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Hepatic myomatous angiomyolipoma diagnosed preoperatively from specific imaging features: A case report

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ABSTRACT

INTRODUCTION: Hepatic angiomyolipoma is a rare tumour and is difficult to obtain the accurate diagnosis preoperatively because the imaging features are similar to hepatocellular carcinoma.

PRESENTATION OF CASE: We present a case study of an 80-year old woman with a liver tumour measuring 6.2 cm × 6.0 cm. We were able to diagnose the tumour preoperatively as a rare hepatic myomatous angiomyolipoma based on the presence of early venous return evident on angiography and small low-intensity areas corresponding to fat within the tumour revealed by out-of-phase EOB-MRI. The tumour was removed by minimally invasive surgery and our preoperative diagnosis was confirmed by positive immunoreactivity for both angiomyolipoma-specific human melanoma black 45 and smooth muscle cell positivity for melanin.

DISCUSSION: We consider that the information obtained in this case will be useful for preoperative diagnosis of other hepatic angiomyolipomas, thus facilitating more appropriate and less invasive surgery and improving the overall outcome.

CONCLUSION: Hepatic myomatous angiomyolipoma is a rare tumour. We illustrated the two specific imaging features to diagnose it preoperatively.

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1. Introduction

Angiomyolipoma (AML) is a unique and rare tumour containing varying percentages of blood vessels, smooth muscle, and adipose cells [1]. These tumours generally arise in the kidneys [2], but can also affect several other organs, including the liver, albeit less frequently [3,4]. Hepatic AML was first reported by Ishak et al. in 1976 [5], and the first case in Eastern Asia was reported in Japan by Kawarada et al. in 1983 [3]. Around 120 cases have now been reported worldwide, and the majority have been defined as benign, only a few showing marginal malignant potential [6,7]. Despite this, invasive liver resection for potentially malignant lesions can rarely be avoided for hepatic AML because its clinical characteris-

tics demonstrated by preoperative imaging are similar to those of malignant hepatocellular carcinoma (HCC). Generally, differential diagnosis can only be achieved postoperatively based on immunohistochemical staining of tissue sections for human melanoma black 45 (HMB-45) and melanin (Melan-A) [8,9]. More accurate and effective imaging techniques would be highly valuable for characterization of focal solid hepatic lesions, and in fact a few case studies have reported that more informative surgical decisions can be made on the basis of magnetic resonance imaging (MRI) and angiography in this area [10–12]. Here we present a rare case of hepatic myomatous AML, which we identified preoperatively based on certain features demonstrated by MRI and angiography. The work has been reported in line with the SCARE criteria [13].

2. Presentation of case

An 80-year-old woman was admitted to our institute with a feeling of abdominal distension. Abdominal ultrasound (US) revealed a large liver tumour located in the lateral segment. The patient had no history of liver disease or hepatitis and did not drink alcohol. A range of diagnostic tests and examinations were carried out to evaluate the possibility of malignancy. Hepatitis B

Abbreviations: AML, angiomyolipoma; HMB-45, human melanoma black 45; Melan-A, melanin polyclonal antibody; EOB-MRI, gadolinium-ethoxybenzyl-diethylenetriamine pentaacetic acid- enhanced magnetic resonance imaging; MRI, magnetic resonance imaging; US, ultrasound; AFP, alpha-fetoprotein; PIVKA II, vitamin K absence or antagonist-2; IVC, inferior vena cava.

