

Case Report

Intrahepatic cholangiocarcinoma with aberrant hepatic artery presenting as hepatic abscess: a case report

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ABSTRACT

Intrahepatic cholangiocarcinoma (ICC) is a primary hepatic malignancy that originates from epithelial cells of bile duct. Lack of diagnostic measures and therapies leads to an increasing number of deaths from ICC worldwide. Here we described a case of 61-year-old Chinese female, who initially presented with right upper quadrant pain, combined with the results that a low density mass accompanied by multiple nodules occupied the right liver lobe by CT-scan, which also showed an aberrant right hepatic artery that participated in the right liver lobe and originating from the superior mesenteric artery, this patient was clinically considered as hepatic abscess (HA). The patient's right upper quadrant pain was alleviated after been treated with the infusion chemotherapy of the aberrant right hepatic artery (ARHA) via percutaneous femoral arterial catheterization by Seldinger technique (Meropenem 7 days) following the failure of the liver-puncture drainage. However, the right upper quadrant pain occurred again 6 days later, serum CA19-9>1000.0 U/ml, which indicated the possibility of hepatic malignancy, so we performed laparotomy. The histopathological result of intraoperative frozen section demonstrated cholangiocarcinoma, unfortunately, it was unresectable. Finally, right lower lung pneumonia and pleural empyema happened to her and she succumbed to respiratory failure 22 days following surgery. In this report, we will discuss the case with reference to the literature.

Keywords: Aberrant right hepatic artery, Hepatic abscess, Intrahepatic cholangiocarcinoma

INTRODUCTION

Intrahepatic cholangiocarcinoma (ICC) is the second most common primary liver malignancy that originates from epithelial cells of bile duct, the lack of effective diagnostic measures and therapies causes a poor prognosis for it.¹⁻³ Some cases, though extremely rare, show that ICC shares similar initial symptoms to hepatic abscess (HA), such as right upper quadrant pain and fever, which is still a potential pitfall.⁴⁻⁶ Moreover, the aberrant hepatic arteries made this case worse, so preoperative comprehensive understanding and analysis of these variations was needed, which might avoid disastrous complications caused by unexpected vascular injury. In this report, a case of ICC was described with

the aberrant right hepatic artery (ARHA) originating from superior mesenteric artery (SMA), presenting as HA. This report aims to discuss the potential pitfalls in the diagnosis and treatment of ICC.

CASE REPORT

A 61-year-old lady was admitted for right upper quadrant pain without jaundice. On examination, fist percussion over the liver and right upper quadrant pain upon palpation were noted. Serum HBV and HCV were both negative. Abdominal CT-scan with arterial and portal phase (Siemens Medical Solutions, Forchheim, Germany) showed low density mass with multiple nodules occupied and detected an aberrant right hepatic artery which ran to

the right liver lobe and originating from the superior mesenteric artery (Figure 1). To our knowledge, hepatic abscess was considered. Percutaneous drainage was performed, but we fail to find tumor cells and bacteria in the puncture fluid. Then we successfully performed the infusion chemotherapy of the ARHA via percutaneous femoral arterial catheterization by Seldinger technique (Meropenem 7 days) (Figure 2). After 7 days of treatment, the patient's initial symptom was alleviated.

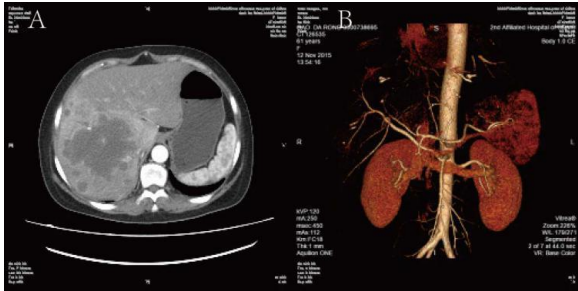


Figure 1: (a) CT of the abdomen axial image shows the low density in the right hepatic lobe (b) 3D reconstruction and this image shows aberrant right hepatic artery originating from the superior mesenteric artery.

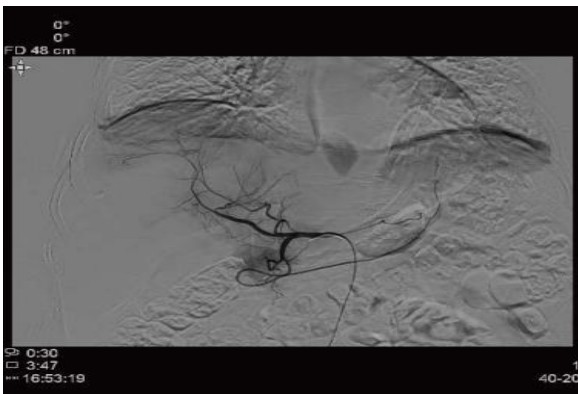


Figure 2: Selective hepatic arteriography and this image shows aberrant right hepatic artery and proper hepatic artery.

