C A S E R E P O R T

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Pleural hydatidosis secondary to intrapulmonary echinococcosis

Pleuralna hidatidoza kao posledica intrapulmonalne ehinokokoze

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Abstract

Introduction. Hydatid cysts are most often found in the liver and lungs, while other locations are rarely involved. Pleural hydatidosis, whether primary or secondary, is rarely seen in clinical practice. **Case report.** In this article, we present a case of a 33-year-old woman who presented with cough and dyspnea. Initial chest X-ray showed a large right-sided pleural effusion. Pleural hydatidosis was suspected based on the findings of the computed tomography scan of the chest. Right thoracotomy and cystectomy were performed. Albendazole was administered postoperatively for 6 months, and during this period, liver function tests and abdominal ultrasonography showed no abnormalities. **Conclusion.** This case emphasizes that pleural hydatidosis should be suspected in patients with pleural effusions and large cystic formations found on imaging, especially in endemic areas.

Keywords:
albendazole; diagnosis; echinococcosis; montenegro; pleura; tomography, x-ray computed.

Apstrakt

Uvod. Hidatidne ciste se najčešće nalaze u jetri i plućima, dok su druga mesta retko zastupljena. Pleuralna hidatidoza, bilo primarna ili sekundarna, retko se viđa u kliničkoj praksi. **Prikaz bolesnika.** U ovom radu prikazana je bolesnica stara 33 godine, koja se javila na pregled zbog kašlja i otežanog disanja. Prvi rendgenski snimak grudnog koša pokazao je veliki pleuralni izliv sa desne strane. Na pleuralnu hidatidozu posumnjalo se na osnovu nalaza kompjuterizovane tomografije grudnog koša. Na desnoj strani grudnog koša izvršene su torakotomija i cistektomija. Albendazol je primenjivan postoperativno tokom šest meseci, a tokom tog perioda testovima za ispitivanje funkcije jetre i ultrazvukom abdomena nisu utvrđene abnormalnosti. **Zaključak.** Ovaj slučaj naglašava da na pleuralnu hidatidozu treba posumnjati kod bolesnika sa pleuralnim izlivima i velikim cističnim formacijama uočenim na radiološkim snimcima, naročito u endemskim područjima.

Ključne reči:
albendazol; dijagnoza; ehinokokoza; crna gora; pleura; tomografija, kompjuterizovana, rendgenska.

Introduction

Hydatidosis is an endemic parasitic disease most often caused by *Echinococcus* (*E.*) *granulosus* [1]. People accidentally become infected by consuming contaminated food or through direct contact with a diseased host [2, 3]. The most commonly affected organs are the liver and lungs, but the parasite can be found in almost any organ in the body [4]. Pleural involvement is of rare occurrence. Primary pleural echinococcosis is an exceptionally rare entity within the spectrum of human cystic echinococcosis (less than 1%) [5]. In clinical practice, it is imperative to clearly distinguish between the primary and secondary forms of the disease to eliminate diagnostic ambiguity. Unlike secondary pleural echinococcosis, which occurs as a complication following the rupture of fertile pulmonary or hepatic hydatid cysts (HCs) and the subsequent release of their contents into the pleural space, the primary form involves the direct development of cysts within the pleural cavity itself [5–7]. In this manifestation, the echinococcal development occurs *de novo*, without prior involvement or concurrent pathological changes in the lung or liver parenchyma. This pathophysiological distinction is essential for understanding the pathogenesis of the presented case and underscores its unique clinical significance. HCs are often accidentally detected during radiological examinations. Spontaneous or traumatic cyst rupture most of the time leads to symptoms such as cough, fever, chest pain, dyspnea, and rarely anaphylactic shock [8].

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We present a case of a patient with pleural hydatidosis, which occurred as a complication of spontaneous cyst rupture into the pleural cavity.

