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Pulmonary cystic echinococcosis in contrast-enhanced ultrasound – a case report

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Keywords
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Abstract

Aim of the study: Pulmonary cystic echinococcosis is a parasitic infection transmitted by dogs and occurring in livestock-raising areas. It is included among the neglected tropical diseases, according to the World Health Organization. Imaging plays a pivotal role in the diagnosis of this disease. While cross-sectional imaging modalities such as computed tomography and magnetic resonance imaging are preferred, lung ultrasound may be another feasible technique. **Case description:** We report a case of pulmonary cystic echinococcosis in a 26-year-old woman who was examined by contrast-enhanced ultrasound, which showed marked annular enhancement around the hydatid cyst, mimicking a superinfected cyst. **Conclusions:** Contrast-enhanced ultrasound examination in pulmonary cystic echinococcosis should be studied in a larger population to determine the value of additional contrast administration. In the present case report, no superinfected echinococcal cyst was seen despite marked annular contrast enhancement.

Introduction

Pulmonary cystic echinococcosis (CE) is a zoonotic disease with a low prevalence in central Europe. Also known as "hydatid disease," it is caused by the canine tapeworm (*Echinococcus granulosus*), which is transmitted primarily by dogs and dog-like animals. To complete the life cycle, *E. granulosus* requires a further mammalian host. Via fecal-oral transmission, the ova are digested by natural intermediate hosts (especially ruminants). Humans are infected accidentally by ingestion of these eggs. The larvae migrate from the small intestine via blood or lymphatic vessels, primarily to the liver (70–80%) and to the lung (15–20%)[1]. In up to one in six patients with pulmonary CE, a concomitant liver manifestation is reported. Here, the larvae mature and form single or multiple expanding cysts. The hydatid cyst is formed by three layers and intraluminal hydatid fluid: a vascularized connective tissue capsule (pericyst), derived from the external host, the middle cuticula (exocyst), building an acellular defense shield, and an internal germ layer (endocyst), producing cystic fluid and protoscolices.

Clinically, these pulmonary cysts are usually asymptomatic and diagnosed incidentally (uncomplicated cysts). Complicated cysts present with respiratory symptoms such as cough, respiratory distress, chest pain, or hemoptysis. Hence, imaging is crucial for diagnosis, and further workup with additional serological testing is necessary. It has been shown that B-mode ultrasound (US) is helpful for diagnosing pulmonary CE[2]. This case report describes perfusion patterns observed via contrast-enhanced ultrasound (CEUS) in a case of pulmonary cystic echinococcosis.

