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Case report

An unusual manifestation of hydatid disease: A case in a 20-year old male patient☆

Jonathan Minagogo Hart[a,*], Fiker Eshetu[b], Seyoum Kassa[c]

[a] Department of Surgery Black Lion Hospital, College of Health Sciences, Addis Ababa University, Ethiopia
[b] Department of Surgery Menelik II Referral Hospitals, Addis Ababa, Ethiopia
[c] Department of Surgery Menelik II Referral Hospitals, Addis Ababa University, Ethiopia

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ABSTRACT

Echinococcosis is a re-emerging public health issue in developing countries as most communities are homes to dogs and other domesticated animals while cattle raising is a major habit of most sub-Saharan countries. The usual presentations include liver and lung cysts. While few documentations and publications have been made on extra-pulmonary intrathoracic hydatid cysts in other parts of the world, there has been no published document on extra-pulmonary intrathoracic hydatid cysts consisting of eighty or more cystic masses in a single patient in Ethiopia. We present a case of right sided extra-pulmonary intrathoracic hydatid cysts in a 20-year old Ethiopian male patient with compressive respiratory symptoms and significant social history of cohabiting with dogs throughout childhood. The patient was treated by surgery via a posterolateral thoracotomy and had a smooth Post-operative course and discharged home on post-operative day 15 on albendazole and analgesics. Intraoperative findings included approximately 1500 ml of cystic fluid, eighty cystic masses and collapsed lung.

Introduction

Echinococcosis (hydatid disease) is among the most neglected tropical diseases of the world. It is a zoonotic disease caused by an infection with cestode of the genus Echinococcus. It has been known to human since the time of Galen and Hippocrates but was described by Thebesius in the 17th century [3,2,1]. It is prevalent in sheep and cattle-raising regions of the world. It has two hosts and three developmental stages. A definite host-carnivores- and intermediate host-omnivores. Humans are accidental intermediate hosts. The developmental stages involve the adult tapeworm in the definite host, the eggs in the environment, and the metacestode in the intermediate host. The mode of human transmission involves ingestion of the eggs after contamination of the hands by handling dogs and/or contaminated vegetables. Oncospheres hatch in the duodenum, penetrate the intestines, get carried via the bloodstream, through the portal system to the liver. About 70% of hydatid cysts lodge in the liver, mostly in segments 7 and 8 [4]. Those that pass the liver travel via the right side of the heart to the lungs. Finally, a few embryos pass through the lungs and lodge in the systemic distribution. Clinical manifestations vary widely depending on parasite load, status and location of the cyst. The presenting symptoms are cough, followed by chest pains of varying severity. Ruptured cyst may present with productive cough, repetitive hemoptysis, fever, pleural effusion, or anaphylactic shock [5,1].

Extra-pulmonary intrathoracic hydatid disease constitutes 7.4% of all hydatid diseases [4]. Cysts in such sites can lead to fatal complications including bronchial rupture, fistulas to the pleural and pericardial cavities, and severe bleeding [1]. The combination of imaging and serology test usually enables diagnosis. There is no “best” treatment option for echinococcal infections: watch and wait, anti-parasitic treatment, percutaneous treatments, and surgery are available treatment modalities [8,7,6].

Case presentation and management

We present a case of a 20-year-old Ethiopian male presented to Menelik referral hospital with a six-month history of progressive dry cough followed by progressively worsening dyspnea, pleuritic chest

Abbreviations: POD, Post-Operative Day; CXR, Chest X-rays; CT, Computed Tomography; PAIR, puncture of the cyst wall, aspiration of cyst contents, instillation and re-aspiration of the scolicidal agent; PO, Per-os; VATS, Video-Assisted Thoracoscopic Surgery

☆ Categories: Gastroenterology, Pulmonary and infectious disease.

* Corresponding author.
E-mail addresses: jmhart206@gmail.com (J.M. Hart), fikereshetu7@gmail.com (F. Eshetu), seyoumkassa@gmail.com (S. Kassa).