Case report

A 33-year-old female patient from the Roma population of the central region of Montenegro was admitted to the Department of Thoracic Surgery, Clinical Center of Montenegro, Podgorica, Montenegro, due to progressive dyspnea lasting one year prior to hospitalization and a persistent non-productive cough lasting 6 months prior to hospitalization. The patient was an active smoker (3.5 pack-years) with no prior comorbidities. Upon admission, physical examination of the chest revealed dullness to percussion and the absence of breath sounds on the right side. Laboratory findings were as follows: erythrocytes 4.47 × 10^{12}/L [reference range (RR): 3.90–5.20 × 10^{12}/L], hemoglobin 139 g/L (RR: 120–153 g/L), leukocytes 23.84 × 10^{9}/L (RR: 3.7–10.0 × 10^{9}/L), neutrophils 22.23 × 10^{9}/L (RR: 1.60–5.80 × 10^{9}/L), eosinophilic granulocytes 0.02 × 10^{9}/L (RR: 0.02–0.50 × 10^{9}/L), platelet count 257 × 10^{9}/L (RR: 150–410 × 10^{9}/L), erythrocyte sedimentation rate 68 mm/h [normal value (NV) < 20 mm/h], glucose 5.5 mmol/L (RR: 4.6–6.4 mmol/L), urea 1.9 mmol/L (RR: 3.5–7.2 mmol/L), creatinine 46 μmol/L (RR: 44–80 μmol/L), aspartate aminotransferase 12 U/L (NV: < 31 U/L), alanine aminotransferase 10 U/L (NV: < 33 U/L), lactate dehydrogenase 138 U/L (RR: 135–225 U/L), creatine kinase 43 U/L (NV: < 190 U/L), C-reactive protein 117.91 mg/dL (NV: < 5 mg/dL).

Serological testing [enzyme-linked immunosorbent assay (ELISA) echinococcus immunoglobulin (Ig) G > 15 U/mL] was positive (NV: negative < 9, equivocal 9–11, positive > 11 U/mL), confirming the suspicion of a parasitic etiology. Forced spirometry: forced vital capacity (FVC) 1.58 L (47%) (NV: 3.36 L; ≥ 80%), forced expiratory volume in 1 sec (FEV1) 1.39 L (48%) (NV: 2.90 L; ≥ 80%), which indicated severe damage to lung function.

The bronchoscopy finding indicated that the vocal cords, trachea, and main carina appeared healthy (normal). Except for signs of extramural compression in the right principal and the *bronchus intermedius*, starting from the main carina, which was deviated to the left, the rest of the endoscopic findings were normal.

An initial chest X-ray and then a computed tomography (CT) scan were performed three weeks before hospitalization. Chest X-ray showed uniform opacity of the right hemithorax with a leftward mediastinal shift, indicating a massive pleural effusion (Figure 1). A subsequent contrast-enhanced multislice CT (MSCT) scan of the chest (Figure 2a, b) revealed a complex radiological presentation: a small pleural effusion 10 mm in the right pleural cavity containing multiple cystic formations. Additionally, three oval cystic formations were visualized within the parenchyma of the upper and middle lobes of the right lung. The largest cyst (6 cm in diameter) was located in the upper lobe, and a smaller one (4.5 cm) in the middle lobe was in direct contact with the visceral pleura. The liver was markedly enlarged, with a 5.5 × 4 cm cystic lesion in segment VI (Figure 3).

Abdominal ultrasound indicated the presence of a cyst in segment VI of the liver (6 × 4 cm), which was drained percutaneously at the same time, followed by sclerosing with ethoxysclerol. The aspirate samples were sent for microbiological and serological testing. Given the extent of the lesions, surgical exploration *via* right lateral thoracotomy was indicated. Intraoperatively, a small serous pleural effusion was identified (biochemical profiling was consistent with exudate, and cytology was negative for malignant cells).

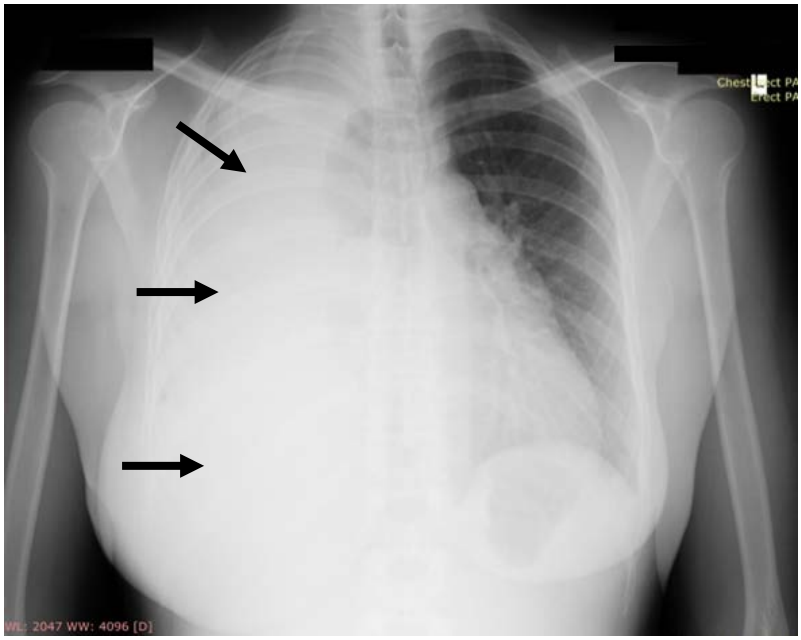

Fig. 1 – Chest X-ray showing uniform opacity of the right hemithorax (arrows).

