

An Unusual Cause of Diastolic Restriction and Pulmonary Hypertension: Pulmonary Hydatid Cyst Associated to Pulmonary Embolism- A Rare Association

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Abstract

Pulmonary hydatid disease, attributable to *Echinococcus granulosus**, is prevalent in various regions, notably among rural populations in Mediterranean countries. The condition frequently manifests with respiratory symptoms, but can also result in systemic issues, particularly when cysts exert pressure on neighboring structures, including the heart. This report examines an uncommon correlation between pulmonary hydatid cysts, pulmonary embolism, diastolic dysfunction, and pulmonary hypertension.

Keywords: pulmonary hypertension; pulmonary embolism; diastolic dysfunction; respiratory symptoms

Introduction

A 57-year-old woman from a rural community with a documented history of pulmonary hydatid disease presented to our clinic with chronic dyspnea, persistent cough, unintentional weight loss, and clinical indications of right ventricular dysfunction. Imaging studies, comprising chest X-ray, CT scan, and transthoracic echocardiography, identified bilateral pulmonary embolism alongside substantial mediastinal hydatid cysts that were compressing the heart. These cysts induced hemodynamic alterations, characterized by increased pulmonary pressures and diastolic dysfunction.

Management and Outcome:

The patient was started on anticoagulation therapy with Apixaban to address the pulmonary embolism, and albendazole was recommended for the management of the hydatid cysts. Unfortunately, despite the proposed medical treatments, the patient refused surgical intervention and sadly passed away one month following the diagnosis.

This case underscores the rare and intricate relationship between pulmonary hydatid cysts and pulmonary embolism, which can lead to significant cardiovascular implications, including diastolic dysfunction and pulmonary hypertension. Timely diagnosis and suitable management—especially surgical options when feasible—are essential for enhancing patient prognosis. This case underscores the necessity of considering hydatid disease in the differential diagnosis of pulmonary embolism, particularly in endemic regions.

Human hydatid disease is endemic in Morocco and remains a health concern problem in Mediterranean countries. It occurs when the tapeworm *Echinococcus* develops through its metacystode stage to form larvae. The

majority of human infections are caused by *E. granulosus* which leads to cystic echinococcosis [1].

The primary hosts of the worm include dogs, wolves and coyotes, while sheep, cattle, and deer are considered as intermediate hosts. Humans can become infected through exposure to contaminated water, food, and direct contact with carnivorous animals: however, they do not participate in worm's biological cycle. Residing in rural area is a significant risk factor that warrants further investigation.

The initial type is primary echinococcosis, which can manifest in any organ; the second type is secondary echinococcosis, characterized by the dissemination of metacystodes from primary locations to other areas [1].

The lung is the second organ most frequently affected, following the liver [2]. Intrathoracic extrapulmonary localizations may involve various structures, including the pleura, pericardium, diaphragm, and chest wall. [3].

Hydatid cysts can also be located in other locations such as the kidneys and spleen, which occurs in approximately 2% of cases [4]. Additionally, bones, muscles and the brain may also be affected by the infection.

The diagnosis of hydatid disease relies on characteristic clinical and radiological findings.

In this report, we present a case of a pulmonary hydatid cyst that led to compressive cardiac tamponade, accompanied by pulmonary embolism which resulted in diastolic restriction and pulmonary hypertension. We aim to elucidate the clinical and imaging manifestations with this condition.

Case Presentation

We present the case of a 57-year-old woman residing in a rural area. Her medical history includes a pulmonary hydatid infection that occurred twenty years prior, which was managed medically with a positive outcome.

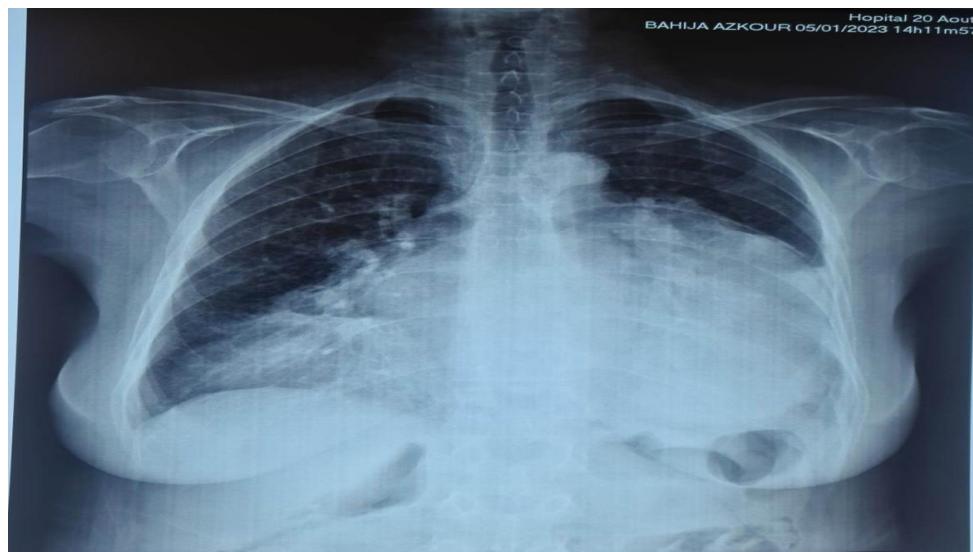
The patient was admitted to our hospital presenting respiratory symptoms that included chronic dyspnea, a cough accompanied by hydatidoptysis, basithoracic pain, and a sensation of heaviness. Additionally, she lost 4 kg of her weight over the course of one-month, nocturnal sweats, asthenia and significant severe anorexia.

