

Post-Traumatic Rupture of a Pleural Hydatid Cyst: A Case Report

Oussama El Abbassi (MD)^{1,2*}, Zakariae Missaoui (MD)^{1,2}, Arghal Mohammed (MD)^{1,2}, Diouri Lamyae (MD)^{1,2}, Siham Nasri (Ph.D)^{1,2}, Imane Kamaoui (Ph.D)^{1,2,3}, Imane Skiker (Ph.D)^{1,2}

¹Faculty of Medicine and Pharmacy, Mohammed Ist University, Oujda, Morocco

²Department of Radiology, Mohammed VI University Hospital Mohammed I University Oujda Morocco

³Vascular Anomalies Research Unit, Medical Science Research Laboratory Mohamed VI University Hospital Faculty of Medicine Oujda Morocco

DOI: [10.36347/sjmcr.2024.v12i02.022](https://doi.org/10.36347/sjmcr.2024.v12i02.022)

Received: 10.01.2023 | Accepted: 13.02.2024 | Published: 26.02.2024

*Corresponding author: Oussama El Abbassi

Faculty of Medicine and Pharmacy, Mohammed Ist University, Oujda, Morocco

Abstract

Case Report

Introduction: The preferred site of the hydatid cyst is the liver followed by the lung [1]. The extrapulmonary intrathoracic locations are usually the mediastinum, pleura, pericardium and chest wall. Most reported cases of intrapleural hydatid cysts are secondary; the primary pleural location is rare [2]. **Case presentation:** We report the case of a 16-year-old girl, operated two years ago for a pulmonary hydatid cyst, admitted to the emergency room in acute respiratory distress in the aftermath of a minimal trauma, responsible for the rupture of a pleural hydatid cyst in the pleural cavity. **Discussion:** A primary pleural hydatidosis ruptured in the pleural cavity was retained by a thoracic scan. Despite its rarity, our case raises the suspicion of primary pleural hydatidosis in the presence of a large pleural cyst and small daughter vesicles floating in the pleural cavity with deviation of the mediastinal structures and pleural effusion.

Keywords: Echinococcosis, Hydatid cyst, Pulmonary, Pleural.

Copyright © 2024 The Author(s): This is an open-access article distributed under the terms of the Creative Commons Attribution 4.0 International License (CC BY-NC 4.0) which permits unrestricted use, distribution, and reproduction in any medium for non-commercial use provided the original author and source are credited.

INTRODUCTION

Hydatid disease is a zoonotic disease, a parasitic infection by a tapeworm of the genus *Echinococcosis*, which accounts for 95% of reported cases of human hydatid disease [3].

Herbivores are the intermediate hosts of the parasite, while carnivores represent the definitive hosts. Humans are not part of the biological life cycle, accidentally infected by ingestion of echinococcosis eggs [4].

The preferred sites of hydatid cysts are mainly the liver and lung, extra thoracic intra thoracic localisation is very rare, the characteristics and management of which should be discussed [2]. This case report presents a secondary intra pleural hydatid cyst ruptured in the pleural cavity.

CASE PRESENTATION

We present the case of a 16-year-old girl with a history of a large ruptured pulmonary hydatid cyst in the pleural cavity that was enucleated two years ago with pleural packing without any particular complications. The history of the disease included persistent chest pain

with rest dyspnoea after minimal trauma, which prompted the patient to consult the emergency department.

On admission to our institution, clinical examination found a conscious, tachycardic patient with a HR of 105 beats per minute, polypnoeic with a FR of 28 cycles per minute, a room air saturation of 80% and a fever of 38.1 C. The cardiovascular and neurological examination was normal.

The biological work-up was disturbed with a very positive infectious work-up with leukocytes at 21,000 with PNN predominance, CRP = 300, PCT at 33.

A first standard radiograph showed a large left pleural effusion collapsing the lung parenchyma opposite with a slight deviation of the mediastinal structures towards the contralateral side as well as a blunting of the right cul-de-sac without any associated post-traumatic lung parenchymal or bone anomaly (figure 1).

A thoracic CT scan without and with contrast injection was performed (Figure 2) showing:

- Collapse of the left lower lobe with a hypodense area, corresponding to the old residual cavity.

- A homolateral oblong pleural formation with heterogeneous fluid content, covering the entire pleural cavity, containing locally calcified and locally discontinuous proliguous membranes, extending for about 17 cm. This formation scallops out the lung parenchyma.
- It is associated with other well-limited cystic pleural formations with regular contours and a spontaneously dense wall, the largest of which measures 23 x 16 mm apically.
- Pleural effusion of medium to great abundance.
- The whole exerts a mass effect on the mediastinal structures with a discreet deviation towards the right side
- Right pleural effusion blade.