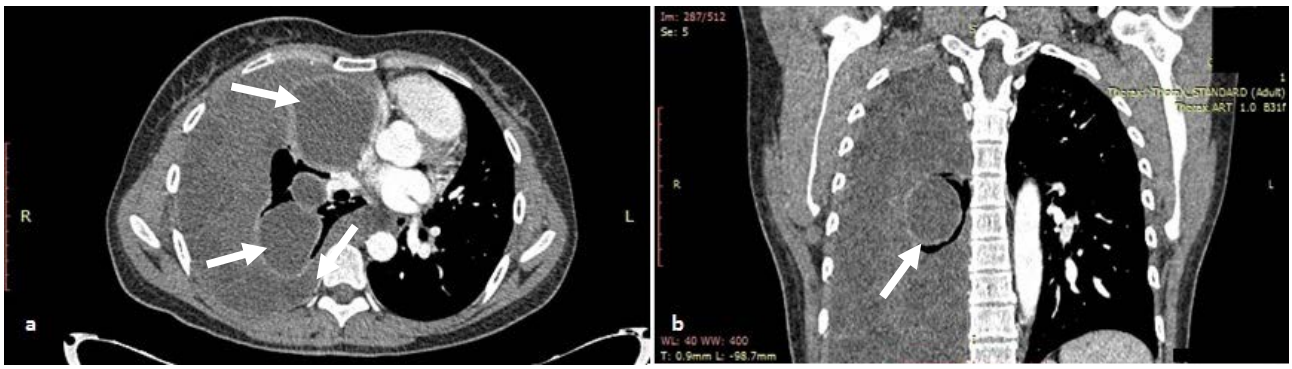

Fig. 2 – Chest computed tomography scan: a) axial view and b) coronal view, showing cystic formations in the right pleural space and in the upper and middle lobes of the right lung (arrows).

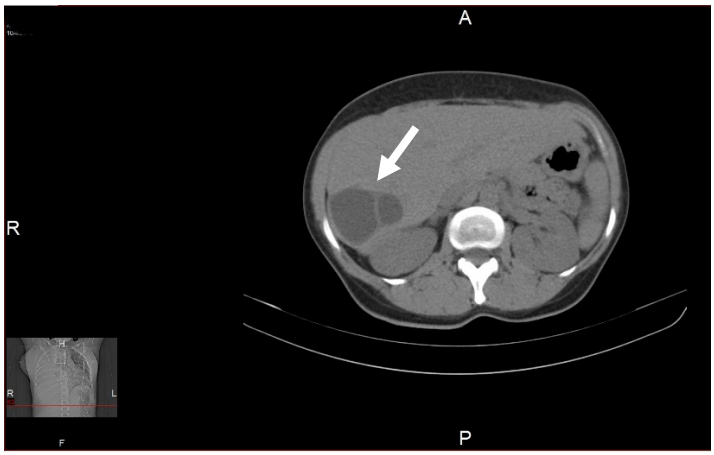

Fig. 3 – Chest computed tomography and upper abdomen scan showing cystic formation in segment VI of the liver (arrow).

