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Case Report

Multidisciplinary Management of Alveolar Echinococcosis With Bilateral Pulmonary Involvement: A Case Report

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Abstract

Alveolar echinococcosis (AE) is a rare, chronic, and invasive parasitic zoonosis caused by the larval stage of the helminth Echinococcus multilocularis, which can often mimic malignancy. While this disease particularly affects the liver, pulmonary involvement is less commonly seen. Bilateral pulmonary involvement has been reported in a very limited number of cases in the literature.

In this study, we present a 45-year-old male patient with liver and bilateral pulmonary involvement. Imaging studies of the patient who presented with abdominal pain revealed an irregularly bordered mass in the right hepatic lobe, a lobulated contoured lesion approximately 2x3 cm in size with calcified areas in the center located subpleurally in the right upper lung lobe, and a lesion approximately 1x2 cm in size in the left lower lung lobe were detected. Under multidisciplinary council management, uniportal VATS wedge resection was applied to the left lower lobe, and the pathology was confirmed as echinococcus alveolaris. Subsequently, wedge resection was performed to the right upper lobe and right hepatectomy was performed for liver involvement.

Following surgical treatment with multidisciplinary approach, the patient was discharged with recovery.

Keywords: Echinococcus Alveolaris, Alveolar Cyst, Pulmonary Cyst, Minimally Invasive Surgery, Zoonotic Disease, Major Hepatectomy

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Alveolar echinococcosis (AE) is a parasitic zoonosis caused by the metacestode (larval) stage of Echinococcus multilocularis, a helminth from the cestode class.[1]

It is more commonly encountered in the Eastern Anatolia region of Türkiye. The global incidence is estimated at 0.03-1.2/100,000 per year, but this rate is higher in endemic areas.

Following infection, parasite larvae primarily settle in the liver through portal circulation and form the main disease focus there. From the liver, it can spread to other organs through hematogenous, lymphatic, or direct invasion routes. Primary pulmonary involvement is quite rare; in most cases, hematogenous spread of infection that initially begins in the liver is involved.[2,3] Pulmonary involvement is seen in approximately 20% of cases, while bilateral pulmonary involvement is quite rare and has been reported in a limited number of cases in the literature.

AE develops as a lesion that progresses chronically, is characterized by a long asymptomatic period (5-15 years), and develops in the form of an invasive malignancy-like, multiple, and exogenously budding lesion, so its detection is

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usually incidental or diagnosis is made when symptoms appear in advanced stages.[4] Due to this characteristic, it is also called "parasitic cancer."

Imaging methods play an important role in diagnosis. Ultrasonography, computed tomography (CT), and magnetic resonance imaging (MRI) are frequently used modalities. Radiologically, pulmonary AE, which is observed as nodular lesions containing calcifications, irregularly bordered, harboring necrotic areas, and having an infiltrative character, can be confused with primary lung malignancies or metastatic lesions.[5,6] Therefore, pulmonary tuberculosis, lung carcinoma, metastatic lesions, and other infectious pathologies should be considered in differential diagnosis. Serological tests, especially in endemic areas, can be helpful in diagnosis. Enzyme-linked immunosorbent assay (ELISA) and Western blot techniques are the most commonly used serological methods.[2,5]

In this case, we present an alveolar echinococcosis case with both hepatic and bilateral pulmonary involvement that mimics malignancy.

Case Report

A 45-year-old male patient with no known medical history, engaged in farming and living in a rural area, presented to an external center with complaints of right upper quadrant pain that had been continuing for 3 months and had intensified in the last 2 weeks. The patient was diagnosed with E. alveolaris with accompanying hepatic and bilateral pulmonary involvement, and due to biliary obstruction, a stent was placed in the right bile duct via ERCP and he was referred to our center for further treatment.

Upon evaluation at our hospital, physical examination revealed tenderness in the right upper quadrant and hepatomegaly, with no other pathological findings. Respiratory system examination showed no pathological findings. The patient's temperature was 36.8°C, pulse 78/min, blood pressure 130/80 mmHg, respiratory rate 16/min, oxygen saturation 97%, blood type A Rh (+), height 170 cm, and weight 66 kg.

Laboratory tests revealed leukocyte count 9,470/mm^3, eosinophil ratio 1.5%, hemoglobin 12.9 g/dL, platelet count 550,000/mm^3, ALT 48 U/L, AST 54 U/L, GGT 99 U/L, ALP 264 U/L, total bilirubin 0.77 mg/dL, direct bilirubin 0.25 mg/dL, sedimentation rate 42 mm/hour, and C-reactive protein 16.9 mg/dL.

Abdominal ultrasonography revealed a heterogeneous, irregularly bordered, hypoechoic solid lesion in the right hepatic lobe. Abdominal computed tomography showed a mass lesion with heterogeneous structure, irregular borders, and areas of calcification and necrosis filling the right hepatic lobe, and a stent was observed in the right bile duct (Fig. 1a).

