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Case Report

Hepatic Invasive Echinococcosis Masquerading Angio-Invasive Hepatocellular Carcinoma — A Case Report

Faisal Ali[1]*, Muhammad Naseer[2], Ibrahim Ahmed[3], Ahmad Faruqui[3], Muhammad Rehan Javed[3]

[1]Resident Gastroenterology, Shifa International Hospital, Islamabad, Pakistan.

[2]Consultant Gastroenterology, Shifa International hospital, Islamabad, Pakistan.

[3]Medical officer, Shifa International Hospital, Islamabad, Pakistan.

***Corresponding Author:** Faisal Ali, Resident Gastroenterology, Shifa International Hospital, Islamabad, Pakistan.

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Abstract

Hepatic invasive echinococcosis (HIE) also known as hepatic alveolar echinococcosis (HAE) is caused by the larval stage of Echinococcus multilocularis tapeworm. It's transmitted through food or water contaminated with the eggs or through close contact with foxes or dogs. a case of 31 years old female presented with epigastric pain, nausea and vomiting from past several years. Abdominal imaging reveal multiple liver lesions which were biopsied. Histopathology of liver lesions showed chronic caseating granuloma, after which antituberculosis drugs was started. Her symptoms did not improved and on repeat imaging the size of the liver lesions had increased. On repeat biopsy of the liver lesions and lab workup, she was diagnosed with invasive echinococcosis. Albendazole 400mg BID was started. Her symptoms improved and the size of the lesions and cysts remained stable. She kept on regular follow ups with repeat abdominal imaging and lab workup. Hydatid cyst in the liver may be underdiagnosed and need careful evaluation with careful history, physical examination and appropriate imaging investigations.

Keywords: *Echinococcosis, Hepatic, Albendazole, Chronic Granulomatous Diseases*

Introduction

Hepatic invasive echinococcosis (HIE) also known as hepatic alveolar echinococcosis (HAE) is caused by the larval stage of Echinococcus multilocularis tapeworm. It's transmitted through food or water contaminated with the eggs or through close contact with foxes or dogs. It can spread to other organs such as the lungs, liver, spleen, kidneys, bone, and brain, leading to progressive destruction of tissue. Prompt diagnosis and treatment is paramount considering the morbidity of this disease. In the liver, common symptoms may include abdominal pain, jaundice, weight loss, and fatigue. In the lungs, symptoms may include chest pain, shortness of breath, or cough and in severe cases, anaphylaxis, or bleeding.

The disease is most found in Europe, Asia, and North America. Treatment includes a combination of surgery and long-term anti-parasitic medication to prevent recurrence. If not possible, needle aspiration, percutaneous drainage, or chemotherapy may be considered. Prevention of invasive echinococcosis involves proper hygiene and sanitation practices should be implemented. Travelers to high-risk areas are advised to avoid eating unwashed fruits and vegetables or drinking unfiltered water. Mortality is more than 90% in untreated patients within 10-15 years from the diagnosis (1). The important factor to improve the prognosis of HIE is the early diagnosis when complete resection is feasible (2). Owing to the long clinical latency, there are still some sporadic cases not diagnosed until progressing to the advanced stage when the lesion cannot be resected completely. The advanced stage is characterized by extensive intrahepatic lesions, invasion of caval vein or hepatic hilum, extended invasion of the diaphragm and retroperitoneal space, and distant metastasis. The management of advanced HIE especially the necessity of aggressive operations, like palliative resection and orthotopic liver transplantation, remains controversial (3-5).

Case Presentation

A 31 years old female with no previous co-morbid presented with the complaint of epigastric pain, nausea, decrease oral intake, and vomiting since 2016. On abdominal imaging, she had liver lesions. Baseline investigations like complete blood count, liver function tests, creatinine, and international normalized ratio were normal. Her ESR was 120 mm/hr and her tumor markers and viral serology were negative.

