Biliary cystadenocarcinoma complicated by intralesional haemorrhage

Introduction

Biliary cystadenocarcinomas are rare, malignant, cystic neoplasms of the liver arising from the biliary epithelium. They commonly tend to be incidentally discovered on imaging because they often present with vague symptoms and signs. The imaging features overlap with other pathologies and therefore the differential diagnosis can be extensive. This case presents an exceedingly rare complication of a biliary cystadenocarcinoma of acute intralesional haemorrhage. The patient initially underwent successful radiological embolization of the feeding vessel to control the haemorrhage and finally underwent formal resection of the lesion. Histopathological examination confirmed the diagnosis. The patient had an uncomplicated postoperative period. Unfortunately, a year later he died from metastatic oesophageal carcinoma.

Discussion

Biliary cystadenocarcinoma is a rare neoplasm arising from the biliary epithelium, accounting for only small percentage of the cystic hepatic neoplasms. They most frequently occur in middle-aged women. Presenting features may be the result of local mass effect but are more often discovered incidentally during radiographical evaluation for other clinical indications (Manouras et al, 2006).

The differential diagnosis of a biliary cystadenoma or biliary cystadenocarcinoma ranges from a simple cyst, hydatid cyst, post-traumatic or infective cysts, spectrum of ductal plate malformations, and neoplastic pathologies such as cystic metastasis and undifferentiated embryonal sarcoma. This highlights the need for referral to a specialist unit and discussion in a multidisciplinary setting.

The presence of an irregular thickened wall, mural nodules or papillary projections indicate possible malignant cystadenocarcinomas rather than benign cystadenomas (Manouras et al, 2006). Coarse calcification along the wall and/or septation also suggests a malignancy (Korobkin et al, 1989). The above features are best illustrated by a combination of ultrasound, magnetic resonance imaging and multidetector computed tomography. Angiography is not essential in the preoperative investigation of cystic liver lesions but signs raising the suspicion of malignancy are attenuated intracystic vessels, stretching of thin arteries and hypervascular mural nodules (Frick and Feinberg, 1982; Sato et al, 2003). The authors postulate that in the current case one of these thin, attenuated, irregular vessels ruptured, resulting in the intralesional haemorrhage. Tani et al (2008) assumed that in their case the anaemia was secondary to intracystic bleeding. The unique feature of this case is the fact that the haemorrhage has been depicted and con-

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Figure 1. Multidetector computed tomography of the upper abdomen. Arrow indicates active contrast extravasation, i.e. haemorrhage. The cyst is of high attenuation in keeping with haemorrhage.

Figure 2. Multidetector computed tomography of the upper abdomen post coiling. Arrow indicates coil.

Case Report

A 48-year-old man underwent a cardio-oesophagectomy for a T2N1 distal oesophageal tumour 4 years ago. Histology showed that the tumour was neuroendocrine in origin. A preoperative staging multidetector computed tomography of the chest, abdomen and pelvis demonstrated no focal liver lesion.

However, a follow-up multidetector computed tomography scan 2 years after his cardio-oesophagectomy revealed a new hepatic cystic lesion for which he was referred to the authors' unit for further assessment and management. No extrahepatic disease was identified. At the time of referral he was asymptomatic. His liver function tests and tumour markers were essentially unremarkable. A preoperative diagnosis of a biliary cystadenoma or biliary cystadenocarcinoma was made. The differential diagnosis included a cystic metastasis. Hydatid serology was negative. The management plan was for surgical excision.

While awaiting a clinic appointment he presented acutely with right-sided chest and upper abdominal pain. He was haemodynamically stable and was initially thought to have a pulmonary embolus. Multidetector computed tomography scan revealed no features of pulmonary embolus but active contrast extravasation into the cystic focal liver lesion indicating haemorrhage (*Figure 1*). He was transferred to the authors' unit and went on to have an angiography and coiling of the feeding artery to the cyst (*Figure 2*). Following this he underwent a formal right hemihepatectomy. Histologically, the lesion was confirmed to be a biliary cystadenocarcinoma (*Figure 3*). The patient was discharged home 12 days after the operation. Unfortunately, 1 year later he died from metastatic oesophageal carcinoma.