Upon clinical assessment, the patient presented as hemodynamically fairly stable. However cardiovascular examination revealed signs of right

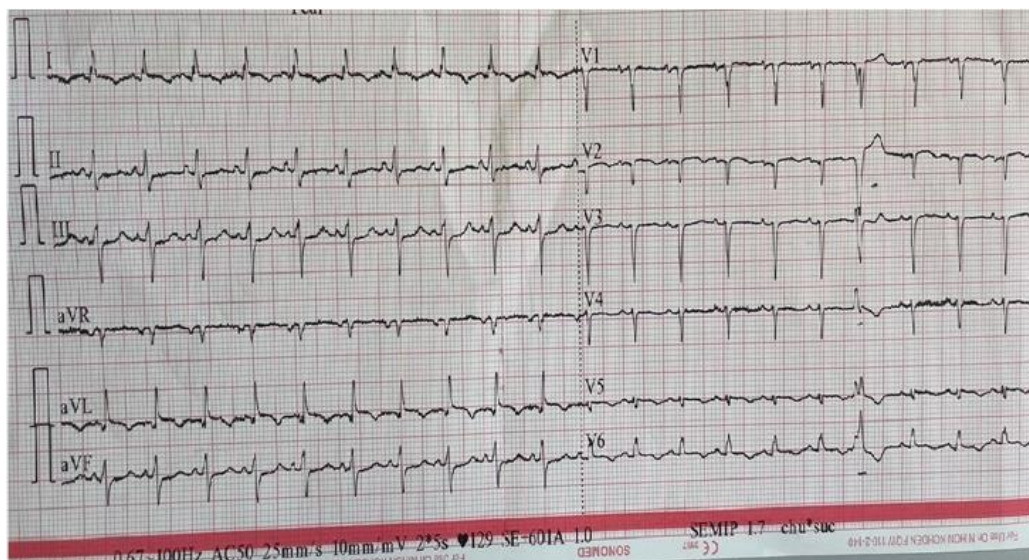
ventricular dysfunction, along side notable finding of painful hepatomegaly comprising 35 finger spans, hepatojugular reflux and lower extremity edema. The pleuropulmonary examination indicated the presence of sub-crepitations in the left basithoracic region.

The routine hematological examination returned normal results; however, the D-dimer assay indicated elevated levels.

