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Original Article

A tumor-mimicking parasitic disease: Radiological findings of alveolar Echinococcosis

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Abstract

Introduction: Alveolar echinococcosis (AE) is a tumor-mimicking parasitic disease caused by Echinococcus multilocularis. In this study, we aimed to present the radiological imaging findings of AE characterized by malignant features.

Methods: The radiological findings of 20 patients who were admitted to our hospital in Van province between years 2014 and 2020 and were diagnosed with AE were retrospectively evaluated. Of these patients, 30% had been operated due to a preliminary diagnosis of a tumor. The study was conducted retrospectively by examining the drawback radiological images of patients from the radiological data archiving system. The diagnosis of AE was confirmed when lesions manifested the following radiological imaging findings: irregular margins, no contrast uptake in dynamic contrast images or late peripheral enhancement, presence of calcifications, and contralateral lobe hypertrophy.

Results: The mean age of the patients at the time of diagnosis was 50.95 (12-86) years. Of the patients, 15 (75%) were women and 5 (25%) were men. Typical histopathological and radiological imaging findings were present in 8 (40%) and 17 (85%) patients, respectively. Only 12 (60%) out of 20 patients had seropositivity by ELISA. The remaining 8 (40%) patients were seronegative.

Conclusion: Radiological imaging examinations are essential in diagnosis and evaluation of the extent of the disease. Different radiological techniques can be used to differentiate AE from other diseases. Early diagnosis is important for avoiding unnecessary or inadequate operations due to a misdiagnosis and preventing potential complications.

Introduction

Alveolar echinococcosis (AE) is a parasitic disease that can lead to serious complications in endemic areas. The definitive host is fox; the intermediate hosts are rodents, and humans are accidental hosts. Lesions can mimic tumors at different stages, invading the respective and neighboring organs and even manifesting distant metastases. The geographical area of Van province and its surroundings are endemic for Echinococcus granulosus (EG) and Echinococcus multilocularis (EM). Alveolar echinococcosis is an infection caused by EM.

Although AE is seen most commonly in the liver. It can involve many organs and body systems as the parasite can invade adjacent organs and migrate through the body by the systemic circulation and the lymphatic circulation. The disease can cause acute and chronic complications. Furthermore, the infection can behave invasively, mimicking a malignant tumor. Therefore, early diagnosis and treatment diminish morbidity and mortality associated with AE.

Clinicians and radiologists should keep AE in mind in the differential diagnosis of cystic lesions in endemic areas and should be familiar with the clinical and radiological findings of AE. In this study, radiological imaging findings of AE have been presented. Of the cases presented in this study, nearly one-third were preliminarily considered to have a malignant tumor.

Materials and Methods

Radiological images of the patients who had referred to our center from Van and surrounding provinces due to abdominal pain or other complaints in the period between 2014 and 2020 and who had received a diagnosis of AE based on the results of diagnostic tests, were examined. Twenty-six patients who had a diagnosis of AE based on serological, histopathological, or radiological tests and examinations were found eligible to be included in the study. All 26 patients had abdominal computed tomography (CT) images. Of these 26 patients, 6 patients who lacked a diagnosis or who did not receive a definite diagnosis in their subsequent evaluations and follow-up imaging were excluded from the study. Histopathology

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was accepted as the gold standard in the diagnosis of AE. Finally, a total of 20 patients were included in the study. The study was conducted retrospectively by examining the drawback radiological test images of patients from PACS (Picture Archiving and Communication Systems) radiological data archiving system. The abdominal CT images of the patients were evaluated. Using a 16-slice multislice device (Siemens, medical solution); CT images had been obtained with a slice thickness of 3mm, a dose of 120 kV, and by administering an average of 90 ml of contrast material intravenously. Axial images drawbacks from the PACS (karpacswiever v.1.0) system were examined for the study. The diagnosis of AE was confirmed when lesions manifested the following radiological imaging findings: irregular margins, no contrast uptake in dynamic contrast images or late peripheral enhancement, presence of calcifications, and contralateral lobe hypertrophy. Patients who had received the diagnosis of AE based on histopathological examinations of tissue biopsy samples or who were diagnosed postoperatively were included in the study. All patients, three patients, and one patient had images or reports of CT imaging, magnetic resonance imaging (MRI), and ultrasonography (USG), respectively. Hypointense lesions in T2-weighted images in MRI, lesions with no contrast uptake in dynamic images, and any characteristic findings associated with lesion margins were considered favouring the diagnosis of AE. Eight patients had undergone surgery and received a diagnosis of AE based on the pathological examinations of the biopsy specimens. Patients who were diagnosed with EG infections, and were not diagnosed with AE, or had radiological image findings incompatible with AE were not included in the study.

