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Case Report

Ruptured pulmonary hydatid cyst presenting as hemoptysis in TB endemic country: A case report

Raju Prasad Pangeni [a], Pradeep Raj Regmi [c], Sulochana Khadka [b,*], Bibek Timilsina [b]

[a] HAMS Hospital Kathmandu, Nepal
[b] Nepalese Army Institute of Health Sciences, College of Medicine, Kathmandu, Nepal
[c] Tribhuvan University Teaching Hospital

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ABSTRACT

Introduction: Hydatid disease is a major zoonotic disease. After the liver, the lungs are the common site of involvement. Clinical manifestations of the disease depend on the site and size of the cysts as well as the presence of complications. The majority of the cases remain asymptomatic. Complicated pulmonary cysts can present as chest pain, cough, shortness of breath, and hemoptysis. Sometimes, imaging is not sufficient for diagnosis and histopathological evaluation can provide the confirmatory diagnosis.

Case presentation: A 32-year-old female presented with the complaints of episodic hemoptysis for the duration of two years. Radiological imaging could not provide a confirmatory diagnosis. Bronchoscopy was planned further. An endobronchial biopsy was taken for histopathological evaluation after seeing the whitish membranous structure within the right middle lobe bronchus. Hence, the diagnosis of ruptured cystic pulmonary hydatidosis was made.

Clinical discussion: Echinococcus granulosus is the cause of cystic pulmonary hydatid disease which is transmitted through contamination by the infected definitive host, mostly dogs. Most cases remain asymptomatic for a long period. Hydatid disease has many clinical and radiological forms which should be recognized and included in the differential diagnosis of many pulmonary problems. Sometimes, imaging and serological studies may not confirm the diagnosis, histopathological evaluation may be required.

Conclusion: Uncomplicated hydatid cysts are most commonly diagnosed incidentally in the adult population. Complicated hydatid cyst can present with various clinical manifestations. Episodic hemoptysis is one of the manifestations in our case. Clinicians should be aware of the typical as well as atypical manifestations of pulmonary hydatid disease.

1. Introduction and importance

Hydatid disease is a major zoonotic disease worldwide. Among six recognized species of echinococcosis and four being of a public health concern *Echinococcus granulosus (E. granulosus)* causing cystic echinococcosis (CE) is the most common followed by *Echinococcu smultiloculari*s causing alveolar echinococcosis, and *Echinococcus vogeli* and *Echinococcus oligarthrus* causing polycystic echinococcosis [1]. Hydatid disease has significant public health and economic impact in Nepal as well [2].

CE is the most common presentation in humans, contributing to more than 95% of the estimated 2–3 million global cases [3]. The most common organ involved in an adult is the liver followed by the lungs with respective frequencies of 60% and 20–30% [4]. The clinical features of hydatid cysts depend on the site and size of the cyst and the presence of complications. Uncomplicated small, peripherally located cysts often remain asymptomatic and are discovered incidentally on chest radiography as Shehatha et al., 2008 reported asymptomatic cysts in 37% of 763 cases [5]. Clinical symptoms occur when the cysts grow large enough to exert mechanical effects on the adjacent structures or to develop complications like rupture [6].

The treatment of hydatid lung cysts often requires surgical removal combined with chemotherapy (albendazole and/or mebendazole). Here, we present our case with the history of hemoptysis for two years, diagnosed through histopathological evaluation and managed with oral albendazole. This case report has been reported in line with the SCARE

* Corresponding author.
E-mail address: sulochanakhadka1@gmail.com (S. Khadka).

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2020 criteria [7].

2. Case presentation

A 32-year-old married female from Banke district presented to our OPD with the complaint of episodic hemoptysis for 2 years with the most recent episode 1 week back. Hemoptysis was sudden in onset, occurred once or twice in a month and sometimes was continuous for about 1 month which was usually a handful in amount, both bright and dark in color, with no diurnal variation, aggravating and relieving factors. It was preceded by a cough. There was no history of fever, significant weight loss, difficulty breathing, palpitations, abnormal body movements, or loss of consciousness. Her bowel habits were normal and she had her last menstrual period a couple of weeks ago. There was no family history of a similar illness, no past surgical history, and no significant psychosocial history. She was a non-smoker and non-alcoholic.