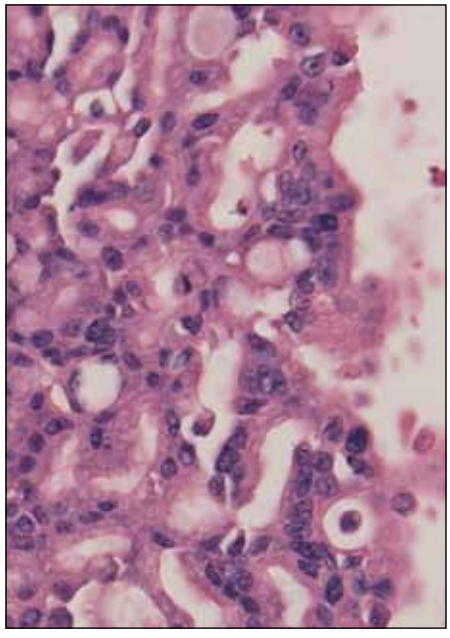

Figure 3. High power view of cyst lining showing epithelial stratification with nuclear dyspolarity, pleomorphism and atypia, features which diagnose cystadenocarcinoma.

firmed by radiological investigation. Despite the above imaging features distinction between biliary cystadenomas and biliary cystadenocarcinomas is notoriously difficult on preoperative investigation and ultimately the final diagnosis is made on a histological basis.

Surgical resection is the treatment of choice especially since differentiation of cystadenomas and cystadenocarcinomas can not be reliably made preoperatively despite the use of multimodality imaging. However, overall these cases have a good prognosis. BJHM

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LEARNING POINTS

- A high index of suspicion requiring a multidisciplinary approach is needed to make the diagnosis of a biliary cystadenoma or biliary cystadenocarcinoma *vs* other cystic liver lesions.
- Distinction between a biliary cystadenoma and biliary cystadenocarcinoma is not crucial since resection is the treatment of choice.
- Intralesional haemorrhage may indicate biliary cystadenocarcinoma.
- Appropriate use of interventional radiology may stabilize a patient before definitive treatment.

Testicular capillary haemangioma

An 80-year-old presented with a painless, solid abnormality in his left testicle. Scrotal ultrasound reported a lesion suggestive of a malignant tumour (*Figure 1*). Tumour markers were normal. He underwent a left radical orchidectomy and histology confirmed a testicular haemangioma.

Testicular haemangiomas are rare and occur in children and young adults. This benign condition presents as a palpable testicular abnormality and cannot be differentiated clinically or radiologically from malignancy (Mazal et al, 2000).

Histopathological examination is diagnostic (*Figure 2*). Immunohistochemistry for vascular markers confirm the diagnosis.

This case highlights the preoperative diagnostic limitations with benign testicular masses and raises a question about considering intraoperative examination and frozen section study with an organ-sparing approach especially in patients with solitary testis, atypical features and extremes of age, given the practical feasibility and accuracy of frozen section study to identify malignant and benign lesions (Elert et al, 2002). BJHM

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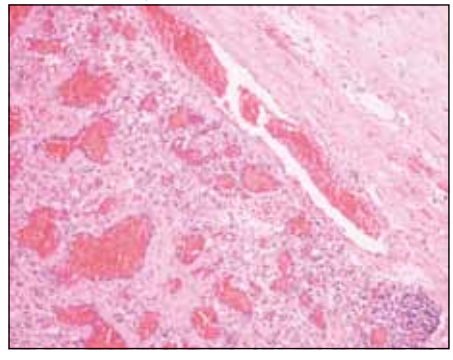
Figure 1. Scrotal ultrasound showing the focal lesion (arrow) in the upper pole of the left testis.

Figure 2. Photomicrograph showing a testicular haemangioma with non-anastomosing capillary sized vessels and lacking atypia (haematoxylin and eosin x100).



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Mazal PR, Kratzik C, Kain R et al (2000) Capillary Hemangioma of the Testis. *J Clin Path* **53**: 641–2