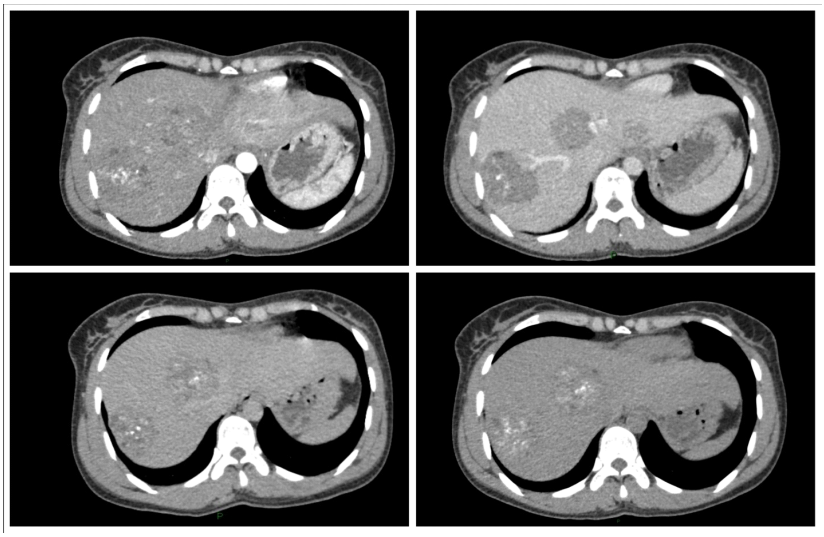

Figure 1. *Contrast Enhanced CT Abdomen showing multiple liver lesions with internal hyper densities and calcifications with surrounding hypo densities in both lobes.*

Ultrasound-guided core biopsy of the liver lesion was done using an 18 G needle, which showed large foci of calcification and necrosis, surrounded by epithelial cells and lymphocytes giving an appearance of chronic caseating granulomatous inflammation suspecting hepatic tuberculosis. She took anti-tuberculosis drugs for 9 months but her symptoms did not improve. Later she presented in a clinic in 2023 with fever, right upper quadrant pain, and vomiting. On repeat radiological scan, there was an interval increase in the size of large multiple coalescing masses in the liver, having an increase in the degree of blooming central calcification and surrounding hypo dense zone. Right lobe mass partially projecting into peri-porta distribution causing biliary obstruction and biliary dilation. There was hepatic vein thrombosis. There is a lollipop sign with a segmental branch of the hepatic and portal vein. The liver showed a nutmeg appearance with background nodularity and heterogeneity. There was an interval increase in peri-pancreatic and retroperitoneal lymph nodes.

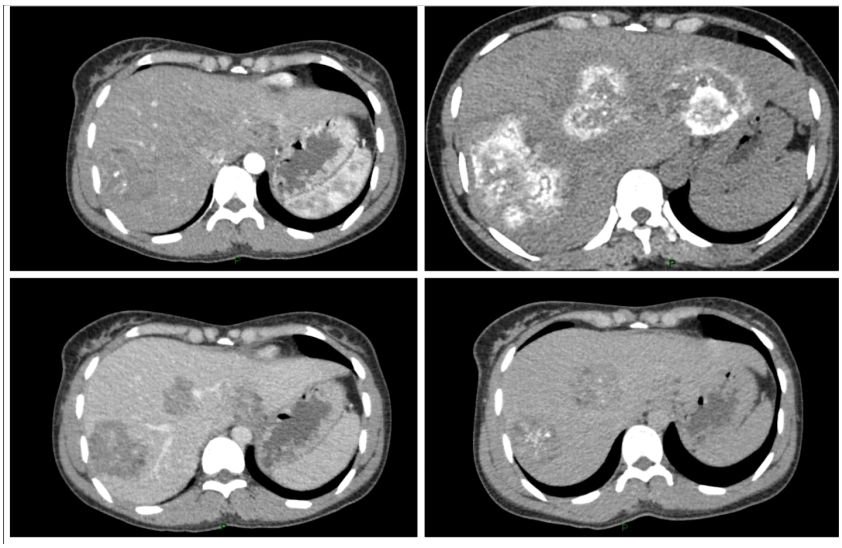

Figure 2. *Contrast enhanced CT abdomen in 2023 showing re-demonstration of multiple mixed hepatic lesions.*