Results

The mean age of the patients at the time of diagnosis was 50.95 (12-86) years. Fifteen (75%) patients were women and five (25%) were men. At the time of diagnosis, 8 (40%) patients had typical findings in histopathological examinations and 17 (85%) patients had typical radiological imaging findings. Only 12 (60%) of 20 patients were seropositive by ELISA. Eight (40%) patients were diagnosed with AE despite seronegativity.

Discussion

Hepatic AE is a rare parasitic disease caused by E. multilocularis, which is endemic in some regions of central and northern Europe, northern Asia, Japan, China, Turkey, and North America. The disease mimics a slowly growing and progressing life-threatening tumor in the liver of an accidental intermediate host, delaying diagnosis or causing misdiagnoses.[1] In our study, 6 (30%) patients had received a preliminary diagnosis of a tumor but they were diagnosed with AE later after histopathological examinations.

The infection can spread secondarily by migrating in the body via hematogenous or lymphatic dissemination, involving any other body system or organ including adrenal glands, lungs, and the brain. Consistent with this information, cranial metastasis was present in one of our patients. Besides, AE can directly invade adjacent organs.[2] AE is more sporadic than the hydatid form caused by EG but still deserves to receive considerable attention as it exhibits distinctive manifestations such as infiltrative and massive growth.[1] The hydatid disease occurs as a benign mass and usually causes compression to neighbor structures; but AE manifests itself in the form of a slowly growing malignant mass. A cystic component is more common in the hydatid disease, also called cystic echinococcosis. The typical macroscopic appearance is an infiltrating mass of multiple small cysts.[3] In all of our cases, the lesions were hypodense with irregular margins and showed no contrast uptake. They were infiltrative invading neighbouring vascular structures. The absence of contrast uptake is an important finding to make a differential diagnosis of lesions from tumors.

Imaging procedures are essential in the diagnosis and evaluation of the extent of AE. For diagnosis and identification of complications, the following steps can be suggested to be used in an algorithm, respectively: abdominal USG, CT, and MRI. In addition, endoscopic retrograde cholangiopancreatography (ERCP) is an interventional method that can be used in both the diagnosis and treatment of the complications of the biliary tract.[4]

USG is the first-line diagnostic method because it is inexpensive, non-invasive, easily accessible, and is useful in the diagnosis of cysts and disease stages. Typical findings include the presence of an amorphous, heterogeneous, and hyperechoic mass with central pseudo-fluid necrosis. The lesions can sometimes be hypoechoic with an irregular pattern. Reactive fibrotic tissue causes the lesion to have a hyperechoic pattern. Many lesions contain irregular calcifications that can be nodular or in the form of a 'plaque'. Signs of vascular or biliary invasion may occur.[5] In the retrospective analysis of our cases, we observed that the lesions were reported as hypoechoic with irregular margins. The lesions were described as solid masses with an indefinite blood supply. Dynamic imaging with upper abdominal CT or MRI was recommended as advanced imaging techniques.

Despite the risk of radiation exposure, CT provides detailed information about the location, size, number, and internal structure of the cyst. In CT images; the presence of calcifications can be detected as an important sign to make a diagnosis of AE.[6]

CT is performed because it is always associated with high sensitivity (94%). However, radiation exposure and exposure to intravenous contrast material are the disadvantages of CT. Determining the presence of any vascular, biliary, or extrahepatic dilation and thus, determining the resectability of the lesion is important.[7-8]

Classically, an infiltrative tumor-like hepatic mass with irregular borders and a calcified or cystic component may be detected on CT images (Figure 1).

None of our cases had accompanying vascular invasion with calcifications or central contrast uptake. The latter is an important parameter in differentiating the lesion from tumors in dynamic series.