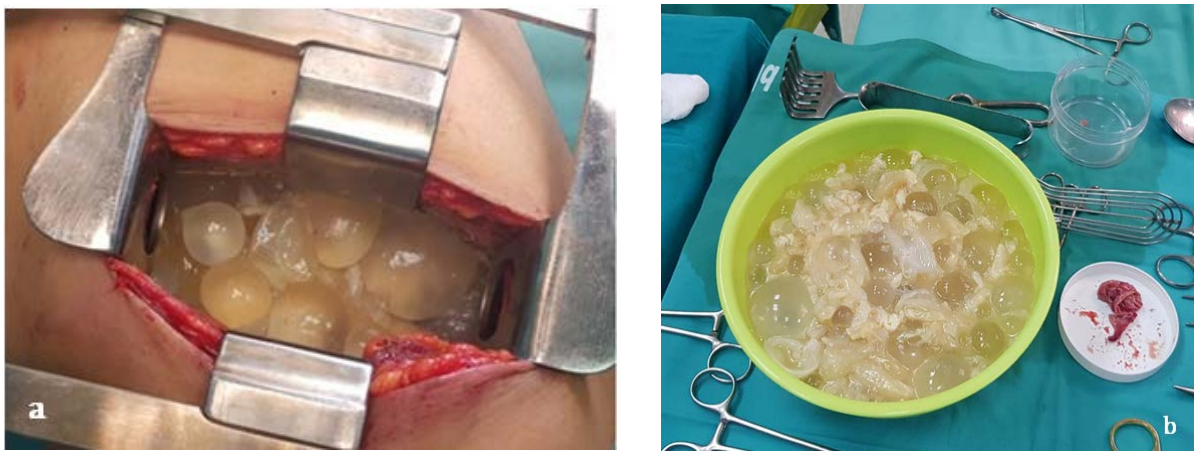

Fig. 4 – Open lateral thoracotomy showing numerous hydatid cysts: a) within the pleural space and b) evacuated from the pleural space.

Multiple intact, firm cystic formations (over 150) were found within the pleural space (Figure 4a, b) and were subsequently evacuated (Figure 5). A thorough inspection of the lung identified three intraparenchymal cysts in the upper and middle lobes. The largest cyst in the upper lobe showed evidence of prior rupture into the pleural space, with several cystobronchial fistulas, which were repaired using 4-0 polydioxanone sutures. Endocystectomy, partial pericystectomy, and lung decortication were performed. Histopathological analysis confirmed the presence of fertile elements (scolices). The postoperative recovery was uneventful. Adjuvant therapy with albendazole was initiated at a dose of 400 mg twice daily for 28 days, followed by a 14-day drug-free interval, for a total of six

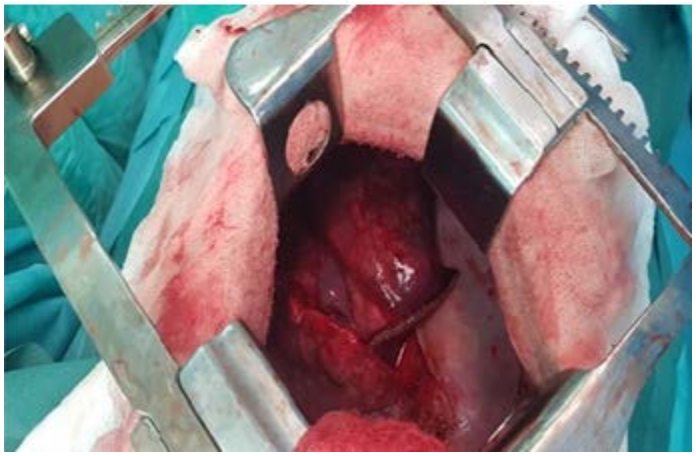

Fig. 5 – Open lateral thoracotomy after removal of hydatid cysts and capitonnage.

cycles.During the treatment period, liver function tests and abdominal ultrasonography were performed monthly. The initially described liver cysts (type CE1, up to 6 × 4 cm) showed regression and transitioned to inactive stages (types CE4/CE5) after 6 months of therapy. A follow-up chest MSCT scan at 6 months demonstrated complete lung re-expansion with no signs of disease recurrence.