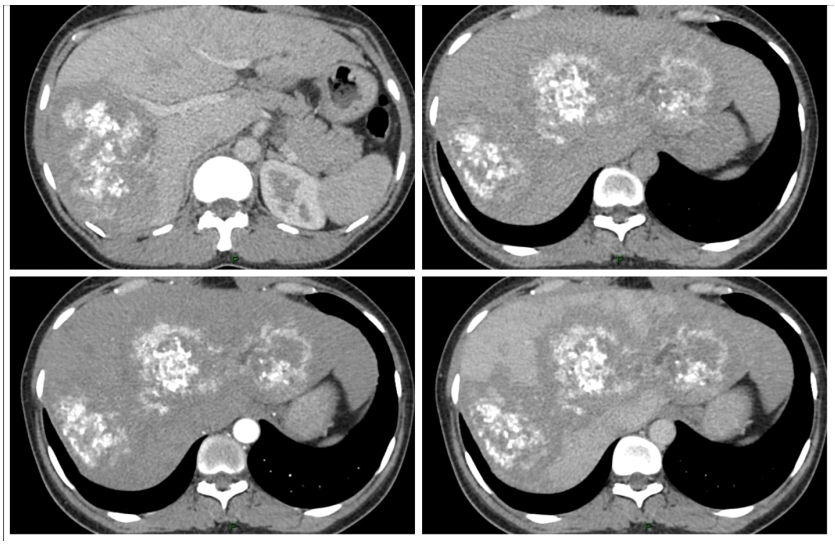

Figure 3. *Contrast Enhanced CT Abdomen showing multiple liver lesions with thrombosis of posterior division of portal vein.*

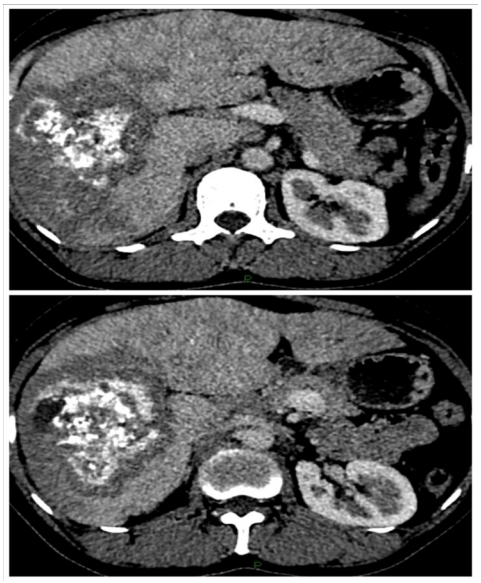

Figure 4. *Contrast Enhanced CT abdomen showing nutmeg appearance of liver with background nodularity and heterogeneity.*

Liver lesion biopsy was done. The biopsy showed chronic caseating granulomatous inflammation secondary to a hydatid cyst. Sections showed numerous granulomas. They contain a palisading cluster of epitheloid cells peripherally rimmed by lymphocytes with central necrosis. Within the necrosis, there are scattered irregular cyst fragments with acellular laminated membrane, proto-scoliosis, and hooklets. Foci of dystrophic micro-calcification are seen within the cyst wall.

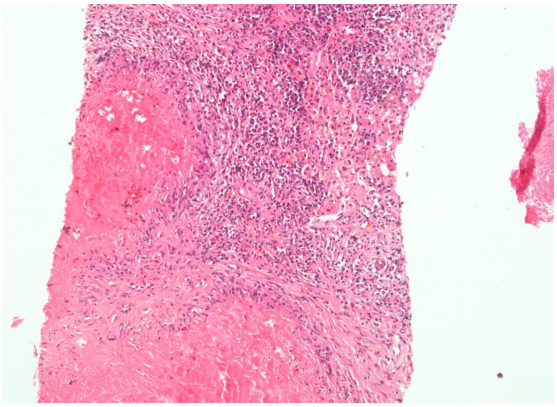

Figure 5. *Liver core biopsy showing granuloma, epitheloid cells, histocytes with central necrosis zone. (20x magnification with H&E staining)*