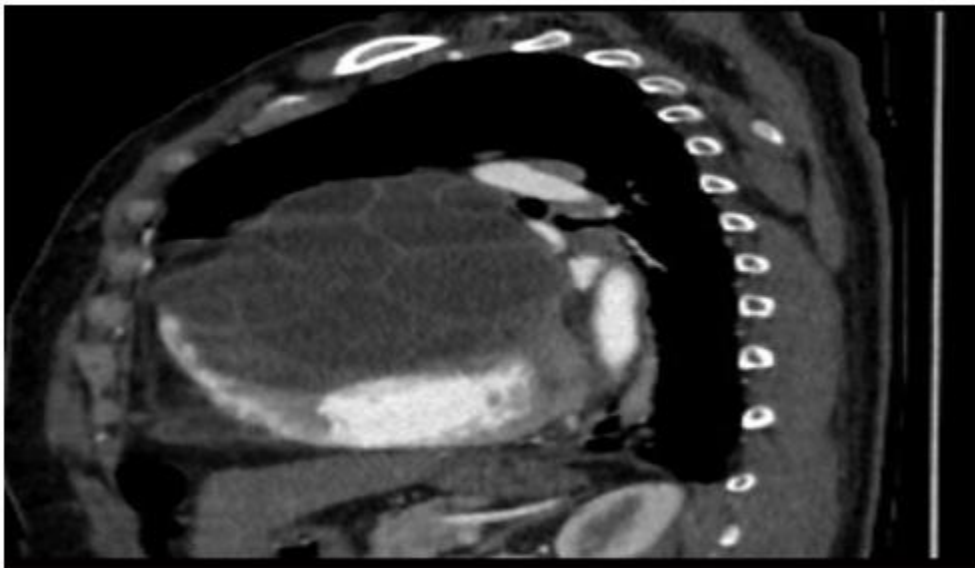
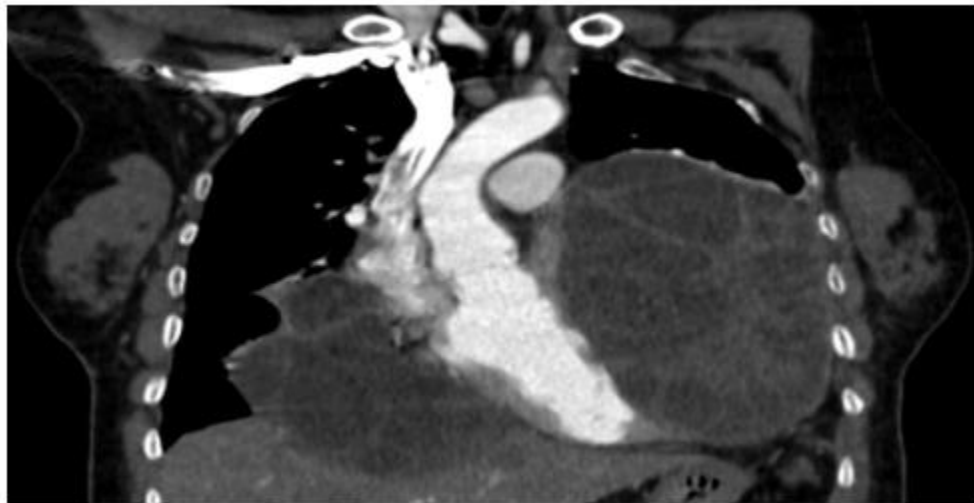
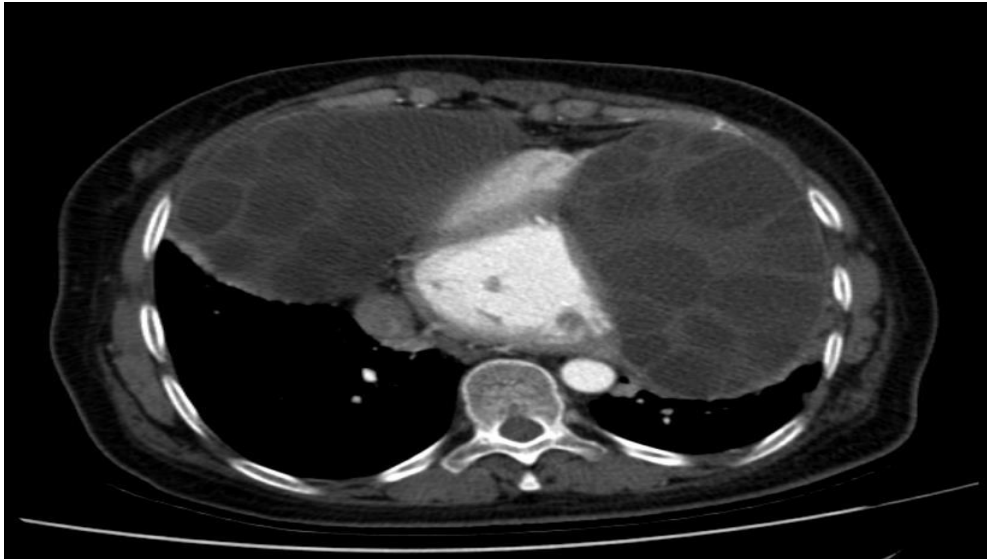
A chest X-ray revealed a significant rounded opacity in the left hemithorax, which obscured the outline of both the left mediastinal border and the ipsilateral diaphragmatic dome, consistent with the silhouette sign and indicating its antero-inferior positioning. Additionally a similar opacity was identified in the right lower hemithorax. (Figure 1).