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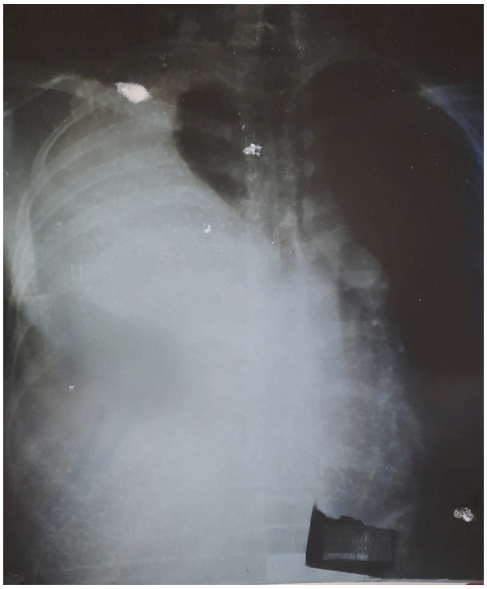

Fig. 1. CXR revealed diffused homogenous opacity of the right thoracic cavity with collapsed lung, apical pneumothorax, and shifting of the mediastinal structures as well as hyper inflected left lung.

pain localized to right lower hemithorax, and progressive weight loss. Associated symptoms included fatigability, palpitations, low-grade intermittent fever, loss of appetite, nausea and significant history of being raised in an environment with dogs and other domesticated animals. Otherwise, he has no history of smoking or being a passive smoker. He did not use alcohol or other illicit substances and he works at a mill. Other clinical and family history were unremarkable. For this condition, he was admitted at local clinics repeatedly and treated with IV antibiotics and analgesics for three months before his admission to this facility.

Physical examination revealed a chronically ill-looking male patient in mild respiratory distress otherwise stable with T: 36.6 °C, HR: 88b/m, BP: 110/70 mmHg, RR: 24 cy/m, and SPO_2: 92% at room air. The chest was asymmetrical, remarkable on the right, with decreased movement, dull percussion note with absent breath sound over the ipsilateral side. No signs of jaundice, or palpable lymph nodes. The rest of the examinations was unremarkable.

Chest X-ray and CT scan revealed features diagnostic of hydatid cyst (Figs. 1, 2 & 3). Abdominal ultrasonography and echocardiography were unremarkable. With the above imaging findings, the patient was diagnosed with multi-loculated cystic mass of the right lung 2° Hydatid cyst. Differential diagnosis included complex infected pleural effusion. Baseline laboratory work was significant for the absence of leukocytosis.

Albendazole 400 mg q12hrs for four days was initiated and informed consent signed. Preoperative preparations were done and the patient was taken to the operation room. Under general anesthesia and paravertebral nerve block, a right posterolateral thoracotomy was performed. Intraoperative findings included: (a) 1500 ml of cystic fluid, (b) thickened parietal pleura, (c) eighty cystic masses with the majority located between the parietal and visceral pleura followed by the intrapulmonary fissures and few within the pericardium, and (d) collapsed right lung (Fig. 4).

Cysts evacuation and cystectomy performed followed by decortication and pleurectomy. The thoracic cavity was lavage, air leak

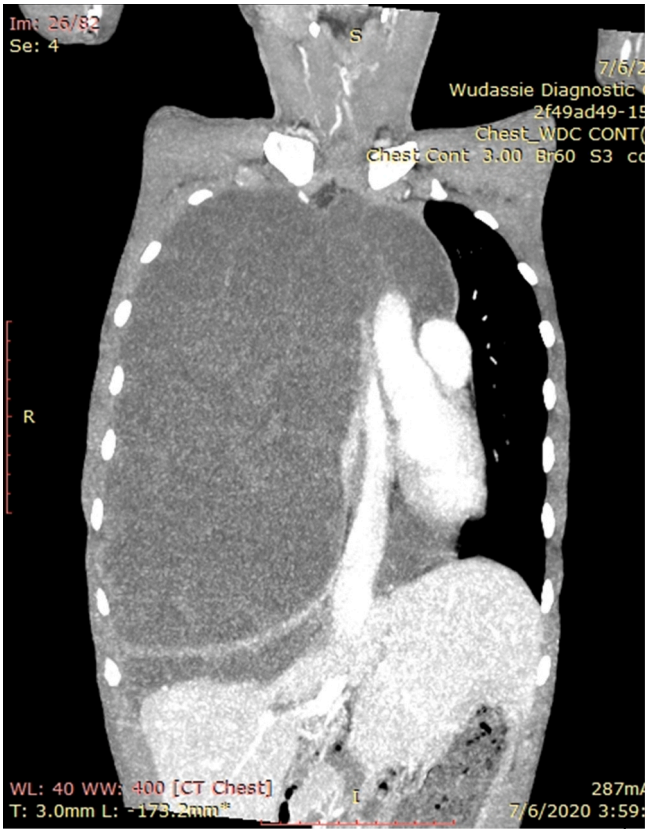

Fig. 2. Right hemi thorax homogenous opacity displacing the thoracic structures medially and posteriorly towards the vertebra.

