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Alveolar Echinococcosis of the Liver: A Diagnostic Problem in a Nonendemic Area

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Alveolar echinococcosis is a parasitic disease primarily invading the liver. Owing to its aggressive nature, it invades the adjacent structures and can even metastasize to distant organs. The appearance of hepatic involvement on computed tomographic scan is characteristic, but not specific, with areas of calcification seen within a hypoenhancing mass. Although magnetic resonance imaging may better define the extent of the disease, it often misleads the radiologist, especially if the lesion is devoid of cystic component(s) and if it occurs in nonendemic areas. Knowledge of the imaging appearance may prompt serological evaluation and aid in making an early diagnosis and planning appropriate treatment of this uncommon fatal disease, especially in nonendemic areas.

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Introduction

Alveolar echinococcosis is caused by the larval stage of *Echinococcus multilocularis*. It primarily affects the liver and typically consists of multiple small cysts (giving alveolar appearance) with surrounding inflammation.[1] The lesion grows slowly over years and may show surrounding organ infiltration, making it difficult to differentiate from neoplastic lesions. Diagnosis on imaging is often difficult in nonendemic areas because of the rarity of the disease and unusual imaging appearances.[2] We describe the imaging features, including magnetic resonance imaging (MRI), of a patient with infiltrative liver lesion that was misdiagnosed as an inflammatory pseudotumor on imaging but was confirmed as hepatic alveolar echinococcosis after surgical resection.

Case Report

A 45-year-old man presented with long-standing history of vague right upper quadrant pain with recent onset of progressive jaundice for 2 months. There was no history of fever or weight loss. Clinical examination revealed a firm lump in the right hypochondrium, measuring approximately 3×4 cm^2. There were no signs of portal hypertension or extremity edema. The total leukocyte count was 5600/μL (normal: 4000-11,000/μL). Liver function tests showed a bilirubin level of 3.5 mg/dL (normal: 0.1-1.2 mg/dL), alkaline phosphatase level of 1015 IU/L (normal: 30-240 IU/L), alanine aminotransferase level of 90 IU/L (normal: 0-40 IU/L), aspartate aminotransferase level of 59 IU/L (normal: 0-40 IU/L), and albumin level of 3.4 g/dL (normal: 3.5-5.5 g/dL). The serum alpha-fetoprotein level was within the normal range. Findings on upper gastrointestinal endoscopy were normal, with no varices. Ultrasonography revealed a large heterogeneous mass in the right lobe of the liver with infiltration of the gall bladder with mild dilatation of intrahepatic biliary radicles (IHBR). The splenoportal axis and spleen were normal. Contrast-enhanced computed tomographic (CT) scan, performed with a provisional diagnosis of carcinoma of the gall bladder, showed a large hypodense mass in segments 4, 5, and 8 of the liver with involvement of the gall bladder and the common hepatic duct, causing dilatation of IHBR (Fig. 1A-C). Peripheral amorphous areas of calcification were seen. Review of CT scan performed 6 years earlier showed a smaller and less-invasive mass (Fig. 1D). As imaging appearances were inconclusive, MRI was performed. It showed a large hepatic mass appearing isointense to hyperintense on T1-weighted and isointense to hypointense on T2-weighted (T2W) images, involving the gall bladder with dilated IHBR (Fig. 2). Multiple tiny T2W hyperintense foci were seen within the mass, which were better depicted on magnetic resonance cholangiopancreatography images (Fig. 2C). The mass was hypointense on diffusion-weighted images without restriction of diffusion (Fig. 3). Only mild peripheral contrast enhancement was noted (Fig. 4). With these imaging appearances, a provisional diagnosis of inflammatory pseudotumor of the liver was made. Subsequently, fine-needle aspiration cytology was attempted under ultrasound guidance, which did not yield any material. This was followed by ultrasound-guided percutaneous core biopsies (taken twice) from the mass, which were also inconclusive. Subsequently, extended right hepatectomy with Roux-en-Y left cholangiojejunostomy was performed. At surgery, the mass was hard, and on cut section it

