Adenosquamous carcinoma presenting as liver abscess

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ABSTRACT Adenosquamous carcinoma (ASC) has a nonspecific presentation. We report a patient with ASC who presented with fever and epigastric pain. Computed tomographic findings and clinical features were suggestive of a liver abscess, while ultrasonography showed a complex space-occupying lesion. Biopsy revealed a lesion with malignant glandular and squamous components, a finding that was compatible with ASC. Complex space-occupying lesions should raise the suspicion of a mass lesion. Subtle imaging clues of a metastatic disease were reviewed retrospectively.

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INTRODUCTION

Adenosquamous carcinoma (ASC) is defined as a tumour containing both the malignant glandular and squamous components. It is associated with an aggressive course and poorer prognosis compared to common cholangiocarcinoma of pure adenocarcinoma (AC) histology.⁽¹⁾ We describe an uncommon presentation of ASC as an abscess in this report and discuss the imaging findings of our patient. Similar cases have rarely been reported in the literature.

CASE REPORT

A previously healthy 60-year-old Chinese woman was admitted for fever. She complained of epigastric pain lasting three months and constitutional symptoms such as weight loss, fatigue and poor appetite. Physical examination revealed epigastric tenderness; however, the patient had no stigmata of chronic liver disease. Blood tests showed anaemia, and elevated white cell count, alkaline phosphatase and bilirubin levels. The patient tested negative for hepatitis serology.

Urgent computed tomography (CT) of the abdomen (Figs. 1 & 2) showed a large hypodense lesion with enhancing rim and multiple internal septa occupying the left lobe of the liver. In keeping with the clinical findings of fever and elevated white cell count, the above findings were suggestive of an abscess. Multiple intraductal stones and dilated bile ducts were observed in the left lobe, which may have been the source of infection. Stones were also observed within the common bile duct. Multiple hypodense hypo-enhancing lesions were also present on both lobes of the liver, which could represent multifocal abscesses or metastatic lesions. A small amount of peritoneal fluid was present and enlarged pericardial lymph nodes were seen.

Endoscopic retrograde cholangiopancreatography (ERCP) showed a dilated common duct with multiple filling defects,

in keeping with stones, which were removed. Multiple filling defects were also observed at the left intrahepatic duct, but selective cannulation failed (Fig. 3). Bile culture was positive for enterococci. The patient was treated with antibiotics.

In view of the fever and CT findings that were suggestive of abscesses, ultrasonography-guided drainage was booked. Ultrasonography of the abdomen (Fig. 4) performed before drainage showed a large heterogeneous space-occupying lesion with multiple intraductal stones in the left lobe of the liver. Multiple nodules with hypo-echoic rim were also observed in the right lobe of the liver. The common bile duct was dilated, with multiple intraductal stones. Ultrasonography-guided biopsy instead of drainage of the liver lesions was performed. Histology showed tumour cells with both squamous and glandular differentiation; the diagnosis was ASC. Ca 19-9 level was elevated to 130 U/mL. The patient refused active oncology or surgical treatment and subsequently succumbed to the disease.

DISCUSSION

Ultrasonographic images from our patient showed a solid liver lesion with a small amount of intralesional vascularity and no definite cystic component. These features were suggestive of an inflammatory mass, such as an early abscess or an infected tumour. Multiple lesions with hypo-echoic halo were also observed in the rest of the liver (Fig. 4c). Liver lesions with a sonolucent halo may be associated with malignancy and an aggressive growth pattern.^(2,3) This sonographic appearance is termed a 'target sign' or 'bull's eye'; it is non-specific and may be seen in cases of metastatic disease, hepatocellular carcinoma and fungal infection.⁽⁴⁾ In this patient, the multiplicity of target lesions and absence of background liver disease or immunocompromised status were suggestive of a metastatic disease. Upon review of the CT images, subtle nodularities were seen

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Fig. 1 Pre-contrast images show (a) intraductal stones (arrowhead) and dilated ducts (arrow) in the left lateral segment of the liver; and (b & c) the large lobulated lesion with multiple enhancing septa and mild rim-enhancement in the left lobe of the liver. An enlarged pericardial lymph node is also present (1b, arrow).



Fig. 2 Pre-contrast images show (a) the presence of another intra-ductal stone within the dilated bile duct in the left lobe of the liver; and (b) lesions with peripheral enhancement in the left lobe (arrowhead). Another small hypo-enhancing lesion is seen in segment 7 (arrow).