checked, instruments and gauze counted, a single chest tube inserted and the wound closed in layers. The patient was placed on IV antibiotics, analgesic and albendazole. Cardiopulmonary exercises resumed on POD1 and control CXR taken (Fig. 5). The patient remained stable throughout the first five postoperative days with a drain output of less than 200 ml/24hrs and on POD5 a second CXR was taken which revealed complications (Fig. 6) and chest tube manipulated. On POD15 a third CXR was taken (Fig. 7) and the patient discharged thereafter with PO analgesics and asked to return in two-weeks. Follow up visit was unremarkable (Fig. 8).

Discussion

Extra-pulmonary intrathoracic hydatid cysts are rare entities with cysts located within the parietal pleura (18%); intrapulmonary fissures (15%); the chest wall (14%); the mediastinum (4.5%) and; additional 4.5% in the diaphragm [4].

In this case, we described a 20-year-old male patient with typical clinical manifestation diagnostic of extra-pulmonary intrathoracic hydatid cysts with majority located within the parietal pleura and fissures.

Rupture of hydatid cysts into the pleural cavity causes pneumothorax, pleural effusion, empyema, and anaphylactic reaction [9,10]. Unlike the literature, the only features of ruptured cysts were pneumothorax and pleural effusion. Our explanation for the absence of these features was repeated use of IV antibiotics and weaker immune response.

