Single Case

Sump Syndrome: A Rare Long-Term Complication of Choledochoduodenostomy

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Keywords

Sump syndrome · Choledochoduodenostomy · Complication

Abstract

Biliary sump syndrome is a rare condition. It is seen as a rare long-term complication in patients with a history of a side-to-side choledochoduodenostomy. In the era before endoscopic retrograde cholangiopancreatography, side-to-side choledochoduodenostomy was a common surgical procedure for the management of biliary obstruction. In the setting of a side-to-side choledochoduodenostomy, the bile does not drain through the distal common bile duct anymore. Therefore, the part of the common bile duct distal from the choledochoduodenostomy anastomosis consequently transforms into a poorly drained reservoir, making this so-called “sump” prone to accumulation of debris. These patients are prone to cholangitis. We present a 64-year-old man with a history of side-to-side choledochoduodenostomy who presented with manifestations of cholangitis. An endoscopic retrograde cholangiopancreatography confirmed a diagnosis of sump syndrome. The etiology, clinical manifestations, and treatment of biliary sump syndrome are discussed in this article.

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Introduction

Sump syndrome is a rare long-term complication of side-to-side choledochoduodenostomy (CDD), a common surgical procedure done with cholecystectomy to improve biliary drainage done in the era before endoscopic retrograde cholangiopancreatography (ERCP) [1]. CDD is usually done for retained biliary stones or a dilated common bile duct (CBD). After surgery, pneumobilia is the only sign for functioning biliary-enteric anastomosis. Clinical sign of ascending cholangitis, decades after the initial CDD procedure and pneumobilia, leads to the suspicion of sump syndrome. It is diagnosed by ERCP and patients recover quickly after endoscopic debris extraction, sphincterotomy, and antibiotic treatment. In the ERCP era, little is known about CDD and its long-term complications like sump syndrome.

Sump syndrome is a rare complication of biliary enteric anastomosis after cholecystectomy [2]. After side-to-side CDD, the CBD between the anastomosis and the ampulla of Vater becomes a potential sump (a recess or reservoir serving as a drain for liquids) [3]. Accumulation of debris, stones, and static bile occurs in this poorly drained reservoir and hence acts as a nidus for bacterial proliferation, thereby obstructing normal biliary drainage leading to complications like recurrent cholangitis, pancreatitis, or biliary obstruction. The treatment of choice is a sphincterotomy done through ERCP [4].

We present the case of a 64-year-old man with sump syndrome who presented with clinical signs of ascending cholangitis. The detailed medical history that was taken in combination with the presence of pneumobilia led to the suspicion of sump syndrome.

Case Description

A 64-year-old Hispanic man with a past medical history of type 2 diabetes, hypertension, dyslipidemia, benign prostatic hypertrophy, Parkinson's disease, multiple hepatic abscesses, alcohol abuse, history of cholecystectomy and revision of CBD (in 1980 in Mexico) presented to the emergency room with sharp epigastric pain radiating to the right upper quadrant of the abdomen, loss of appetite, fever, and recurrent vomiting for 2 days. The vital signs were significant for high-grade fever (104°F), tachycardia (heart rate of 102 beats per minute), and hypotension (blood pressure of 95/65 mm Hg). Physical examination was significant for tenderness over the right upper quadrant of the abdomen. The rest of the physical examination was within normal limits. Laboratory studies revealed a leukocytosis (WBC count of 14,600 cells/μL) with bandemia, normal total bilirubin, and elevated alkaline phosphatase (446 units/L). There was mild increase of transaminases (ALT of 60 units/L and AST of 61 units/L), and normal amylase and lipase. The INR and PTT were normal. He was started on intravenous fluids and broad-spectrum antibiotics. CT of the abdomen revealed pneumobilia (arrow in Fig. 1). A few hours after admission to the hospital, the patient developed severe hypotension, hypoxia, and confusion. He was immediately transferred to ICU and was intubated. Assuming that a side-to-side CDD had been performed several decades before in the context of a revision of the CBD due to complicated cholecystectomy, an urgent ERCP was performed to evaluate biliary drainage. The endoscopic examination of the duodenum revealed a small orifice proximal to the papilla of Vater. Air insufflation into this orifice induced distinct pneumobilia, whereas contrast injection showed a normal proximal CBD and drainage of the contrast dye via the anastomosis, proving the presence of CDD and its functional efficiency. In addition, filling defects of the distal CBD revealed impaction of abundant debris and drainage of pus. The findings of ascending cholangitis in the context of

a former CDD procedure together with the endoscopic picture of filling defects in the distal CBD (arrow in Fig. 2) led to the diagnosis of sump syndrome. Sphincterotomy and stenting was performed through ERCP (cross and arrow in Fig. 3). His WBC count and the liver function tests improved. His hospital course was complicated by hospital-acquired pneumonia and severe alcohol withdrawal. He also had gram-negative bacteremia (*Aeromonas*) secondary to cholangitis which was treated with antibiotics (levofloxacin) based on culture and sensitivity. The patient was extubated on the second day and clinically, he improved after 4 days of stay in the ICU. He was then transferred to the general medical floor and later on he was discharged home.