The patient's posteroanterior chest radiograph showed irregularly bordered nodules in the right middle lung zone and left lower lung zone (Fig. 1b), and thoracic tomography revealed a lobulated contoured lesion approximately 2x3 cm in size with calcified areas in the center located subpleurally in the right upper lung lobe and an irregularly bordered lesion approximately 1x2 cm in size in the left lower lung lobe (Fig. 1c, d). These findings suggested primary lung malignancy or metastatic lesions.

Serological tests showed positive Anti Echinococcus multilocularis antibodies by ELISA.

In the multidisciplinary tumor council, the patient was evaluated as having E. alveolaris filling the right hepatic lobe with suspected inferior vena cava invasion and also bilateral pulmonary involvement. It was decided that lung lesions would first be resected by thoracic surgery, followed by treatment of the liver lesion by general surgery. For the patient with extensive hepatic involvement, it was decided to perform ICG R15 testing and calculate remaining liver volume to evaluate resectability.

First, wedge resection was applied to the lesion in the left lung via uniportal VATS (Video Assisted Thoracoscopic Surgery), then to the right lung. Intraoperatively, a subpleurally located, hard, white-yellow colored, irregularly bordered

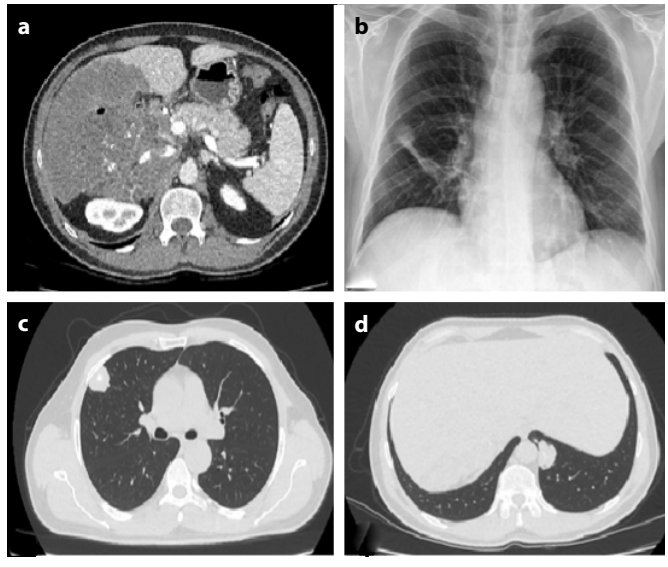

Figure 1. (a) Preoperative tomographic image of Echinococcus alveolaris in the right hepatic lobe. **(b)** Preoperative posteroanterior chest radiograph image of Echinococcus alveolaris. **(c)** Preoperative axial section tomographic image of Echinococcus alveolaris in the right lung. **(d)** Preoperative axial section tomographic image of Echinococcus alveolaris in the left lung.

lesion was observed. Histopathological examination revealed the diagnosis of echinococcus alveolaris with laminar membrane structures and alveolar pattern showing the development of characteristic daughter vesicles. Immunohistochemical staining yielded positive reactions against parasite antigens. Two weeks later, wedge resection was also applied to the lesion in the right upper lung lobe, and the histopathology of this lesion was also compatible with echinococcus alveolaris.

Two months after lung surgery, the patient was re-evaluated by general surgery for liver treatment. Volumetric examination calculated FRL (left lobe) as 630cc, GRWR >0.8%. ICG testing could not be performed due to technical reasons. The patient, who was suitable for major hepatectomy, underwent surgery. Right hepatectomy was completed with placement of a T-tube in the choledochus. Histopathological examination of the liver specimen was also reported as compatible with echinococcus alveolaris.

In the postoperative period, the patient developed bile leak and biloma. The biloma was percutaneously drained and the bile leak was controlled with percutaneous and endoscopic methods. Albendazole treatment was started (at a dose of 10-15 mg/kg/day for 2 years) and the patient was called for regular follow-ups (Fig. 2). Recurrence was not detected in control imaging at 18 months postoperatively.

Discussion

Alveolar echinococcosis is a rare parasitic disease that primarily affects the liver and has a mortality rate of up to 90% if left untreated.[1,3] Delays in diagnosis may occur due to its long asymptomatic course and incidental onset. Primary pulmonary involvement is quite rare. Studies have reported primary pulmonary involvement in less than 2% of AE cases.[2] Bilateral pulmonary involvement, as in our case, is reported even more rarely in the literature.