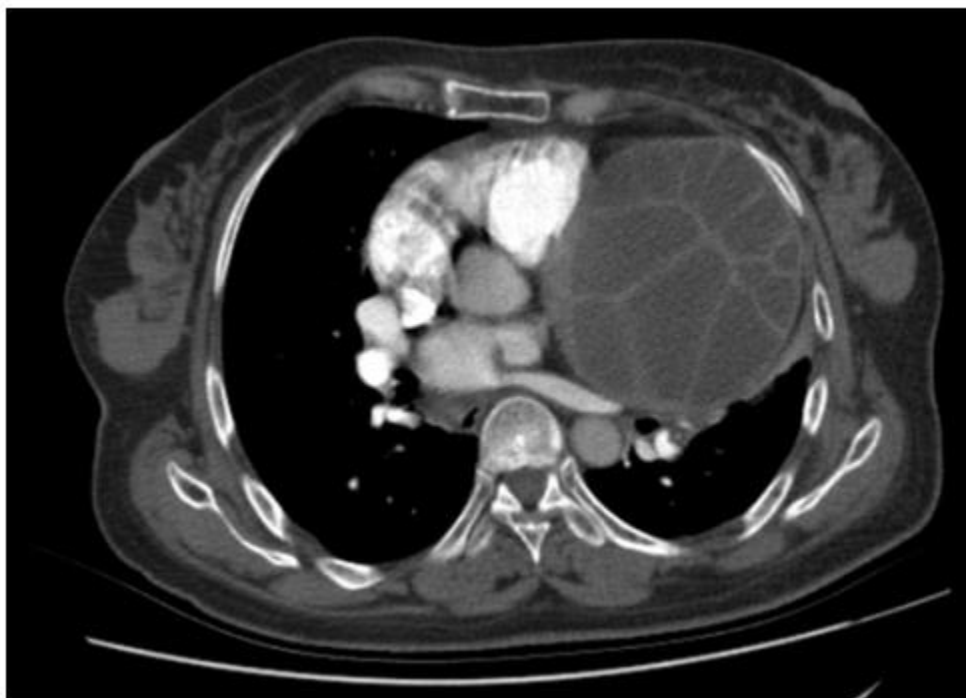
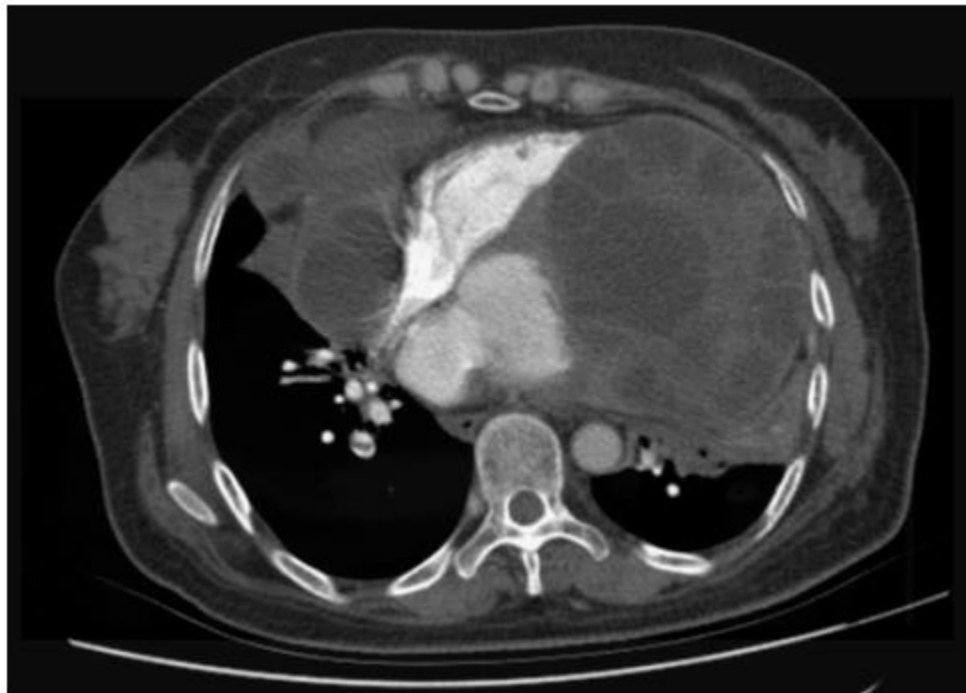


On a 12-lead electrocardiogram (ECG), sinus tachycardia accompanied by a secondary repolarization disorder was observed, along with the presence of ventricular premature complexes (Figure 2)



A computed tomography (CT) chest scan revealed (Figures 3-7):





- Evidence of bilateral lobar and segmental pulmonary embolism on the right in addition to segmental pulmonary embolism localized to the left side. The seembolic changes exhibited a chronic appearance without any indication of pulmonary parenchymal infarction. (Figures 6-7). These finding scorrelated with the observed increase in D- dimer levels.

- 2 large bilater al anterior mediastinal and para- cardiac formations, likely of hydatid origin, are causing extrinsic compression of the cardiaccavities (Figures 3 – 5):

The structures in question are characterized by multi-walls, a regular outline, distinct margins, and an enhanced wall definition following the injection of contrast medium.

