the importance of considering late complications following hydatid cyst surgery and demonstrates the utility of multimodal imaging in diagnosis. Early recognition and appropriate management are crucial for optimal patient outcomes.

2. Introduction

Hepatoduodenal fistula represents an abnormal communication between the liver and duodenum and is extremely rare, with an incidence of less than 0.1% of all gastrointestinal fistulas [1]. These complications are predominantly iatrogenic, occurring as sequelae of hepatobiliary procedures, particularly hydatid cyst surgery [2]. The clinical presentation is often nonspecific, leading to diagnostic delays and potential complications [3]. The occurrence of hepatoduodenal fistula is extraordinarily rare, and the presentation of multiple simultaneous hepatoduodenal fistulas represents an even more exceptional clinical entity. While single hepatoduodenal fistulas have been sporadically reported in the literature, mostly in association with aortoduodenal fistulas or post-cholecystectomy complications [4,5], the simultaneous presence of multiple discrete fistulous communications between the hepatic parenchyma and duodenum following hydatid cyst surgery has not been previously described in the published literature. A comprehensive review of case reports over the past two decades reveals that most hepatobiliary fistulas are singular communications, with multiple fistulas being exceedingly uncommon and typically associated with complex inflammatory conditions or extensive surgical trauma [6]. This case therefore represents a unique contribution to the literature, documenting the first reported instance of multiple hepatoduodenal fistulas as a delayed complication of hydatid cyst surgery.

3. Case Presentation

A 60-year-old female presented with a 2-week history of abdominal pain, fever, and mild jaundice accompanied by vomiting. She had undergone hydatid cyst surgery of the left hepatic lobe five years previously with an uneventful postoperative course at that time.The patient reported epigastric pain radiating to the back, associated with low-grade fever and progressive jaundice. There was no relationship to meals, and she denied weight loss or changes in bowel habits. physical examination revealed normal vital signs with mild jaundice. Abdominal examination showed an ill-defined swelling in the epigastric region with a

surgical scar from the previous procedure in the epigastric and right hypochondriac regions. Complete blood count was normal. Liver function tests showed elevated serum bilirubin (2.3 mg/dL), alkaline phosphatase (324 U/L), and transaminases (SGOT/SGPT: 88/67 U/L). Inflammatory markers including CRP and ESR were elevated. MRCP demonstrated dilated intrahepatic biliary ducts with irregularity of the left hepatic duct and ill-defined luminal defects. The posterior branch of the right hepatic duct was not clearly defined. Multiple hyperintense rounded lesions were seen in the right lobe. The D1 part along with the pylorus appeared to be pulled toward the liver hilum, while the common bile duct appeared normal (Figure 1). CECT findings with intravenous and oral contrast revealed pneumonia with oral contrast visualized in the left lobe of the liver, showing a 6×5.5 cm ill-defined hypo attenuated area (Figure 2). Upper GI endoscopy findings revealed three fistulous openings at the duodenal level with mild inflammatory changes at D1 and mild luminal narrowing (Figure 3). Integration of clinical, endoscopic, and radiological findings confirmed the diagnosis of multiple hepatoduodenal fistulas with cholangitis and liver abscesses, with gastric contents in the liver suggesting a reverse-functioning fistula.A multidisciplinary team approach was employed, involving hepatobiliary surgeons, gastroenterologists, and interventional radiologists. After extensive discussion of conservative versus surgical management options, the patient was counselled about treatment alternatives and consented to initial conservative management with close monitoring.The patient was started on broad-spectrum antibiotics for cholangitis, nutritional support, and symptomatic management. Conservative measures included bowel rest, parenteral nutrition, and treatment of associated infections. Endoscopic Treatment Options along with surgical approaches were discussed as second stage options which are elaborated in the discussion below. A long-term follow-up plan was established with serial imaging and clinical assessments to monitor for spontaneous closure or need for escalation to endoscopic or surgical intervention.

