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# Case Report

# Anaphylactic shock due to ruptured pulmonary hydatid cyst in a young patient from Iran

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#### ARTICLE INFO

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#### ABSTRACT

*Introduction:* and importance: Hydatidosis is a zoonotic condition caused by contact with the tapeworm Echinococcus granulosus metacestode. The liver and lungs are the most prominent locations for cysts. This is a rare case of pulmonary hydatid cyst (PHC) rupture in a young woman following a severe cough. *Case presentation:* On July 9, 2018, a 20-year-old woman presented to our hospital in northern Iran with a

complaint of cough with excessive sputum, dyspnea, drooling, symptoms of nausea and vomiting, itching and urticarial. Imaging findings showed evidence of a large, space-occupying mass in the right lung. She underwent a thoracoscopic wedge resection (TWR) as a minimally invasive technique, to remove the wall and contents of the cyst. Also, anaphylactic shock occurred in the patient. Due to hemodynamic disorders and heart and respiratory failure, unfortunately, the patient expired.

*Clinical discussion:* The rupture of a hydatid cyst may result in irreversible damage. PHC rupture is a serious complication that causes excessive coughing and chest pain. A rupture into the pleural or pericardial cavity is a serious and potentially fatal disease.

*Conclusion:* Anaphylactic shock should be considered as one of the serious complications of PHC, particularly in young ones. Therefore, early diagnosis and appropriate treatment are essential to prevent severe complications such as anaphylactic shock.

#### 1. Introduction

Hydatidosis is a zoonotic condition caused by contact with the tapeworm *Echinococcus granulosus* metacestode. It is found all over the world. The liver and lungs are the most prominent locations for cysts, but they may also appear in the spleen, brain, eye, heart, bone marrow, and kidney [1]. Hydatidosis is mostly asymptomatic until the cyst spreads and develops for many years following diagnosis, due to the gradual growth of cysts. In some cases, it is discovered by chance during imaging experiments [2]. Following the rupture of the cyst, symptomatic hydatidosis of the lungs is common [3,4]. Cyst breakup may occur

naturally or as a result of trauma. Symptoms of pulmonary hydatidosis include a sudden onset of cough, fever, hemoptysis, and anaphylactic shock. In this status, if it is left undiagnosed at an early stage, the chance of mortality is increased. Here we describe anaphylactic shock due to a ruptured pulmonary hydatid cyst (PHC) in a patient from Iran.

# 2. Case presentation

On July 9, 2018, a 20-year-old woman presented to Imam Khomeini hospital, Sari, Mazandaran Province, northern Iran, with a complaint of cough with excessive sputum, shortness of breath, drooling, symptoms

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of nausea and vomiting, itching and urticarial. She was admitted with vital signs (BP: 110/70, HR: 88, RR: 22, T: 37.7). On clinical examination, icteric sclera was not observed, and the patient's heart rate was normal. But on hearing in the lungs, there was a sharp decrease in sound in the right lung. The patient's abdomen was soft and no signs of tenderness or organomegaly were reported in the liver and spleen. After chest radiography and a computed tomography (CT) scan, evidence of a large and space-occupying mass that caused the right lower lobe of the lung (126.57\*99.71 mm) to disappear with a compressive effect on the diaphragm and the main bronchus was observed in the right hemithorax (see Fig. 1). The patient was prescribed albendazole 400 mg. For the laboratory diagnosis of hydatid cyst, a serological test was requested and the titer of anti-Echinococcus antibody was detected in the patient's serum. The patient was transferred to the operating room for surgery. Unfortunately, before intubation, the cyst ruptured due to a severe cough. The secretions and contents of the cyst were immediately sucked out and, after intubation, she underwent a thoracoscopic wedge resection (TWR), a minimally invasive technique, to remove the wall and contents of the cyst. Also, following anaphylactic shock in the patient, a cardiopulmonary resuscitation (CPR) was performed immediately due to hemodynamic disorders and heart and respiratory failure. Unfortunately, the patient expired.

The patient was then transferred to the ICU and underwent bronchoscopy due to urotracheal secretions. During the course of treatment, the patient developed subcutaneous emphysema and heart failure, after which a bilateral chest tube was implanted for the patient. Due to hemodynamic disorders and heart and respiratory failure, unfortunately, the patient expired after a week. The work has been reported in line with the STROCSS criteria [5]. This study is registered with the Research Registry, and the UIN is research registry 6919 (https://www.researchr egistry.com/register-now#home/addregistration/register-research-st udy-please-note-it-costs-99-to-register-payment/60d30450b5d71d0020 bd1c32/).