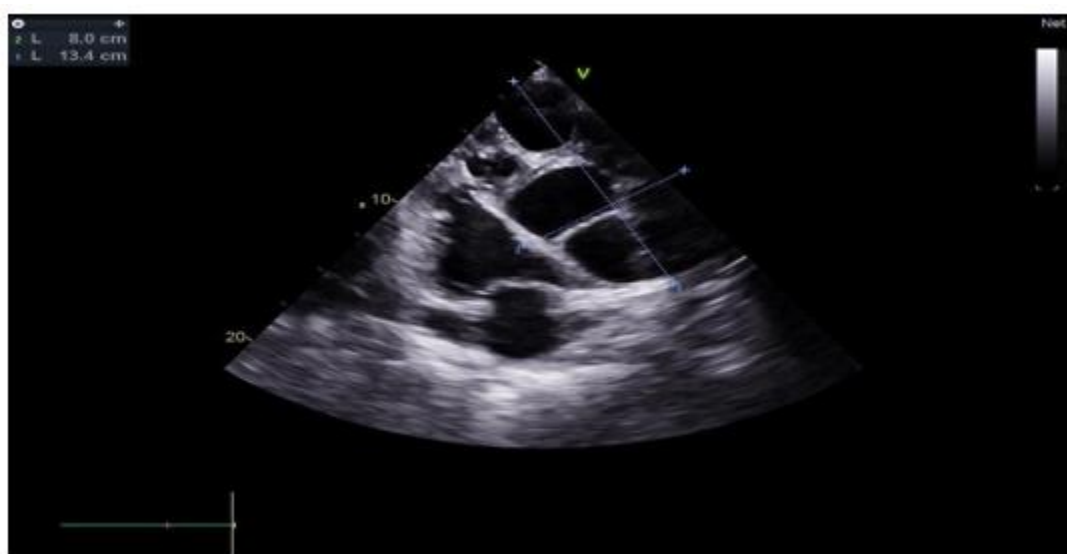
The dimensions on the right are 93 x 114 mm and extend over 119 mm; while on the left, the measurements are 119 x 78 mm extending over 140 mm.

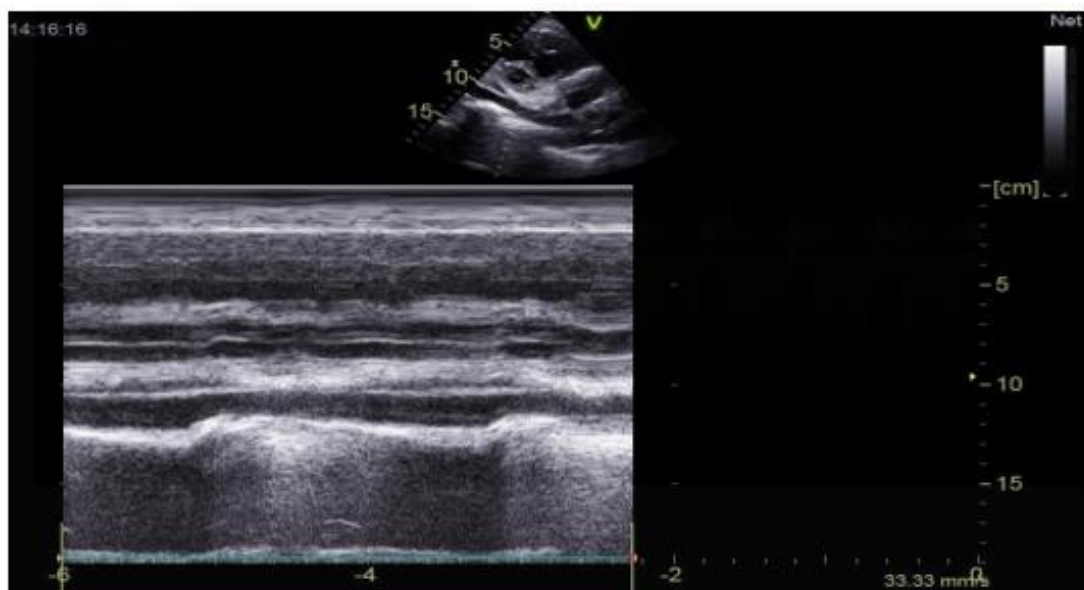
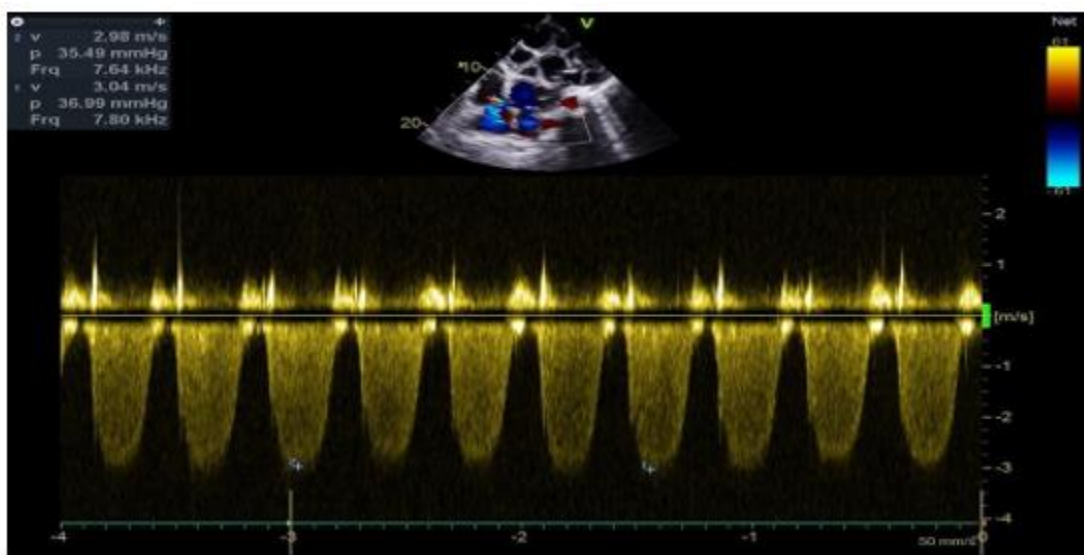
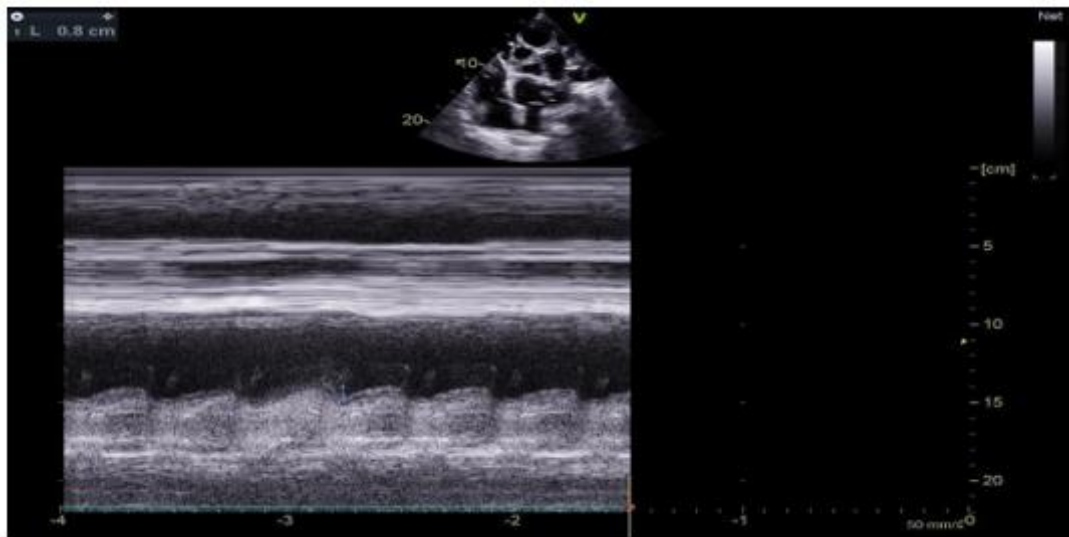
Posteriorly, these structures exert compression on the adjacent lung parenchyma resulting in passive collapse.