She was averagely built, cooperative, and well oriented to time, place, and person. Her pulse rate was 110 beats per minute with a regular rhythm, normal volume, and blood pressure of 100/70 mmHg, with normal temperature, respiratory rate, and oxygen saturation. On systemic examination, her chest was clear and other examinations were non-significant.

Complete blood count showed a total WBC count of 6340 cu/mm with Hemoglobin of 10.5 gm/dl. Her ESR was 25 mm/hr (0–9 mm/hr). Sputum examination for AFB stain was performed which was negative for acid-fast bacilli, and KOH examination was negative for fungal stain. USG abdomen and pelvis was not indicative of any significant finding. However, her Chest X-Ray showed a small opacity in the middle zone of the right lung.

Then, a CT scan was ordered for further diagnosis. HRCT finding was assertive of well-defined thin-walled cavitary lesion of size approximately 2.3 × 1.5 cm in right middle lobe with intracavitary solid component with lobulated margin and air crescent in the anterior aspect of the lesion (Fig. 1). CECT chest showed a solid cystic lesion in the right middle lobe with non-enhancing solid component in the dependent aspect and an air crescent in the anterior aspect (Fig. 2). Differential diagnosis of tuberculosis, aspergilloma, and focal bronchial atresia were made. However, it could not elicit a definitive diagnosis.

Other blood investigations like ANA, ANCA, PT/INR, and Echinococcus serology were also sent. The reports were found to be negative (see Table 1). She was then planned for bronchoscopy by a pulmonologist for further examination. Bronchoscopic wash from the right middle lobe of the lung was sent for cytology, gram stain, KOH, C/S, AFB, and geneXPERT. On collected specimen from the bronchoscopic wash, cytology was negative for malignant cells. No fungal element was seen on KOH preparation and acid-fast bacilli in Acid Fast Bacteria stain. Gram stain showed 30 WBC/HPF, 2–4 epithelial cells/HPF, and was negative for bacterial organisms and yeast cells. Similarly, no pathogenic organism was isolated in culture for 48 hours.

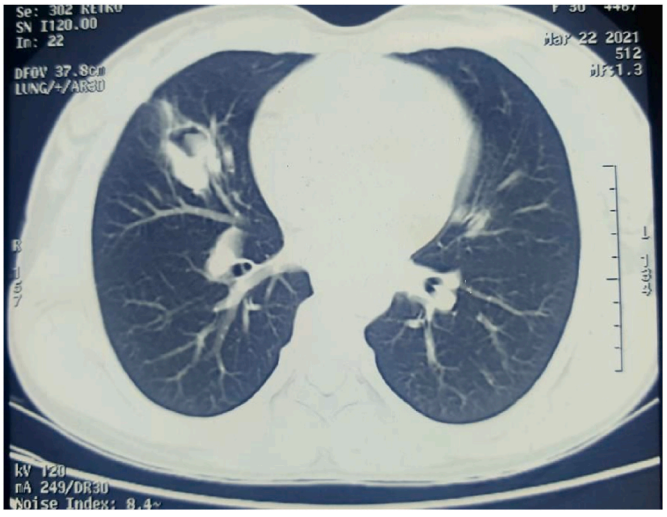

Fig. 1. HRCT chest axial image shows a well-defined thin-walled cavitary lesion in the right middle lobe with an intracavitary solid component with a lobulated margin. Air-crescent is seen in the anterior aspect of the cavity. The surrounding lung parenchyma appears normal.