Fig. 3 ERCP shows filling defects in the lower common bile duct and left hepatic duct (arrow) due to stones.

over the capsular surface of the left lobe of the liver and the left hemidiaphragm (Fig. 5). Another tiny nodule was seen abutting the spleen. These features are also suggestive of malignancy with peritoneal deposits.

Primary ASC is a rare tumour of the liver and a rare subtype of cholangiocarcinoma. It has slightly more aggressive clinicopathological features compared to common cholangio-carcinoma of pure AC.⁽¹⁾ Nakajima and Kondo compared the clinicopathological features of ASC and AC and reported that patients from the ASC group had slightly larger tumours and shorter survival compared to the AC group (4.0 ± 1.2 months) vs. 6.9 ± 1.2 months). They also reported that metastases to the lymph node, remote organs and within the liver were also more commonly observed in the ASC group, with 100% intrahepatic metastases in their ASC group of 11 cases.⁽¹⁾

Cholangiocarcinoma may present with abdominal pain, jaundice, constitutional symptoms and fever. It was reported to be the cause of liver abscess in 3.3% of cases in the series by Jan et al.⁽⁵⁾ We found two cases of ASC where the patients presented with fever and were initially diagnosed with liver abscess; one case was reported by Kwon et al,⁽⁶⁾ and the other by Suzuki et al.⁽⁷⁾ The CT image of Kwon et al's patient showed a liver lesion with peripheral enhancement suggestive of an abscess, and ultrasonography showed a complex echoic liver mass. Ultrasonography-guided biopsy was performed in view of a complex liver echoic liver lesion, and pathology revealed



Fig. 4 Ultrasonography images of the abdomen show (a) a space occupying lesion about 7 cm in diameter in the left lobe of the liver. Small amount of intralesional vascularity is observed. There is no cystic component within the lesion; b) calcification of about 2 cm within the left lobe of the liver, representing intraductal stones; and c) several small lesions with hypo-echoic rim and target appearance in the right lobe of the liver.



Fig. 5 CT images show (a) a large liver lesion with rim-enhancement and multiple internal septa. Multiple enhancing nodules are seen adjacent to the capsular surface of the left lobe and the left hemidiaphragm (arrow); (b) well-demonstrated lesions by coronal reformat images (arrow); and (c) another small peritoneal nodule adjacent to the spleen.

AC. The patient was treated with extended left lobectomy, and AC and squamous cell carcinoma were present in the surgical specimen, which confirmed the diagnosis of ASC. The patient received adjuvant chemotherapy with 5-fluorouracil (5-FU) and cisplatin. Follow-up abdominal CT did not show any signs of cancer recurrence. Suzuki et al's patient presented with fever, and ultrasonography revealed a space-occupying lesion in his liver. He was initially treated with antibiotics. Tumour with abscess formation was suspected in view of persistent fever, and a needle biopsy showed AC. Subsequent CT showed a liver lesion with central and peripheral necrosis. The patient was treated with left hepatic lobectomy. Both the AC and squamous cell carcinoma components were observed in the surgical specimen. CT at three months post operation showed recurrence within the liver. Intrahepatic arterial infusion of cisplatin and 5-FU was performed. The recurrent tumours initially showed reduction in size, but the patient eventually succumbed to the disease 15 months after the operation.

Surgery should be the treatment of choice for ASC; however, many cases are invasive and thus, complete resection is not always possible.⁽⁸⁾ Nakai et al reported a patient of ASC who had tumour invasion of the inferior vena cava and the diaphragm, and the tumour was too advanced for complete resection. Partial hepatectomy and partial resection of the diaphragm were performed. The patient was then given repeated transcatheter arterial infusion chemotherapy with cisplatin and 5-FU mixed with lipiodol, followed by degradable starch microspheres. The microspheres caused transient reduction in blood flow and prolonged retention of the chemotherapy agents within the tumour.⁽⁸⁾ The patient remained alive and well 12 months post laparotomy.

In conclusion, ASC has a nonspecific clinical presentation. While our patient had clinical and imaging features suggestive of an abscess, there were also clues of a metastatic disease. Similar cases of ASC presenting as liver abscess have previously been reported. Biopsy should be performed when the clinical and imaging features are not in keeping with simple liver abscesses. Complete surgical resection may not be possible due to the invasive nature of ASC; in such cases, systemic chemotherapy and transarterial chemoembolisation may be considered as other possible treatment options.

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