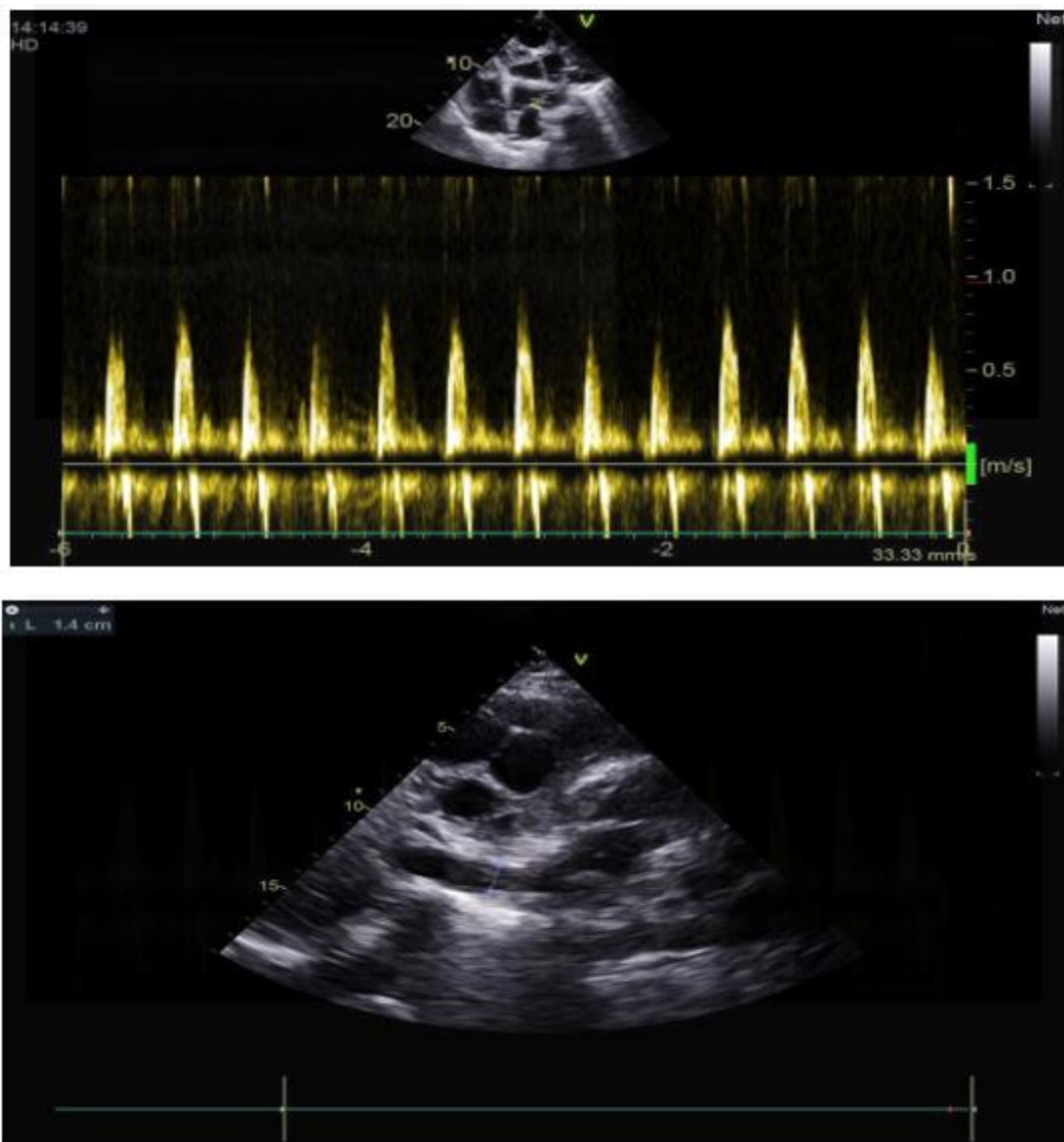
- Absence of pleural effusion.