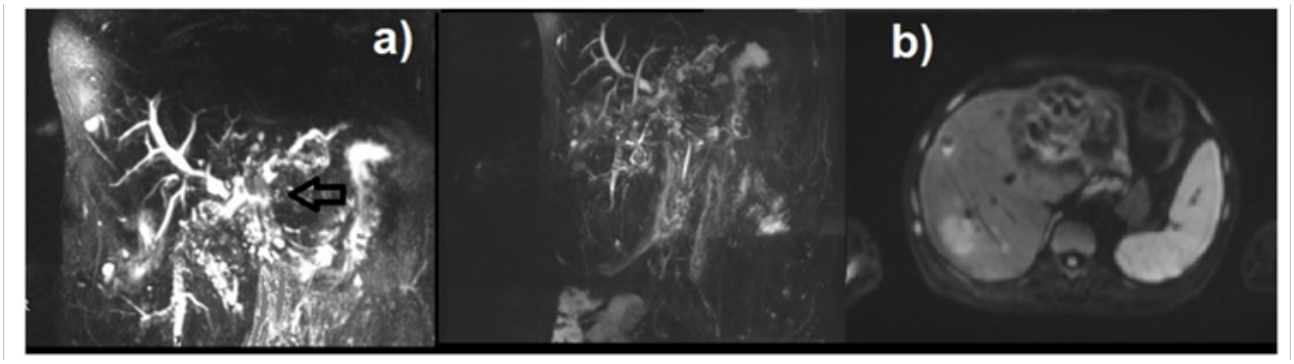

Figure 1: a):MRCP coronal image showing irregular dilated left hepatic duct with non-filling of right hepatic ducts with narrowing at hila with pulled up bulb(arrow). b) Plain diffusion MR axial image showing multiple biliary abscesses.

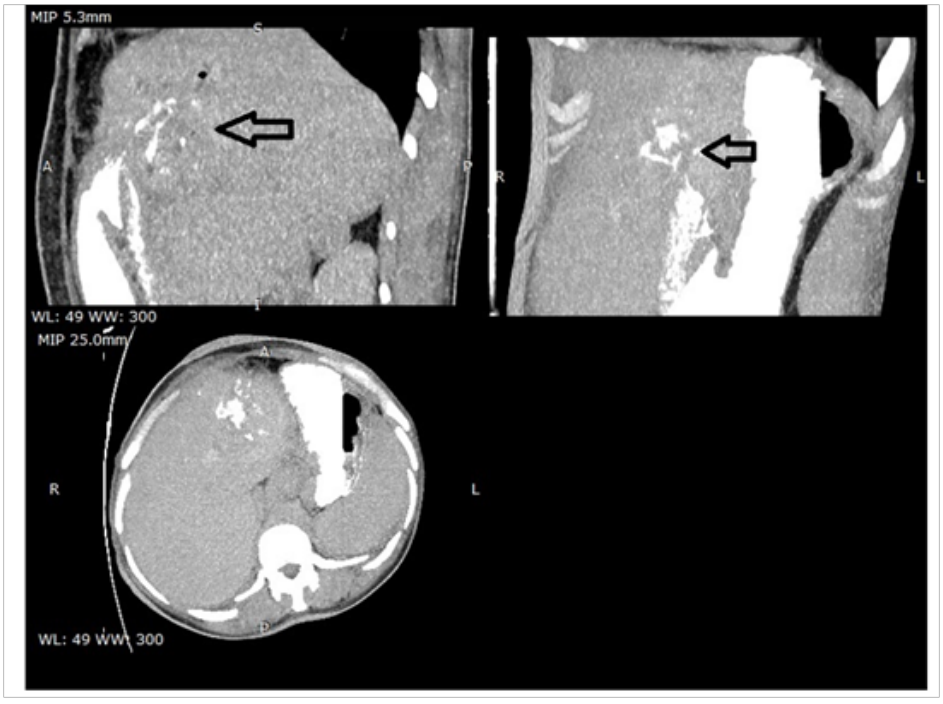

Figure 2: Multiplanar images of CECT with oral contrast showing oral contrast in the left lobe of liver with biliary extension(arrows).

