Multiple Biliary Stones: a Rare Long-term Complication after Whipple Procedure

Múltiplos cálculos biliares: uma complicação rara a longo prazo após o procedimento de Whipple

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ABSTRACT

A hypertensive, diabetic woman underwent a successful Whipple procedure at the age of 84 due to carcinoma of the ampulla of Vater. She presented an extremely rare complication 24 months after the surgery, consisting of acute cholangitis due to multiple biliary lithiases associated with a bilioenteric anastomotic stricture. The diagnosis was confirmed with computed tomography, magnetic resonance cholangiopancreatography, and cholangiography. The patient was successfully treated with multiple percutaneous transhepatic cholangioplasties.

Keywords: Biliary lithiasis, Long-term complication, Whipple procedure, Anastomotic stricture, Cholangioplasty, Ampullary cancer.

RESUMO

Uma mulher hipertensa e diabética foi submetida a um procedimento bem-sucedido de Whipple aos 84 anos devido a um carcinoma da ampola de Vater e apresentou uma complicação extremamente rara 24 meses após da cirurgia, consistindo em colangite aguda devido à presença de litíase biliar múltipla associada com estenose da anastomose bilio-entérica. O diagnóstico foi confirmado com tomografia computadorizada, colangiopancreatografia por ressonância magnética e colangiografia.O paciente foi tratado com sucesso com múltiplas colangioplastias transhepáticas percutâneas.

Palavras-chave: Cálculos biliares, Complicações pós-operatórias, Pancreaticoduodenectomia, Neoplasia periampular.

INTRODUCTION

The carcinoma of the ampulla of Vater represents 0.5% of all gastrointestinal tumors and 7% of all periampullary tumors, with approximately 6 cases per million population per year [1,8]. This neoplasm has a resectability percentage of 80% and 5-year survival of 30-70% [2,3,8].

Pancreaticoduodenectomy is the treatment with curative intent for patients with pancreatic head and periampullary tumors. The most common postoperative complications include pancreatic fistula, pneumonia, delayed gastric emptying, abdominal abscesses, infections, and anastomotic leak [3]. However, few studies have been developed on possible long-term complications. The presence of stones in the pancreatic and biliary ducts represents one of the rare post-pancreaticoduodenectomy complications, with only a few cases reported in the literature [4,5,6].

Clinical Case

An 86-year-old diabetic and hypertensive woman with a history of Whipple procedure due to a carcinoma of the ampulla of Vater with extension to the pancreatic head and peripancreatic fat presented two years later with a 1-week history of jaundice and fever.

Laboratory values showed elevated levels of total bilirubin (9.90 mg/dL), conjugated bilirubin (7.75 mg/dL), gamma-glutamyl transpeptidase (324 U/L), alkaline phosphatase (279 U/L), CA 19-9 (208.1 UI/mL), and leukocytes (15.5 x10^3 mm^3.) Likewise, computed tomography (CT) scan of her abdomen revealed multiple isodense ovoid lesions in the dilated common hepatic duct, suggestive of lithiasis (Fig. 1). The diagnosis was later confirmed with magnetic resonance cholangiopancreatography (MRCP), which showed multiple calculi in the right hepatic duct

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and the common hepatic duct, as well as a significant bilioenteric anastomotic stricture (BAS), with no signs of a neoformative process (Fig. 1).

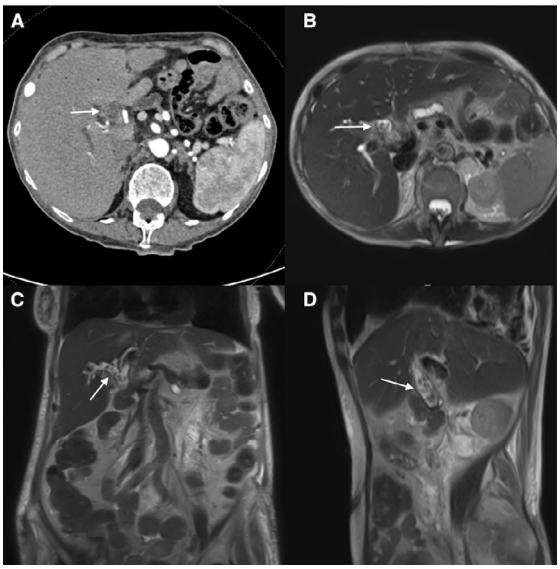

Figure 1: CT scan (A) showing multiple isodense ovoid lesions in the common hepatic duct compatible with lithiasis. Transverse (B), sagittal (C) and coronal (D) views of MRCP showing multiple stones in the right and common hepatic bile ducts, as well as a significant bilioenteric anastomotic stricture.