Discussion

Hydatid disease is a parasitic disease that usually occurs in younger patients and is most often caused by *E. granulosus* [9]. Echinococcosis is a zoonosis with two hosts, a transitional host (e.g., sheep) and a definitive host (e.g., dogs) [2]. The life cycle of *E. granulosus* does not necessarily involve humans. People are accidentally infected by eating contaminated food or by direct contact with an infected host [2, 3]. HCs usually involve the liver or lungs, but simultaneous involvement of both organs has also been described [4, 10, 11]. After being introduced into the organism orally, the embryo enters the portal circulation. It is carried by the bloodstream to the liver, which is, in most cases, the first destination of the parasite [6]. The possibility of hematogenous or lymphogenous dissemination means that the parasite can be found in almost any other organ. By entering the lymphatic vessels of the stomach, the parasite can reach the thoracic lymphatic duct and so directly travel into the pleura, resulting in primary pleural hydatidosis, which is very rare (< 1%) [11, 12]. Secondary pleural hydatidosis is most often caused by transdiaphragmatic contamination of the pleural space by cysts located in the right upper lobe of the liver or by rupturing of the peripheral lung cysts, and can manifest as pleural HCs or as a parasitic pleural effusion, which is rare in clinical practice [2]. In both cases, HCs can survive and grow within the avascular layers of the pleura due to the specific structure of their membranes, which enable diffusion and active transport of substances necessary for metabolic processes [1]. HC of the lung that is close to the pleural surface can cause thinning of the adventitia and can rupture into the pleural cavity [11]. This may initially go clinically unnoticed, but over time, as the cyst grows, symptoms gradually appear. Pleural involvement, whether primary or secondary, is rarely seen in clinical practice [12]. Clinical manifestations are variable and depend on the size, location, and pressure of the cyst on the surrounding lung tissue [8]. The most common symptoms are cough, dyspnea, fever, and chest pain [8, 12, 13]. In some rare cases, HC perforation can clinically manifest as anaphylactic shock. The most common complications are pleural effusion, empyema, and pneumothorax [2]. In most cases of pleural effusion or pneumothorax, significant mediastinal compression and displacement are observed, while empyema, one of the most serious complications, is associated with bacterial infection of a perforated cyst [12]. The diagnosis of pleural hydatidosis is not always easy. Differential diagnosis includes benign tumors, inflammatory masses, pathological conditions with formation of aero-liquid levels in the lungs, inflammatory processes, metastatic diseases, and malignancies. A standard chest radiograph is usually used as the first diagnostic method [6]. The most valuable diagnostic method for the detection of pleural HCs is a chest CT scan. CT findings can range from typical images of intact cysts that appear as round, homogeneous, and dense, through simple pleural effusions, to complicated empyema, pneumothorax, or hydropneumothorax [6, 10]. Sometimes it is possible to see calcifications of the cyst wall referred to as “eggshell calcifications” [5]. A chest CT scan also plays an important role in surgery planning. Lung ultrasound is rarely used for diagnosing pleural HCs; however, it can sometimes provide valuable information, particularly in the presence of complex cystic lesions, and it is safe to use in pregnant women [13]. In addition to radiological methods, various serological tests (i.e., ELISA, latex agglutination, and indirect hemagglutination test), as well as pathological examinations, are used for diagnosing hydatidosis [10]. These tests can be used in postoperative monitoring of patients, and also for screening. Some tests (skin tests, complement fixation, eosinophil count) have a tendency towards false positive results, so one should be careful when interpreting the results. Recent research

suggests that interleukin-4 could be useful for the identification of active cysts [14]. Fine-needle aspiration of HCs is not recommended for the diagnosis of pleural hydatidosis due to the high risk of complications such as pneumothorax, cyst rupture, and anaphylaxis [2]. Definitive diagnosis is made by thoracotomy and direct visualization of the cysts [12]. Surgical removal of intact HCs is the treatment of choice. However, it is necessary to completely remove the cysts without opening them; otherwise, it could lead to intraoperative dissemination of the parasites [15]. Intraoperative administration of 1% formaldehyde or hypertonic saline can prevent that by deactivating the cysts [9]. Pericystectomy is a common procedure, but other procedures like simple drainage, capitonnage, marsupialization, or resection of the lung can also be used, depending on the location and condition of the cyst [15]. According to Yalçinkaya et al. [9], cystotomy and capitonnage of the residual cavity are the methods of choice. Aribas et al. [6] state that cystotomy with capitonnage or cystectomy with capitonnage are the most preferred types of operations. However, some authors believe that capitonnage is not necessary after cystectomy and that only bronchial openings should be sutured and the cavity left open, arguing that capitonnage causes lung parenchyma deformation, prolongs surgery time, and increases morbidity [2]. In most cases, decortication is also performed. Radical lung resections should only be performed when the lung parenchyma around the cyst is destroyed and/or the lungs cannot expand [6]. Recurrence rates of 2–12% have been reported in the literature, while the operative mortality rate is 0.5–4% [9, 12]. Most authors suggest pharmacological treatment after surgery [2, 6]. During the administration of these drugs, regular monitoring of laboratory findings is required. Some authors suggest that percutaneous therapy of pulmonary hydatid disease is an effective alternative to surgical treatment in patients who have failed medical therapy [16, 17].

Conclusion

Hydatid cysts most commonly affect the liver and the lungs, while other locations are rarely involved. Pleural hydatidosis, although rare, most often occurs as a complication of rupture of pulmonary hydatid cysts. The diagnosis is based on radiological findings and serological tests, but intraoperative detection of the cyst is crucial. The treatment modality with the greatest therapeutic potential is surgical removal of the cysts. In countries where hydatid disease is endemic, prevention is of utmost importance. Our case highlights the importance of detailed radiological and intraoperative evaluation in distinguishing between the forms of pleural echinococcosis. Timely surgical intervention, supplemented by an adequate dose of albendazole, ensures a complete cure and prevents serious complications such as empyema or anaphylactic shock.

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