Introduction

Biliary stones located above the biliary confluence represent an uncommon event in the western areas and is defined as hepatolithiasis. It creates not simple problems in terms of diagnosis and treatment. In recurrent or non-treated cases several complications may arise, up to severe hepatic atrophy and biliary cirrhosis.

Conservative percutaneous transhepatic approaches need a large experience but, in selected cases, may be successful with limited morbidity. Surgical resection is indicated in case of affected hepatic lobe fibrosis or atrophy or dubious neoplastic strictures.

The Authors describe a 68 yrs patient with massive left intrahepatic lithiasis secondary to common bile duct lithiasis and lithiasic cholecystitis with acute pancreatitis. He successfully underwent conservative treatment with...
Integrated treatment of secondary hepatolithiasis. Case report

Case report

In January 2007 the Authors observed a 68 yrs old man (w 65 kg; h 177 cm) suffering since 20 yrs from cholelithiasis with recurrent biliary pain but never jaundice, cholangitis or pancreatitis. No other disease was referred for. In December 2006 the patient was admitted for acute pancreatitis; amylases were 1706 UL (n.r.=28-100), slight increase of bilirubin 1,7 mg/dl (n.r.= 0.2-1.1) and increase of cholestatic indexes ( gGT x4, AP x2). A cholangio-MRI described left (II,III, IV segments) massive intrahepatic lithiasis with some branches of the VII segment involved, cefalic and body acute pancreatitis, scleroatrophic cholecystitis and normal size of the common bile duct with several stones. The left biliary tree above the confluence was dilated (Fig 1).

Following medical therapy a new cholangio-MRI was performed and a 6cm fluid collection in the body of the pancreas with Wirsung duct dilatation was described as well as a persistent dilatation of the left biliary tree. After a month patient's general situation was better, no pain or fever were present and amilasys, lipasis, bilirubin and transaminasis were normal. Cholestasis indexes were instead still abnormal. Tumour markers ( CEA, CA 19.9, CA 125) were in the normal range as well.

Patient referred to an other instution for hepatic surgery, refused the proposed surgery (left hepatectomy with caudate resection). He therefore underwent to transhepatic percutaneous cholangiography: the biliary tree was approached through the hepatic branch of the III segment. A dilatation of left main biliary duct at the confluence with a 10mm baloon was performed; removal of the stones from the main branches and pneumatic dilatation with a 10mm baloon of the papilla with removal of choledocal stones followed. Then choleunctomy was performed to remove the cystic stones. Eventually an external-internal transhepatic percutaneous drain (14F) of the left biliary tree was left in situ. Subsequent repeated cholangiographic sessions using MTBE (metil ter butil etere) and occluding the distal baloon in the choledocus, allowed the selective chatearteris of all biliary ducts with complete removal of the stones (Fig 2). Successively injection of saline in the cleaned ducts (30-50 cc at 12-15 cc/sec) with an automatic device was performed.

One month later patient underwent laparoscopic cholecistectomy, the biliary drainage being still in situ. Strong adhesions were described between the liver and the abdominal wall and of the gallbladder with the duodenum and large omentum. Hepatic biopsies at the V segment were performed as well as resection of a 2 cm module (with a stone inside) of the free margin of the III segment. Histology confirmed intrhepatic lithiasis with chronic cholecystis. Discharge occurred 48 hours later with prescription of antibiotic therapy (amoxicillin/clavulanic acid 1grx3die), selected on antibiogram positive for Klebsiella pneumoniae. Three weeks after the operation (3 months from the initial diagnosis) the patient underwent transhepatic percutaneous cholangiography showing a complete clearance of the biliary tree and a stricture of the Wirsung duct. It was dilated at the confluence with a 3.5 mm baloon and the biliary drainage was removed. MRI control showed a complete resolution of the intraheptic lithiasis, a normal biliary tree and no trace of the peri pancreatic fluid collection (Fig 3).

Discussion

Intrahepatic lithiasis or hepatolithiasis is characte-
lation of the biliary tree. In the secondary forms, more frequent in the western literature, a retrograde migration of stones from the gallbladder to the main ducts is observed, sometimes also the choledocus is involved. Associate strictures are often described (40-80%), sometimes multiple and responsible of relapses (3,4); it is difficult to prove if they cause or are induced by the lithiasis. Some Caroli’s cases present with intrahepatic lithiasis. The left biliary tree is often involved and for unknown reasons notably in the initial phase of the disease (5). In the Chen (6) series of 103 hepatic resections for hepatolithiasis, 86% was in the left biliary tree. Tsunoda proposed a classification of the disease in 4 types on the basis of the site of the stones and the presence of strictures and dilatations (7).

