HEPATOBILIARY CYSTADENOCARCINOMA RESECTION USING RADIOFREQUENCY COAGULATION – A CASE REPORT

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Hepatobiliary cystadenocarcinoma (HC) is a rare entity: fewer than one hundred cases have been reported worldwide (1). The demonstration of benign epithelium in the majority of cases of HC makes it probable that these arose from previously benign cystadenoma (1). Clinical symptoms are mostly related to a mass effect. We report a case of HC located in the left lobe of the liver and treated by left lateral sectionectomy after radiofrequency (RF) coagulation of the liver parenchyma. Epidemiology, diagnosis and treatment of HC are discussed.

CASE REPORT

A 75-year-old woman was referred to our hospital complaining of sporadic epigastric pain, anorexia and weight loss. She had no past medical history. The physical examination demonstrated a large palpable epigastric mass. Routine blood analyses including liver function were normal. Serum dosages of tumour markers showed an increased carbohydrate antigen (CA) 19.9 level up to 172 U/ml (normal <31) and a normal carcinoembryonic antigen (CEA) level. Abdominal sonography showed a well circumscribed and multilocular cystic mass in the left lobe of the liver measuring eight centimetres in diameter containing septations. An unenhanced and contrast enhanced magnetic resonance (MRI) of the abdomen showed a well marginated, septated and multilocular cystic mass of the left lobe, highly suspected of malignancy (fig. 1). The lesion bulged into the stomach without any sign of frank tumoral invasion. The portal region was not displaced and there was no evidence of bile duct dilatation or invasion of the portal vein. We performed a left lateral sectoricetomy following radiofrequency coagulation of the liver's paren-
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Fig. 1. Abdominal MRI showing a well marginated, septated and multilocular cystic mass of the left lobe of the liver.

Chyma under the control of peroperative sonography (fig. 2). A control of Glissonian’s pedicles of segments 2 and 3 was first performed. We used the Radionics® (Burlington, USA) system: a peristaltic pump is used to infuse saline at 0°C into the lumen of a hollow electrode to maintain the electrode tip temperature at 20-25°C. Heat is created using a 500 kHz generator resulting in coagulation of the parenchyma. The cooled electrode prevents overheating of the tissue adjacent to the electrode, thus preventing charring. A RF electrode was applied in the liver parenchyma every centimetre along the plane of the discoloured surface for one minute. Hepatectomy was achieved using the monopolar electrocautery knife. A five millimetre edge of coagulated liver was left in place (fig. 3). Blood loss was less than 50 millilitres.

The histopathology analysis revealed a biliary mucinous cystadenocarcinoma. A positive tumoral marker for CA-19.9 was found on immuno-histochemical analysis. The CA-19.9 serum dosage dramatically decreased at day 2. The postoperative course was uneventful and the patient was discharged on postoperative day 12.

DISCUSSION

Cystadenoma and cystadenocarcinoma of the liver are rare and account for less than 5% of intrahepatic cystic masses of biliary origin (2). Hepatobiliary cystadenocarcinomas (HC) are exceptional malignant tumours of the liver with an incidence of 0.41% among all hepatic malignant epithelial tumours, including autopsy cases (3). Extrahepatic tumours have been rarely described; they usually arise from the extrahepatic bile duct or the gall-bladder (2). Women and men are affected equally (1). Cysts represent dilated intrahepatic bile ducts invaded by tumour cells with a colloid carcinoma-like pattern. HC is not associated with any predisposing factor and its exact pathogenesis remains unclear; one of the hypothesis is based on histological findings which suggest that some HCs arise from previously benign cystadenomas (4).

Fig. 2. Application of the RF ‘s electrode

Fig. 3. Specimen of cystadenocarcinoma of the left lobe
It is argued that two types of HCs exist; one presents an “ovarian-like” stroma and concern women exclusively (1). The other type lacks ovarian-like stroma and concern both men and women (1). Clinical symptoms are usually related to the mass effect of the lesion and consist of intermittent abdominal pain, anorexia, nausea, abdominal fullness and weight loss (5). In addition, a cystadenocarcinoma may open whether in the biliary tree leading to a biliary obstruction (5), or in the adjacent organs as the duodenum, causing intratumoral gas bubbles at sonography (6). Diagnosis of HC is suggested upon viewing radiological findings.