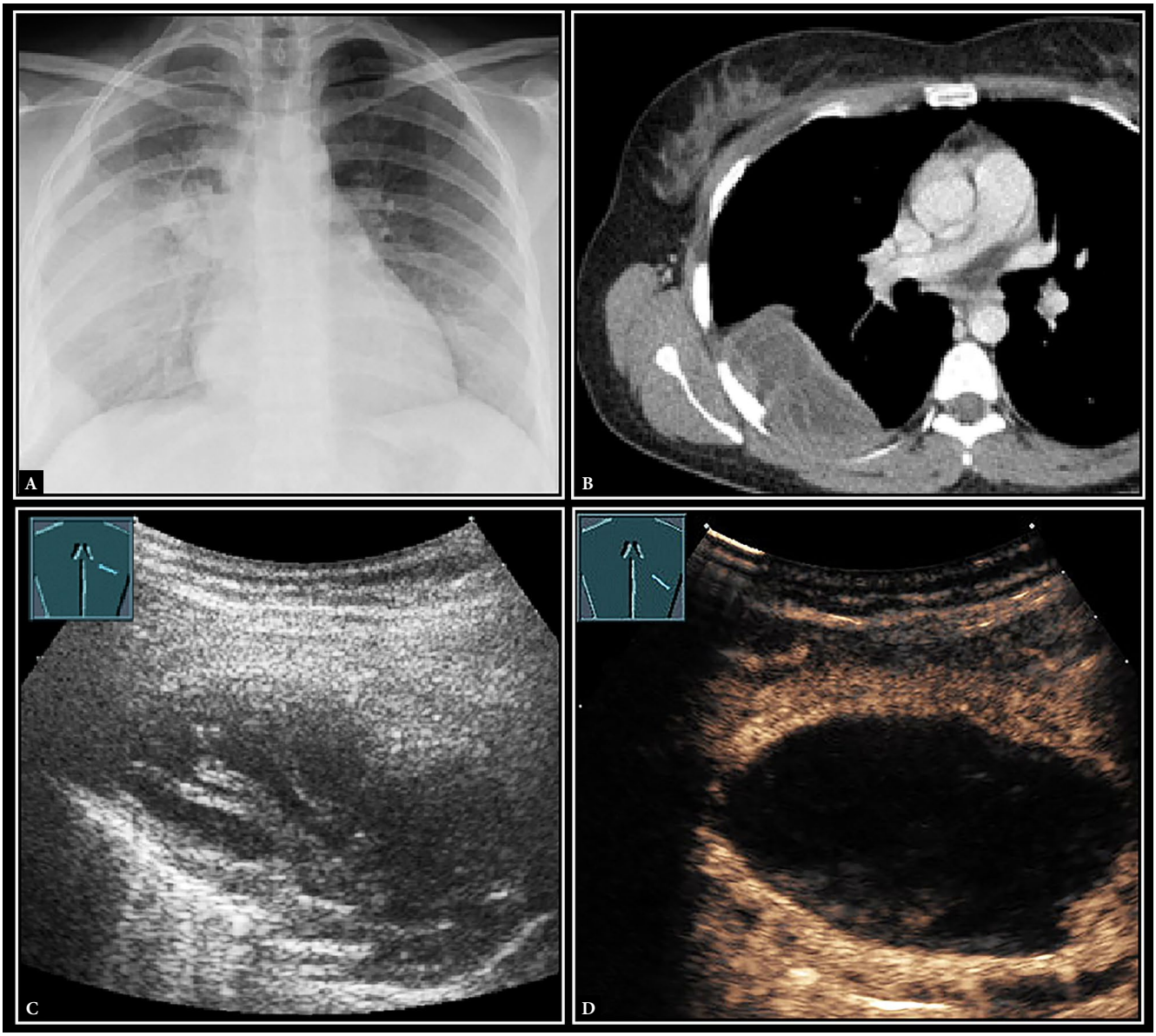

Fig. 1. A. *Corresponding X-ray shows right-sided pulmonary shadowing.* **B.** *CT of the chest demonstrates the right dorsal lung consolidation with cystic and septated structures (courtesy of Prof. Dr. A. Mahnken, Department of Diagnostic and Interventional Radiology at Marburg University Hospital).* **C.** *On B-mode US, an echocomplex, septated, cystic lung consolidation in the rightdorsal region was seen, corresponding to stage CE2.* **D.** *On CEUS, homogeneous, marked bronchial arterial enhancement in the periphery of the consolidation was seen after 12 s, with complete non-perfused central area*

Case report

A 26-year-old and otherwise-healthy female was admitted with subacute respiratory symptoms to an external hospital. The diagnosis of community-acquired pneumonia was made, and antibiotic treatment was established. Her condition failed to improve, so further diagnostic workup was performed. The woman had no preexisting conditions. Originally from Iraq, she had been living in Germany for several years. The CT scan of the chest showed three cystic lesions with contact to the pleura (Fig. 1B).

Thus, serological testing was performed, and treatment with albendazole and voriconazole was initiated. The patient was discharged after symptom relief, with no definitive diagnosis, but presented again, only a few days later, with increasing complaints, to another hospital. Pleural paracentesis was performed on the suspicion of a pulmonary abscess. A small amount of transparent fluid was drained, but the procedure resulted in an anaphylactic reaction. The patient was stabilized and transferred to our university hospital with suspected pulmonary echinococcosis.

On arrival, the patient showed normal vital parameters, besides breathing 2l of oxygen via nasal cannula. The physical examination was unremarkable, and laboratory results demonstrated slightly elevated inflammation markers, and mild normochromic normocytic anemia.

Ultrasound examination was performed (Siemens, ACUSON Sequoia 512, USA) with a convex probe (4C1). In B-mode US, some oval-shaped consolidations were visualized in the right lung. The most prominent lesion in the right upper-posterior region of the lung