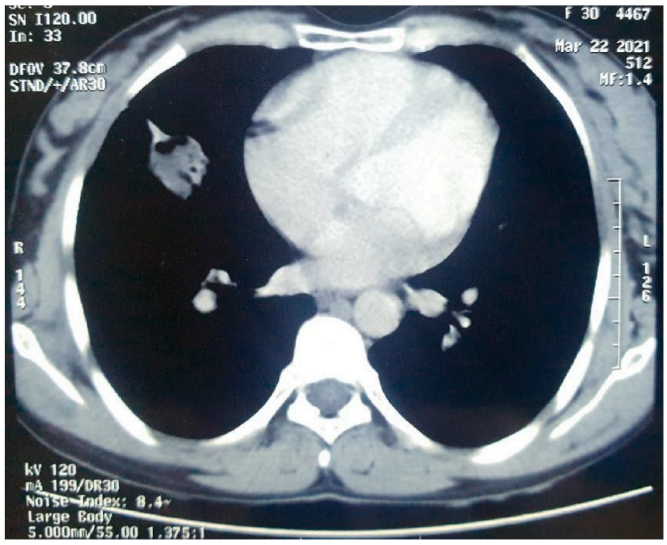

Fig. 2. CECT chest axial image shows a solid cystic lesion in the right middle lobe with an *air crescent sign* in the anterior aspect.

Table 1
Blood investigations in patient.

WBC	6340/cu.mm
Neutrophils	49%
Lymphocytes	44%
Eosinophils	02%
Monocytes	05%
Basophils	00%
Hemoglobin	10.5 gm/dl
Platelet count	275,000/cu.mm
MCH	25.6%
MCHC	32.1%
RBC	4.1 10^6/ul
ESR	25 mm/hr
PT	14.1 sec
INR	1.1 sec
Echinococcus serology	Negative
ANA	Negative
ANCA	Negative

However, her bronchoscopy findings showed a mucus plug with a creamy white thick membranous surface completely occluding the segment bronchi of the lateral segment of RML. The membrane sloughed out on suctioning leaving behind distal segments filled with mucus and similar membranous structures in the right bronchial tree (see Fig. 3 and Fig. 4). It was suspected to be a ruptured hydatid disease. Subsequently, endobronchial biopsy/membranous tissue was sent for histopathology.

The histopathology report eventually demonstrated eosinophilic laminated membranous structures in the specimen suggestive of hydatid disease (see Fig. 5 and 6). Therefore, the diagnosis of cystic pulmonary hydatidosis was made.

The patient was explained about the medical treatment and the need for surgical management. She was prescribed an oral tablet of albendazole 200 mg twice daily for the duration of 3 months after which her symptoms improved. She agreed to comply with the medications and surgical management such as lobectomy if required. Currently, she is in a good state and has regular follow-ups.

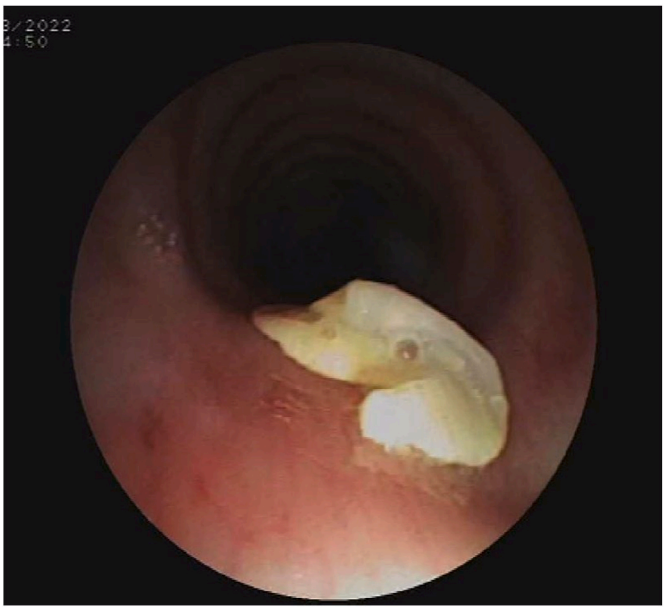

Fig. 3. Bronchoscopy finding showing a creamy white membranous structure in the distal tracheal lumen.