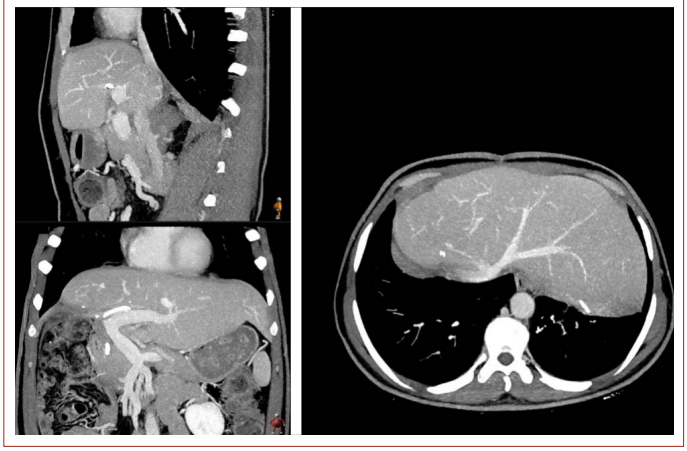

Figure 2. Post-hepatectomy 6 month images.

The main approach in AE treatment is surgical excision and long-term antiparasitic chemotherapy. Radical resection is recommended when surgical excision of lesions is possible, as in our case.[3,5] In the WHO guidelines published in 2010, surgical treatment is recommended as the first choice in cases where R0 resection can be performed in AE treatment. With the development of minimally invasive surgical techniques in recent years, resection can be performed with approaches such as VATS in appropriate cases. This method was preferred and successfully applied in our case.

Antiparasitic chemotherapy can be applied as a complement to surgical treatment or alone in inoperable cases. Benzimidazole derivatives (albendazole and mebendazole) are the most commonly used agents. Albendazole is preferred because it shows better bioavailability than mebendazole.[1,7] Albendazole treatment is generally recommended at a dose of 10-15 mg/kg/day for two years or longer.

Brunetti et al.[1] emphasized the importance of a multidisciplinary approach in AE treatment in the WHO guidelines they published. In our case, successful treatment results were obtained through coordinated work of thoracic surgery, general surgery, radiology, and hepatology departments.

Early diagnosis and appropriate treatment approach are determining factors in the prognosis of alveolar echinococcosis. In a study by Tichý et al.,[7] they reported that the 10-year survival rate in cases undergoing R0 resection was over 90%. Curative surgery can be performed in 35% of cases, and in unresectable cases, the only curative surgery is liver transplantation.[8] In inoperable cases, 5-year survival is around 80% with long-term albendazole treatment. Radical surgical approach was applied in this patient and albendazole treatment was started. No recurrence was detected at the 18-month postoperative control, but long-term follow-up is recommended due to the risk of late recurrence of AE.

Our study has several important clinical and scientific features. First, it is an AE case with bilateral pulmonary involvement, which is rarely seen. Second, having a radiological appearance that mimics malignancy shows that AE should also be considered in differential diagnosis. Third, successful treatment results were obtained with minimally invasive surgical techniques. Finally, it emphasizes the importance of a multidisciplinary approach in both treatment planning and post-treatment management in cases with liver and lung involvement.

Conclusion

In this case of primary hepatic E.A. accompanied by bilateral pulmonary involvement, lung lesions were first treated minimally invasively, and subsequently, the highly complicated case was curatively treated with major hepatectomy. Management of E.A. cases should be multidisciplinary.

Disclosures

Informed Consent: Written informed consent was obtained from the patient while maintaining confidentiality of patient identity.

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References

1. Brunetti E, Kern P, Vuitton DA. Writing Panel for the WHO-IWGE. Expert consensus for the diagnosis and treatment of cystic and alveolar echinococcosis in humans. Acta Trop. 2010;114(1):1-16.
2. Bulakçı M, Kartal MG, Yılmaz S, et al. Multimodality imaging in diagnosis and management of alveolar echinococcosis: An update. Diagn Interv Radiol. 2016;22(3):247-256.
3. Özdemir A, Bozdemir ŞE, Akbiyik D, et al. Pulmonary alveolar echinococcosis: a case report. Turkiye Parazitol Derg. 2021;45(2):147-150.
4. Bresson-Hadni S, Delabrousse E, Blagosklonov O, et al. Imaging aspects and non-surgical interventional treatment in human alveolar echinococcosis. Parasitol Int. 2006;55 Suppl: S267-S272.
5. Kayaalp C, Dirican A, Aydin C. Primary hepatic alveolar echinococcosis: a critical review of the literature. World J Gastroenterol. 2022;28(2):226-239.
6. Kern P, Bardonnet K, Renner E, et al. European echinococcosis registry: human alveolar echinococcosis, Europe, 1982-2000. Emerg Infect Dis. 2003;9(3):343-349.
7. Tichý E, Torbašinović I, Kolářová L, et al. Surgical treatment outcomes of alveolar echinococcosis: A 25-year single-centre experience. PLoS Negl Trop Dis. 2022;16(5):e0010408.
8. Ozdemir F, Ince V, Barut B, Onur A, Kayaalp C, Yilmaz S. Living donor liver transplantation for Echinococcus Alveolaris: single-center experience. Liver Transpl. 2015;21(8):1091-5.