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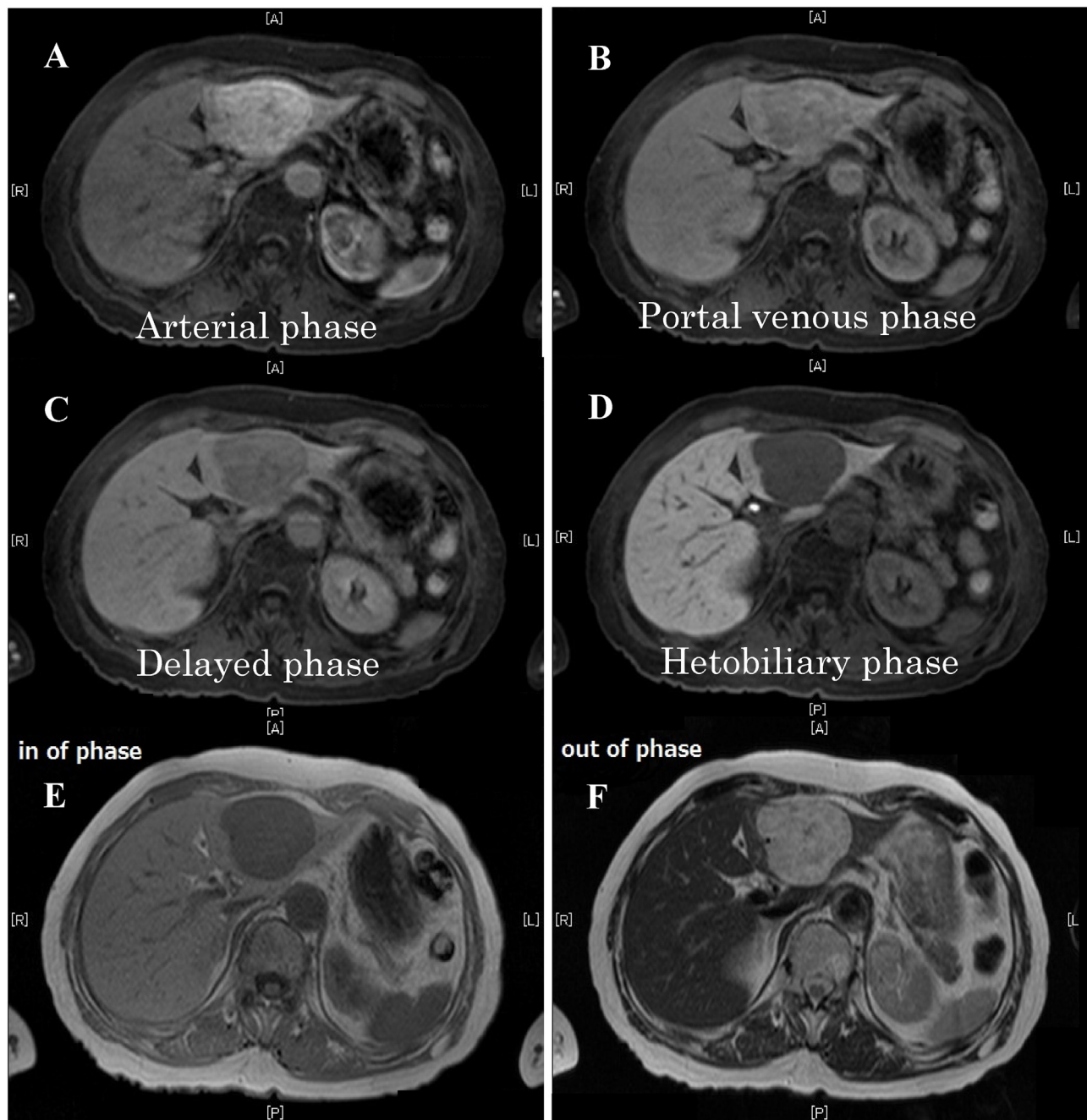


Fig. 1. EOB-MRI showed a hepatic mass with early-phase hyperattenuation and portal-phase hypoattenuation with gradually wash out the medium (Fig. 1A and B). This was followed by a decreasing the tumour signal in the delayed phase. The hepatobiliary phase showed a wash-out pattern and low signal, which was hypoattenuation, measuring 6.2 cm × 6.0 cm (Fig. 1C and D). The opposed phase of EOB-MRI revealed high intensity of whole the tumour with small low intensity inside the tumour.

surface antigen and anti-hepatitis C antibody were negative. Laboratory tests revealed a white blood cell count of 3000/mm³, haemoglobin 12.5 g/dL, platelet count 17.9 × 10⁴/mm³, albumin 3.2 g/dL, total bilirubin 0.5 mg/dL, and direct bilirubin 0.3 mg/dL. Serum levels of transaminase, alpha-fetoprotein (AFP) and protein induced by vitamin K absence or antagonist-2 (PIVKA II) were within the normal ranges (Table 1). Gadolinium-ethoxybenzyl-diethylenetriamine pentaacetic acid enhanced magnetic resonance imaging (EOB-MRI) showed a hepatic mass with early-phase hyperattenuation and portal-phase hypoattenuation with gradual wash-out in the lateral segment (Fig. 1A and B). This was followed by a decrease of the tumour signal in the delayed phase. The hepatocyte phase showed a typical wash-out pattern and low sig-

nal, indicating a hypo-enhanced lesion, measuring 6.2 cm × 6.0 cm (Fig. 1C and D). In addition, out-of-phase EOB-MRI revealed high intensity in the whole tumour (Fig. 1E), with several remarkable small areas of low intensity (Fig. 1F). This pattern indicated that, overall, the tumour contained a minimal amount of fluid but a small amount of fat, suggesting either hepatic AML or hepatocellular carcinoma including some fat components. Subsequent angiography revealed the tumour as a well circumscribed hypervascular mass (Fig. 2A) with central vessels, and notably a drainage vein from the tumour to the inferior vena cava (IVC) (Fig. 2B). This latter feature is known as early venous return and is specific to AML [14]. Therefore, on the basis of imaging alone, we diagnosed this tumour as AML containing a small amount of fat. However, given the large size of

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