The transthoracic echocardiographic examination revealed (Figures 8 – 18):









- A lobulated cystic image exerting severe compression on both the right and left cardiac chambers, resulting in hemodynamic disturbance and considerable fluctuation in respiratory flows (Figures 12 – 13,17).
- Left ventricle showed no sign of dilatation. Although the assessment of contractile function was influenced by compression from pulmonary hydatid cyst, it appears to maintain functionality.
- Elevated left ventricular filling pressure were noted.
- The right ventricle exhibited no dilation (Figure 11) despite the presence of longitudinal systolic dysfunction (Figures 14).
- Grade II tricuspid insufficiency was identified, with an estimated systolic pulmonary arterial pressure of 47 mmHg (calculated as $37 + 10$ mmHg), indicating the presence of pulmonary hypertension.
- The inferior vena cava demonstrated no dilation (Figure 18) and was noted to be non-compliant (Figure 16).
- No pericardial effusion was present.

No additional localized findings were identified during the abdominal ultrasound examination.

We maintained the diagnosis of cardiac pre-tamponade secondary to a large pulmonary hydatid which led to an a diastolism alongside pulmonary embolism.

The patient's respiratory and haemodynamic conditions were monitored, and she was initiated on anticoagulant therapy with APIXABAN.

The Surgical intervention was presented as a critical option to both the patient and her family; however, they ultimately decided against undergoing the procedure. Consequently, we suggested a medical treatment regimen including ALBENDAZOLE, despite the fact that the hydatid cyst had not ruptured.

Regrettably, the patient passed away one month following the diagnosis.

All procedures performed in this study were in accordance the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013).

Discussion

Hydatid disease is a widespread zoonotic infection that remains a significant public health concern, particularly in rural regions. It is primarily caused by the parasites *Echinococcus granulosus* and *Echinococcus multilocularis*. This infection is endemic in various geographical areas, including the Mediterranean region, Africa, South America, the Middle East, Australia and New-Zealand. The most prevalent manifestation of hydatidosis in humans is cystic echinococcosis (CE). In this transmission cycle, dogs serve as definitive hosts while sheep act as intermediate hosts.

The liver serves as the primary line of defense in humans, accounting for involvement in approximately 75 % of cases. In contrast, the lungs are

implicated in 15% of cases, as was observed in our patient, while other sites of involvement make up the remaining 10% [2].

Literature indicates that mediastinal localization predominantly occurs in the posterior mediastinum, with anterior and upper mediastinal presentations being comparatively rare. Notably, this type of localization tends to progress to calcification more frequently than pulmonary hydatid cysts, primarily due to the lack of ventilation-related factors [5].

This condition is predominantly asymptomatic; however, it can present with a range of symptoms due to its compressive impact on nearby vital structures. Common manifestations include dyspnea, retrosternal chest pain, cough, dysphagia, back pain, and superior vena cava syndrome [6]. In the case of our patient, the symptoms were primarily a result of compression on the cardiac chambers.

The patient exhibited pulmonary hypertension as a result of pulmonary embolism and diastolic dysfunction due to compression from a hydatid cyst, despite such occurrences being exceptionally linked to *Echinococcus granulosus* infection. Symptoms such as cough, dyspnea, dysphagia, and chest pain are frequently documented in similar cases, mirroring the presentation of our patient [7].

The most significant complications include invasion of the aorta and the potential onset of anaphylactic shock resulting from an allergic reaction to the cyst [8].

Hydatid serology can offer valuable diagnostic insights in numerous cases and facilitate the assessment of treatment effectiveness [9].