The evolution was marked by a worsening of the clinical situation with signs of shock requiring her to

be sent to an intensive care unit for further treatment. An evacuation puncture was performed draining 300cc of frank pus. Then the patient benefited from a surgical exploration which found:

- A very intimate basal sheathing with issue of hydatid daughter vesicles on aspiration.
- A thickened parietal pleura of 5mm whitish fibrotic with deposit of false whitish membranes reminding the content of a ruptured hydatid cyst
- The visceral pleura is thickened enveloping the entire lung
- After lavage and drainage of the pleural cavity a pleurectomy was performed (figure 3).
- The evolution was good after the operation. The thoracic radiography showed a clear improvement, with a good re-expansion of the lung (figure 4).



Figure 1: A standard radiograph showed a large left pleural effusion collapsing the lung parenchyma opposite with a slight deviation of the mediastinal structures towards the contralateral side as well as a blunting of the right cul-de-sac

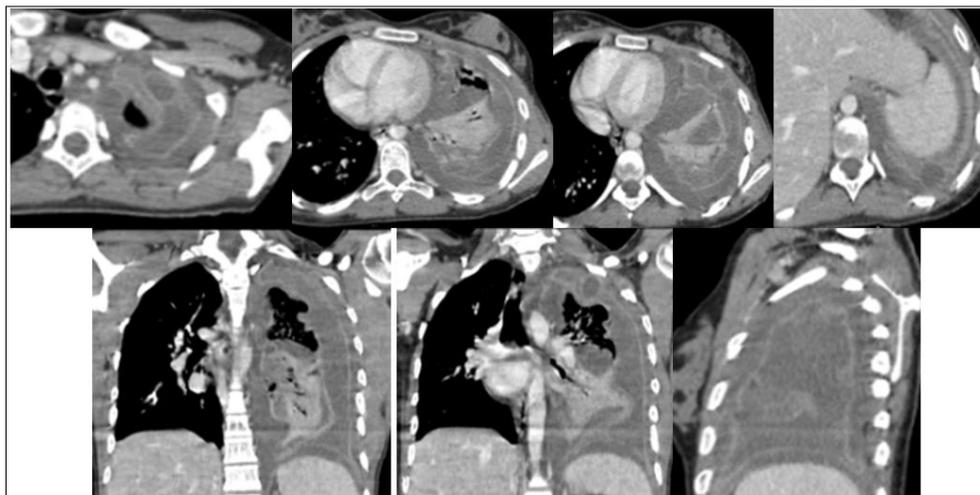


Figure 2: CT scan after injection of axial, coronal and sagittal contrast medium showing Collapse of the left lower lobe with a hypodense area, corresponding to the old residual cavity. A homolateral oblong pleural formation with heterogeneous fluid content, covering the entire pleural cavity, containing locally calcified and locally discontinuous proliguous membranes, extending for about 17 cm. This formation scallops out the lung parenchyma and it is associated with other well-limited cystic pleural formations with regular contours and a spontaneously dense wall, the largest of which measures 23 x 16 mm apically. And pleural effusion of medium to great abundance



Figure 3: The pleurectomy operative piece



Figure 4: Control chest radiograph showing good re-expansion of the left lung parenchyma

DISCUSSION

Hydatid disease is caused by *Echinococcus granulosus* and is endemic in many parts of the world, including Morocco [1]. Human contamination occurs through ingestion of parasite eggs through contaminated food, water or direct contact with the host [2].

This parasite can produce cysts in almost every organ of the body, with a predilection for the liver and lung. However, the cyst tends to appear in different sites and sometimes in unusual areas [1]. In most cases the lung is the most affected organ in childhood [5]. Pleural hydatidosis is a very rare disease, the pleural location is often secondary to rupture of a supra- or sub-diaphragmatic hydatid cyst [6].

Primary pleural hydatid cysts are included in the category of extra-pulmonary intrathoracic cysts, along with those found in the parietal pleura, mediastinum, pericardium, diaphragm, fissures and chest wall, by lymphatic or haematogenous dissemination [2]. They are extremely rare, Erkoç *et al.*, presented a ruptured pleural hydatid cyst in this unusual location [4].

The pleural layers are avascular and a hydatid cyst can form and grow in this region because the layered membrane structure of the cyst is permeable to calcium, potassium, chloride, water and urea. Thus, these and other nutrients that may be useful to the parasite can pass through the membrane by diffusion. Active transport may be involved in this process [7]. Its initial phase is usually asymptomatic and latency periods of several decades can be observed. This explains the late presentation of symptoms [4].

In our case, the patient had a history of a ruptured pulmonary cyst in the pleura two years ago, which led to dissemination of the disease into the pleural cavity, despite appropriate treatment. This disease affects mostly young adult males. Clinically, the pleural hydatid cyst is characterised by its total latency and therefore its fortuitous discovery on the chest X-ray [8].

More rarely, it may manifest with a non-specific and discrete clinical symptomatology: chest pain, dyspnoea, dry cough, etc. Exceptionally, there may be signs of mediastinal compression, depending on the organ involved [8]. Halezaroglu and colleagues noted that cysts larger than 10 cm would be seen in young people due to the greater elasticity of the tissue [5]. Our patient presented with a minor trauma with respiratory distress

Biological diagnosis of hydatidosis is based on various immunological tests which are often negative in isolated uncomplicated pleural cysts [8]. The sensitivity of immunology increases markedly in cases of complication or associated liver cysts (7). CBC may reveal hyper-eosinophilia ($>500/\text{mm}^3$) [9].