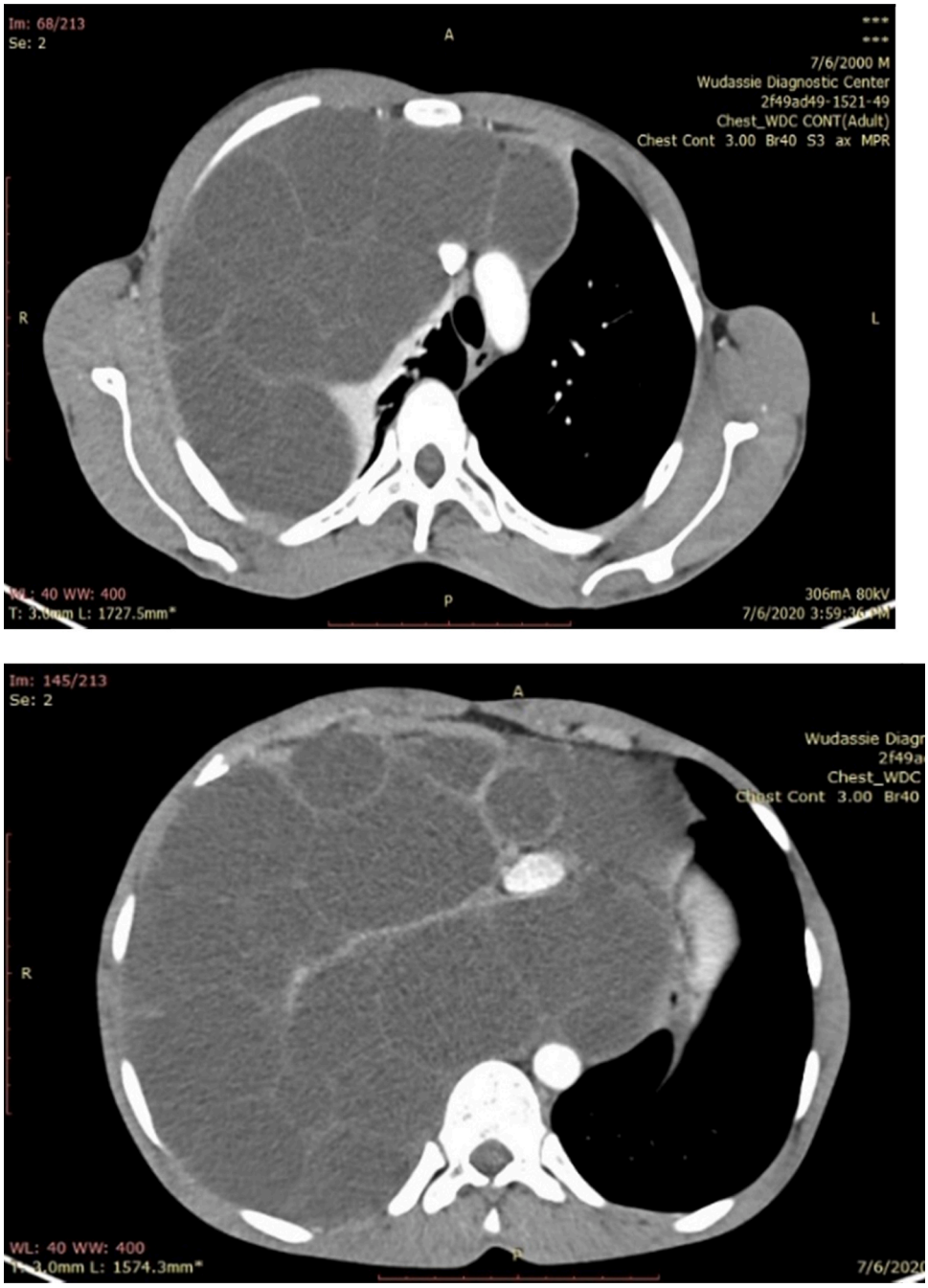

Fig. 3. These images show huge, well defined, round and thick-walled cyst with multiple daughter cysts occupying the right hemithorax with mass effect displacing the adjacent structures; aortic arch to the left and main pulmonary artery anteriorly with the right lung collapsed and displaced.

Diagnosis is made by physical findings, imaging, and serological testing [4]. In our case, the diagnosis was by history and chest imaging.

Treatment options involve non-surgical and surgical. Non-surgical management include medical treatment (benzimidazoles), PAIR, and interventional radiology techniques [11,12]. Medical management (chemotherapy) is indicated in patients with primary liver or lung cysts that are inoperable, patients with cysts in two or more organ and peritoneal cysts, and patients with liver transplants [11,12]. Response rates from medical treatment varies; 30% having the cysts disappearing, 30%– 50% have a decrease in the size of the cyst and 20%– 40% have no change [13,14,15].

The indications for interventional radiology techniques include inoperable patients, patients refusing surgery, patients with multiple cysts in segments I, II, and III of the liver, and relapse after surgery or chemotherapy. Contraindications include early pregnancy, lung and

