Respiratory Medicine Case Reports 30 (2020) 101054

Contents lists available at ScienceDirect

Respiratory Medicine Case Reports

journal homepage: http://www.elsevier.com/locate/rmcr

Case report

Massive pulmonary embolism due to hydatid cysts: A rare postoperative complication of liver echinococcosis

Sotirios Kotoulas [a], Konstantinos Grapatsas [a,b,c,*], Vasileios Leivaditis [d], Ioannis Panagiotou [a,b], Emmanouil Spiridakis [a], Uyen-Thao Le [c], Thomas Osei-Agyemang [c], Christophoros Kotoulas [a,b]

[a] Cardiothoracic Surgery Department, "Iaso" General Hospital, Athens, Greece
[b] Cardiac Surgery Department, 401 General Military Hospital of Athens, Athens, Greece
[c] Department of Thoracic Surgery, Medical Center-University of Freiburg, Faculty of Medicine, Germany
[d] Department of Cardiothoracic and Vascular Surgery, Westpfalz-Klinikum, Kaiserslautern, Germany

ARTICLE INFO

Keywords:
Hydatid disease
Echinococcosis
Hydatidosis
Pulmonary embolism
Surgery

ABSTRACT

Pulmonary embolism due to hydatid cysts is a very rare pathology. A 55-year-old male was transferred to our hospital due to respiratory failure, as a result of left pulmonary artery obstruction by cystic lesions. His medical history included multiple operations for abdominal echinococcosis. He was urgently operated for the extraction of the proximal hydatid cysts with the use of cardiopulmonary bypass with an excellent outcome and six months later through a right thoracotomy for the distal right pulmonary artery, the cysts were extracted. Meanwhile, he followed additional medication treatment with albendazole, preoperatively as well as postoperatively. Today, 5 years later, he has no evidence of hydatidosis recurrence.

1. Introduction

Echinococcosis is a chronic parasitic infection which is caused from Echinococcus granulosus tapeworm. The majority of cases are detected in the liver primarily and secondarily in the lungs. However, the detection frequency in the Cardiovascular system is less than 2% [1]. We present a rare case of a 55-year old male with massive pulmonary embolism due to hydatid cysts (HC), investigating the cause of pathology.

2. Case presentation

A 55-year-old Caucasian male was presented with orthopnea, tachypnea (saturation 70% on air). Blood testes revealed eosinophilia. He had a history of abdominal echinococcosis with hepatectomy, splenectomy and other cyst resections from the bowel, 8 years ago in a different center. Urgent chest CT-scan revealed cysts within both pulmonary arteries, occlusion of left main pulmonary artery and a left diaphragmatic hernia as a complication of the previous operations (Fig. 1). No liver HC were detected.

Using median sternotomy and cardiopulmonary bypass, in a hypertonic environment, the left main pulmonary artery cyst was extracted, as well as all the cysts from its branches. As the cyst was occluding the artery, the content was aspirated, and the cyst wall was removed safely. This was not possible for the right pulmonary artery cysts due to their distal location (Fig. 2a). The patient's condition improved dramatically. He followed medication with albendazole in four cycles of 28 days with an interval of 14 days. Six months later, cysts and clots from the distal portion of the right pulmonary artery were removed through right thoracotomy (Fig. 2b). He continued medication with albentazole in the same protocol. Five years later, he is disease-free, with no evidence of hydatidosis recurrence and with no other operations for it.

3. Discussion

HPE is an extremely rare clinical entity. Possible mechanisms are the rupture of a hepatic cyst or the release of daughter cysts in hepatic veins or directly in the inferior vena cava or a direct embolism from a ruptured cyst of the right cardiac chambers(1,2). It can occur not only automatically due to the cyst pressure to the neighboring tissues, but also intraoperatively due to the surgical manipulations. Cysts sited in pulmonary arteries can be asymptomatic for a long time since they grow slowly within the pulmonary artery, as the pulmonary perfusion is maintained via the bronchial arteries. Excessive growth in size of these cysts and finally total obstruction of the pulmonary artery may result to

* Corresponding author. Department of Thoracic Surgery Medical Center-University of Freiburg Hugstetter Strasse 55, 79106, Freiburg, Germany.
E-mail address: grapatsaskostas@gmail.com (K. Grapatsas).

https://doi.org/10.1016/j.rmcr.2020.101054
Received 22 February 2020; Received in revised form 4 April 2020; Accepted 4 April 2020
Available online 20 April 2020

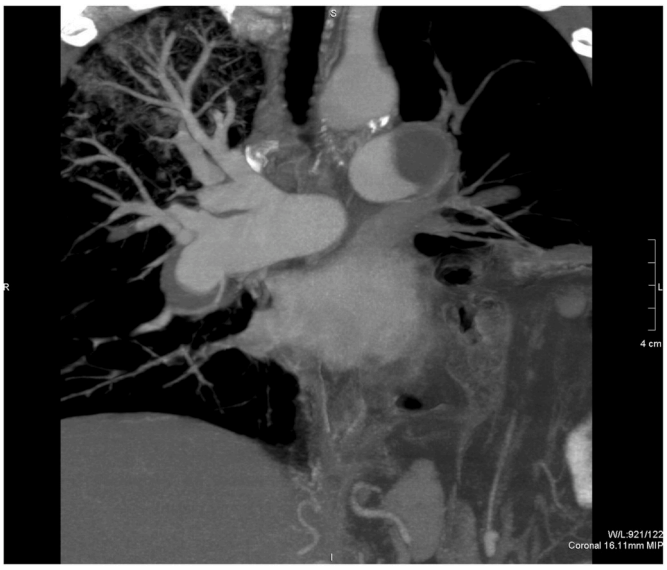

Fig. 1. CT-scan reveals the presence of the hydatid cysts.