Some radiological malignant criteria have been described but a reliable differentiation between biliary cystadenoma and cystadenocarcinoma can be difficult; at sonography, HCs appear as cyst-like multilobular, hypoechoic lesions with internal septa and small nodules at the level of the cystic wall (7). Although distinction between cystadenoma and cystadenocarcinoma cannot be achieved with radiological findings alone, this is not of grave importance since surgical excision is considered the therapy of choice due to the premalignant nature of biliary cystadenoma. However, differentiation from other pathological entities such as a hepatic abscess and a hydatid cyst of the liver is of crucial importance. These infectious diseases can be evaluated using laboratory findings.

Some hemorrhagic congenital hepatic cysts can mimic HC because both are slow growing lesions filled with a low-attenuating fluid (mucin, blood, bile). Furthermore, both may present with septa on abdominal CT or magnetic resonance imaging (MRI). However, HCs generally are solitary lesions, whereas congenital hepatic cysts tend to be multiple. On CT or MRI findings, malignancy may be suspected by the presence of large pedunculated excrescences into the cyst lobules and septal calcifications, although papillary areas and polypoid projections have been reported in cystadenomas without frank malignancy (7). After a contrast media injection, the septa, mural nodules and pedunculated projections show enhancement (7). The differential diagnosis includes other very rare cystic neoplasms such as multilocular cystic hepatocellular carcinoma without cirrhosis, mimicking mucinous cystadenocarcinoma (8). The incidence and prognosis of this rare entity is unknown. It has been suggested that high serum and cystic level of the tumoral marker CA-19.9 may be of value in the diagnosis of cystadenoma but not cystadenocarcinoma (9). However, a normal serum level and a very high cystic fluid level of CEA and CA-19.9 have been reported in a liver cystadenoma (9). Although an elevated level of CA-19.9 in the cyst fluid may orient one towards cystadenoma or a cystadenocarcinoma (9), puncture of the cyst should be avoided because low cystic fluid levels of CA-19.9 associated with a normal serum level of CA-19.9 have been found in a cystadenocarcinoma without ovarian-like stroma (10). In addition, even fine needle puncture may be responsible for peritoneal tumour dissemination (5). For the same reason, an “en bloc” resection of the tumour is usually recommended, with care taken to not break the tumour. Moreover, complete resection is the treatment of choice because of difficulty in accurately predicting malignancy (5) and because there is a high likelihood of recurrence with procedures other than total ablation (11).

In the present case, we performed a left lateral sectoriectomy after coagulation of the liver’s parenchyma using a radiofrequency device because this technique has been reported to be safe and blood-sparing (12). No transfusions were needed during or after the operation. As usual, a peroperative sonography was mandatory to locate the position of the RF’s needle in the liver’s parenchyma so that burn lesions related to RF effects on major Glissonean’s pedicles would be avoided. Furthermore, perioperative use of sonography allows for visualization of possible excrescences that may arise from the tumour so that a complete resection could be performed. These circumstances may modified the planned hepatectomy.

**CONCLUSION**

Hepatobiliary cystadenocarcinoma of the liver remains a very rare entity. Women and men are equally affected. Despite the use of sonography and computed tomodensitometry, preoperative assessment of malignancy is a difficult challenge. Furthermore, if surgery remains the only chance for cure, resection should be complete and perioperative sonography is mandatory to identify possible extra-tumoral projections leading to an extended and unexpected hepatectomy. Although our experience with RF in liver resection is limited to segmentectomies or bisegmentectomies, we think...
that when surgery is undertaken, one must be able to undertake major hepatectomy. In accordance with other authors (12), this case outlines advantages of radiofrequency coagulation of the parenchyma as a safe technique to be used before liver resection.

REFERENCES