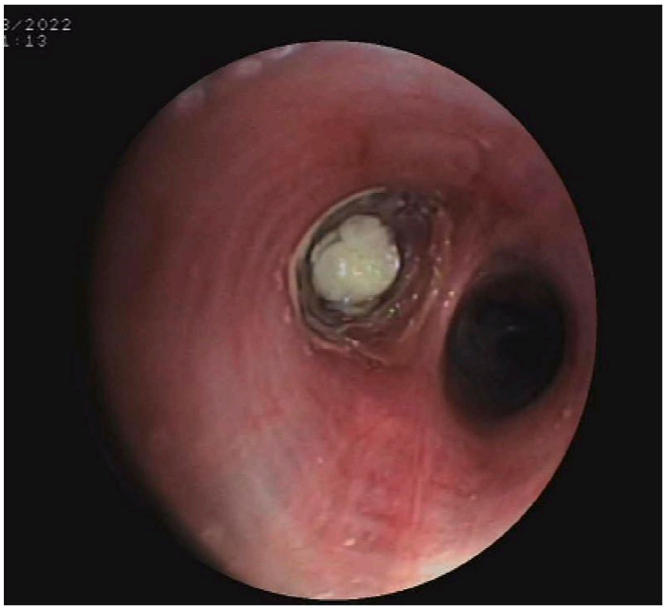

Fig. 4. Bronchoscopy finding showing complete occlusion of the subsegment of lateral segment of the right middle lobe.

3. Clinical discussion

Human infection with E. granulosus leads to the development of one or more hydatid cysts located most often in the liver and lungs, and less frequently in the bones, kidneys, spleen, muscles, and central nervous system. A study done by El khattabi et al., 2012 showed that the mean age of occurrence of hydatid disease was 35 years with male predominance (53%) [8]. Our case was a female with the age of 32 years.

The asymptomatic incubation period of the disease can last many years until hydatid cysts grow to an extent that triggers clinical signs [9]. Hydatid cyst is usually transmitted to humans through the ingestion of the food or water contaminated by an infected definitive host, most commonly dogs [10]. Hydatid cyst disease has many clinical and radiological forms which should be recognized and included in the

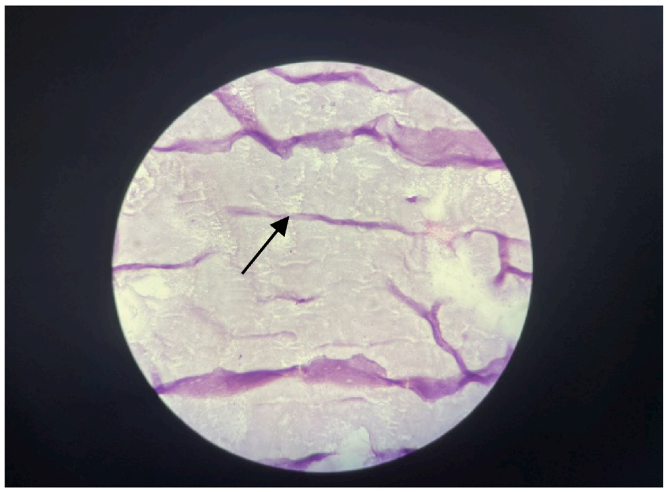

Fig. 5. Histopathological picture showing lamellated membranes (black arrow in Fig. 5).

