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.

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].

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

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).

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).
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

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

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Authors’ contributions
All authors contributed to the concept, collection and analysis of the data for this study and participated in drafting the manuscript.
All authors did a critical revision for intellectual content.

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