

OUTCOMES OF LIVER RESECTION FOR LOCALISED INTRAHEPATIC STONES IN A LOW INCIDENCE COUNTRY

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Abstract

Background: Intrahepatic stones (IS) are very common in far eastern patients but relatively rare elsewhere in the world. The aim of this study is to report the short-term and mid-term outcomes of liver resection indicated for primary intrahepatic stones in a low incidence country.

Results: From January 2010 to December 2014, 5(2,7%) among 182 consecutive liver resections were performed for intrahepatic stones (IS) in the Surgical Department A at IbnSina Hospital (Rabat, Morocco). Three patients had IS secondary to Caroli's disease and 2 patients had IS secondary to benign postoperative biliary stricture. Liver resections consisted of 2 right hepatectomies and 3 left hepatectomies. Hepaticojejunostomy was associated in 2 patients. Postoperative mortality was not reported. Two patients experienced postoperative morbidity (1 biloma and 1 severe cholangitis). No recurrence of symptoms occurred after a median follow-up of 36 months.

Conclusion: Liver resection can provide definitive resolution of symptoms in selected patients with localized intrahepatic stones.

Keywords: Intrahepatic Stones, Liver Resection, Outcome.

Introduction.

The presence of stones at the level and/or above the biliary confluence can result from [1] migration of gallbladder stones into intrahepatic biliary ducts, [2] intrahepatic development of stones proximal to biliary stricture or inside cystic biliary dilatation such as Caroli's disease and/or [3] primary intrahepatic formation of stones in presence of risk factors such as biliary stasis and bacterial infection. This last mechanism is very common in East Asia (intrahepatic stone disease) but rare elsewhere in the world. Intrahepatic stones typical clinical manifestation is recurrent cholangitis but the evolution of the disease can lead to life threatening complications such as secondary biliary cirrhosis and/or cholangiocarcinoma. Partial liver resection is considered today as the most rational treatment for localized intrahepatic stones as it permits to

remove stones but also biliary strictures, cystic dilatations and atrophic liver parenchyma. These challenging liver resections are associated with high postoperative morbidity rates including bile leaks and/or sepsis. The aim of this study is to report the short-term and mid-term outcomes of liver resection indicated for intrahepatic stones in a low incidence country.

Patients and methods:

From January 2010 to December 2014, 5 (2.7%) among 182 consecutive liver resections included in a prospective database were performed for intrahepatic stones (IS) in the Surgical Department A at Ibn Sina Hospital (Rabat, Morocco).

Liver resection was indicated during a multidisciplinary meeting in patients with symptomatic and localized IS as showed by a

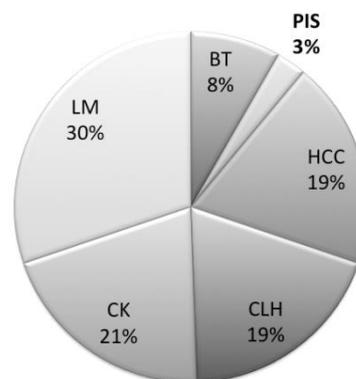
complete imaging assessment (ultrasound, computed tomography and magnetic resonance imaging). The timing of surgery was delayed 1 month at least from any episode of cholangitis. Extent of anatomic liver resection was planned in order to remove stones, biliary strictures, biliary cystic dilatations and atrophic liver parenchyma. Roux-en-Y hepaticojejunostomy was considered in association with liver resection [1] when the biliary confluence was resected because of a stricture and/or [2] when subsequent percutaneous biliary interventions were considered because of incomplete stone clearance.

Clamp crushing technique and ligation of vascular and biliary pedicles was used to perform liver parenchyma transection. Final hemostasis of the rough parenchymal surface was achieved by bipolar cautery. Intermittent clamping of the pedicle was used selectively.

