Liver resection in the treatment of intrahepatic lithiasis. Immediate and long-term results in a single-center series

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RESUMEN

Objetivo: la litiasis intrahepática (LIH) es una entidad poco frecuente en nuestro medio. Cuando se asocia a enfermedad de Caroli o a estenosis de los radicales biliares puede ser necesaria la resección hepática para su resolución definitiva. Presentamos los resultados a corto y largo plazo en una serie española de pacientes con resección hepática como tratamiento de la LIH.

Pacientes: entre enero de 1996 y diciembre de 2007 realizamos una resección hepática en 8 pacientes por LIH. En 3 casos la LIH se asoció a enfermedad de Caroli y en los 5 casos restantes se asoció a estenosis de radicales segmentarios. La LIH se manifestó como colangitis aguda en 5 casos, como cólico hepático en dos casos y como pancreatitis recidivante en un caso. La técnica quirúrgica fue una hepatectomía derecha, 2 hepatectomías izquierdas y 5 resecciones segmentarias.

Resultados: no existió mortalidad intra- ni postoperatoria. La morbilidad fue del 25%. En el seguimiento a largo plazo se produjo la muerte por colangiocarcinoma en un paciente (12%) con enfermedad de Caroli que había desarrollado la enfermedad malignizada. El resto de pacientes (88%) no ha habido recurrencia de la enfermedad a lo largo de un seguimiento medio de 62 ± 2 meses (rango 31-106). En los 7 pacientes restantes, no se ha producido la muerte por enfermedad de Caroli.

Conclusiones: en nuestra experiencia, la resección hepática es el tratamiento indicado en aquellos pacientes con LIH asociada a enfermedad de Caroli o con estenosis de radicales biliares, consiguiendo una resolución completa de la enfermedad con baja morbimortalidad.


ABSTRACT

Objective: intrahepatic lithiasis (IHL) is an uncommon entity in our environment. When associated with Caroli’s disease or stenosis of the biliary radicals it may be necessary to perform liver resection to provide definitive resolution. We present immediate and long term results in a Spanish series of patients with hepatic resection to treat hepatolithiasis.

Patients: between January 1996 and December 2007 we performed a liver resection (LR) in 8 patients for IHL. The IHL was associated with Caroli’s disease in 3 cases and with stenosis of segmentary radicals in the other 5 cases. It manifested itself as acute cholangitis in 5 cases, as biliary colic in 2 cases and recurrent pancreatitis in one case. The surgical technique was a right hepatectomy, 2 left hepatectomies and 5 segmentary resections.

Results: there was no intra- or postoperative mortality. The morbidity rate was 25%. One patient (12%) with Caroli’s disease which had malignised to cholangiocarcinoma died in the follow-up period. The remaining 7 patients have had no IHL recurrence after a mean follow-up of 62 ± 2 months (range: 31-106).

Conclusions: in our experience liver resection, either lobar or segmentary, is the treatment for patients with IHL associated with stenosis and dilatation of the bile duct, as it provides complete resolution of the disease with low rates of morbidity and mortality.

Key words: Intrahepatic lithiasis. Liver resection. Cholangiocarcinoma. Caroli’s disease.

INTRODUCTION

Intrahepatic lithiasis (IHL), or the presence of stones in the intrahepatic bile ducts, particularly affects women and is very common in Southeast Asian countries (10-15% of patients undergoing biliary surgery for lithiasis) (1-3) and less so in South America (0.5-2%) and Europe (4); reports in our country are scarce with few cases (5-8). This condition may be associated with complications such as cholangitis, liver abscesses, hepatic insufficiency, and even malignant progression to cholangiocarcinoma (CCC). Stones associated with no underlying pathology and located in the main intrahepatic biliary radicals can be removed by cholecdochothy, endoscopy, or external
radiological drainage (9-11). In cases where IHL is associated with stenosis or dilated intrahepatic bile ducts (Caroli’s disease) the treatment of choice is liver resection to prevent recurrence and complications (12,13).

The aim of this article is to discuss 8 patients with IHL associated with stenosis or biliary dilatations who underwent liver resection for definitive resolution.

PATIENTS

Between January 1996 and December 2007 we performed 475 liver resections in our liver surgery and liver transplantation unit, the indication for which was IHL in 8 patients. Mean patient age was 42 ± 3 years (range: 29-77), and there was a predominance of males over females (6:2). Seven patients were Caucasian and one woman was Asian. Mean follow-up was 62 ± 2 months (range: 31-106).