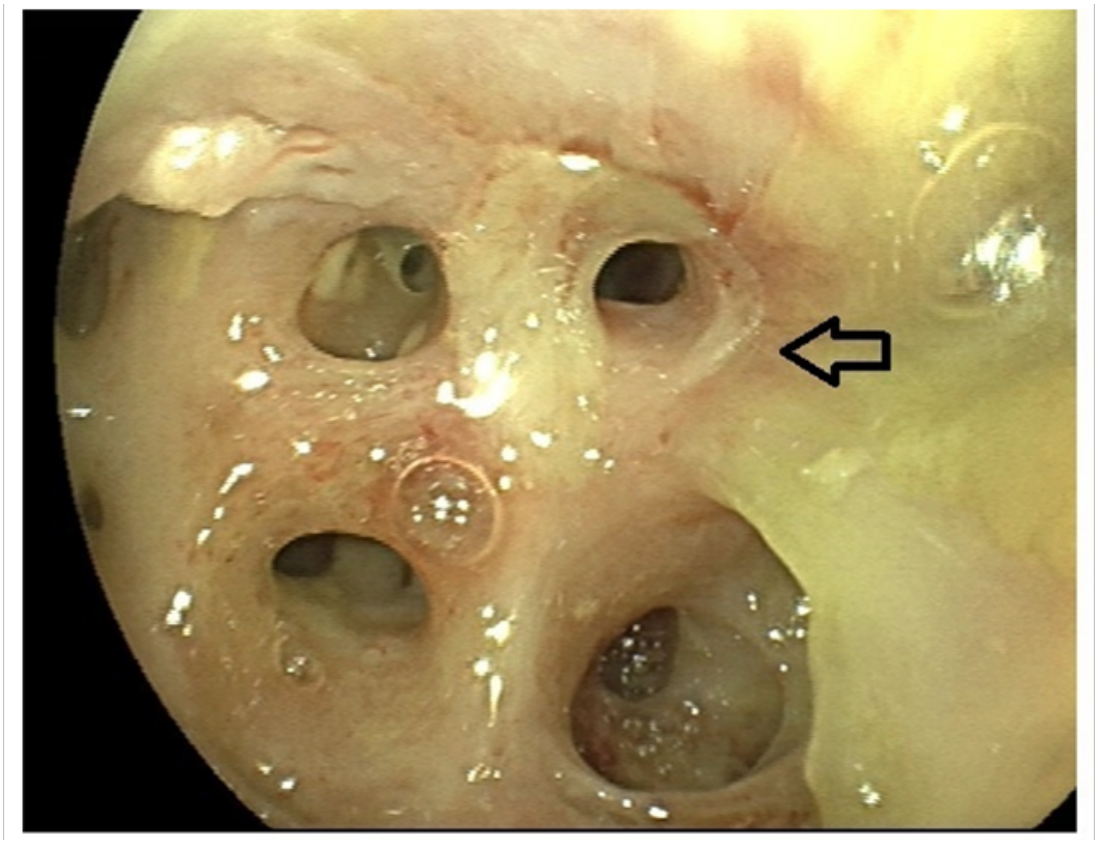

Figure 3: Endoscopic image showing multiple openings of fistulas in the Duodenal bulb(arrow).

4. Discussion

Hepatoduodenal fistula following hydatid cyst surgery represents one of the rarest complications in hepatobiliary surgery. The case highlights the need to discuss the possible etiology and pathophysiological considerations. The etiology and risk factors include foremost being: Iatrogenic causes including post-surgical complications following hepatobiliary procedures, hydatid cyst surgery complications, and liver resection complications [7]. Non-iatrogenic causes encompass penetrating trauma, inflammatory conditions, and malignancy, though these are rare [8]. The suggested predisposing factors include previous hepatic surgery, postoperative complications such as infection or bile leak, and the anatomical proximity of the liver to the duodenum [9]. Patholphysiologic considerations in our case leading to the development of hepatoduodenal fistula following hepatic hydatid cyst surgery involves several interconnected pathophysiological processes:Primary mechanisms include surgical trauma with direct injury to hepatic parenchyma during cyst evacuation, disruption of normal anatomical planes, and creation of potential spaces for abnormal communication [10]. The increased intracystic pressure theory suggests that when pressure exceeds ductal or tissue pressure, gradual erosion occurs at contact points between the cyst wall and adjacent organs [11]. Secondary contributing factors involve adhesion formation creating abnormal tissue planes, ischemic changes compromising blood supply, and secondary bacterial infection leading to abscess formation with erosion into adjacent organs. Kumar et al. reported a similar case of hepatoduodenal fistula as a complication of liver abscess, emphasizing the rarity of this condition [9]. The delayed presentation in our case, occurring five years post-surgery, is particularly noteworthy and highlights the importance of long-term surveillance following hydatid cyst procedures. The delayed formation of hepatoduodenal fistulas following hydatid surgery involves complex mechanisms. Akbulut and Yagmur described how incomplete cyst evacuation can lead to chronic inflammation and eventual erosion into adjacent organs [10]. In our case, the presence of multiple fistulous openings suggests extensive inflammatory processes, possibly related to residual hydatid material or secondary bacterial infection. The delayed presentation five years post-surgery in this case exemplifies the chronic inflammatory pathophysiology underlying hepatoduodenal fistula formation. The initial hydatid cyst surgery likely created microscopic tissue injury and altered the normal anatomical planes between the liver and duodenum. Over time, several interconnected pathophysiological processes contributed to fistula development. First, residual hydatid material or incomplete cyst wall removal may have triggered a persistent foreign body inflammatory response, characterized by chronic granulomatous inflammation with macrophage activation and release of inflammatory mediators. This chronic inflammation leads to progressive tissue breakdown through upregulation of matrix metalloproteinases (MMPs), particularly MMP-2 and MMP-9, which degrade extracellular

