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Beyond the Liver: A Rare Case of Bilateral Pulmonary Cystic Echinococcosis

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ABSTRACT

Background: Pulmonary hydatidosis, a zoonotic infection caused by the larval stage of Echinococcus granulosus, is an uncommon manifestation of cystic echinococcosis. While the liver is the primary site of involvement, pulmonary involvement in relatively rare, making these cases relatively diagnostically challenging.

This case report presents the clinical features, diagnostic workup, and successful treatment of a young man with isolated pulmonary hydatid disease, emphasizing the importance of considering this rare etiology in the differential diagnosis of cystic lung lesions.

Case Presentation: In early 2024, the patient was admitted to a secondary level hospital with asthenia, weight loss, and hemoptysis. A chest CT scan revealed two fluid-filled cystic lesions. Blood tests showed eosinophilia and elevated inflammatory markers. Serological testing for *E. granulosus* resulted positive, and surgical intervention was recommended. Parasitological examination of intraoperative specimens confirmed the presence of E. granulosus, which was also verified histologically.

Following two surgical procedures and prolonged pharmacological treatment, the patient achieved complete clinical remission.

Conclusion: Timely and accurate microbiological diagnosis is essential in the management of echinococcosis. The treatment of pulmonary cysts poses significant challenges, both surgically, due to the risk of rupture and dissemination, and therapeutically, given the limited data on optimal treatment duration.

Keywords: Hydatid disease; Echinococcosis; Pulmonary disease; Parasites

INTRODUCTION

Cystic Echinococcosis (CE), or hydatidosis, is a globally distributed zoonosis. Endemic regions include South and Central America, the Middle East, sub-Saharan Africa, Russia, China, Australia, and New Zealand. In the United States and Central Europe, most cases occur in immigrants from endemic areas [1].

CE is caused by the larval stage of a cestode belonging to the genus Echinococcus. The adult worm parasitizes the small intestine of canids, particularly dogs. Its life cycle involves the formation of larval cysts in various organs (e.g., liver, lungs) of intermediate hosts such as sheep, cattle, pigs, goats, and horses. Humans become accidental intermediate hosts through direct contact with infected dogs or ingestion of food contaminated with feces containing parasite eggs [2].

In humans, ingested eggs hatch in the intestine, releasing oncospheres that penetrate the intestinal wall, enter the bloodstream, and potentially migrate to any organ; these cysts may contain daughter capsules and numerous infectious protoscoleces.

In recent years, the incidence of the CE has progressively increased. The most frequently affected organ is the liver, with a rate of 60% to 80% and the second most common is the lung with a rate of 20% to 30% [3-6].

The majority of CE cases involve a single organ with the presence of a solitary cyst. Pulmonary CE is often secondary to hepatic involvement; however, an exclusively pulmonary presentation is possible, most commonly affecting one lung, and rarely, both lungs. Pulmonary CE is more frequent in children compared with adults. Small pulmonary CE lesions are often asymptomatic, whereas large cyst can result in acute life-threatening complications such as anaphylactic shock, pneumotorax, empyema and pulmunary abscess with secondary infection.

Diagnosis of CE is based on a combination of epidemiological history, clinical findings, imaging, serology, and analysis of cyst fluid [7].

Clinical manifestations vary depending on the number, size, and location of the cysts.

The treatment of pulmonary CE is both medical and surgical. Although some studies reported the benefit of exclusive antiparasitic treatment in selected patients, surgical intervention is the main treatment of choice. Some studies recommend avoiding the use of preoperative antiparasitic therapy, suggesting it should be initiated immediately after the surgical procedure; however, this approach remains a matter of debate. Moreover, the timing of postoperative medical treatment is not clearly established, particularly in the presence of intraoperative complications that increase risk of dissemination and disease recurrence [3,8-10].

CASE PRESENTATION

We report the case of a 27-year-old male from Mali who arrived in Italy in 2014 after traveling through Libya and Algeria. He worked as a labourer, lived alone, and had no significant medical history prior to 2023. In early 2024, he was admitted to the secondary level Guglielmo da Saliceto Hospital with asthenia, weight loss, and hemoptysis.