However, the right upper quadrant pain occurred again 6 days later, serum CA19-9>1000.0 U/ml, which indicated the possibility of hepatic malignancy, so we performed laparotomy. Multiple, firm, nodules on the surface of the right hepatic lobe were found during operation unfortunately, what's more, the histopathological result of intraoperative frozen section demonstrated malignant cholangiocarcinoma, which was unresectable, therefore Transcatheter Arterial Chemoembolization (TACE) was performed after operation. However, due to her dyspnea, caused by the right lower lung pneumonia and pleural empyema which was detected by CT, she was sent to ICU. Finally, both medical and surgical treatments were fail to eliminate the infection, the patient succumbed to respiratory failure 22 days following surgery.

DISCUSSION

The normal hepatic arterial anatomy is classically described as common hepatic artery arising from celiac trunk, dividing into gastroduodenal artery and proper hepatic artery (PHA), which then divides into right (or/and medial) and left hepatic arteries. Otherwise they are considered as variant. A relatively large number of variations in hepatic artery have been well described by Michels, after studying 200 cadavers, he proposed classification of the hepatic artery variations into 10 types, and based on where gastroduodenal artery and PHA arising.⁷ The hepatic artery variations were simplified into internationally recognized 6 types by Hiatt after reviewed records of 1000 patients who underwent liver harvesting for orthotopic transplantation.⁸ And many operative complications may be seen because of anatomical variations in hepatic arteries, such as ARHA, originating from the right edge of SMA.⁹

However, the incidence of ARHA may be much higher than that in the previous documents because the aim of the previous documents was not to diagnose ARHA. Recent documents suggested that hepatic arterial variants could be discovered easily by multi-detector CT even during regular examinations.¹⁰ With the development of surgery, accurate depiction and definition of the hepatic arterial variants are needed, because the incidence of ARHA, as mentioned above, may be much higher than we thought. We have to fully understand the variation of hepatic artery, so that we can insert into the target vessel precisely.

In general, aberrant tumor-feeding arteries have close relationship with the site of the tumor. For example, the ARHA originating from SMA usually supply the right hepatic tumor, while the ARHA originating from left gastric artery usually supply the left hepatic tumor. In this case, ARHA and the right hepatic tumor were detected by multi-detector CT, and we were vigilant about such aberrant hepatic arteries to avoid potentially preoperative and postoperative complications. However, the relationship between such hepatic arterial variants and intrahepatic cholangiocarcinoma mimicking HA, and whether these arterial variants play crucial roles in haemodynamics of intrahepatic cholangiocarcinoma are still unclear, further studies are needed.

Studies suggested that patients with HA should be treated by minimally invasive chemotherapy first, and surgery should be reserved for those who have failed to respond to medication.^{11,12} In this case, although the patient's right abdominal pain alleviated by infusion chemotherapy of the ARHA via percutaneous femoral arterial catheterization by Seldinger technique (Meropenem 7 days), sufficient evidence is still needed to exclude tumor. In this situation, biopsy of liver by fine needle aspiration may be more helpful to confirm the disease than pleural fluid culture. Primary empirical anti-infectious treatment of HA may delay the treatment of

cancer. Surgical resection in time is the most effective way for resectable intrahepatic cholangiocarcinoma mimicking HA.¹³

Previous studies suggested that patients with HA have a consistently high incidence of pulmonary abscess and pleural empyema, purulent pericarditis and pericardial tamponade.^{14,15} For patients with liver lesions of an undetermined origin, the final diagnosis depends on pathological and immunohistochemical examination of biopsy specimens, in addition, laparoscopic surgery is a useful tool for differential diagnosis.¹⁸ In this case, pleural empyema occurred after transdiaphragmatic rupture of HA, which led to patient's respiratory failure and death 22 days following surgery.^{16,17}

Cases of patients with ICC present as HA have been reported in the past decades, but pitfalls in the diagnosis are still existing. ICC is highly malignant, Li et al reported four cases of ICC that presented as HA, all of them died with 1 year.¹⁹ Therefore, it's necessary to pay attention to the identification of ICC and HA, so that appropriate treatment measures can be taken for ICC patients in time.

CONCLUSION

A case of ICC was presented associated with right hepatic artery originating from the superior mesenteric artery. And the potential pitfalls in the diagnosis and treatment of intrahepatic cholangiocarcinoma with ARHA have been discussed above, suggesting that primary empirical anti-infectious treatment of HA is not recommended, and surgical treatment needs to be performed in time when intrahepatic cholangiocarcinoma mimicking HA is considered. In addition, as the number of cases is still limited, further clinical studies are needed to explore the relationship between ARHA and intrahepatic cholangiocarcinoma mimicking HA.

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