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Multicystic biliary hamartoma

Hamartoma multiquístico de vías biliares

Multicystic biliary tract hamartoma is a rare cystic disease of the liver, of which only 8 cases have been published worldwide as of 2012[1]. It is usually asymptomatic, and, in most published series, it is diagnosed incidentally or as a result of autopsy studies[2].

We present the case of a 45-year-old woman, with no relevant personal history, with clinical symptoms of epigastric abdominal pain over several months and no other associated symptoms. An abdominal ultrasound scan was performed, followed by magnetic resonance imaging (MRI), which revealed a polylobulated cystic liver lesion with internal septa, located between segments VII–VIII, measuring approximately 6.5 × 6 × 6.5 cm. The peripheral loculations showed fluid content, while the central loculation showed heterogeneous haemorrhagic content. An angio-CT scan was also performed (Fig. 1), with findings compatible with biliary cystic neoplasia, and a PET/CT scan, with no evidence of SOL uptake. Laboratory results showed no abnormalities, and CEA and Ca 19.9 values were normal.

It was decided to perform elective surgery via right subcostal laparotomy, showing a hepatic SOL in segments VII and VIII with apparent infiltration of the right suprahepatic vein, without free fluid or other hepatic or peritoneal lesions. A right hepatectomy was performed with resection of diaphragmatic pastille of approximately 5 × 5 cm due to infiltration by the tumour. A sample of the intracystic fluid was taken, which was sterile and showed cytological findings of extensive tumour necrosis and few atypical cells suspicious of adenocarcinoma.

The final anatomopathological analysis showed a right hepatectomy specimen with multiple cysts lined with biliary epithelium, without atypia, with the presence of haemorrhagic content, thrombosis and partial recanalisation in the largest cyst, without identifying ovarian-like stroma, all indicative of multicystic biliary hamartoma (Fig. 2). No other abnormalities were identified in the rest of the liver parenchyma.

The patient progressed favourably and was discharged home on the fifth postoperative day without complications.

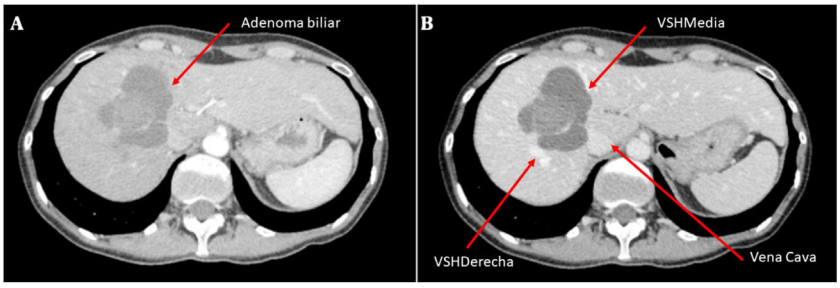

Fig. 1 – (a) 3D angio-CT reconstruction showing hypervascularised tumour in the left carotid bifurcation. (b) Embolisation by direct puncture with Onyx®.

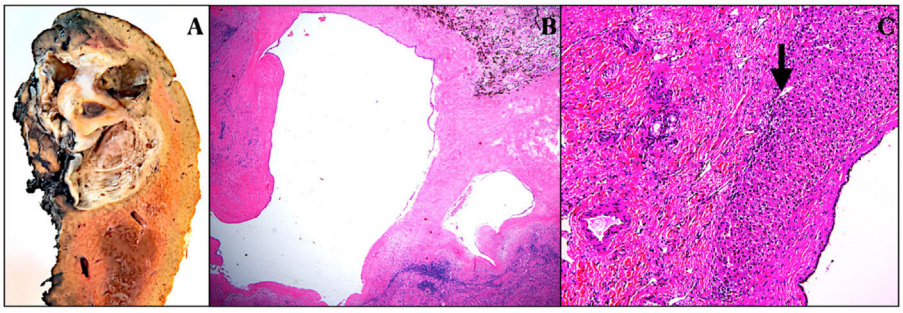

Fig. 2 – (A) Right hepatectomy specimen with identification of a multicystic lesion with cavities between .3–4 cm in diameter with abundant haematic content inside, located immediately proximal to the right SHV. (B) Multiple cysts lined with cylindrical or cuboidal biliary epithelium without atypia, with areas of haemorrhage at the periphery. H&E staining. 10× magnification. (C) Cysts lined by cuboidal biliary epithelium without atypia and interposed liver tissue (arrow). No ovarian-type stroma is observed in the lamina propria of any of the cysts. H&E staining. 10× magnification.

Multicystic biliary hamartoma was first described in 2006[1] as a rare lesion of unknown aetiology[2], characterised by irregular and branched bile ducts with a malformed appearance.

It is considered a benign liver malformation originating as a result of a disturbance in the bile duct cells triggered by a disruptive or ischaemic factor during bile duct lamina remodelling[3], which is an embryonic structure generated by the development of the distal ducts[2].

As mentioned above, it is usually asymptomatic, although it can sometimes be associated with episodes of pain in the right hypochondrium, as well as bloating and a sensation of abdominal mass[2].