The percutaneous transhepatic (PT) cholangiography and external biliary drainage were first performed due to the patient's acute cholangitis; during the procedure, it was possible to visualize multiple calculi in the bile duct, as well as a BAS (Fig. 2). Three days later, a PT cholangioplasty was performed, in which the BAS was dilated with an 8-mm balloon catheter, and then the stone extraction was carried out using a Fogarty balloon-tipped catheter, which pushed the stones into the jejunum (Fig. 3-4).

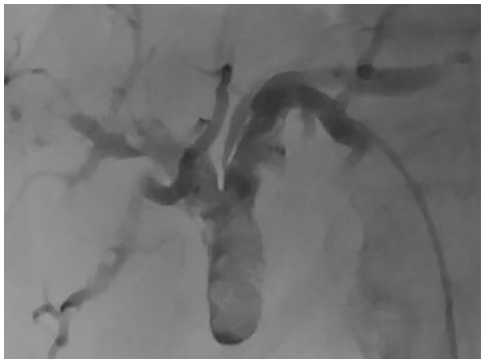

Figure 2: The first PT cholangiography showing multiple filling defects consistent with lithiasis and a significant bilioenteric anastomotic stricture.

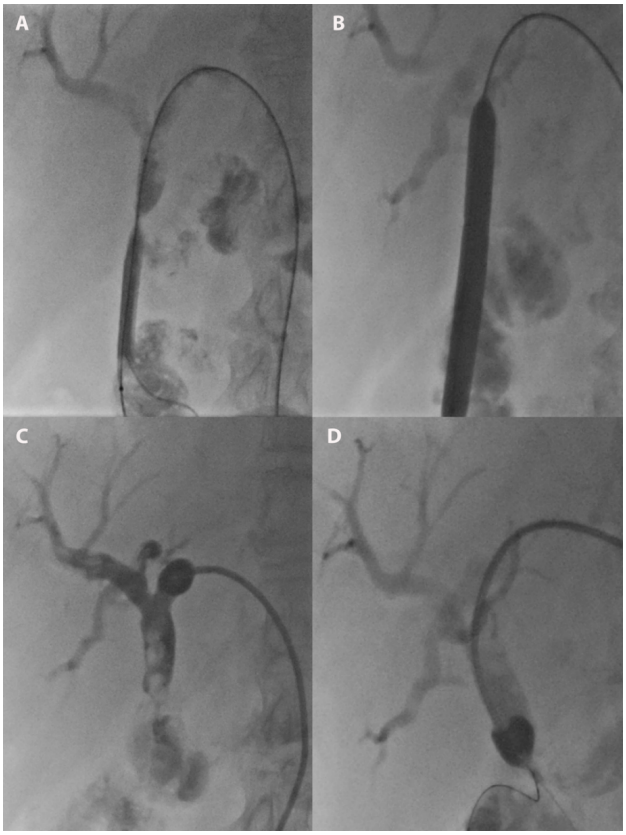

Figure 3: The first PT cholangioplasty with an 8-mm balloon catheter (A, B). First stone extraction with Fogarty balloon-tipped catheter (C, D).

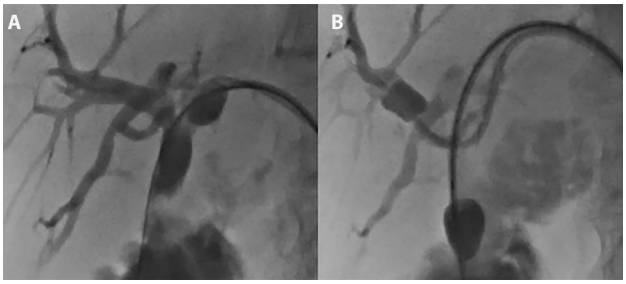

Figure 4: Second stone extraction using two Fogarty balloon-tipped catheters.