matrix components and weaken tissue integrity at the hepatoduodenal interface [11]. The presence of pneumobilia and oral contrast within the hepatic parenchyma in our patient indicates established epithelialized fistulous tracts, suggesting that the inflammatory process progressed through distinct phases: initial tissue injury, chronic inflammation with progressive erosion, and finally epithelialization of the abnormal communication. The reverse-functioning nature of the fistulas, with gastric contents flowing into the liver, indicates that intraluminal duodenal pressure exceeded the resistance of the weakened hepatic tissue, creating a pressure gradient that maintained and expanded the fistulous communications. This pathophysiological sequence explains the delayed clinical presentation and the challenging nature of spontaneous closure in such cases [12]. The molecular basis involves an inflammatory cascade with release of pro-inflammatory cytokines (TNF-α, IL-1β, IL-6) and activation of matrix metalloproteinases causing tissue breakdown [3]. Abnormal collagen deposition creates weak tissue planes, while epithelialization of the fistulous tract prevents spontaneous closure [7]. The nonspecific presentation of hepatoduodenal fistulas often leads to diagnostic delays. Pedrosa et al. emphasized the importance of cross-sectional imaging in identifying hydatid disease complications [13]. In our case, the combination of MRCP, CECT, and endoscopy provided comprehensive visualization of the fistulous communications.The presence of pneumobilia on CT imaging is a key diagnostic finding, as described by Gharbi et al. in their seminal work on hydatid disease imaging [14]. The visualization of oral contrast within the hepatic parenchyma confirmed the abnormal communication between the gastrointestinal tract and liver. Since our patient had cholangitis and chronic biliary abscesses conservative management was planned as the first step however there are interventional treatment strategies which are worthwhile discussing. There are endoscopic techniques which have revolutionized the management of gastrointestinal fistulas, offering less invasive alternatives to surgical repair. Endoscopically deployable stents, endoscopic suturing devices, through-the-scope and over-the-scope clips, sealants, and fistula plugs can be used to treat fistulae. These therapies demonstrate enhanced effectiveness when used in combination [15].Over-the-Scope Clip (OTSC) Technology has emerged as a promising technique for hepatoduodenal fistula closure. The OTSC system provides superior compression force compared to through-the-scope clips and can achieve full-thickness tissue approximation [16]. The procedure involves endoscopic identification of the fistulous opening, followed by precise clip deployment to achieve complete closure. Advanced endoscopic suturing devices allow for multi-layer closure of fistulous defects. These endoscopic techniques include gastrointestinal stents, endoscopic suturing, cardiac septal occludes, endo-sponge, vacuum therapy and others. The Apollo OverStitch system enables precise tissue approximation with full-thickness suturing capability, particularly useful for larger defects [17]. Fully covered self-expanding metal stents provide barrier coverage while maintaining luminal patency. However, stent migration remains a concern, occurring in 26-30% of cases, necessitating endoscopic fixation techniques [18]. Biologic Sealants and Plugs. These devices promote natural healing through scaffold formation and can be deployed endoscopically under fluoroscopic guidance [19]. Vacuum-assisted sponge closure is another emerging technique that has demonstrated effectiveness in the closure of postsurgical leaks. EVT involves placement of an open-cell sponge with attached vacuum suction to promote granulation tissue formation and accelerate healing [20]. The available alternative surgical Management Approaches include: Primary Repair Techniques. Direct surgical repair remains the gold standard for complex hepatoduodenal fistulas, particularly when endoscopic approaches have failed [21]. Fistula Resection and Reconstruction Resection with primary anastomosis of the section or sections of bowel containing the fistula is the optimal procedure. This approach involves complete excision of the fistulous tract, followed by primary repair of both hepatic and duodenal defects [22]. Hepatic Resection Strategies For extensive hepatic involvement, formal hepatic resection may be necessary. Roux-en-Y hepaticojejunostomy is the extreme therapeutic option for both conditions. Left hepatic lobectomy or segmental resection can be performed depending on the extent of hepatic damage [23]. Duodenal Repair Techniques Repair of duodenal fistulae by a serosal patch procedure offers an alternative to primary closure in cases where duodenal wall integrity is compromised. The serosal patch technique involves mobilization of healthy jejunal serosa to cover the duodenal defect [24]. Biliary Reconstruction Cholecystectomy with a cheiloplasty is the most frequent treatment of primary fistulas, whereas the bile duct drainage or the endoscopic stenting is the best choice in case of minor iatrogenic bile duct injuries. Complex biliary reconstruction may require Roux-en-Y hepaticojejunostomy to ensure adequate biliary drainage [25]. Minimally Invasive Approaches Most cases of cholecystoduodenal fistula could be dealt with laparoscopically. Laparoscopic repair techniques have been successfully applied to hepatoduodenal fistulas, offering reduced morbidity and faster recovery compared to open procedures [26]. Lastly is the Damage Control Surgery In unstable patients or those with extensive inflammation, damage control approaches may be preferred. The safer procedure in these instances is transection of the intestinal segment proximal and distal to the fistula, leaving the fistula tract in place. This staged approach allows for resolution of acute inflammation before definitive reconstruction [27]. The case also highlights the need to take meticulous surgical care while doing hydatid cyst removal to prevent future fistula formation. Sozuer et al. emphasized the importance of complete cyst evacuation and adequate irrigation to