His medical history revealed that in October 2023, during a stay in Mali, he underwent a pulmonology consultation in Bamako for persistent cough, dyspnea, and pleuritic chest pain. Sputum testing for acid-fast bacilli was negative,

while an interferon-gamma release assay resulted positive. Thoracentesis revealed exudative pleural fluid with 45% lymphocytes, but no imaging was performed. Empirical antitubercular therapy was initiated based on a presumptive diagnosis of pleural tuberculosis, consisting of isoniazid, rifampicin, ethambutol, and pyrazinamide for two months (from November 2023), followed by rifampicin and isoniazid from January 2024.

Despite treatment, the patient's condition deteriorated. Upon returning to Italy, he was urgently hospitalized. Chest and abdominal CT scans revealed two large pulmonary cysts with fluid density and thick walls, showing homogeneous contrast enhancement (right: ~15.5 cm × 12 cm × 17.5 cm; left: ~13.5 cm × 10 cm × 15 cm), causing compression and atelectasis of both lower lobes (Figure 1). No abdominal abnormalities were detected. Blood tests (Figure 2) showed eosinophilia, and serology for *E. granulosus* was positive (IgG Index: 1.54, Positive cut off: >1,1). Antitubercular therapy was discontinued, and surgical intervention was planned.

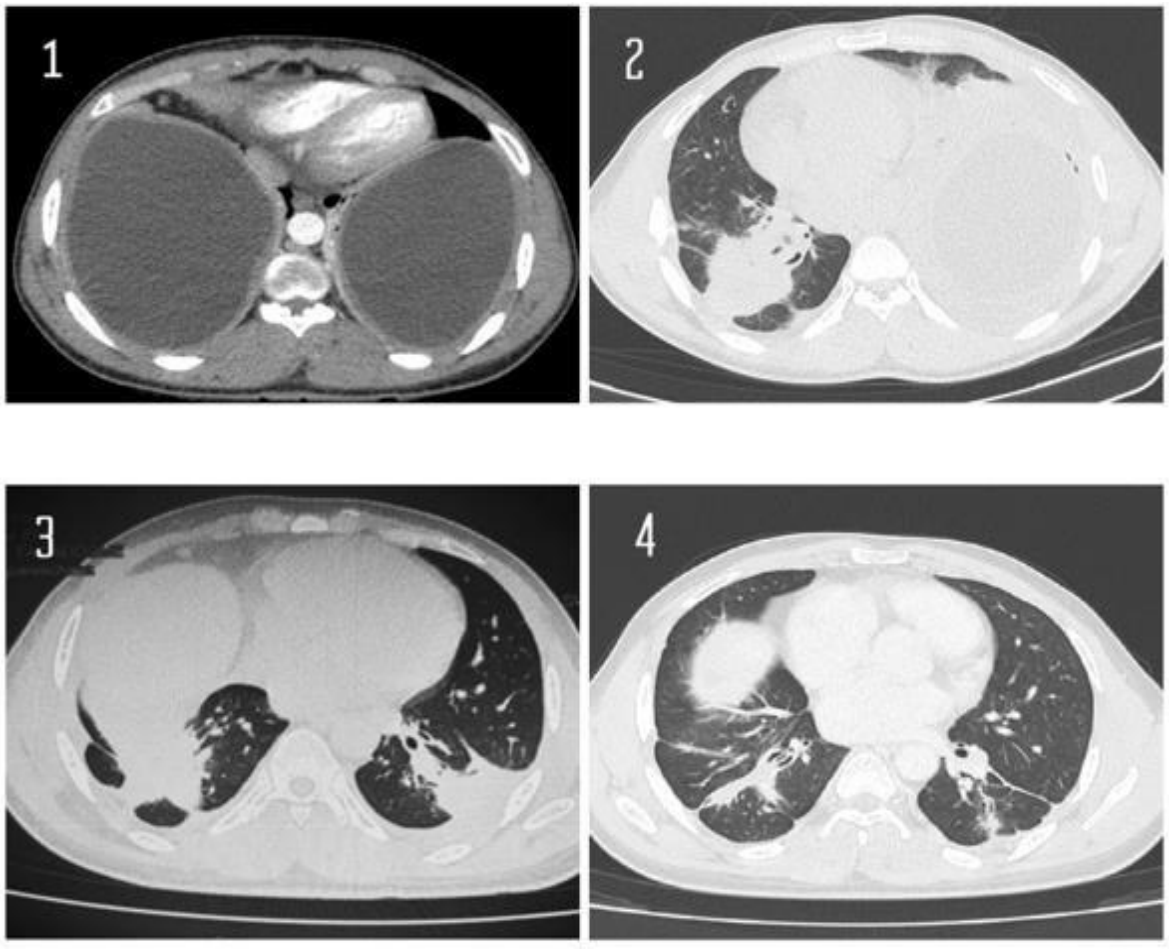

Figure 1: Radiological progression from diagnosis to the end of antiparasitic treatment. The first image (1) shows the baseline findings, followed by CT-scan before the second surgery (2), and the radiological follow up at six (3) and nine (4) months after diagnosis.