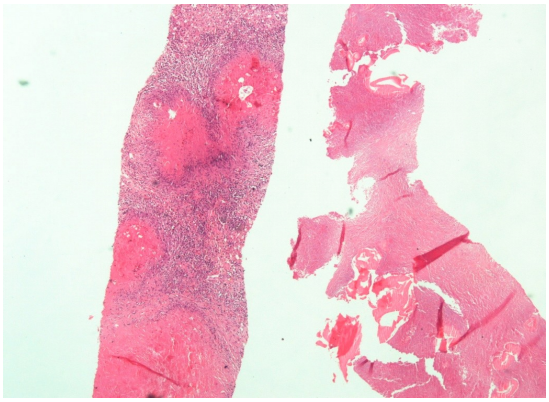

Figure 6. *Liver granuloma with necrotic debris (4x Magnification with H&E staining).*

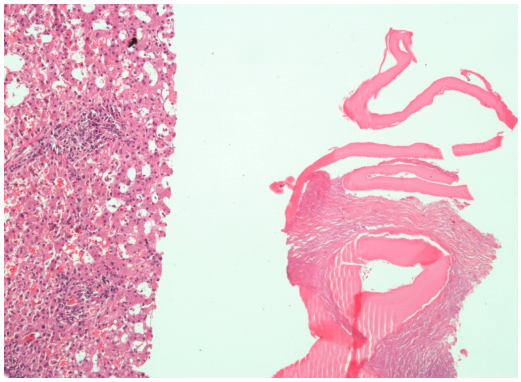

Figure 7. *Hydatid cyst with lamellated acellular layer (40x magnification with H&E staining).*

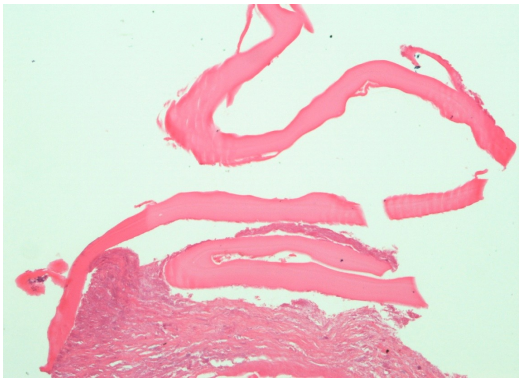

Figure 8. *Hydatid cyst acellular layer.*

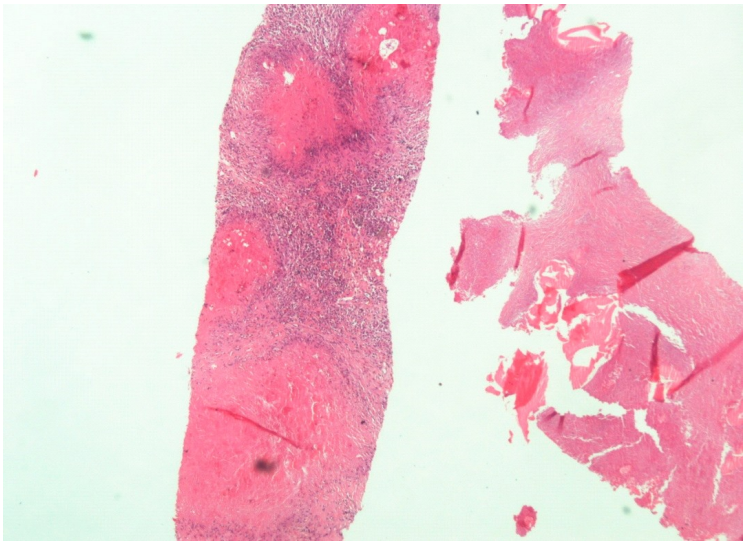

Figure 9. *Granuloma with caseous necrosis and epithelioid histocytes.*

Echinococcus serology was sent which came back positive. Albendazole 400mg Bid was started. The radiology scans were discussed in a multidisciplinary meeting; the findings were consistent with invasive echinococcosis. Patient was kept on regular follow ups. Her symptoms improved and the size of the cystic lesions remains stable. The liver function tests remain normal. We kept the patient on Albendazole therapy and regular follow ups with imaging and liver function test.

