

Case Report

Spontaneous and Post-Choledochoduodenostomy Sump Syndrome: About Two Cases and Literature Review

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Abstract

Sump Syndrome (SS) is recognized as an infrequent and late complication of choledochostomy. It results from the reflux of biliary and enteric contents into the distal segment of the common bile duct leading to complications like recurrent cholangitis, pancreatitis or hepatic abscesses. Endoscopic sphincterotomy is recommended as the treatment of choice. Spontaneous cases of sump syndrome are exceedingly rare and only one case has been published in literature. A case of cholangitis revealing a spontaneous SS and a classic case of SS complicating choledochoduodenostomy are described in the present report.

Keywords: Choledochoduodenostomy; Endoscopic Sphincterotomy; Sump Syndrome

Introduction

Biliary sump syndrome is a rare long term complication of biliary enteric anastomosis, usually side-to-side Choledochoduodenostomy (CDD) and much less commonly choledochojejunostomy and Roux-en-Y hepaticojejunostomy [1]. In the pre-ERCP era, CDD was a common surgical procedure for the management of biliary obstruction. Nowadays, CDD is much less commonly performed and has been widely replaced by ERCP. However, we are still faced with its consequences and long term complications especially the Sump Syndrome (SS) [2,3]. This term was first coined in 1978 [4]. Classically, the distal segment of the Common Bile Duct (CBD) acts as a reservoir ‘‘sump’’ where lithogenic bile, debris, stones as well as refluxed duodenal contents accumulate giving rise to biliary and/or pancreatic complications. Endoscopic Sphincterotomy (ES) has been recommended as the

primary treatment modality in the SS. We herein report two cases of this uncommon syndrome.

Case Reports

Case 1

A 70-year-old-woman presented to our department with a 3 months’ history of episodic right upper abdominal quadrant and epigastric pain without associated jaundice or fever. Her past medical history included a cholecystectomy with side-to-side CDD carried out for choledocholithiasis 34 years ago. Physical examination revealed tenderness over the epigastrium and the right upper abdominal quadrant. Laboratory evaluation was significant for a mildly elevated alkaline phosphatase (195 UI/l), gamma-glutamyltransferase (103 UI/l). White Blood Cell Count (WBC), C-reactive protein, total bilirubin, transaminases and lipase levels were within the normal limits. Abdominal ultrasound and Magnetic Resonance Cholangiopancreatography (MRCP) showed a grossly dilated CBD as well as dilated intrahepatic ducts containing sludge and debris (Figure1).

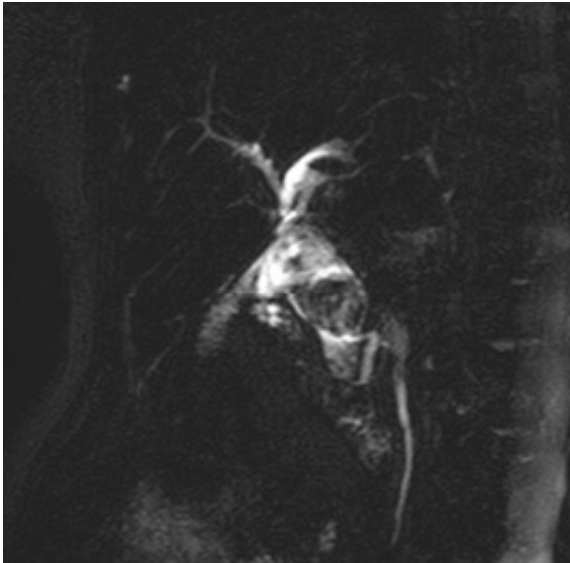


Figure 1: 3D-MRCP image showing a grossly dilated CBD within debris, stones and sludge.

An Endoscopic Retrograde Cholangiopancreatography (ERCP) was performed to evaluate biliary drainage. The CBD was cannulated through the ampullary orifice after passing a guide wire. Contrast injection revealed 21-mm-wide CBD with contrast extravasation through the anastomosis proving the permeability of the CDD. Filling defects of the distal CBD revealed a large amount of food debris which were extracted after an ES using a balloon catheter besides continuous saline irrigation followed by CBD clearance (Figure 2). After the procedure, the patient was relieved from her symptoms. On 1 year follow up, she is doing well.



Figure 2: ERCP: Food debris extraction using a balloon catheter.

Case 2

We present the case of a 69-year-old-man who presented to our department with a sump syndrome. His past medical history included a cholecystectomy carried out 13 years ago. He was also admitted many years ago for an acute cholangitis. Imaging showed multiple hepatic and splenic abscesses as well as a thrombosis of the left branch of the portal vein and the right segmental branches with CBD stones. He received antibiotic treatment and anticoagulants with subsequent clinical and radiological improvement. ERCP was not performed. He had been suffering for 7 years after that from recurrent cholangitis requiring several hospital admissions. After he developed the full clinical picture of acute cholangitis, he was referred to our tertiary medical center. On examination, he was mildly icteric with some epigastric tenderness. Laboratory tests showed increasing serum gamma-glutamyltransferase (425U/l), aspartate aminotransferase (93 U/l), alanine aminotransferase (110 U/l), total bilirubin (38,8 mg/l), C-reactive protein (119 mg/l) and a slight leukocytosis (10190 elements/mm³).