Hepatolithiasis is frequent in the eastern countries with incidence varying from 4,1% of Japan to 47,3% of Taiwan (8); high rates of relapses are described. Western countries show instead rates of incidence from 0,6 to 1,3% (9,10). Thus two different types of diseases are described and different are the approaches to be used.

The primary intrahepatic lithiasis, typical in eastern countries and of unclear etiology, may derive from a recurrent bacterial infection of the bile duct with a bacterial traslocation secondary to a lesion of the intestinal mucosa. It may be related to poor hygienic conditions and probably to specific contents of the diet, often rich in carbohydrates and poor in proteins. Additional cause factors are represented by genic proneness, parasitic infections, congenital abnormalities (11). The association of biliary stagnation and infection is determinant in the stone formation and moreover a specific enzyme activity is able to precipitate conjugate bilirubin with calcium salts. Thus the primary type occurs irrespectively of sex in young patients, often less than 30 yrs of age, and may cause a subtle and asymptomatic evolution to cirrhosis or presents with the typical symptoms (biliary colic, fever, jaundice, cholangitis).

In secondary forms patients are older and have a clinical history of biliary colics by cholelithiasis.

It is reported also the association between hepatolithiasis and cholangiocarcinoma, maybe related to the chronic cholangitis, with a variable incidence from 2,3 and 10% (12,13). In such cases diagnosis of the nature of the strictures is extremely complex even with MRI or direct cholangiography. Serum CEA may be useful in these patients (14). Hepatic resection in unclear cases is recommended.

Early treatment is mandatory to avoid complications. The introduction of cholangio-MRI among the diagnostic tools has given to transhepatic percutaneous or endoscopic approach a therapeutic sense. A multidisciplinary protocol involving surgeon, endoscopist, interventional radiologist is still controversial. Main goals of treatment are the infection eradication, resolution of biliary stasis with adequate reopening of the biliary tree and salvage of healthy hepatic parenchima. There fore each single case has to be dealt with in referral centre for hepatobiliary disease.

Surgical options include cholecodotomy with associate sphincteroplasty, biliary derivation, hepatic resections up to liver transplantation in case of severe secondary biliary cirrhosis (15).

Surgical resection carries today low morbidity and mortality rates in referral centers and represents a radical solution for lithiasis and related strictures but it can be extremely complex in case of bilateral multi-segmental involvement. Among the derivative procedures associated or not to resection it is particularly interesting the hepatic-jejunostomy with a chance of percutaneous access to a redo-cholangioscopy (16) and also a defunctioned loop Anastomised to the duodenum for a possible future endoscopic trans-duodenal access (17).

Patients who underwent hepatectomy with complete removal of stones have a better prognosis in case of massive lithiasis and related strictures compared with those treated without surgery. Despite this, the “conservative” approaches, now widespreadly used, show a comparable efficacy in clearing the stones but maybe carry a higher rate of relapse (11,18). They include: percutaneous trans-hepatic cholangiographic procedures; endoscopic procedures; percutaneous trans-hepatic choledoscopy that is effective in a high number of cases (11,19) and particularly useful to biopsy dubious strictures; laser or hydraulic lithotomy or chemical dissolution. They all allow a resolution of the pathology up to 90% of ca-
Hepatic fibrosis or atrophy, recurrent cholangitis with topographic alterations of the biliary tree or the suspiciousness of a cholangiocarcinoma are absolute indications to hepatic resection (even a major resection), often a left lobectomy. Hepatectomies also may have unsatisfactory results and require reoperation (28).

Conclusion

In conclusion, treatment of intrahepatic lithiasis is still a matter of debate, basically on the comparison between surgical and conservative approaches. Also the role of resection compared to biliary derivation is controversial. Percutaneous approach seems more effective than endoscopic treatment and is well indicated in case of unilateral disease without complex strictures. Biopsy under visual control is the unchallenged advantage of cholangioscopy; however, it carries a higher morbidity. Multiple strictures with biliary distortion, hepatic fibrosis or atrophy and a suspected cholangiocarcinoma are absolute indications to resection. Integration of therapeutic protocols in multidisciplinary teams offers the best long-term results.

References

22. Otani K, Shimizu S, et al. Comparison of treatments for hepato-