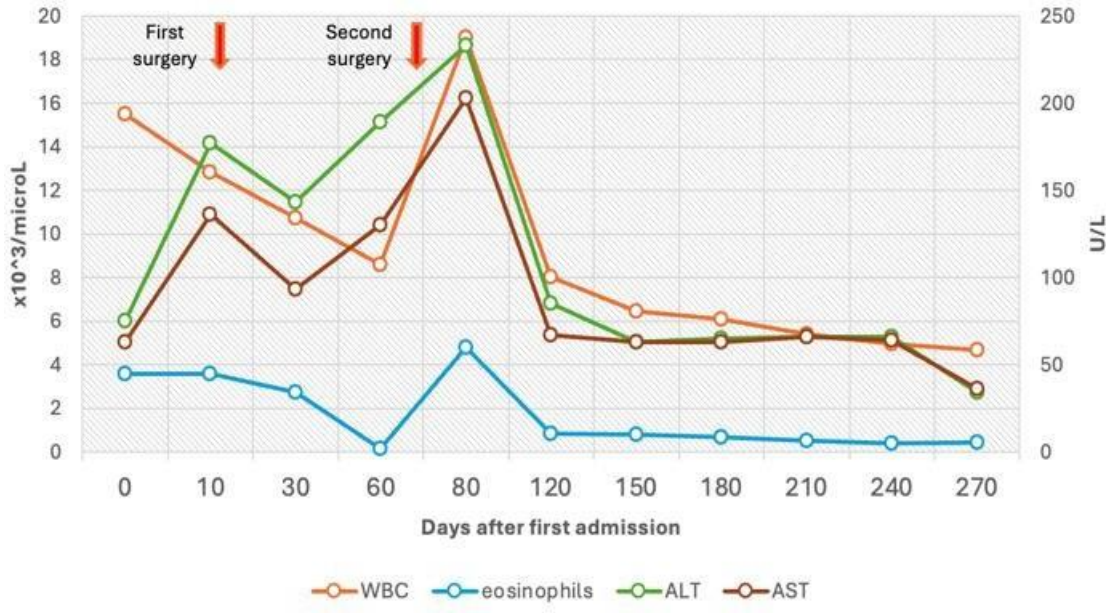

Figure 2: Trend of blood test results from diagnosis to the end of antiparasitic treatment.
WBC: White Blood Count; AST: Aspartate Aminotransferase; ALT: Alanine Aminotransferase

Approximately two weeks later, the patient underwent right thoracotomy. A large hydatid cyst was identified in the lower pleural cavity, with extensive adhesions. During dissection, the cyst ruptured, triggering an anaphylactic reaction and preventing removal of the contralateral cyst. The patient was transferred to the intensive care unit for monitoring.

Parasitological analysis of the cystic fluid revealed hooklets and protoscoleces of *E. granulosus* (Figure 3), and histological examination confirmed the diagnosis. Perioperatively and postoperatively, the patient was treated with albendazole (400 mg twice daily) and praziquantel (40 mg/kg weekly, for a total of four weeks). Despite mild cholestasis, his condition improved, and he was discharged on albendazole.

Two months later, while awaiting surgery for the left-sided cyst, the patient developed acute respiratory failure. CT imaging showed detachment of the cyst wall (12.5 cm × 8.5 cm) and pleural effusion. Emergency surgery was performed, and the left-sided cyst was successfully removed. This time, the cyst was punctured and aspirated prior to extraction, avoiding complications.

The patient continued albendazole therapy despite elevated liver enzymes, without gastrointestinal symptoms. Three months later, CT imaging showed bilateral parenchymal consolidation with some liquefaction areas. Albendazole was extended to six months post-second surgery (nine months total). Radiological improvement, normalization of eosinophilia, and liver function were achieved.