Abdominal and endoscopic ultrasound revealed pneumobilia and an echogenic material within dilated bile ducts. Empiric antibiotic therapy (ceftriaxone with metronidazole) and fluid resuscitation were started. An ERCP was immediately carried out which revealed a fistulous orifice draining some debris proximal to the papilla of Vater. After cannulation of the CBD, contrast injection showed a 31-mm-wide CBD with filling defects (Figure 3). Multiple food debris and sludge were removed using a dormia basket and a balloon catheter with saline irrigation. The diagnosis of a spontaneous sump syndrome was established. After the procedure, the patient recovered quickly and later on was discharged home.

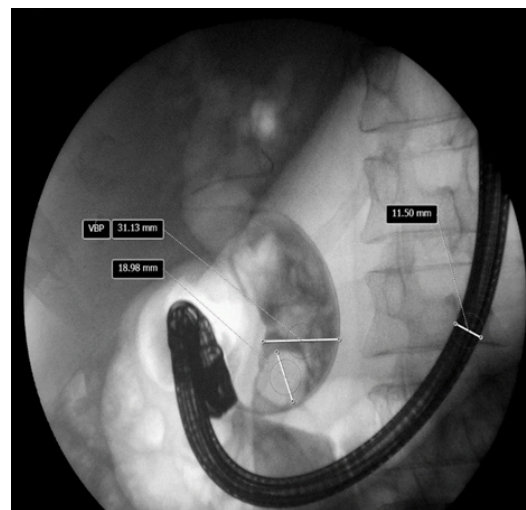


Figure 3: ERCP cholangiogram showing a 31-mm-wide CBD with filling defects.

Discussion

A sump is defined as “a pit, well, or reservoir in which water or other liquid is collected” [5]. After side-to-side CDD, the distal segment between the anastomosis and the ampulla is excluded from the flow of the bile. Occasionally, biliary secretions and enteric debris (refluxed through the anastomosis) are able to exit (via the ampulla or the enterostomy). Consequently, accumulation of lithogenic bile, cholesterol crystals and food residues occurs in this nonfunctional reservoir “sump” and hence acts as a nidus for bacterial proliferation, stasis and perhaps dysplastic changes in the bile duct mucosa as well as in new stone formation. Thus, intermittent or complete obstruction of the enterostomy lead to complications in the biliary tract as well as in the pancreas [1,6]. Several factors could explain this complication: Long length of the sump segment, dysfunctional papilla, stomal stenosis and stone retention [7,8].

According to Marbet et al, 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 SS [10]. SS was first described in 1976 with a wide prevalence ranging from 2.5 to 15.7% after CDD. We are facing a re-emergence of this rare entity due to global migration from under developed countries where CDD is still performed to manage biliary obstruction instead of ERCP [2,9]. The syndrome is expressed by a variety of symptoms including recurrent episodes of colicky pain, fever, jaundice, chills, nausea and vomiting [6]. The most commonly reported complications include cholangitis, pancreatitis, hepatic abscesses and secondary biliary cirrhosis. Rarely, malabsorption and steatorrhea caused by a lack of intestinal bile salts could be the initial sign [3,6]. Abdominal ultrasound may show pneumobilia, bile duct dilatation, biliary stones, changes of pancreatitis, cholangitis, pancreatic duct dilatation and liver abscesses, echogenic material within the ducts.

Debris and stones in the distal CBD are the most frequent and the most indicative CT and MRCP findings of the SS. CT also shows prior surgical changes, thickening and/or enhancement of the bile duct walls as a result to supportive cholangitis or adjacent stone, dilated bile and/or pancreatic ducts, liver abscesses and pancreatitis [3,6,9]. In the second case, the patient developed spontaneous SS as a result of the formation of choledochoduodenal fistula. The CBD segment between the fistula and the ampulla of Vater became a sump. To the best of our knowledge, only one case of spontaneous SS has been described in literature and this the first case describing splenic abscesses and portal vein thrombosis as a complication of SS [11]. The principal therapeutic approach is to improve the biliary drainage of the choledocus distal to the anastomosis either endoscopically or by surgery [10]. Formerly, the recommended treatment for the SS was the surgical repair. More recently, ERCP with Endoscopic Sphincterotomy (ES) was

introduced as the treatment of choice for the SS based on several reports [1,7]. The use of ES to drain the sump was first reported in 1977 [12]. Blair et al described endoscopic dilatation of the narrow choledochoduodenal anastomosis if ES was not possible. However, this procedure results only in a temporary drainage according to some authors [13].

