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Letters to the Editor

Choledochocele complicated with pancreas carcinoma in an old patient. Endoscopical diagnosis and treatment

Key words: Choledochocele. Pancreatic adenocarcinoma. Endoscopic treatment.

Dear Editor,

The choledochal cyst (CC) is an unusual congenital pathology, which is usually diagnosed in childhood (1-4). According to the classification of Todani, there are 6 different types (5,6) and one of the less frequent one is type III CC or choledochocele (2% of CC) (7). Choledochocele is defined as dilation in the distal bile duct that *protrudes* in the duodenum, covered by normal biliary mucosa (8). In the present case, we report a new patient with choledochocele with some peculiarities which make it singular.

Clinical case

An 81 year old male patient was admitted in our hospital in February 2008, with non painful jaundice and anorexia of 2 weeks of evolution. His background history referred a progressive dementia, diabetes mellitus, high arterial tension, atrial fibrillation, chronic bronchitis, abdominal aortic aneurysm and biological mitral prosthesis.

In the physical exploration, it was remarkable a bad general state with jaundice drowsiness and disorientation. The temperature was 38.5 ºC and a distended gallbladder was palpable. In the cell blood count a lekocytosis with neutrophilia was observed. In the general biochemistry the total bilirubin was 22.5 g/dl and the conjugated bilirrubin was 15.6 g/dl. Other liver enzymes were also raised: alkaline phosphatase 287 U/1 (normal < 129), GGT 472 U/1, AST 134 U/1 and ALT 227 U/1.

The abdominal ultrasonography (US) showed a dilated gallbladder with several stones and a dilatation of the intrahepatic and extrahepatic biliary tree. In the abdominal CT scan a mass of 3 cm of diameter in the pancreatic head was found, which obstructed the choledochal and the pancreatic ducts and invaded the gastroduodenal artery. The intrahepatic biliary tree was dilated with hydropic gallbladder. The diagnosis was pancreatic neoplasm with bile duct obstruction and vascular invasion. The choledochal cyst was not identified with this image technique.

In the endoscopic retrograde cholangiopancreatography (ERCP), we observed in the second duodenal portion a 3 cm diameter round formation, soft and loose to the contact with the sphincterothome. The papilar hole was identified on the top right edge. The contrast injection simultaneously opacified the cyst, the biliary tract and the Wirsung duct (Fig. 1). The pancreatic duct appeared amputated at the union between head and body. In the *choledochus,* at the same level of the pancreatic wound, a clear stricture *was identified*. The image was characteristic of a pancreatic neoplasm with choledochal stricture, associated to a choledochocele. An endoscopic sphyncterothomy was made and an 8 cm long uncovered self expandable metal stent was placed (Figs. 2 and 3).

The patient followed a favorable fast outcome with the disappearance of the fever and the jaundice. Obviously surgical treatment was rejected. Three months later the patient was asymptomatic and the total bilirubin was 1.5 g/dl with normal AST and ALT levels, alkaline phosphatase of 184 U/I and GGT 172 U/11. Nine months later he was still alive and has not needed a stent change.

Discussion

The case of choledochocele that we have presented unites it is short frequency to other unusual characteristics. The first showy thing is that in the eleven cases of CC which we have reported there are 2 type III CC. This is not the usual proportion.

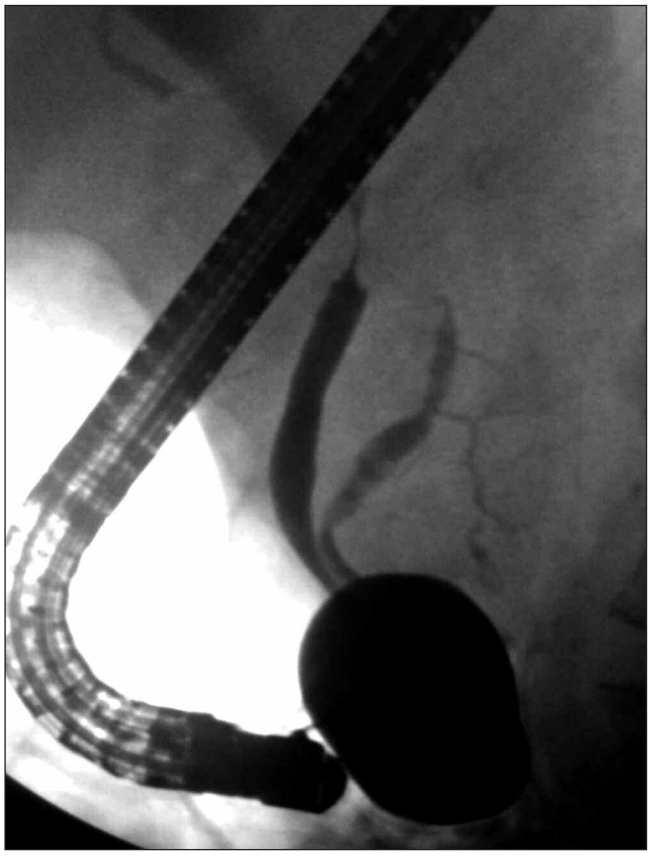

Fig. 1.