One year after diagnosis and three months after completing antiparasitic therapy, the patient remained clinically stable and asymptomatic. Follow-up imaging showed no evidence of disease recurrence.

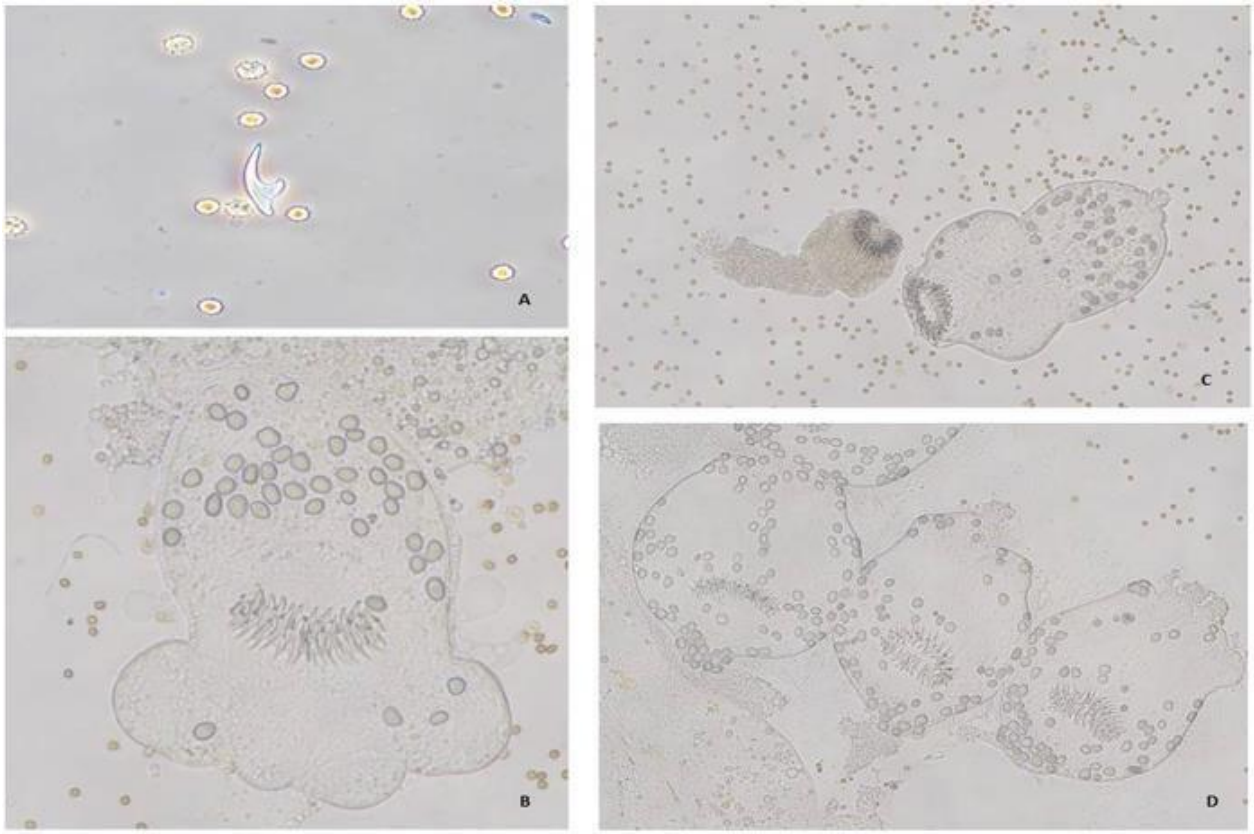

Figure 3: Wet mount of cystic fluid from pulmonary cyst showing hooklets (A) and protoscoleces (B-C-D) of Echinococcus granulosus by microscopic examination.

DISCUSSION

Echinococcosis is classified as a Neglected Tropical Disease (NTD) despite its global distribution and significant public health burden. Humans are incidental hosts, and individuals living in rural and ranching areas are at higher risk of infection.

In this case report we presented the clinical features, diagnostic workup, and successful treatment of a young man with a rare isolated bilateral pulmonary CE.