#### 3. Discussion

Hydatid disease is prevalent in developing countries, especially in areas where sheep are raised, such as Iran [6,7]. Hydatidosis has a wide range of clinical symptoms that are dependent on the place, position, and pressure effect of the slowly enlarging cyst in the involved organ [1, 4]. Differentiating hydatid cysts from benign cysts, mycoses, abscesses, caviar tuberculosis, and benign or malignant neoplasms is crucial. In patients with hydatidosis, the liver is the most affected organ, followed by the lungs (in nearly 25% of cases) [8]. Ruptured PHC splits into the pleural cavity and/or bronchus [9]. A cyst ruptured into the bronchus in the present situation.

PHC bronchus rupture is a serious complication that causes excessive



Fig. 1. Lung CT scan showing a space-occupying mass in the right lower lobe.

cough and chest pain [10], as seen in this case. Rupture into the pleural or pericardial cavity is a serious, potentially fatal disease that may result in bronchopleural fistula, stress pneumothorax, simple pneumothorax, collapsed lung, pleural thickening, empyema, and broad residual cavity [9,11,12].

Our patient had hives on his skin and a serious cough, according to the article. Hydatidosis has been linked to rare cases of sudden shortness of breath and rash [13]. The respiratory signs and rash vanished following treatment with albendazole in a 7-year-old child with ruptured PHC. Eosinophilia is present in 25% of contaminated patients with hydatidosis, according to research [8]. Furthermore, when a cyst ruptures, the number of eosinophils grows sharply [10,14]. Moderate eosinophilia was found in this situation (6%).

The most important diagnostic instruments for lung hydatidosis are imaging methods such as simple X-ray and CT scan [3,15–17]. Intact cysts have a round or oval appearance with sharp-edged homogeneous densities on plain chest radiographs, but ruptured cysts have meniscus sign, water-lily sign, cavity with or without air–fluid volumes, pneumonitis, and atelectasis of the lung. In patients with complicated cysts, the occurrence of an air–fluid level is the most common radiologic finding [11]. On a CT scan, a large mass with abnormal air fluid levels was found in the patient's right lower lobe of the lung. In addition, adjacent parenchymal infiltration, pleural thickening, and mild pleural effusion were observed in the right hemithorax.

Sputum specimen analysis is a non-invasive method for diagnosing suspicious PHC in a limited number of cases, and hooklet/scolices can be seen in smears. In some circumstances, this method may be used instead of a biopsy procedure [18,19]. PHC is mostly treated surgically, with chemotherapy used as a supplement. This technique is the best option for removing all cysts and preventing recurrences [9,11,20]. Removal of the cyst membrane, closing of the bronchial openings, and capitonnage are the most popular surgical procedures, although in more complex hydatid cysts, decortication, segmentectomy, TWR, and lobectomy may be essential [9,19]. To extract all the cysts in our patient, due to the emergency situation and the occurrence of anaphylactic shock to save his life, it was decided to perform immediate surgery with the TWR technique. Anaphylactic shock is a serious allergic reaction that starts quickly and can be fatal. Symptoms following a ruptured hydatid cyst include: itchy rash, swollen throat or tongue (angioedema), flushing or shortness of breath, wheezing or stridor, vomiting, tachycardia and hypotension, lightheadedness and loss of consciousness. Following surgery, there may be risks to the patient, including an unexpanded lung, empyema, pneumothorax, bronchobiliary fistula, and hydatid content spillage. However, unfortunately, our case suffered from a ruptured cyst wall and leakage of fluid into the airways due to a severe cough before surgery. Finally, she died because therapeutic work-up should begin prior to any irreversible and critical complications. Therefore, early diagnosis and appropriate treatment are essential to prevent severe complications such as anaphylactic shock.

# 4. Conclusion

Anaphylactic shock should be considered as one of the serious complications of PHC, particularly in young ones. Furthermore, imaging approaches, particularly chest radiography and CT scans and their findings, could be useful for early diagnosis and preventing irreversible complications while a large space-occupying mass exists.

#### Provenance and peer review

Not commissioned, externally peer-reviewed.

# Availability of data and materials

The raw data belonged to the present study cannot be made publicly available, because the disclosure of personal data was not included in the research protocol of the present study.

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# Ethical approval

The study was approved by our local ethics committee.

# **Consent for publication**

Written informed consent was obtained from the patient parents 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.

# Authors' contributions

ZZ and MF designed the study, wrote the manuscript, and analyzed and interpreted the data. AS, MS and AZ collected the data and provided critical comments. EB involved in interpretation and editing the manuscript. All authors read and approved the final version of the manuscript.

# **Registration of research studies**

6919.

### Guarantor

Zakaria Zakariaei.

#### Declaration of competing interest

None.