Liver parenchyma adjacent to the mass is typically atrophied due to capsular retraction caused by the invasion of the biliary tract or blood vessels. Thus, such lesions can be misdiagnosed as cholangiocellular tumors, originating from the bile ducts of the liver. AE can show a progressive and peripheral contrast enhancement pattern in dynamic contrast-enhanced CT images. Because the clinical picture is not specific, diagnosis is mainly based on findings obtained from imaging and serological tests, and histopathological examinations. Serological testing for E. multilocularis is based on detection of a specific antibody response with a potential for cross-reaction with E. Granulosis.[4] However, as shown in our study, the absence of an antibody response does not exclude the diagnosis of AE. In our study, 8 out of 20 patients (reaching a 40% rate, which is a high percentage) were diagnosed with AE based on radiological and histopathological findings, although they were seronegative.

In cases, where a definite diagnosis cannot be made, MRI can be used to confirm the diagnosis and view the lesion in different planes. The best imaging procedure to show a cystic component is an MRI examination.

MRI is superior in revealing especially the small cystic components of AE lesions. MRI examinations can be used as problem-solving modalities when it is difficult to make a diagnosis. Characteristic MR imaging findings of AE include the presence of a heterogeneous infiltrative mass with irregular margins, low or moderate signal intensity on T1-weighted images, and heterogeneous signal intensity on T2-weighted images due to necrosis in the center.[9] (Figure 2). Similarly, the parenchyma were hypointense and heterogeneous in our three cases in T2 images.

Besides extrahepatic dilatation, vascular or biliary tree involvement can be observed, as well. The lesion is usually defined as having ill-defined and irregular margins and partially solid and partially cystic components. Growth of the lesion and the increase in the invasion rate in the subsequent images suggest the presence of a tumor-like lesion. Kodama et al.[10] categorized MRI images of AE into five different types as follows: Multiple small round cysts without a solid component were categorized as type 1 (4%), multiple small round cysts with a solid component were categorized as type 2 (40%), a solid component associated with irregular large cysts were categorized as type 3 (46%), a completely solid component without any cysts was categorized as type 4 (4%), and a large cyst without a solid component was categorized as type 5 (6%). In our study, only one patient had MRI images and the findings were categorized as type 4 according to the Kodama classification.

When a heterogeneous mass with calcifications and peripheral enhancement is observed in the late venous phase on dynamic CT and MRI images, it should be considered as a characteristic finding for AE.[9] In our study, it was possible to make a diagnosis by these imaging modalities. In our 8 cases, the diagnosis was made based on histopathological examinations because of the incompatibility of the findings obtained by clinical serology and imaging modalities.

The retrospective design and the small number of our

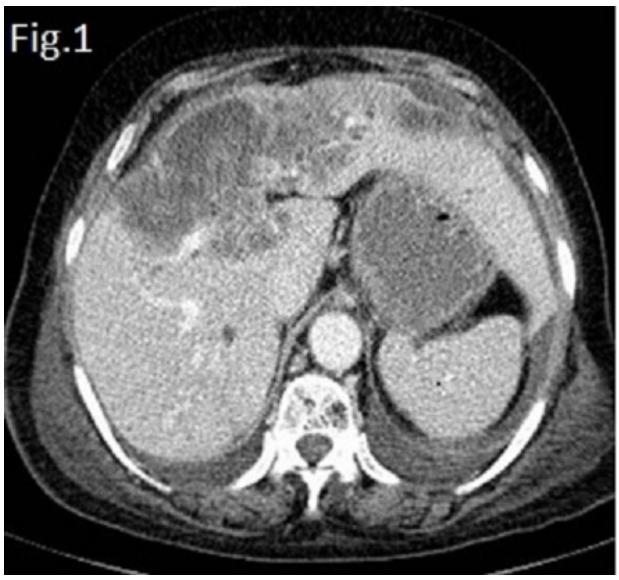

Figure 1. A 56-year-old female patient diagnosed with alveolar echinococcosis; the CT image shows a malignant-like hypodense lesion infiltrating the right and left lobes of the liver.