Two of these 8 patients had been previously cholecystectomized (10 and 12 years before). The initial diagnosis was cholangitis in 5 patients and biliary colic in 2 patients; disease onset in the remaining case was related to recurrent pancreatitis. All cases had altered liver enzymes, and mean bilirubin was 3.6 g/dl (range: 0.8-8.6). Preoperative ultrasound was the initial diagnostic technique in all 8 cases, and detected IHL in 6 patients; it also revealed choledocholithiasis in 4, and dilated biliary radicals in 3 cases (Caroli’s disease). Of the two remaining cases in which IHL was not detected, one had abscesses in segments V and VI, and one had cholelithiasis with choledocholithiasis. Computed tomography (CT) and magnetic resonance imaging (MRI) confirmed the presence of intrahepatic stones in all cases. Four patients also had an endoscopic retrograde cholangiopancreatography (ERCP), which showed IHL and biliary dilatations in the right liver lobe (RLL) in one case, amputation of the left biliary radical in another, and choledocholithiasis in the remaining 2 cases, stones in the latter case being removed after papillotomy. Two patients underwent transpapillary cholangiography (TPHC), which revealed IHL and stenosis of the biliary radical in segment VI in one case, and Caroli’s disease in left liver lobe (LLL) in the other case (Table I).

The initial therapeutic approach in five patients was right hepatectomy (due to associated Caroli’s disease), 2 left hepatectomies (one of them associated with Caroli’s disease), 1 resection of segment VIII, and 1 left lateral bisegmentectomy (associated with Caroli’s disease). In the latter case we detected an incidental CCC in the pathological study; the patient underwent re-operation to extend the surgical margin with segmentectomy of segments IV and I, resection of the common bile duct, and hepatojjunostomy reconstruction.

The initial operation for the remaining 3 patients was cholecystectomy and choledochocholangiograpy in 2 cases, and choledochochotomy with closure over a T-tube in the third case. The outcome in these 3 patients was torpid, and they presented with further symptoms of cholangitis due to recurrent disease; they underwent re-operation with liver resection as the definitive technique (segmentectomy VI and VIII, bisegmentectomy V and VI, and in the third case segmentectomy V and VIII) (Table II).

RESULTS

Early morbidity and mortality

There was no intra- or postoperative mortality. None of the patients required intra- or postoperative transfusion. Two patients (25%) had a subphrenic abscess with detection of polymicrobial flora in the culture, which was resolved with radiological drainage and antibiotics.
Long-term results

Only one patient who revealed an incidental CCC died 24 months after surgery due to non-resectable liver recurrence (12% mortality). The remaining 7 patients have shown no IHL recurrence after a mean follow-up of 62 ± 2 months (range 31-106). It should be noted that the 5 patients who underwent liver resection as their initial surgery had no recurrence. However, all 3 cases that underwent only a bile duct approach for stone removal as initial surgery presented with further symptoms of cholangitis and required liver resection for a definitive resolution of their disease.

DISCUSSION

IHL can be a serious problem, as it has a morbidity rate of 20-30% (occasionally requiring several operations for resolution), and a mortality rate of 1-10% (9-22).

The origin of intrahepatic stones may be twofold: Primary intrahepatic stones and stones emigrating from the gallbladder to the common bile duct, and from here to intrahepatic radicals (secondary) (23). In Southeast Asia IHL is usually associated with intrahepatic biliary radical stenosis, and is believed to be secondary to parasitical or bacterial infection, which conditions prestenotic dilatation, bile stagnation due to overinfection, and primary stone formation (12,20,23-25). In Western countries IHL is uncommon and usually secondary to lithiasis in the common bile duct, and subsequent stone migration into the liver. Primary intrahepatic stones (12,15,26,27) exist in Caroli’s disease, sclerosing cholangitis, cholangiocarcinoma, post-traumatic or post-surgical biliary strictures, etc. In our series stones were intrahepatic and primary in 3 cases, which had Caroli’s disease. The remaining 5 cases had a secondary origin associated with choledocholithiasis.

Serious forms of clinical manifestation have been described (12,21,26) (suppurative cholangitis, hemobilia, multiple liver abscesses, secondary biliary cirrhosis with portal hypertension and esophageal varices, acute liver failure, and even cholangiocarcinoma). There were reduced symptoms of acute cholangitis in five of our patients, who were managed easily with antibiotic treatment, and in one patient who had an incidental cholangiocarcinoma detected in the histological study, and who died from tumor recurrence after 24 months.