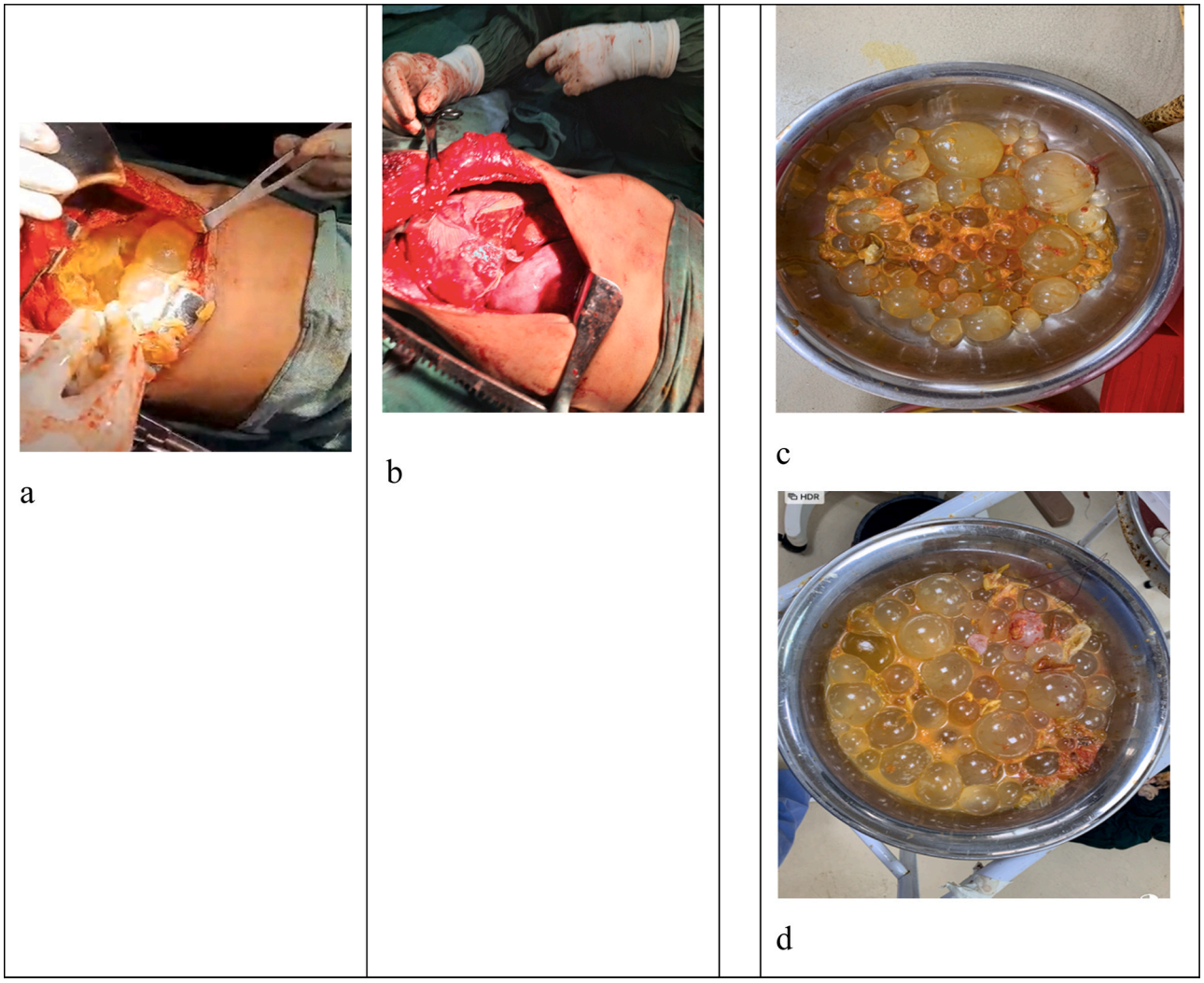

Fig. 4. Intraoperative finding (a) after skin incision; (b) after cystic masses evacuated; (c & d) contents evacuated.

brain cysts, inaccessible cysts, superficially located cysts, type II honeycomb cysts, type IV cysts, and cysts communicating with the biliary tree [13,16,17,18].

Effective treatment of hydatid cysts in organs, especially in the lungs, is achieved by complete excision of the cysts and capitonnage. Surgical management includes minimal invasiveness and thoracotomy. The choice of surgical technique ranges from radical to conservative surgery [13,16,17,18]. The aim of surgery in a pulmonary hydatid cyst is to remove the cyst completely while preserving lung tissue as much as possible. The basic steps of the procedure are protection of the surrounding tissues and cavities, sterilization of the cyst cavity using soclicidal agents and mechanical removal of the cysts and capitonnage [19]. Resection of pulmonary parenchyma is only indicated when the adjacent tissue is seriously damaged or infected, when the atelectatic areas are presumably irrecoverable or when a big cyst or numerous cysts had destroyed a certain anatomical substrate [13,16,17,18]. Additional medical treatment with PO benzimidazoles should be instituted for 4–6 months postoperatively [11].

Indications for surgical approach include large cysts with/without daughter cysts, superficially located single liver cysts, liver cysts with biliary tree communication and pressure effects on vital organs, cysts in lungs, brain, kidneys, eyes, and bones. Contraindications include general contraindication to surgery, multiple cysts in multiple organs, calcified cysts, and very small cysts. Postoperative complications include pleural effusion, prolonged air leak, empyema, bronchopulmonary fistula and recurrence.