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# References

- A. Rostami, M. Mozafari, M. Gholipourmalekabadi, H.H. Caicedo, Z. Lasjerdi, M. Sameni, et al., Optimization of fluoride-containing bioactive glasses as a novel scolicidal agent adjunct to hydatid surgery, Acta Trop. 148 (2015) 105–114.
- [2] D.M. Smith, L.S. Rickman, Echinococcosis: a drop of water—review on human disease, diagnosis, and management, Infect. Dis. Clin. Pract. 10 (7) (2001) 355–359.
- [3] P.A. Koul, A.N. Koul, A. Wahid, F.A. Mir, CT in pulmonary hydatid disease: unusual appearances, Chest 118 (6) (2000) 1645–1647.
- [4] S. Santivanez, H.H. Garcia, Pulmonary cystic echinococcosis, Curr. Opin. Pulm. Med. 16 (3) (2010) 257.
- [5] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, A. Kerwan, A. Thoma, et al., The SCARE 2020 guideline: updating consensus surgical CAse REport (SCARE) guidelines, Int. J. Surg. 84 (2020) 226–230.
- [6] A. Rostami, M. Ebrahimi, S. Mehravar, V.F. Omrani, S. Fallahi, H. Behniafar, Contamination of commonly consumed raw vegetables with soil transmitted helminth eggs in Mazandaran province, northern Iran, Int. J. Food Microbiol. 225 (2016) 54–58.
- [7] A. Rostami, M. Taheri, M. Gholizadeh, S.J. Seyyedtabaei, S. Raeghi, S. Fallahi, Scolicidal effect of some herbs on Echinococcus granulosus protoscoleces: a systematic literature review. Herbal Medicines. Journal 1 (1) (2016) 53–59.
- [8] P. Moro, P.M. Schantz, Echinococcosis: a review, Int. J. Infect. Dis. 13 (2) (2009) 125–133.
- [9] A.E. Balci, N. Eren, Evval Eren Ş, R. Ülkü, Ruptured hydatid cysts of the lung in children: clinical review and results of surgery, Ann. Thorac. Surg. 74 (3) (2002) 889–892.
- [10] R. Doğan, M. Yüksel, G. Cetin, K. Süzer, M. Alp, S. Kaya, et al., Surgical treatment of hydatid cysts of the lung: report on 1055 patients, Thorax 44 (3) (1989) 192–199.
- [11] O.K. Aribas, F. Kanat, N. Gormus, E. Turk, Pleural complications of hydatid disease, J. Thorac. Cardiovasc. Surg. 123 (3) (2002) 492–497.
- [12] D. Puri, A.K. Mandal, H.P. Kaur, T.S. Mahant, Ruptured hydatid cyst with an unusual presentation, Case reports in surgery (2011), 2011.
- [13] W. Zingg, C. Kellenberger, B. Frey, F. Grimm, C. Berger, A 7-year-old girl with dyspnea and rash, Clin. Infect. Dis. 37 (1) (2003) 129–130.
- [14] G. Basavana, G. Siddesh, B. Jayaraj, M. Krishnan, Ruptured hydatid cyst of lung, J. Assoc. Phys. India 55 (2007) 141–145.
- [15] M. Lightowlers, B. Gottstein, Echinococcosis/hydatidosis: antigens, immunological and molecular diagnosis, Echinococcosis/hydatidosis: antigens, immunological and molecular diagnosis (1995) 355–410.
- [16] S. Rouhani, P. Parvizi, A. Spotin, Using specific synthetic peptide (p176) derived AgB 8/1-kDa accompanied by modified patient's sera: a novel hypothesis to followup of Cystic echinococcosis after surgery, Med. Hypotheses 81 (4) (2013) 557–560.
- [17] E. Şahin, S. Enön, A.K. Cangır, H. Kutlay, Ş. Kavukçu, H. Akay, et al., Single-stage transthoracic approach for right lung and liver hydatid disease, J. Thorac. Cardiovasc. Surg. 126 (3) (2003) 769–773.
- [18] L.S. Garcia, R.Y. Shimizu, D.A. Bruckner, Sinus tract extension of a liver hydatid cyst and recovery of diagnostic hooklets in sputum, Am. J. Clin. Pathol. 85 (4) (1986) 519–521.
- [19] G. Ramos, M. Antonio Orduña, M. Mariano García-yuste, Hydatid cyst of the lung: diagnosis and treatment, World J. Surg. 25 (1) (2001) 46.
- [20] A. Kuzucu, Ö. Soysal, M. Özgel, S. Yologlu, Complicated hydatid cysts of the lung: clinical and therapeutic issues, Ann. Thorac. Surg. 77 (4) (2004) 1200–1204.