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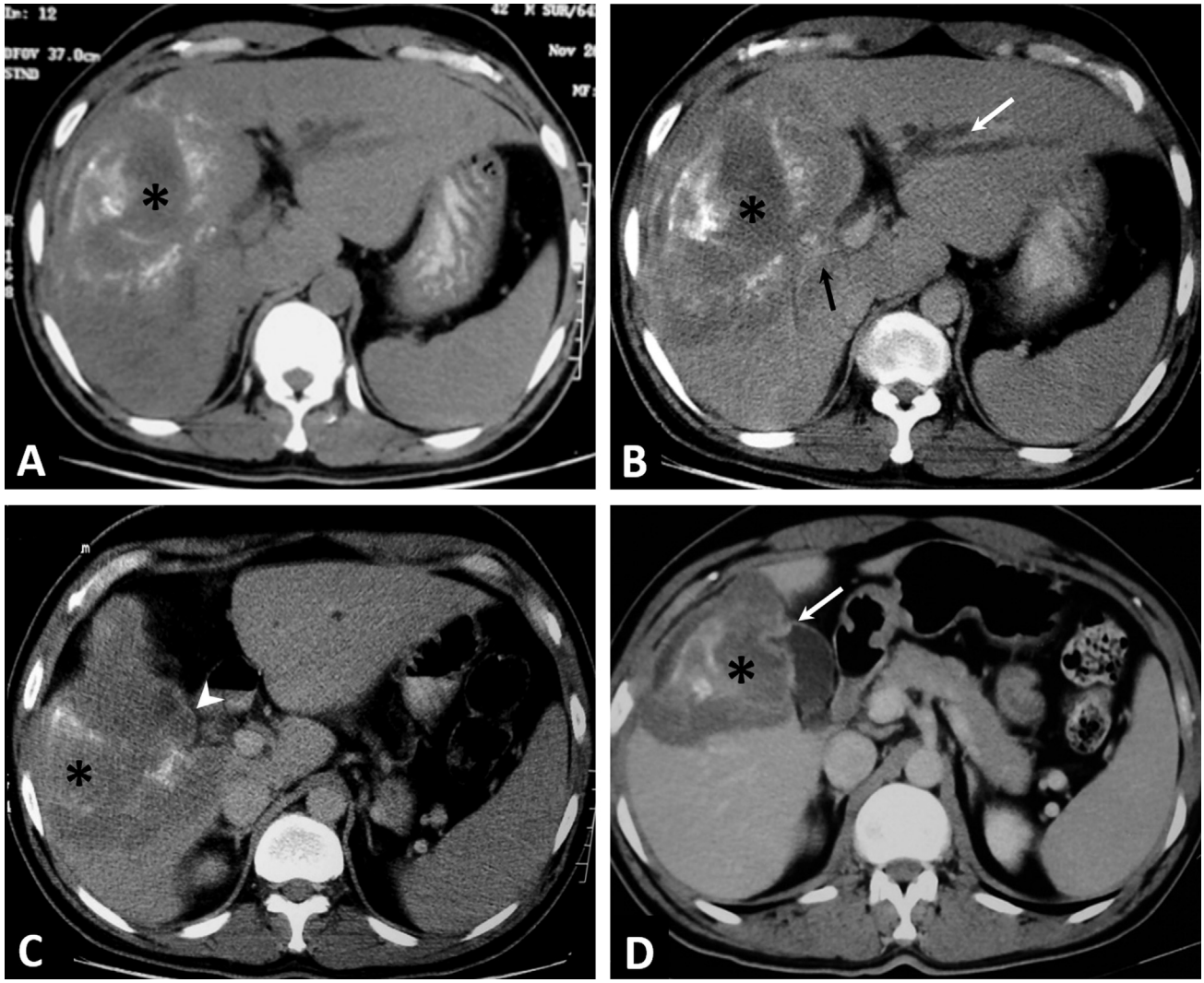

Fig. 1. Non–contrast-enhanced (A) and contrast-enhanced CT scans (B and C) of the patient at presentation show an ill-defined hypoenhancing lesion in segments 4B, 5, and 8 of the liver, with amorphous calcification (asterisk), with invasion of the gall bladder (arrow head in C) and the right portal vein (black arrow in B) and dilatation of the intrahepatic biliary radicles of the left lobe (white arrow in B). (D) Contrast-enhanced CT scan performed 6 years earlier shows a smaller sized lesion (asterisk) with calcification and gall bladder involvement (arrow), but without biliary dilatation.

was infiltrating the surrounding liver parenchyma and the gall bladder. Histopathology revealed multiple cysts with laminated membranes and extensive fibrosis, suggestive of *E. multilocularis*. The patient recovered well in the postoperative period and was discharged.

Discussion

Hepatic alveolar echinococcosis is a rare disease that is fatal if left untreated. The definitive hosts of *E. multilocularis* are wild foxes, domestic dogs, or cats and intermediate hosts are rodents. Humans are accidental hosts who become infected through contact with definitive hosts or through contaminated food and water.[3] The ingested ova develop usually in the liver and rarely in the lungs or the brain.[2] They grow in the form of multiple cysts measuring 1-30 mm, giving alveolar appearance.[4] A strong inflammatory reaction develops surrounding the lesion with formation of granulomas by macrophages and lymphocytes and dense fibrous connective tissue.

Infected patients remain asymptomatic for a long period (5-15 years) as the lesion is slow growing.[1] The patients often present with vague abdominal pain or discomfort, with lump or with symptoms due to mass effect on surrounding structures, such as jaundice (biliary compression), lower limb edema (venacaval compression), or with features of portal hypertension due to liver failure.[5] In long-standing cases, distant metastasis to the lungs, brain, or bones may be seen. Prognosis is poor in untreated cases.

Imaging methods used in diagnosis of alveolar echinococcosis are commonly ultrasonography and CT.[6] On sonography, the lesion appears as a large heterogeneous hypoechoic mass in the liver, which may show echogenic foci of calcification. Cystic areas may be seen uncommonly. Typical sonographic findings of alveolar echinococcosis include a heteroechoic infiltrative mass with calcific foci or an irregular thick-walled pseudocystic lesion with central necrosis. Uncommonly, the lesion may appear completely cystic, entirely echogenic, or calcified.[4,6] CT scan plays a major role in evaluation of disease morphology and extent. The lesion is typically seen as a large nonenhancing or hypoenhancing mass with peripheral irregular calcification.[7] The lesion typically shows irregular and ill-defined margins with infiltration of surrounding structures, such as the gall bladder, portal vein, hepatic artery, or bile ducts. Only mild contrast enhancement is seen because of extensive fibrosis and necrosis. The lesion may also show cystic or necrotic areas, central punctuate calcification, or scattered dense calcification. The lesion may appear cystic with irregular mildly

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