It is not usually associated with analytical abnormalities[2] and radiologically it is very similar to malignant lesions, such as liver metastases[4], making differential diagnosis by imaging tests alone difficult. MRI-cholangiography is probably the most important imaging test in establishing the differential diagnosis[3].

Radiological features include a honeycomb appearance and the presence of normal liver parenchyma intermingled within the nodular lesion, usually in the peripheral portion of the lesion[5]. It can be differentiated from biliary cystadenoma by its shape, as multicystic hamartoma has a honeycomb-like appearance and the cysts are relatively uniform in size, although they may vary if haemorrhage occurs, whereas this does not occur in cystadenoma or biliary cystadenocarcinoma[5].

The definitive diagnosis is established by biopsy, which should be taken by wedge or core needle biopsy[2], as fine needle aspiration biopsy is usually nondiagnostic and leaves more uncertainty[6].

Macroscopically, there are superficial, subcapsular nodulations, .1 to .5 cm in diameter, with no proven lobar predilection, although there are cases that suggest that there may be a greater frequency in the right lobe[7].

It is a lesion that is usually located around the hepatic capsule, near the fissure of the falciform ligament, protruding from the liver. It is usually composed of ductal structures, periductal glands and fibrous connective tissue, which contain biliary material in these structures[8].

It is treated conservatively, although it is usually operated due to diagnostic uncertainty, and cases of cholangiocarcinoma have been reported almost anecdotally[2].

Authorship

Victoria Carmona: drafting of the article; critical review and approval of the final review.

Iago Justo: data collection and acquisition; critical review and approval of the final review.

Yolanda Rodríguez-Gil: data collection and acquisition.

Alberto Marcacuzco: critical review and image selection.

Carmelo Loinaz: approval of final review.

Carlos Jiménez: critical review and approval of final review.

Conflict of interests

The authors have no conflict of interests to declare.

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Resection of carotid paraganglioma after percutaneous embolization with Onyx® under radiological control

Resección de paraganglioma carotídeo previa embolización percutánea con Onyx® bajo control radiológico

Carotid body tumours, glomus tumours or chemodectomas[1] are the most common paragangliomas of the head and neck. They are rare, benign neoplasms, whose most frequent clinical presentation is a slow-growing, asymptomatic mass, which causes compressive symptoms in some cases[1,2].

Clinical history, physical examination, and radiological diagnosis are the cornerstones of diagnosis and treatment[1,3]. Angiography is essential to study the vascular anatomy, particularly the ''lyre sign'', typical of large tumours, which can distort the carotid bifurcation, extending the internal carotid artery (ICA) and external carotid artery (ECA)[1,2].

Management of these tumours is technically challenging due to the size, location and hypervascularised state. They usually receive their blood supply through branches of the ECA[2]. Early surgical excision is considered the primary curative treatment option[1,2]. Advances in endovascular techniques have resulted in endovascular techniques to treat these tumours. Advances in endovascular techniques have enabled hybrid techniques, which involve embolisation of the tumour prior to surgical excision. These techniques are used in large tumours, with the aim of occluding the tumour's nutrient vessels, reducing intraoperative bleeding and, therefore, morbidity and mortality[4–7]. Preoperative embolisation remains controversial, as it is not without risks, such as increased subsequent morbidity and inflammatory effects of the tumour, which may hinder accurate subadventitial dissection[8]. Transarterial embolization is the most commonly used route and is conditioned by the vascular anatomy of the afferent arteries, i.e., their small calibre, tortuosity, or the possibility of vasospasm during embolisation, making the procedure more complex and time-consuming[5,6]. Embolisation by direct puncture of the tumour is a simpler and less risky technique than transarterial embolisation, thus simplifying the surgical procedure and facilitating complete tumour resection by marking the limits of the tumour, with minimal complication rates[5,6,9]. Radiotherapeutic treatment is reserved for bilateral cases or in cases of excessive invasion[10].

A 59-year-old female patient, active smoker as the only history of interest, was assessed by ENT for a left cervical mass of 2 months' duration, associated with vertigo and tinnitus. On examination, she presented a left cervical mass, with vertical displacement and murmur. CT angiography showed a heterogeneous mass measuring 39 × 31 × 24 mm in its anteroposterior and transverse longitudinal axes, respectively, which enhanced intensely with contrast medium, located at the bifurcation of the left common carotid artery, between the ICA and ECA, with displacement of these arteries without obstructive signs (Fig. 1a). It was a Shamblin type II tumour.

Elective surgery under general anaesthesia was decided, after prior embolisation of the tumour by the interventional radiology department. Embolisation of the paraganglioma was performed by direct percutaneous puncture with Onyx®, under radiological control, via the right femoral artery (Fig. 1b), with good subsequent control, where the absence of staining of the lesion was verified, and a repletion defect was visualised in the ECA due to a small amount of Onyx® inside it. After 3 days, surgical resection was performed transcervically, with a longitudinal incision typical of carotid endarterectomy, dissection by planes, with primary control using vessel loops of internal jugular vein, continuing with arterial dissection and subsequent control of common carotid artery, ECA, ICA, and superior thyroid artery. A tumour of approximately 3 cm