Surgical options include *Barrett technique, Perez Fontana method, Ugon technique, Posada method, Figuera technique, segmental resection, Lobectomy,* and needle aspiration [3,2]. For this case, management was done through open thoracotomy with cysts evacuation, decortication, and pleurectomy. This method was chosen over other possible options due to the number of cysts and lack of VATS equipment in the hospital. There was no intraoperative complication

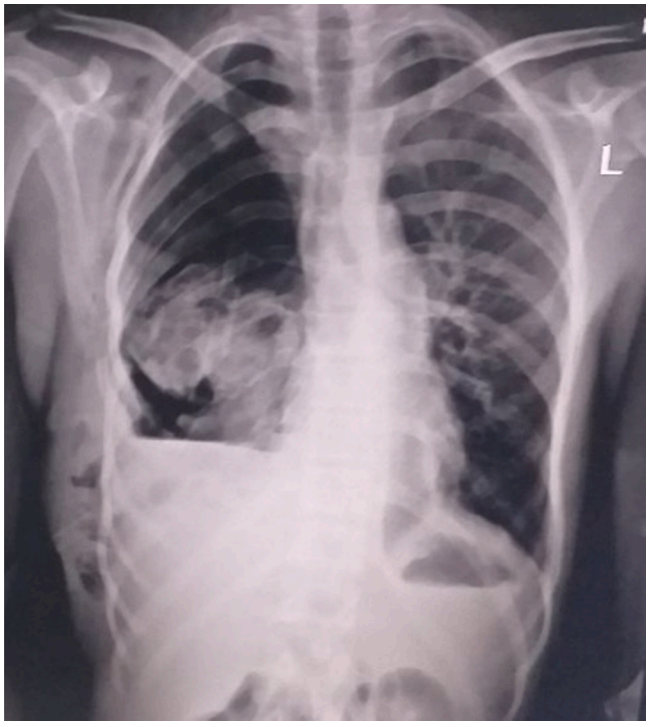

Fig. 5. POD# 1: X-ray showed rt lung with pneumothorax, consolidation and fluid collection.

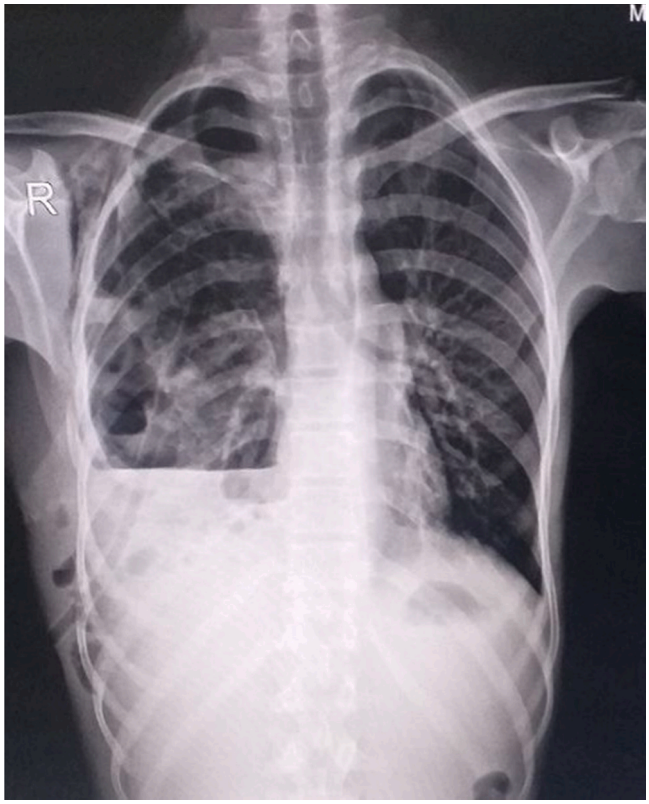

Fig. 6. POD5 X-ray showed relatively expanded right lung with significant fluid collection.

recorded in our patient. However, our patient developed post-operative loculated pleural effusion. Prognosis of our patient is expected to be good.