A PT cholangiography was performed two weeks later, which still revealed multiple stones. The patient underwent a second PT cholangioplasty with a 10-mm balloon catheter, followed by a stone extraction using two balloon catheters, one blocking the entry of stones into the right hepatic duct and the other pushing them through the common hepatic duct into the jejunum (Fig. 5).

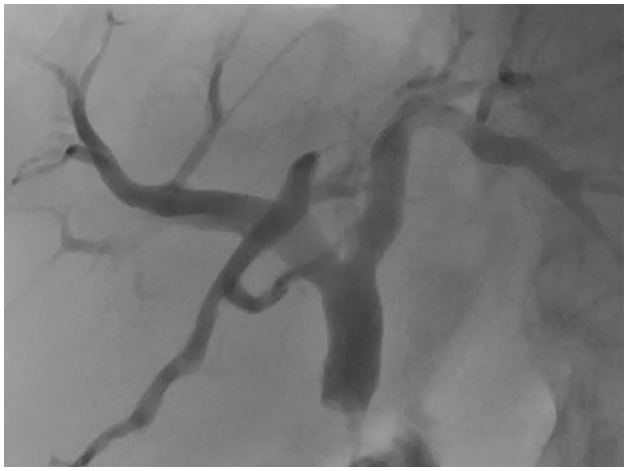

Figure 5: Final cholangiography, which revealed a permeable bilioenteric anastomosis with no signs of calculi in the intra and extrahepatic bile ducts

A final PT cholangiography was performed two weeks later, which revealed a permeable bilioenteric anastomosis with no signs of calculi. The patient was medicated with a dose of 750 mg/day of ursodeoxycholic acid for 8 months to avoid the future formation of bile duct stones. The patient had three medical control in the next three years, one per year, without evidence of new stones.

DISCUSSION

We present the case of an elderly woman who underwent a successful Whipple procedure at the age of 84 due to carcinoma of the ampulla of Vater and presented no complications or tumor recurrence throughout the two following years. However, 24 months after the surgery, the patient presented multiple biliary lithiases associated with a bilioenteric anastomotic stricture.

First off, we would like to comment on the fact that advanced age is no longer a contraindication for performing a Whipple procedure. Lee et al. have proved this surgery to be safe and acceptable for selected octogenarians [7], and the presented case demonstrates an example of a successful pancreaticoduodenectomy in a patient older than 80 years.

Likewise, it is worth mentioning that the presented case has not shown tumor recurrence 24 months after the Whipple procedure. Colussi et al. investigated predictors of recurrence after pancreaticoduodenectomy in ampullary cancer and found that 44% of patients had tumor relapse in 5 years. In addition, the authors found that lymph node involvement was the main prognosis factor for recurrence, which was not present in the reported case [8].

In addition, we would like to comment on the biliary lithiasis and bilioenteric anastomotic stricture. Common postoperative complications of the Whipple procedure include pancreatic fistula, hemorrhage, delayed gastric emptying, abdominal abscess, and infections [3], which were not present in the patient. However, she did develop biliary lithiasis 24 months after the Whipple procedure, which is an extremely rare long-term complication of pancreaticoduodenectomy that has not been well described in the literature. There are few cases reported in the world [4,5,6], the most recent in the study by Zhan et al., in which five patients developed stones in the pancreatic or bile duct an average of 47 months after pancreaticoduodenectomy with an age range between 60-70 years and whose most common symptoms were fever, abdominal pain, jaundice, and vomiting. The present case developed calculi in a shorter time (24 months) than the Zhan et al. study and other previous reports [4,5,6].

The pathogenesis of biliary stones after pancreaticoduodenectomy is not yet clear in the literature; however, anastomotic strictures seem to play a defining role [6,9]. A single-institution review at John's Hopkins Hospital of over 1500 patients with the periampullary disease found that only 2.6% of them developed bilioenteric anastomotic strictures after pancreaticoduodenectomy; the average time of presentation was 13 months after surgery, which is shorter than the presented case. It is worth reminding that the presented case had a history of diabetes mellitus and hypertension. In addition, Kim et al. performed a retrospective analysis of 21 patients treated with percutaneous transhepatic cholangioscopy (PTC) for benign bilioenteric anastomotic strictures, of which 81% of them were complicated by biliary stones [10]. Lastly, other risk factors for forming biliary stones in the context of a bilioenteric stricture include bile stasis, remnant suture materials, and suture clips, which were not present in the reported case, as well as infection and reflux [5].