Discussion

A “sump” is defined as a covered cistern or reservoir. The term “sump syndrome” was coined after the observation of accumulated debris proximal to the papilla, in the distal bile duct reservoir of affected patients [5–7]. In the pre-ERCP era, CDD was a common surgical procedure in patients with biliary tract disease. In the setting of a side-to-side CDD, the bile does not drain through the distal CBD anymore. Therefore the part of the CBD distal from the CDD anastomosis consequently transforms into a poorly drained reservoir, making this so-called “sump” prone to accumulation of debris [1]. Sump syndrome was first described in 1976, with a wide prevalence ranging from 2.5 to 15.7% after CDD [8–11]. Nowadays, CDD is widely replaced by ERCP [12], but we are still faced with its consequences and long-term complications in elderly patients due to global migration of people to the United States from developing and underdeveloped countries where ERCP is still not being used as a standard procedure. Sump syndrome is not precisely defined, but it results from accumulation of lithogenic bile, debris, or calculi as well as refluxed duodenal contents in the distal CBD, leading to biliary and/or pancreatic complications [1]. According to Marbet et al. [13], reduced filling pressure as well as reduced peristalsis and drainage of the distal CBD caused by the upstream anastomosis play an important role in the pathophysiology of sump syndrome. It can present with a variety of symptoms and clinical pictures such as recurrent pancreatitis, colicky pain, jaundice, and cholangitis. Therefore, the diagnosis of sump syndrome is challenging because no characteristic clinical or laboratory finding is highly specific [1]. Taking a detailed past medical history and having a high degree of suspicion appear to be the most important elements for diagnosing sump syndrome. To increase the challenge, sump syndrome often becomes clinically manifest only several decades after the initial CDD procedure [8, 13]. Usually, patients are unaware about the procedure and nonavailability of medical records from the past makes the diagnosis of this disease very challenging. In addition to a detailed past medical history, pneumobilia is another key element in establishing the diagnosis of sump syndrome.

Nowadays, in the ERCP era, CDD is rarely performed and its complications, including sump syndrome, are almost forgotten, especially due to the long interval until clinical manifestations of sump syndrome occur after CDD [12]. In asymptomatic patients with prior CDD, abdominal radiograph shows air in the biliary tree. In symptomatic patients, plain film demonstration of calcification in the right upper quadrant or pneumobilia and debris-filled dilated biliary ducts suggests sump syndrome as clinical diagnosis. Sonogram may show pneumobilia, biliary duct dilatation, biliary stones, changes of cholangitis, pancreatitis, pancreatic duct dilation, and liver abscess. CT shows prior surgical changes, debris, stones in the

distal CBD, and enhancement of duct walls as a result of suppurative cholangitis or adjacent stone [14].

Management of the sump syndrome has been described as early as 1976 by physicians performing ERCP [6]. In a retrospective analysis involving 30 cases of sump syndrome, the most common etiology was the accumulation of food-debris (67% of cases) and calculi (40% of cases) [6]. Sump syndrome can also be treated surgically by creating a Roux-en-Y hepaticojejunostomy besides performing a biliary sphincterotomy and extracting the debris from the CBD though ERCP [6].

Conclusion

It is important to include sump syndrome in the differential diagnosis for patients who present with cholangitis or pancreatitis with a history of cholecystectomy in the pre-ERCP era or if patients are from a developing country. The diagnosis of sump syndrome is challenging as little is known about CDD and its complications. Also, the unknown history and unavailability of previous medical records dating decades back pose a challenge to the diagnosis of sump syndrome.

Statement of Ethics

The authors have no ethical conflicts to disclose.

Disclosure Statement

None of the authors have any conflict of interest financially, either directly or indirectly, with regard to the matter discussed in the manuscript.

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Fig. 1. CT of the abdomen showing pneumobilia (arrow).

Fig. 2. Endoscopic retrograde cholangiopancreatography image showing a sump and a mildly dilated common bile duct (arrow).

Fig. 3. Endoscopic retrograde cholangiopancreatography – endoscopic view showing status post papillotomy (cross) and stent (arrow).