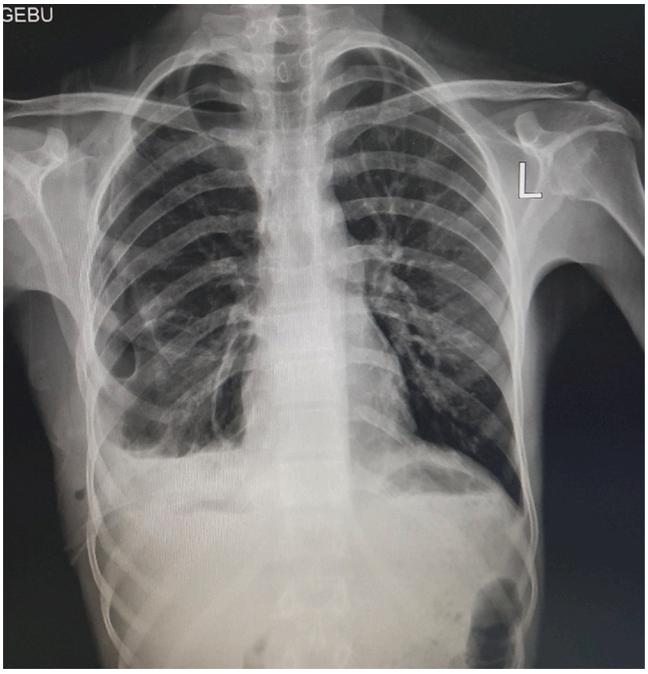

Fig. 7. POD15- expanded right lung with minimal fluid collection.

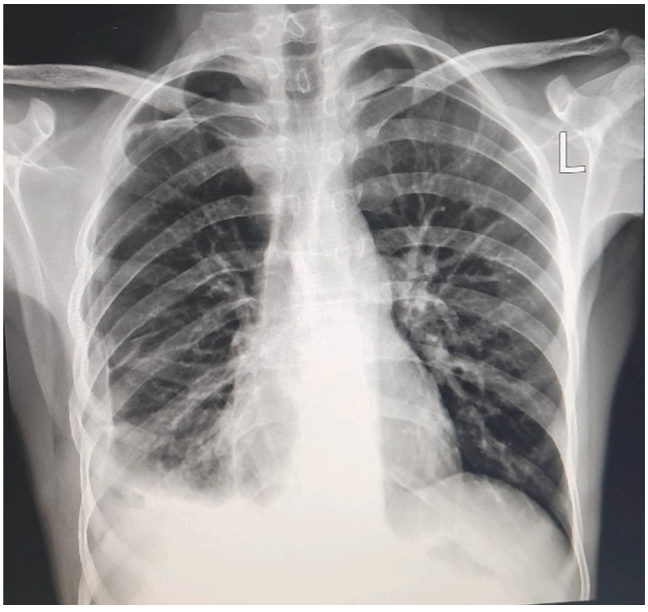

Fig. 8. 3 weeks (following discharge, when patient came for follow-up) - expanded right lung with reduce volume.

Conclusion

Extrapulmonary intrathoracic hydatid cysts are rare and surgery is the mainstream of treatment. The presence of pulmonary hydatid disease should be considered in patients with a well-defined, spherical lung mass or chest imaging features consistent with diagnosis of hydatid cysts, particularly in those who lived in or traveled to endemic areas.

We presented a case of a 20-year-old male patient with complaints of cough, dyspnea, pleuritic chest pain, weight loss, and history of living with lots of dogs. A diagnosis of pulmonary hydatid cysts was made based on chest imaging. The patient was treated by surgical intervention with evacuation of the cystic masses,

decortication, pleurectomy, and chest tube drain. Intraoperative findings included 1.5 L of cystic fluid with approximately eighty cystic masses located within the parietal pleura (majority), intrapulmonary fissures and pericardium with compressive effect. The patient had a relatively smooth post-operative event and was discharged on POD#15. First follow-up visit was unremarkable.

Ethics approval and consent to participate

This case report has obtained approval from the head of the thoracic unit at the department of surgery at the Menelik II Referral Hospital and informed consent from the patients prior to data collection and assembling.

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CRediT authorship contribution statement

J.M. Hart: Conception and design of study, Drafting the manuscript. **J.M. Hart, F. Eshetu:** Acquisition of data. **J.M. Hart, F. Eshetu, S. Kassa:** Analysis and/or interpretation of data. **F. Eshetu, S. Kassa:** Revising the manuscript critically for important intellectual content.

Conflict of Interest

The authors declared that there is no competing conflict of interest best to their individual and collective knowledge.

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