The following data were reviewed and analyzed: sex and age of patients; past history of biliary surgery; clinical manifestation of the disease; preoperative imaging assessment (location of stones in the biliary tract, presence of biliary stricture and/or biliary cystic dilatation); surgical procedure (extent of liver resection, hepaticojejunostomy, duration, intraoperative estimated blood loss and transfusion); pathology examination of the liver resection specimen; postoperative morbidity according to Clavien-Dindo classification; clinical outcome according to Terblanche classification: grade I, no biliary symptoms; grade II, transitory symptoms and no current symptoms; grade III, biliary symptoms requiring medical therapy; and grade IV, recurrent biliary symptoms requiring correction or related to death. Follow-up data were obtained by means of review of hospital records and patients phone calls.

Results:

Between January 2010 and December 2014, 3 women and 2 men were operated for symptomatic intrahepatic stones (Figure 1).



BT: benign tumors; CK: cholangiocarcinomas; CLH: cystic liver hydatidosis; HCC: hepatocellular carcinoma; LM: liver metastasis; PIS: primary intrahepatic stones

Figure 1. Distribution of indications for liver resection in Surgical Department A (N=182; 2010-2014)

Their median age was 50 years [16-70]. Two patients had a history of biliary surgery: cholecystectomy for symptomatic gallbladder stones (n=1) and hepaticoduodenostomy for main bile duct stones (n=1). Patients presented with right upper quadrant abdominal pain (n=5), fever (n=3) and/or jaundice (n=2). Median duration of symptoms before surgery was of 6 months [1-96 months]. Preoperative assessment was consistent with the diagnosis of Caroli's disease in 3 patients (Cases 1 to 3) and the diagnosis of postoperative benign biliary stricture in 2 patients (Cases 4 and 5). No patient had evidence of portal hypertension and/or impairment of liver function. Anatomic liver resection was performed in all patients: 2 right hepatectomies, 2 left hepatectomies and 1 left hepatectomy extended to segment 1. A Roux-en-Y hepaticojejunostomy was associated to the liver resection in 2 patients that presented with jaundice. Postoperative mortality was nil. Postoperative morbidity occurred in the 2 patients that underwent a right hepatectomy: biloma (Clavien Dindo 3a) that required percutaneous drainage in one case and a severe cholangitis (Clavien Dindo 4a) that required intensive care management in the other case. Neither malignancy nor cirrhosis was found on pathological study in all patients. No recurrence of symptoms (Terblanche I) occurred after a median follow-up of 36 months [15-51 months]. Clinical and operative data are presented in Table I and II.

Table I: Clinical and radiological presentation of patients with intrahepatic stones

Case	Year	Sex	Age (Years)	Past biliary surgery (Delay)	Symptoms	Duration of symptoms (Months)	Location of stones	Liver atrophy	Bile ducts abnormalities
1	2013	F	38	-	Pain, jaundice	1	LHD+BC	-	LL cystic dilatation
2	2012	M	51	-	Pain	3	LHD	LL	LL cystic dilatation (Figure 2)
3	2011	F	16	-	Pain, fever	96	RHD	-	RL cystic dilatation
4	2012	F	70	Hepaticoduodenostomy (12mo)	Pain, fever	6	LHD	LL	LHD stricture
5	2013	M	55	Cholecystectomy (184mo)	Pain, jaundice, fever	36	RHD+BC	RL	RHD stricture (Figure 3)

M, male; F, female; BMI, body mass index; LHD, left hepatic duct; RHD, right hepatic duct; BC, biliary confluence; LL, left liver; RL, right liver

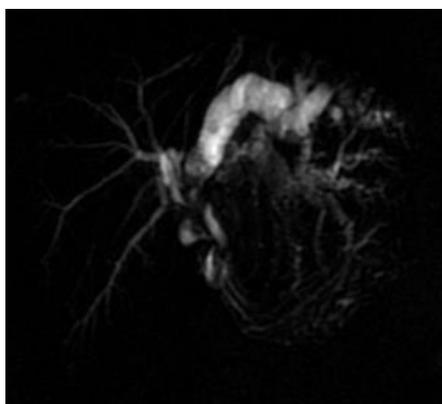


Figure 2. Primary intrahepatic stones inside biliary cystic dilatation of the left liver (MRI, Case 2)



Figure 3: Right liver parenchymal atrophy and intrahepatic stone at biliary confluence (MRI, Case 5)