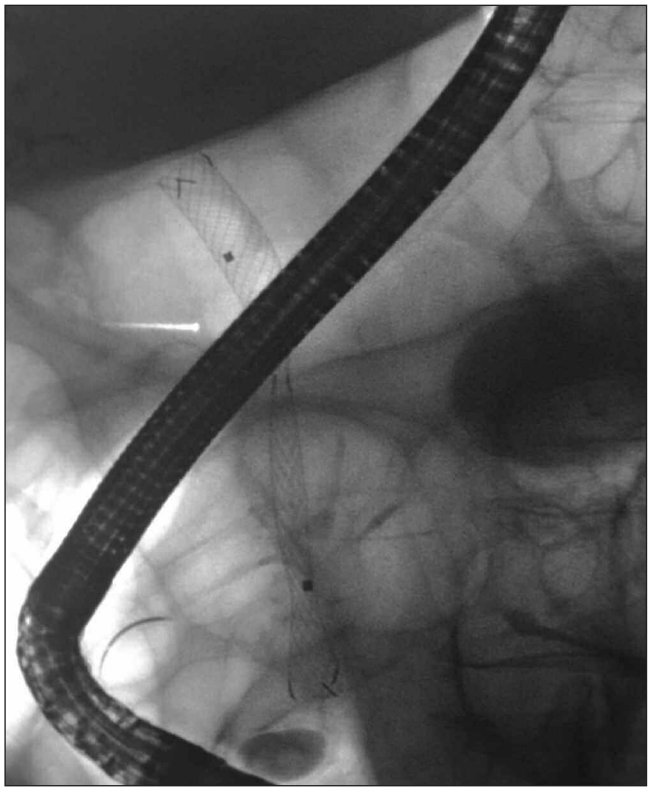

Fig. 2. Self expandable metal stent.

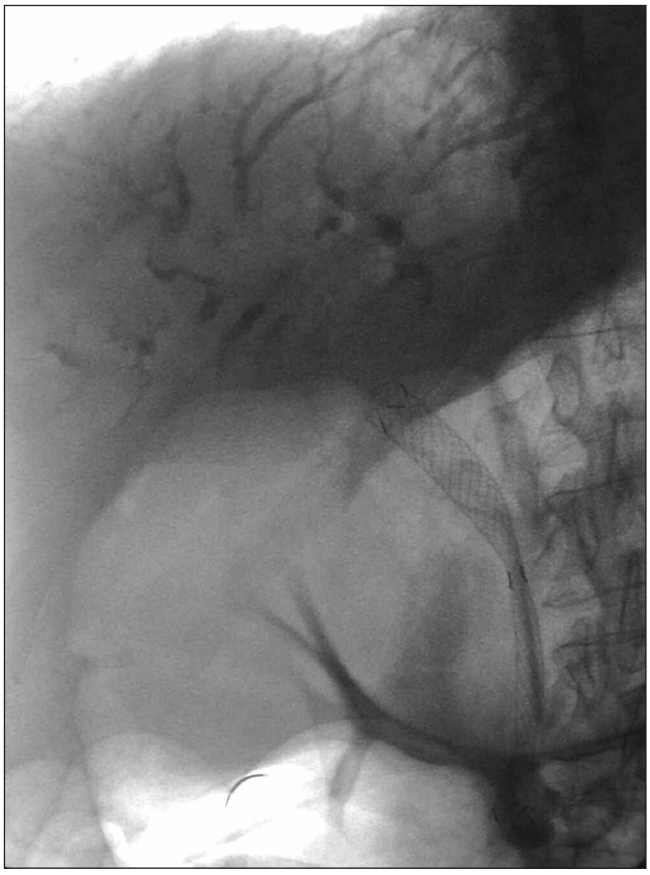

Fig. 3. Self expandable stent: final image.

In the 2 most recent large series of CC report a total of 95 CC and they do not include any type III CC (9,10). The age of presentation is also uncommon. Our two patients with choledochocele are adults in a predominatly pediatric disease (3,4). The late diagnosis probably is because type III CC usually are less symptomatic than other type of CC. Some authors consider that choledochocele is a different disease that should be out of the CC´s classification (9).

Another interesting point is the high malignization rate of those cysts. This risk is much lower in type III CC compared with the rest of types (8-10). When the cholangiocarcinoma appears, arises in the cyst mucosa in 70% of cases. In the remaining 30% it is originated in the biliary tree, gallbladder and pancreas (9), as occurs in this case. In both type III CC presented, ERCP was the main diagnostic and therapeutic tool. Imaging techniques, like abdominal US or CT scan, did not diagnose correctly the cyst.

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