prevent residual disease [25]. Kayaalp et al. reported that larger cyst size and superficial location increased the risk of biliary complications [5]. The WHO classification system for hydatid cysts provides guidance for treatment selection and may help predict complications [26]. Brunetti et al. developed expert consensus guidelines that emphasize the importance of complete treatment and long-term follow-up [27]. This case underscores the need for long-term surveillance following hydatid cyst surgery. Wen et al. emphasized advances in echinococcosis management but noted that surgical complications remain a significant concern [28]. Development of standardized follow-up protocols and early intervention strategies could improve outcomes.Future research should focus on identifying predictive factors for fistula formation and developing minimally invasive treatment approaches. The role of endoscopic interventions in managing heptametric fistulas requires further investigation.

5. Conclusion

Hepatoduodenal fistula is an extremely rare but serious complication that can occur years after hepatic hydatid cyst surgery. This case demonstrates the importance of maintaining high clinical suspicion in patients with previous hepatobiliary surgery presenting with nonspecific gastrointestinal symptoms. Multimodal imaging including endoscopy, CECT, and MRCP is essential for accurate diagnosis. Early recognition and appropriate management, whether conservative or surgical, are crucial for optimal outcomes. Long-term follow-up is mandatory for patients undergoing hydatid cyst surgery to detect late complications.

6. LearningPoints/Take-Home Messages

1.Hepatoduodenal fistula is an extremely rare but serious complication that can occur years after hepatic hydatid cyst surgery

2.High index of suspicion is required in patients with previous hepatobiliary surgery presenting with nonspecific gastrointestinal symptoms.

3.Several features make this case particularly noteworthy: the delayed presentation five years post-surgery, the presence of multiple fistulous openings, and the reverse-functioning nature of the fistulas with gastric contents in the liver. These findings suggest extensive inflammatory remodeling and highlight the potential for late complications following hydatid surgery.

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