Imaging is a key element in positive diagnosis and monitoring. Chest X-ray shows a well-limited homogeneous watery pleural opacity. Rarely, it shows peripheral calcifications that help to orient the diagnosis [9], calcification can also be seen as "eggshell calcification" [4].

Pleural cysts can rupture and release their contents into the pleural cavity, which presents as a pneumothorax or hydro pneumothorax [7], which was the case in our patient who presented on chest X-ray with a white lung following pleural cyst rupture.

Ultrasound, a second-line examination, confirms the liquid nature of the pleural opacity and

evokes a positive diagnosis, particularly in the case of a multivesicular form. It can also detect any associated pleural effusion and look for other cystic localisations, particularly abdominal [10].

The CT scan shows a pleural fluid formation unchanged by the injection of contrast medium, sometimes with fine peripheral calcifications [10]. The differential diagnosis can be made with exudative tuberculosis, loculated empyema and other parasitic infections such as amoebiasis [4].

The main treatment for hydatidosis is surgery followed by medical treatment with antiparasitic drugs [2]. The cyst is carefully dissected from the visceral pleura around the cyst to avoid rupture into the bronchus and complications such as broncho-pleural fistula and persistent postoperative air leakage [7].

The most commonly used medical treatment is Albendazole, while Mebendazole is only used as an alternative because of its side effects. However, antiparasitic treatment is recommended as an anti-infective treatment [2]. Our patient also showed signs of superinfection and was drained and put on antibiotic therapy while awaiting surgical treatment. After surgery, a long follow-up is necessary in case of recurrence and early treatment [4].

CONCLUSION

As can be seen, primary pleural hydatidosis is a rare entity, with only a few cases reported to date.

Cases of primary pleural hydatidosis are very rare but should be considered in patients from endemic areas who are working in occupations that are likely to be exposed to this parasite. Appropriate treatment, both surgical and antiparasitic, can lead to full recovery and a low risk of recurrent disease.

Pleural hydatidosis is often associated with an underlying pulmonary hydatid cyst. We emphasise prevention as the best means of eradicating this disease [6].

Conflict of Interests: The authors declare no conflict of interest.

Authors' Contribution: All authors contributed to the completion of this study.

Financial Support: No financial support was provided.

Informed Consent: The patient's parents agreed with a written informed consent to anonymously publish their daughter's medical informations.

REFERENCES

1. Geramizadeh, B. (2013). Unusual locations of the hydatid cyst: a review from Iran. *Iranian journal of medical sciences*, 38(1), 2.
2. Savu, C., Melinte, A., Grigorie, V., Ilescu, L., Diaconu, C., Dimitriu, M., ... & Bacalbasa, N. (2020). Primary Pleural Hydatidosis—A Rare Occurrence: A Case Report and Literature Review. *Medicina*, 56(11), 567.
3. Aguilar, X., Fernandez-Muixi, J., Magarolas, R., Sauri, A., Vidal, F., & Richart, C. (1998). An unusual presentation of secondary pleural hydatidosis. *European Respiratory Journal*, 11(1), 243-245.
4. Erkoç, M. F., Bilge, A., Alkan, S., & Okur, A. (2014). A rare cause of pleural effusion: ruptured primary pleural hydatid cyst. *BMJ Case Reports*, 2014.
5. Aribas, O. K., Kanat, F., Gormus, N., & Turk, E. (2002). Pleural complications of hydatid disease. *The Journal of thoracic and cardiovascular surgery*, 123(3), 492-497.
6. Masson, E. L'hydatidose pleurale [Internet]. EM-Consulte. [cité 26 déc 2022]. Disponible sur: <https://www.em-consulte.com/article/1023176/1-hydatidose-pleurale>
7. Mardani, P., Karami, M. Y., Jamshidi, K., Zadebagheri, N., & Niakan, H. (2017). A primary pleural hydatid cyst in an unusual location. *Tanaffos*, 16(2), 166.
8. Harzallah, L., Bacha, M., Garrouche, A., Messak, A., Ben Cherifa, L., & Bakir, D. (2007). Kyste hydatique pleural primitif: a propos d'une observation. *RMLG. Revue médicale de Liège*, 62(7-8), 506-508.
9. Nazaroglu, H., Balci, A., Bukte, Y., & Simsek, M. (2002). Giant intrathoracic extrapulmonary hydatid cyst manifested as unilateral pectus carinatum. *Southern medical journal*, 95(10), 1207-1209.
10. Pedrosa, I., Saíz, A., Arrazola, J., Ferreirós, J., & Pedrosa, C. S. (2000). Hydatid Disease: Radiologic and Pathologic Features and Complications 1: (CME available in print version and on RSNA Link). *Radiographics*, 20(3), 795-817.