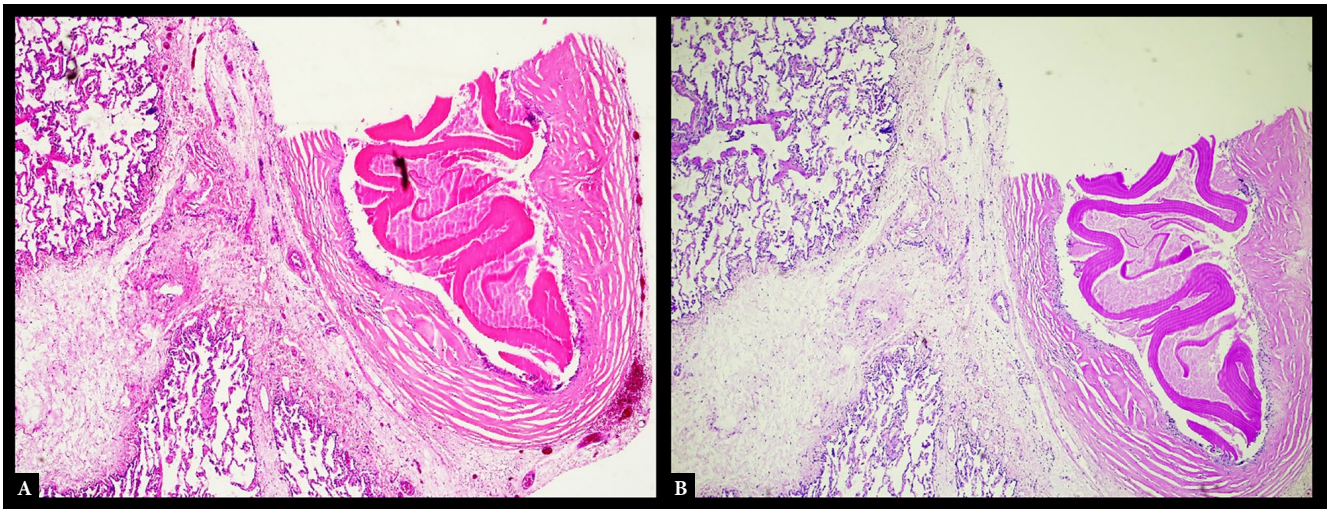

Fig. 2. *Atypical partial lung excision specimen of the right medial lobe, with a 0.2 cm gray-yellow nodal lesion, on histopathological examination (HE staining* **A***), PAS staining,* **B.** *representing a cystic structure (40×: to the right), adjacent to regular lung tissue (40×: to the left), containing PAS-p vositive cuticula, characterizing an echinococcal cyst, surrounded by fibrotic tissue. Protoscolices were not present*

measured 6 × 5 × 2.5 cm and showed a complex pattern, with cystic and septated structures within the lesions (Stage CE2) (Fig. 1C).

A smaller consolidation in the right costodiaphragmatic recess was cystic. The abdominal organs were unremarkable. Contrast-enhanced ultrasound (CEUS) examination was recommended and performed according to guidelines[3]. CEUS showed marked rim-like peripheral bronchial arterial enhancement (time to enhancement: 12 s), with sparing of the central region in both lung consolidations (Fig. 1D).

Positive serological testing for *E. granularis* (enzyme-linked immunosorbent assay (ELISA) and indirect hemagglutination (IHA)) additionally confirmed the diagnosis of pulmonary echinococcosis, so the patient's albendazole treatment was continued. An atypical surgical resection and cyst drainage were performed after two weeks (Fig. 2). Due to the intraoperative findings with pleural dissemination probably caused by the preceding, pleuro-pneumonectomy was recommended after further medical treatment in due course.

Discussion

Even though according to the WHO Informal Working Group on Echinococcosis (WHO-IGWE), ultrasound is the preferred tool for screening and diagnosing hepatic CE[4], pulmonary manifestation is not always detectable by US because of known physical limitations of lung US. Nevertheless, it was shown that B-mode US was able to detect and diagnose pulmonary CE with high specificity by using the wall sign[2]. A previous study has already highlighted the strength of B-mode US in comparison to CT and MRI in the staging of hepatic CE[5].

This observation is critical in clinical decision-making, especially in regions where the diagnostic workup is limited. In addition, in children the disease manifests more often in the lungs than in the liver, so radiation exposure should be kept as low as reasonably achievable[6]. The differential diagnosis of pulmonary CE encompasses solid and cystic conditions, such as primary or secondary lung carcinoma, benign tumors, inflammatory lesions, and lung cysts[7].

Our case showed a bronchial arterial, peripheral, annular enhancement of the cystic lesions, with complete absence of enhancement in the inner parts. Due to MRI and CT imaging, we know that the pericyst is perfused by contrast media agent[8,9]. Some authors interpret marked peripheral enhancement as a radiological sign of a superinfected cyst or abscess[10], which occurs in up to 7% of cases, while others report no additional diagnostic benefit[9].

Despite the marked annular contrast enhancement by CEUS, the presented case did not show any superinfection. Therefore, in cases of pulmonary CE, annular enhancement by CEUS should not be overinterpreted, and a cyst puncture should be indicated very cautiously because of possible adverse events (as seen in the present case). *E. granulosus* triggers an inflammatory host response in which a pericyst forms with an infiltrate of inflammatory cells, which may be the reason for the marked enhancement. Moreover, the perfusion might depend on the level of fibrosis. Therefore, CEUS should be studied in more detail, using a larger number of cases of pulmonary CE, to determine its diagnostic value.

In summary, in the reported case of hydatid disease, CEUS showed marked contrast enhancement of the pericyst, which should not be interpreted as a sign of superinfection or abscess, but rather as highlighting the wall sign.

Conflict of interest

The authors do not report any financial or personal connections with other persons or organizations which might negatively affect the contents of this publication and/or claim authorship rights to this publication.

Author contributions

Original concept of study: HF, CG. Writing of manuscript: HF. Final approval of manuscript: HF, CG, ESZ. Collection, recording and/or compilation of data: HF, CW. Critical review of manuscript: CT, CG, JK, ESZ.

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