Discussion

Echinococcosis is a parasitic illness occurs when the host is infected by the larvae of the Echinococcus tapeworm. According to studies, the population's health in Central Asia is gravely at risk due to the public health issue of echinococcosis. Currently, the two primary pathogens causing echinococcosis in humans are cystic echinococcosis (CE) and alveolar echinococcosis (AE), which are both widely spread. In western China, the prevalence of echinococcosis is on average 1.08%, with some regions of the Qinghai-Tibet Plateau having a frequency as high as 12.09%. AE is known as "worm cancer" or "second cancer" because it is more likely to enter the liver than other organs such the "lung, brain, bone, or kidney" through blood and lymphatic channels (15). Cystic echinococcosis (CE) is a significant public health problem in South America, the Middle East and eastern Mediterranean, some sub-Saharan African countries, western China, and the former Soviet Union. In endemic regions, incidence rates for cystic echinococcosis can reach more than 50 per 100 000 person-years. Prevalence levels as high as 5%–10% may occur in parts of Argentina, Peru, East Africa, Central Asia and China. Alveolar echinococcosis (AE) has been reported in parts of central Europe, much of Russia, the Central Asian republics, northeastern, northwestern, and western China

Although radical resection was regarded as the first choice for HIE, due to the long clinical latency, many cases were not diagnosed until at an advanced stage when the lesion had no chance to be resected completely. The treatment of the advanced HAE, especially the necessity of aggressive operations, is disputed. Chemotherapy with Albendazole is commonly recommended for the HIE patient, but its role alone in advanced HIE remains unknown. Although there is no doubt that the first choice for treatment of HIE is surgical resection even after curative resection, recurrence can occur (7, 9, 10, 11). It seems to be related to invisible remnant parasitic tissue in the liver. Therefore, 2-year chemotherapy with Albendazole is recommended for most HAE patients even after curative resection (10, 11). Nevertheless, the chemotherapy alone seems little effective in the long run, especially for those with advanced HAE (12, 13, 14).

The effect of palliative resection for the advanced HAE was encouraging in a retrospective study by Kawamura and others (3). They reported the reduction surgery group (N = 63) had higher 10- and 15-year overall survival (97.1% and 92.8%) than that the drainage or exploratory laparotomy group (N = 6) (50.0% and 33.3%).

Orthotopic liver transplantation may offer the only opportunity for survival and cure for the patient with progressive advanced HAE, which is usually marked by a succession of biliary tract infectious episodes, obstructive jaundice, liver abscesses, septicemia, recurrent cholangitis, bleeding caused by portal hypertension, and chronic Budd–Chiari syndrome (1).

Our literature review shows that liver transplant is a feasible treatment option among patients with advanced invasive echinococcosis. Despite the risk of recurrent parasitic lesions induced by immunosuppressive agents, available evidence demonstrated that liver transplant has allowed patients with echinococcosis to survive for more than 10 years (6, 7). Sporadic reports of Liver Transplants for echinococcosis have been published in different countries around the world, including Poland, China, Germany, Spain, Switzerland, Belgium, Kyrgyzstan, United States, and mostly from Turkey as an endemic country in the Middle East (8).

Conclusion

In conclusion, LT appears to be feasible and effective for patients with fatal HAE. The earlier decision for LT, pre and post-transplant benzimidazole therapy, minimal post-transplant immunosuppressive regimen, and regular assessments for early diagnosis of parasite recurrence are crucial to prevent local and distant metastases during the post-transplant course and achieve acceptable long-term survival rates (8).

Conflicts of Interest

The authors declare no conflicts of interest to this study.

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