From a diagnostic point of view preoperative ultrasounds is the imaging technique that should raise suspicion for IHL (5,27-30); useful techniques for confirming this diagnosis include CT, MRI, and particularly cholangio-resonance, which may prevent the need for invasive tests. ERCP is useful when there are stones in the common bile duct, as these can be removed (as occurred in two of our patients). Occasionally it allows a visualization of intrahepatic biliary dilatations and IHL. However, indications for ERCP should be individualized due to the risk of associated infectious complications. TPHC allows visualizing biliary dilations, IHL, and stenosis, as occurred in two of our patients. In three of our patients US and CT-MR suggested the presence of IHL, although the surgeon did not attach importance to this finding and only acted on the gallbladder and common bile duct in the first operation; all of them required a second operation to resolve IHL (Table II).

The treatment of IHL may be surgical or instrumental. The latter can be done under percutaneous radiological control (percutaneous transhepatic cholangioscopy, T-tube approach, and others) or under endoscopy with or without associated lithotripsy (3,12,16,29,31,32). The surgical treatment of stones can be done via choledochotomy, hepatotomy, or liver resections (4,5-8,12,15,17,19,24,26,27,33-35).

The indications for one or another form of treatment depend on IHL characteristics: Stones secondary to choledochal lithiasis without stenosis or underlying hepatic pathology are often easily removed via choledochotomy or endoscopic or radiological approaches; however, in patients whose IHL is associated with stenosis and dilatations, or with an underlying hepatic pathology (Caroli’s, cholangiocarcinoma, etc.) simple stone re-

### Table II. Surgical technique for the 8 patients in our series

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<tr>
<th>Case 1</th>
<th>Case 2</th>
<th>Case 3</th>
<th>Case 4</th>
<th>Case 5</th>
<th>Case 6</th>
<th>Case 7</th>
<th>Case 8</th>
</tr>
</thead>
<tbody>
<tr>
<td>1st intervention</td>
<td>RH</td>
<td>Cholecystectomy, choledochotomy C-D</td>
<td>Cholecystectomy, choledochotomy, T-tube</td>
<td>Segmentectomy VIII</td>
<td>Lateral bisegmentectomy CCC</td>
<td>Cholecystectomy, choledochotomy C-D</td>
<td>Cholecystectomy, LH</td>
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<tr>
<td>2nd intervention</td>
<td>IO-US</td>
<td>IHL in S. VI and VII</td>
<td>IHL in S. V and VI</td>
<td>Free surgical margen</td>
<td>S IV and I</td>
<td>Segmentectomy</td>
<td>IHL, S. V and VIII</td>
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<td>Technique</td>
<td>Segmentectomy VI and VII</td>
<td>Segmentectomy V and VI</td>
<td>Hepaticojejunostomy V and VIII</td>
<td>Liver recurrence death at 24 months</td>
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<tr>
<td>Evolution</td>
<td>Cure</td>
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RH: Right hepatectomy; C-D: Choledochoduodenostomy; IHL: Intrahepatic lithiasis; S: Segment; CCC: Cholangiocarcinoma; LH: Left hepatectomy.
moval is not sufficient because recurrence is universal. In the latter situation, presented by our patients, most authors (5-8,12,15-19,26,29,32-38) consider liver resection the technique of choice, as it not only eliminates IHL and factors responsible for its formation, but also possible sequelae such as associated cholangiocarcinoma (5-8,15,18,19,26,34), as occurred in one of our patients. During surgery intraoperative ultrasound and intraoperative cholangiography (28,30) are the fundamental exploratory techniques for locating stones and ensuring they have all been eliminated.

The indications for liver resection are atrophic lobule secondary to repeated infection, stones limited to one segment or one lobule, and associated pathology (Caroli’s, cholangiocarcinoma, stenosis, etc.). Some authors (11,17,24) obtain good results in bilateral IHL when performing left hepatectomy, dilation, and removal of stones from the right side, then reconstructing with hepatojejunostomy to the right hepatic duct for potential subsequent approaches (36,38). In exceptional and very selected cases in which liver resection does not resolve the problem, transplantation might be considered as a therapeutic option, especially in patients with an associated condition with malignant potential such as Caroli’s disease (39). When Caroli’s disease is limited to the right or left liver lobe, which is uncommon, a right or left hepatectomy is the technique of choice, as this technique can be currently performed with low morbidity and mortality rates. Some authors (11-20) report that after hepatectomy some patients require an external approach (radiological and/or endoscopic) for the removal of retained stones. Some (2,11,12,15,36) recommend that these techniques be associated with biliary derivations to the jejunum associated in turn with jejunostomies to allow future endoscopic operations (16,36), or leaving a T-tube in the common bile duct for potential subsequent external radiological approaches (25).

In conclusion, liver resection is the definitive treatment for patients whose IHL is associated with stenosis and dilations, provided it may be performed with low postoperative mortality and morbidity rates.

REFERENCES