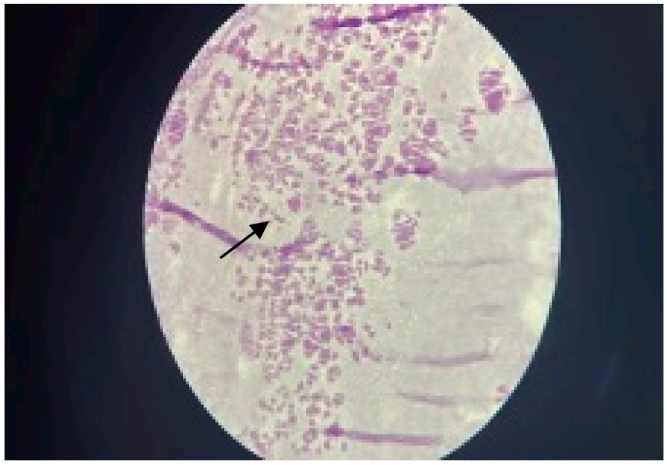

Fig. 6. Histopathological picture showing granular materials (black arrow in Fig. 6).

differential diagnosis of many pulmonary problems. According to Darwish et al. clinical presentations were varied among which 54% had cough, 36% had chest pain, 25% had dyspnea, 19% had hemoptysis, 10% had fever and chills, 10% had hydatidemesis, 5% had pleuritis and 3% had spontaneous pneumothorax [10]. In our case, the patient only had hemoptysis. The ruptured cyst may be contained (only detachment of the pericyst from the endocyst), communicating with the bronchus or into the pleural space. The content of the cyst may have irritated the mucosal lining of the bronchus thereby causing hemoptysis. However, our patient more likely was initially suspected to have other diseases like tuberculosis and aspergillosis which are quite common in our scenario.

In a study done by Ozdemir et al. the diagnosis of the patients was established in the light of the findings obtained from two-sided chest x-ray and CT of the thorax [11]. Along with MRI, CT scan also has high sensitivity and specificity for hydatid disease and is the diagnostic modality in detecting cyst wall or septal/wall calcification, demonstrating internal cystic structure posterior to calcification, assessing complications, and depicting osseous lesions [12]. In our case, CECT was done. The differential diagnosis from the imaging were aspergillosis, pulmonary tuberculosis, and isolated bronchial atresia. The imaging findings were not typical of the hydatid disease. In our scenario, the diagnosis was only considered after bronchoscopy and was confirmed by histopathological evaluation similar to the study done by Lodhia et al. [13]. A

study shows that other modes of diagnosis like serology and blood eosinophil count are not sensitive even in cases of ruptured hydatid cyst [14]. In our patient as well, blood eosinophil count was within normal range and serology for Echinococcus IgG was negative. There was an incidental finding of a whitish thick membranous structure which was confirmed to be a hydatid cyst later on with the histopathological evaluation.

Management includes medical and/or surgical interventions. Indications of chemotherapy include smaller cysts, patients with contraindication for surgery: poor candidate for surgery, a refusal for surgery and multiorgan disease, multiple cysts, recurrent cysts, and patients with risk of intraoperative spillage of hydatid fluid [15]. Our patient was counseled about the medical treatment as well as the need for surgical management. Currently, she is asymptomatic with medical treatment and is in regular follow-up.

Complications of pulmonary hydatid cyst include rupture (50–90% cases), superinfection, and rarely hepato-bronchial fistula or acute pulmonary embolism secondary to rupture [12]. In our case, no such complications were found.

4. Conclusion

Uncomplicated pulmonary hydatid cysts are most commonly diagnosed incidentally in the adult population. Ruptured pulmonary hydatid cyst can present with various clinical manifestations. Episodic hemoptysis is one of the manifestations in our case. Clinicians should be aware of the typical as well as atypical clinical as well as radiological manifestations of the pulmonary hydatid disease.

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Ethical approval

N/A.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contributions

Author 1: Led data collection, concept of study, contributed in writing the case information.

Author 2: Literature review, writing initial draft, revising, and editing the manuscript.

Author 3: Literature review , writing the draft, revising and editing the manuscript.

Author 4: Literature review, revising and editing the manuscript.

All authors were involved in manuscript drafting and revising, and approved the final version.

Registration of research studies

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Sulochana Khadka, Nepalese Army Institute of Health Sciences – College of Medicine, Kathmandu, Nepal.

Email: sulochanakhadka1@gmail.com, Phone: +977–9848559942.

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Declaration of competing interest

None.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.amsu.2022.103836.

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