Imaging techniques such as echocardiography, computed tomography (CT), and magnetic resonance imaging (MRI) are effective in visualizing the cystic characteristics of the mass and its relationship with the cardiac chambers.

Hydatid cysts are categorized into four distinct types based on their imaging characteristics:

- **Type I (Simple cysts):** These appear as well-defined, anechoic masses on ultrasound, which may contain hydatids and/or septa. On computed tomography (CT), they display homogeneous fluid attenuation, while magnetic resonance imaging (MRI) shows low signal on T1-weighted images and high signal on T2-weighted images, typically accompanied by a dark rim on both imaging sequences.
- **Type II (Cysts with daughter cysts and matrix):** Daughter cysts manifest as smaller cystic formations, usually located at the periphery of the primary cyst. These have a lower attenuation on CT and are iso- or hypointense compared to the matrix. In certain instances, larger, irregularly shaped daughter cysts can dominate the interior of the primary cyst.
- **Type III (Calcified/Dead cysts):** These cysts show echogenic calcifications on ultrasound, which are characterized by posterior shadowing. On CT, they are hyperattenuating, and on MRI, they appear hypointense.
- **Type IV (Complicated cysts):** Complications of hydatid cysts may arise from rupture due to degeneration, intervention, trauma, or superimposed infection. In cases of contained rupture, floating membranes can be observed as serpentine structures with low attenuation on CT and low signal on MRI, commonly referred to as the 'Water-lily sign'. The presence of intra-cystic air may indicate a communicating rupture and/or super infection.
- An abdominal ultrasound is a systematic procedure with sensitivity rates ranging from 93% to 97%. This imaging technique is particularly effective for detecting hydatid cysts, which predominantly localize in the liver, while also allowing for the evaluation of other abdominal sites [6,10]. In the case under discussion, the liver appeared normal.
- Although the mortality rate associated with echinococcosis is relatively low, the condition can result in significant disabilities. The prognosis is contingent upon the specific type of infestation.

Morbidity primarily arises from the rupture or infection of cysts, as well as dysfunctions of the affected organs, which may include cirrhosis, biliary obstruction, and bronchial obstruction, among others.

The reported mortality rate associated with this condition ranges from 0.29% to 0.6%. This mortality is primarily attributable to factors such as anaphylaxis, systemic complications (including sepsis and respiratory failure), or the variable characteristics of the infection. These factors can depend on several key variables: (a) the specific organ affected, (b) the size and location of the cyst, (c) the interactions between enlarging cysts and surrounding organs, and (d) complications arising from cyst rupture [11–13]. In the case of our patient, the prognosis was particularly poor due to the severe localization of the condition and its proximity to the cardiac cavities, which led to a potential pre-tamponade situation.

Effective treatment is contingent upon early detection and comprehensive surgical excision. The surgical strategy is determined by the cyst's specific location and the involvement of adjacent structures [6].

The effectiveness of albendazole as a medical treatment continues to be a topic of discussion among health care professionals. This medication is commonly prescribed in the postoperative phase, particularly for patients demonstrating indications of disease dissemination or those with ruptured cysts [5]. Additional scenarios warranting its use include inoperable primary cysts in the liver or lungs, cysts located at multiple sites, and peritoneal cysts [14]. Considering these factors, we proposed an albendazole-based treatment regimen for our patient following a collaborative discussion, especially in light of the patient's unwillingness to proceed with surgical intervention.

This highlights the unfavorable prognosis experienced by our patient, attributable to the delayed diagnosis, which occurred at a late stage. Additionally, the patient's refusal of surgery further contributed to the unfortunate outcome, ultimately resulting in her demise.

Conclusion

This case study illustrates the remarkable correlation between a pulmonary hydatid cyst and pulmonary embolism, resulting in diastolic restriction and pulmonary hypertension. Despite the ongoing prevalence of hydatid disease in various regions, its unusual presentations—particularly those involving cardiac structures—pose considerable diagnostic and therapeutic challenges. The compressive impact of the cyst on cardiac chambers resulted in significant hemodynamic implications, ultimately leading to a pre-tamponade state. Prompt diagnosis and timely intervention are paramount in averting complications and enhancing patient outcomes. Although surgical excision is regarded as the standard treatment for large hydatid cysts, situations involving patient refusal or delayed presentation may require the use of medical therapy with albendazole, which can exhibit variable efficacy. This case emphasizes the necessity for heightened clinical vigilance, particularly in endemic regions, to ensure the swift identification and effective management of complex presentations of hydatid disease.

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Ethics, Consent To Participate, And Consent To Publish:

Ethical approval was not required for this case report. Written informed consent was obtained from the patient (or the patient's legal guardian) for participation and publication.

Clinical Trial Number:

Not applicable.

Ethics Approval:

This study was approved by the Ibn Rochd University Hospital ethics committee.

Informed consent was obtained from the patient and her family.

Data Availability Statement:

The data of this study are available from the authors upon reasonable request.

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