Dill reported a case of SS treated with stent therapy [14]. In a retrospective analysis involving 30 cases of SS, the most common etiology was accumulation of food-debris (67% of cases) and calculi (40% of cases) [3]. ES is safe, effective and efficacious in most of cases. Nevertheless, late recurrences are possible. In another study of 31 patients with SS, all patients were successfully treated by ES. Recurrence was observed in 6 patients (19%) over a median follow-up of 51 months (Range: 18-84 months). All the 6 patients were safely treated with a new sphincterotomy with “sump” clearance [1]. The long term success of endoscopic management, in case of a manifest SS, is significantly impaired by the unchangeable pathological situation of a large choledochoduodenal fistula with consecutive duodenobiliary reflux [15]. When ES or repeated endoscopic intervention is unsuccessful, patients can be managed surgically through a revision of the CDD to a Roux-en-Y hepaticojejunostomy and resection of the distal portion of the CBD [3,14,16].

Conclusion

Sump Syndrome is an uncommon complication of the CDD procedure. The diagnosis is challenging for many reasons: there are no pathognomonic clinical or laboratory findings and symptoms usually appear only decades after the surgical bypass. Furthermore, the former medical records, in order to have a detailed surgical history, may no longer be available [2,10]. Sump syndrome should be suspected in any patient who develops symptoms of cholangitis after a biliary enteric anastomosis. The second case demonstrates that SS may exceptionally occur spontaneously. In the ERCP era, endoscopic sphincterotomy is regarded as the primary modality approach for the SS.

Conflict of Interest

The authors have no conflicts of interest, financial or otherwise, to disclose.

References

1. Christos Mavrogiannis MD, Christos Liatsos MD, Andreas Romanos MD, Spyros Goulas MD, Spyros Dourakis MD, et al. (1999) Sump Syndrome: Endoscopic Treatment and Late Recurrence, *AJG* 94: 972-975.
2. Zeuge U, Fehr M, Meyenberger C, Sulz MC (2014) Mind the sump! Diagnostic challenge of a rare complication of choledochoduodenostomy. *Case Rep Gastroenterol* 8: 358-363.

3. Motaz Qadan, Sharon Clarke, Ellen Morrow, George Triadafilopoulos, Brendan Visser (2012) Sump Syndrome as a Complication of Chole-
dochoduodenostomy, *Dig Dis Sci* 57: 2011–2015.
4. Morrissey PE, Burns GA, Cohn SM (1996) Sump syndrome complicat-
ing Roux-en-Y hepaticojejunostomy: Case report and review of the
literature. *Surgery* 119: 403–405.
5. Siegel JH (1981) Duodenoscopic sphincterotomy in the treatment of
the “sump syndrome”, *Dig Dis Sci* 26: 922-928.
6. Hawes DR1, Pelsang RE, Janda RC, Lu CC (1992) Imaging of the
biliary sump syndrome. *AJR Am J Roentgenol.* 158: 315-319.
7. Caroli-Bosc FX, Demarquay JF, Peten EP, Dumas R, Bourgeon A, et
al. (2000) Endoscopic management of sump syndrome after chole-
dochoduodenostomy: retrospective analysis of 30 cases. *Gastrointest
Endosc* 51: 180-183.
8. Rumans MC, Katon RM, Lowe DK (1987) Hepatic abscesses as a
complication of the sump syndrome: combined surgical and endo-
scopic therapy. Case report and review of the literature. *Gastroentero-
logy.* 92: 791-795.
9. U A Marbet, G A Stalder, H Faust, F Harder, K Gyr (1987) Endoscopic
sphincterotomy and surgical approaches in the treatment of the ‘sump
syndrome’. *Gut* 28: 142-145.
10. Abraham H, Thomas S, Srivastava A (2017) Sump Syndrome: A Rare
Long-Term Complication of Chole-
dochoduodenostomy. *Case Rep
Gastroenterol* 11: 428-433.
11. Hallstone A, Triadafilopoulos G (1990) “Spontaneous sump syndro-
me”: Successful treatment by duodenoscopic sphincterotomy. *Am J
Gastroenterol* 85: 1518-1520.
12. Rey JF, Delmont J (1977) Sphinctérotomie endoscopique dans un cas
de syndrome du cul-de-sac sous anastomotique après cholédoco-
duodénotomie latéro-latérale. *Endoscopy digestive* 11: 121-124.
13. Blair AJ, Leung JWC, Cotton PB (1985) Endoscopic treatment of
stomal stenosis after choledochoduodenostomy: preliminary report.
Surgery 97: 487-489.
14. Dill JE (1993) Endoscopic stent treatment of the sump syndrome. *Am
J Gastroenterol* 88:1295-1296.
15. Ell C, Boosfeld C, Henrich R, Rabenstein T (2006) Endoscopic treat-
ment of the «sump syndrome» after choledochoduodenostomy: a new
technique using an amplatzer septal occluder. *Z Gastroenterol* 44:
1231-1235.
16. Venerito M, Fry LC, Rickes, Malfertheiner P, Mönkemüller K (2009)
Cholangitis as a late complication of choledochoduodenostomy: the
sump syndrome, *Endoscopy* 41: E142-E143.