Pulmonary CE is a potentially life-threatening condition with a variable clinical course and often delayed diagnosis. Its radiologic and clinical polymorphism complicates early recognition, necessitating a multidisciplinary diagnostic approach that includes clinical evaluation, serologic testing, and imaging studies. A widely accepted standard of care for the treatment of CE has yet to be established outside specialized referral centers. This is largely due to the lack of well-designed clinical trials needed to guide evidence-based practice. Additionally, the management of complicated cysts often requires individualized clinical decisions, as there are no specific or universally applicable guidelines to support optimal treatment in such cases.

Surgical excision remains the cornerstone of treatment for pulmonary hydatid cysts. However, the heterogeneity of morphology and anatomical location of cysts requires surgical strategies tailored to each patient. In some cases, particularly when surgery is contraindicated or incomplete, antiparasitic pharmacotherapy plays a crucial additional role.

Three standard modalities are currently recognized in the management of lung CE:

- Surgical intervention
- Antiparasitic therapy with benzimidazoles
- Watch-and-wait approach, particularly for inactive or calcified cysts [8].

Despite surgery is the first-line treatment for pulmonary CE, pharmacological management, especially with albendazole, requires careful planning. Albendazole is typically administered at a dose of 10-15 mg/kg/day, often divided into two doses, and the duration of treatment can vary from several weeks to months depending on the size of the cyst, its location, and surgical outcomes [2].

Albendazole remains the primary pharmacological option for treating CE. Nonetheless, when used in combination with praziquantel, the treatment demonstrates enhanced scolicidal and anti-cyst effects, offering a greater likelihood of cure or clinical improvement than albendazole alone [11].

In this case, an initial treatment with praziquantel and albendazole for one month, followed by prolonged therapy (up to nine months) with albendazole was necessary because of bilateral involvement and intraoperative complications leading to the need for surgery in two stages. Moreover, antiparasitic treatment was initiated perioperatively and not after the diagnosis, since, as indicated in the literature, premature initiation of medical therapy may induce detachment of the parasitic cyst membrane and opening of cystobronchial fistulas [12].

During treatment, monitoring for hepatotoxicity is essential, as increases in liver enzymes are common.

In addition, drug therapy should be undertaken to sterilize the cyst, reduce the possibility of anaphylaxis, decrease cyst wall tension and reduce the rate of post-operative recurrence [13].

Of particular relevance is the geographic origin associated with this case, since the condition was initially misdiagnosed as tuberculosis in the patient's native region. While *E. granulosus* is endemic in several African regions, including North and East Africa, its presence in Mali is less well documented. A 2022 meta-analysis on echinococcosis in Africa reported variable prevalence across the continent, with higher rates in East and North Africa, but limited data from West African countries such as Mali [14]. The disease may therefore have been contracted during the patient’s migratory route; however, this highlights the need to strengthen surveillance and epidemiological studies in underrepresented regions, as well as the importance of conducting thorough travel histories, because underdiagnosis may obscure the true burden of the disease [15,16].

CONCLUSION

In conclusion, in endemic areas, monitoring Echinococcus spp. transmission and implementing control programs are the main public health goals.

In contrast, in low-incidence countries, the management of hydatidosis needs to be less heterogeneous; practical, accessible and shareable guidelines are needed. Targeted treatment must consider the clinic, risk factors, and surgical site assessment.

In conclusion, successful management of pulmonary echinococcosis requires early diagnosis, individualized surgical planning, and prolonged antiparasitic therapy under specialist supervision. Increased awareness and improved diagnostic skills are essential, particularly in regions where the disease may be poorly recognized.

AUTHOR CONTRIBUTION

Conceptualization: M. Degli Antoni, G. Tocci; Original Draft Preparation: G.Tocci and M. Degli Antoni; Methodology: G.Tocci and M. Degli Antoni; Validation: G.Tocci and M. Degli Antoni; Investigation: G. Lo Cascio, V. Lepera, F. Paolillo, A. Zappavigna, G. Palladini, F. Trapani, G.Tocci and M. Degli Antoni; Resources: G. Lo Cascio, V. Lepera, F. Paolillo, A. Zappavigna, G. Palladini, F. Trapani, G.Tocci and M. Degli Antoni; Data Curation: G.Tocci and M. Degli Antoni; Review and Editing: G.Tocci and M. Degli Antoni; Visualization: G. Lo Cascio, F. Trapani; Supervision: G. Lo Cascio.

INFORMED CONSENT STATEMENT

Written informed consent was obtained from the patient for the publication of this paper, along with a privacy form signed in accordance with the guidelines of our Ethics Committee.

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