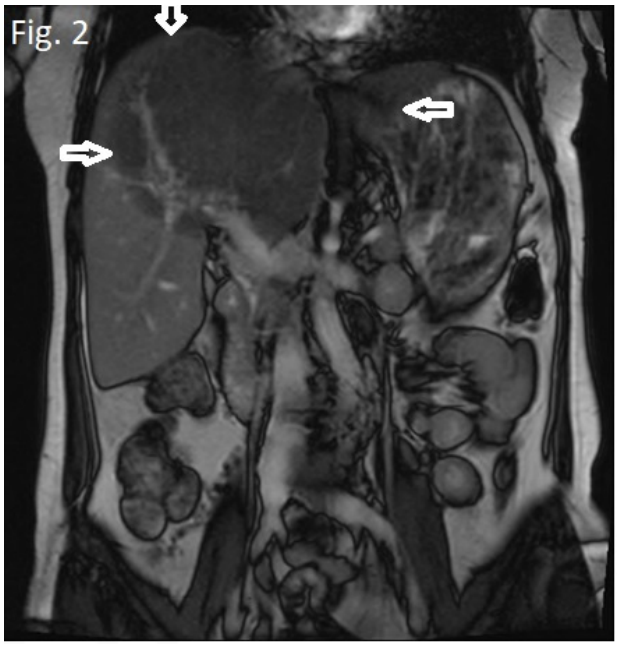

Figure 2. MR image of the patient with the previous CT image; MRI shows an irregularly demarcated infiltrative hypointense lesion with an increased enhancement pattern in the right-left lobe of the liver.

Study Highlights

What is current knowledge?

- The importance of radiological awareness in the diagnosis of AE
-
- ***What is new here?***
- AE disease can act like a tumor with an insidious and progressive growth pattern.

cases can be considered limitations for our study.

Conclusion

AE is a chronic parasitic disease that can slowly progress in several years before the manifestation of the signs and symptoms of the disease. The disease can behave like a tumor with an insidious and invasive growth pattern, making an early diagnosis imperative. We think that the use of radiological imaging can facilitate making the diagnosis of AE, especially in endemic regions.

Conflict of Interest

The authors declare that they have no conflict of interest.

Ethical Approval

Ethics committee approval was received for this research article (Van training and research hospital clinical research ethics committee. 2020/23 Date: 26/11/2020).

Author's Contribution

ST and MÖ contributed to the conception and design of the study and literature review. ST and MÖ collected all data and contributed to data interpretation. ST and MÖ drafted the first manuscript. All authors reviewed and approved the final version of the article.

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References

1. Eckert J, Conraths FJ, Tackmann K. Echinococcosis: an emerging or re-emerging zoonosis? Int J Parasitol. 2000;30(12-13):1283-94. doi: 10.1016/s0020-7519(00)00130-2.
2. Tüzün M, Hekimoğlu B. Pictorial essay. Various locations of cystic and alveolar hydatid disease: CT appearances. J Comput Assist Tomogr. 2001;25(1):81-7. doi: 10.1097/00004728-200101000-00014.
3. Ito A. Serologic and molecular diagnosis of zoonotic larval cestode infections. Parasitol Int. 2002;51(3):221-35. doi: 10.1016/s1383-5769(02)00036-3.
4. Spârchez Z, Osian G, Onica A, Bărbântă C, Tanțău M, Pascu O. Ruptured hydatid cyst of the liver with biliary obstruction: presentation of a case and review of the literature. Rom J Gastroenterol. 2004;13(3):245-50.
5. Macpherson CN, Milner R. Performance characteristics and quality control of community based ultrasound surveys for cystic and alveolar echinococcosis. Acta Trop. 2003;85(2):203-9. doi: 10.1016/s0001-706x(02)00224-3.
6. Eckert J, Gemmell MA, Meslin FX, Pawlowski ZS. WHO/OIE Manual on Echinococcosis in Humans and Animals: A Public Health Problem of Global Concern. Paris, France: World Organisation for Animal Health; 2001.
7. Bresson-Hadni S, Vuitton DA, Bartholomot B, Heyd B, Godart D, Meyer JP, et al. A twenty-year history of alveolar echinococcosis: analysis of a series of 117 patients from eastern France. Eur J Gastroenterol Hepatol. 2000;12(3):327-36. doi: 10.1097/00042737-200012030-00011.
8. Reuter S, Nüssle K, Kolokythas O, Haug U, Rieber A, Kern P, et al. Alveolar liver echinococcosis: a comparative study of three imaging techniques. Infection. 2001;29(3):119-25. doi: 10.1007/s15010-001-1081-2.
9. Kantarci M, Bayraktutan U, Karabulut N, Aydinli B, Ogul H, Yuce I, et al. Alveolar echinococcosis: spectrum of findings at cross-sectional imaging. Radiographics. 2012;32(7):2053-70. doi: 10.1148/rg.327125708.
10. Kodama Y, Fujita N, Shimizu T, Endo H, Nambu T, Sato N, et al. Alveolar echinococcosis: MR findings in the liver. Radiology. 2003;228(1):172-7. doi: 10.1148/radiol.2281020323.