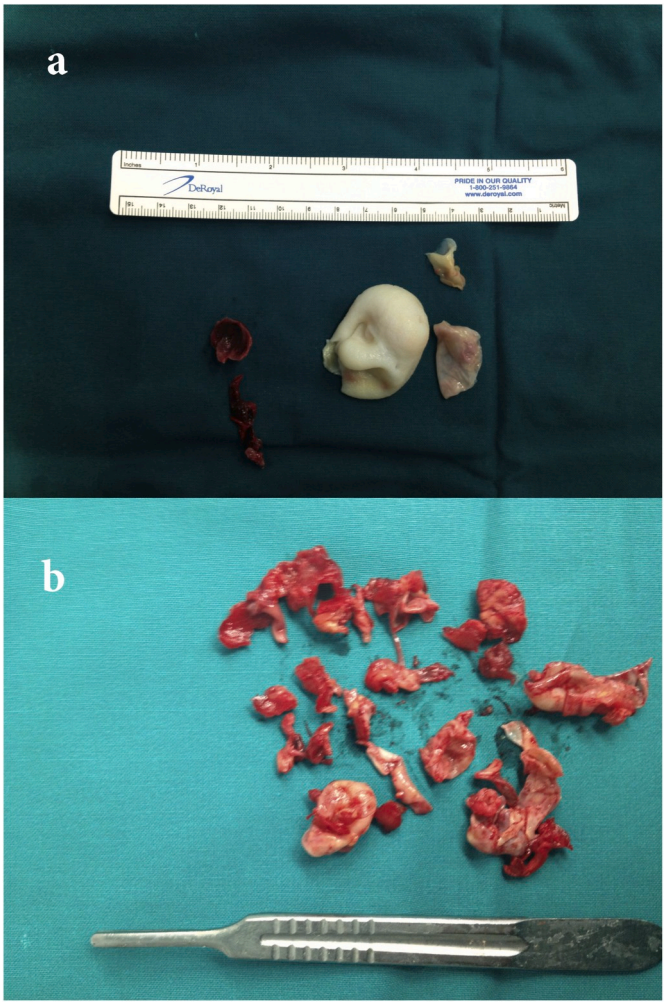

Fig. 2. The total removed content of hydatid cysts and clots from left (a) and right (b) side.

an acute fatal HPE [3]. Chronic HPE can appear as pulmonary hypertension [4]. Herein, we support that surgical manipulations during the multiple abdominal surgeries with the cyst release in the inferior vena cava was the responsible cause.

Important role in the differential diagnosis plays the clinical suspicion based on patient's history of echinococcosis [1]. The Computed Tomography Pulmonary Angiography scan and Magnetic Resonance Angiography are the cornerstone of the diagnosis. Although Lioulias et al. supported that MR angiography comparing to the CT scan could have advantages in the diagnosis of this rare disease as the thoracic anatomical structures are better represented in it, our diagnosis was accomplished through CTPA, because of the emergency of the respiratory failure(1). Transesophageal echocardiography can also be used to exclude cardiac CE [2].

Surgery is the treatment of choice. We preferred the median sternotomy approach due to the ideal and safe access of the proximal part of pulmonary arteries [1,5]. Peripheral embolism can be complicated with clots. Such cases of asymptomatic cysts should also be treated surgically in order to avoid complications, such as cyst rupture, anaphylactic shock and sudden death [3,5]. In our case, a second surgical approach was performed through a thoracotomy because of the similar situations.

The patient was followed medication therapy with albendazole in a special protocol of four 28-day cycles with an interval of 14 days, in order to avoid a secondary seeding or a postoperative recurrence [2,5].

In conclusion, although extremely rare, the HPE should be suspected in presence of unexplained dyspnea in patients with hepatic hydatid disease [1]. CTPA is the gold standard diagnostic tool. Surgery is the treatment of choice, even in asymptomatic patients(3,5).

Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Declaration of competing interest

None declared.

Acknowledgement

None declared.

References

[1] A. Lioulias, C. Kotoulas, J. Kokotsakis, M. Konstantinou, Acute pulmonary embolism due to multiple hydatid cysts, Eur. J. Cardio. Thorac. Surg. 20 (1) (2001) 197–199.
[2] R. Salem, A. Zrig, S. Joober, T. Trimech, W. Harzallah, M.A. Jellali, W. Mnari, J. Saad, B. Hmida, A. Elkamel, M. Golli, Pulmonary embolism in echinococcosis: two case reports and literature review, Ann. Trop. Med. Parasitol. 105 (1) (2011) 85–89.
[3] I. Bakir, Y. Enc, S. Cicek, Hydatid cyst in the pulmonary artery: an uncommon localization, Heart Surg. Forum 7 (1) (2004) 13–15.
[4] W. Bulman, C.M. Coyle, T.E. Brentjens, E.M. Horn, M.L. Dickstein, J.S. Wilt, J. Emond, S.M. Kawut, Severe pulmonary hypertension due to chronic echinococcal pulmonary emboli treated with targeted pulmonary vascular therapy and hepatic resection, Chest 132 (4) (2007) 1356–1358.
[5] H. Yaliniz, A. Tokcan, O.K. Salih, T. Ulus, Surgical treatment of cardiac hydatid disease: a report of 7 cases, Tex. Heart Inst. J. 33 (3) (2006) 333–339.