Table II: Operative data and outcome of liver resection for intrahepatic stones

Case	Extent of Liver resection	Associated procedure	Operative time (Min)	Blood loss (mL)	Transfusion (RBU)	Morbidity (Clavien-Dindo)	Hospital stay (Days)	Follow-up (Months)	Outcome (Terblanche)
1	Left hepatectomy+S1	Hepaticojejunostomy	360	600	2	-	8	36	I
2	Left hepatectomy (Figure 4)	-	390	400	-	-	5	50	I
3	Right hepatectomy	-	210	300	-	Biloma (3a)	9+16	51	I
4	Left hepatectomy	-	180	350	-	-	7	15	I
5	Right hepatectomy	Hepaticojejunostomy	300	1500	3	Sepsis (4a)	21	15	I

RBU: Red blood units

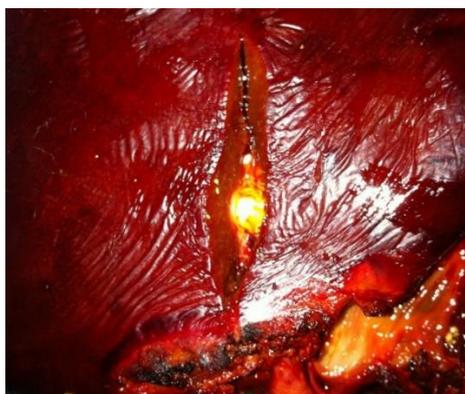


Figure 4: Left hepatectomy specimen with intrahepatic stones (Case 2)

Discussion:

In North Africa, there is no data available concerning intrahepatic stones (IS). This series reports the experience of a Moroccan referral hepatobiliary surgery unit with liver resection for symptomatic intrahepatic stones. During a 5-year study period, only 5 (<3%) liver resections were performed for IS. None of these patients had an East Asia type primary intrahepatic stone disease but [1] IS secondary to localized Caroli's disease (Cases 1- 3) and [2] IS secondary to postoperative benign biliary stricture (Cases 4 and 5).

All biliary symptoms present in patients were related to IS such as right upper quadrant abdominal pain, fever and jaundice. Cholangitis is always present in presence of IS but may vary in severity from simple mild uncharacteristic abdominal pain to septic shock [1,2]. A symptom-based system was proposed by the Hepatolithiasis Research Group in Japan to grade the severity of hepatolithiasis, grade 1 is defined as having no symptom, grade 2 as having abdominal pain, grade 3 as having either transient jaundice or cholangitis, and grade 4 as having either continuous jaundice, sepsis, or cholangiocarcinoma. [3] According to this classification, 4 out of 5 of our patients may be classified \geq grade 3 because of continuous jaundice and/or at least one episode of cholangitis. The recurrence of bacterial cholangitis may lead to secondary sclerosing cholangitis, biliary strictures, parenchymal atrophy and liver abscesses.[2]

In Caroli's disease, a conservative (stone dissolution with ursodeoxycholic acid) and/or interventional (endoscopic retrograde stone removal and biliary drainage) approach may provide momentary remission of the symptoms. However, non-surgical approach to Caroli's disease is associated with constant cholangitis recurrence and potentially lethal complications such as liver abscess and cholangiocarcinoma [4-6]. For these

reasons, liver resection stands as a rational option when the disease is localized to one lobe and that the liver remnant parenchyma is healthy. When hepatic fibrosis or cirrhosis is present, liver transplantation may be considered [7,8]. Neither malignancy nor fibrosis was found in the surgical specimen examination of our 3 patients with Caroli's disease.

Most postoperative benign biliary stricture is secondary to a bile duct injury (BDI). A vascular injury may be associated to the BDI in up to 39% of the patients [9, 10]. In these situations, IS may reveal a complex situation that requires a multidisciplinary management. A liver resection is considered when the biliary stricture is not amenable to a simple repair (secondary biliary confluence) and/or when there is an atrophy of one lobe. [11, 12]

In our experience, IS was always secondary to a biliary abnormality such as a dilation (Caroli disease) or a stricture (postoperative benign stricture). Liver resection was the treatment of choice as it provided definitive resolution in patients with intrahepatic stones and/or stenosis and/or dilatation of the lobar or segmental ducts. These challenging situations should be managed in specialized centers.

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