In addition, diagnosis of biliary lithiasis can be easily performed using imaging examinations, such as computed tomography, magnetic resonance imaging, and cholangiography, which were all used to diagnose the reported case [6].

Lastly, lithiasis and anastomotic strictures can be treated by either surgical or non-surgical procedures. Within the context of non-surgical treatment, it is worth noting that a percutaneous approach is usually implemented to evaluate patients with dilated bile ducts who are not candidates for an endoscopic retrograde cholangiopancreatography (ERCP), such as the presented patient, who had surgically-altered anatomy that did not allow endoscopic biliary access [5,6]. In this case, the lithiasis and bilioenteric anastomotic stricture were treated and resolved using multiple percutaneous transhepatic cholangioplasties. Similar to our case, Hisa et al. demonstrated an anterograde stone expulsion into the jejunum by dilating the anastomotic stenosis through a fistula created by a pancreaticogastrostomy with ultrasound guidance [5].

CONCLUSION

Biliary lithiasis is an extremely rare long-term complication of the Whipple procedure but should still be considered in the differential diagnosis of a jaundiced patient with a history of pancreaticoduodenectomy. Appropriate therapeutic measures should be taken since the patient's surgically-altered anatomy will not permit a regular endoscopic biliary access.

REFERENCES

1. Ahn DH, Bekaii-Saab T. Ampullary cancer: an overview. Am Soc Clin Oncol Educ Book. 2014;112–115.
2. Gaspar B, Beuran M, Paun S, et al. Current strategies in the therapeutic approach for adenocarcinoma of the ampulla of Vater. J Med Life. 2013;6: 260–265.
3. Panzeri F, Crippa S, Castelli P, et al. Mangement of ampullary neoplasms: a tailored approach between endoscopy and surgery. World J Gastroenterol. 2015;21:7970–7987.
4. Hisa T, Momoi T, Shimizu T, Furutake M, Takamatsu M, Ohkubo H. Endoscopic ultrasound-guided antegrade stone removal in a patient with pancreatic stones and anastomotic stricture after end-to-side pancreaticojejunostomy. Pancreatology 2013; 13(4):452-4.
5. Zhan H, Wang L, Zhang G, Zhan H, Hu S, Zhang Z, et al. Rare Long-Term Complications Following Pancreaticoduodenectomy:Biliary and Pancreatic Lithiasis. Current Signal Transduction Therapy. 2016,11(1): 24 -26.
6. Melis M, Marcon F, Masi A, Pinna A, Sarpel U, Miller G, et al. The safety of a pancreaticoduodenectomy in patients older than 80 years: risk vs. Benefits. HPB. 2012, 14: 583–588.
7. Lee MK, Dinorcia J, Reavey PL, Holden MM, Genkinger JM, Lee JA, et al. Pancreaticoduodenectomy can be performed safely in patients aged 80 years and older. J Gastrointest Surg. 2010 Nov;14(11):1838-46.
8. Colussi O, Voron T, Pozet A, et al. Prognostic score for recurrence after Whipple's pancreaticoduodenectomy for ampullary carcinomas; results of an AGEO retrospective multicenter cohort. Eur J Surg Oncol. 2015;41(4):520-526.
9. House MG, Cameron JL, Schulick RD, Campbell KA, Sauter PK, Coleman J, et al. Incidence and outcome of biliary strictures after pancreaticoduodenectomy. Ann Surg. 2006 May;243(5):571-6
10. Oh HC. Percutaneous Transhepatic Cholangioscopy in Bilioenteric Anastomosis Stricture. Clin Endosc. 2016 Nov;49(6):530-532

Informed consent

Patient consents and accepts to participate in the case report.
Attach the documents.

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Declarations

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All authors had full access to the data, contributed to the study, approved the final version for publication, and took responsibility for its accuracy and integrity.

Authors' contributions

All authors contributed to the concept, collection and analysis of the data for this study and participated in drafting the manuscript.
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