MEDICAL SCIENCE

Hydatid cyst masquerading as empyema leading to benign pulmonary neoplasm: A diagnostic challenge

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ABSTRACT

Hydatid cyst (HC) is a disease that occurs due to the parasite Echinococcus (EC) and the most typical infection in humans with hydatid disease is caused by the species granulosus. The most frequent site for cyst development is the liver, followed by the lung which is the second most common location. Therefore, this case report highlights a case of a 75-year-old male who presented with chief complaints of left-sided chest pain, dyspnoea, cough and mucoid expectoration since one month and was initially thought as empyema. A diagnostic assessment was performed involving computed tomography suggested possibility of a hydatid cyst. Serological tests positive for *Echinococcus granulosus*. The treatment parameters involved both surgical and medical approaches. Thoracotomy was performed along with the post-operative medical treatment that mainly involved antihelminthics (Albendazole) and patient recovered well with treatment. This report highlights that large hydatid cysts can be surgically removed with good outcomes and the importance of realizing that the disease is a burden to public health and is much neglected.

Keywords: Hydatid Cyst, *Echinococcus granulosus*, Pulmonary Neoplasm

1. INTRODUCTION

Hydatid cyst (HC) is a disease that occurs due to the tapeworm Echinococcus when it transforms from its metacestode stage to develop into larvae. The most typical infection in humans with hydatid disease is caused by *E. granulosus* (Almutairi and Al-Rajhi, 2018; Gattani et al., 2023). Echinococcosis (EC) can emerge as the main disease in any organ or can spread as metacestodes from the main organ to other organs called as secondary EC (Almutairi and Al-Rajhi, 2018). The most frequent site for hydatid cyst development is the liver contributing 70% of the cases followed by the lungs accounting for 20% of cases (Pakala et al., 2016). HC enlarges more quickly in the lungs due to its elastic structure than in the liver and has the potential to

invade majority of the lobe by enlarging into enormous sizes (Aqqad et al., 2021; Onal and Demir, 2018).

The term HC was first used by Rudolphy, (1808) to describe EC in humans and is commonly seen in areas where farming of sheep and cattle takes place. Dogs act as definitive hosts where adult worms grow in their intestines before releasing their eggs into the faeces. Sheep and other animals contract this disease by consuming contaminated vegetation and when native people living in these infected areas accidently consume the eggs following contamination of the hands, oncospheres hatch in the duodenum enters the intestines and migrate through the circulatory system to different organs (Alloubi, 2013).

In many parts of the world, HC in the pulmonary region is still endemic and the most preferred treatment option is surgery. However, the early surgical method is debatable among many medical professionals and patients due to the progression of the disease into its benign form. The most frequent consequence of HC is perforation, which can result in permanent lung damage that can necessitate resection of the lung (Arroud et al., 2009; Emlik et al., 2015; Onal and Demir, 2018; Yekeler et al., 2013). Hence, this case report emphasizes on the adherence of surgical and medical intervention in an older adult with hydatid cyst that was leading to benign pulmonary neoplasm.

2. CASE REPORT

A 75-year-old male, a farmer by occupation and having exposure to sheep and goats presented with the chief complaints of left-sided chest pain, along with dyspnoea, cough and mucoid expectoration since one month. Additionally, the patient described a past history of tobacco consumption and Tuberculosis ten years back for which he was on anti-tubercular medications for around six months.

During the respiratory examination, it was noted that the left upper lung field had reduced breath sounds with dullness on percussion and tenderness in the left infraclavicular, suprascapular and interscapular areas. His blood sample were sent for laboratory investigations which revealed Eosinophilia and leucocytosis with the majority being neutrophils and as the patient described a past history of tuberculosis 10 years back his sputum was examined for acid-fast bacillus which revealed negative. Chest X-ray (Figure 1) revealed a large defined opacity with well-defined medial margin, with a broad base towards the chest wall and lateral margins merges with the chest wall showing incomplete border sign. Based on clinical and radiological findings patient was provisionally diagnosed as left empyema and was planned for pleural fluid aspiration. Ultrasound screening of lung was suggestive of cystic lesion in left lung so plan for pleural fluid aspiration was abandoned.

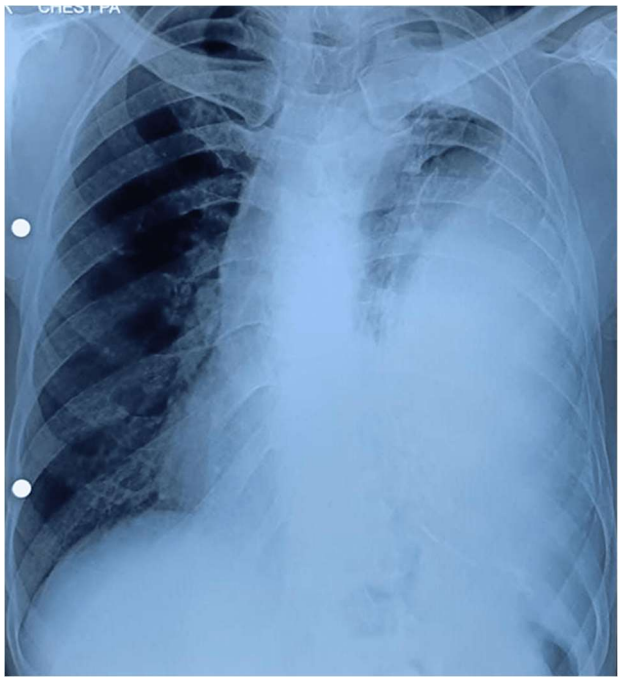

Figure 1 Chest X-ray Postero-anterior (PA) view demonstrating large well-defined opacity in the left lung

High-resolution computed tomography (HRCT) of the thorax (Figure 2) indicated a large solid lesion in the left pleural cavity of the upper lobe measuring 19.2×14.5×9.4 centimetres in maximum craniocaudal, anteroposterior and transverse dimension which appeared hypodense showing thick nodular peripheral calcification and a well-defined calcification in the central part of lesion

along with parenchymal consolidation adjacent to hydatid cyst. Additionally, HRCT thorax revealed the cyst was compressing the left-sided lung with crowding of left-sided ribs along with fibro calcific changes in the apical segment of the left upper lobe which confirmed the hydatid cyst leading to benign pleural neoplasm. As Hydatid cyst is more common in the liver ultrasonography (USG) of the abdomen and pelvis was performed which resulted in no abnormalities. The serological test was found positive for *Echinococcus granulosus* which was performed using an Enzyme-linked immunosorbent assay (ELISA) test that confirmed Hydatid cyst leading to benign pleural neoplasm. Initially after Chest X Ray it was thought to be a case of empyema later on performing HRCT Thorax and serological tests it was confirmed to be a rare case of Pulmonary Hydatid Cyst.

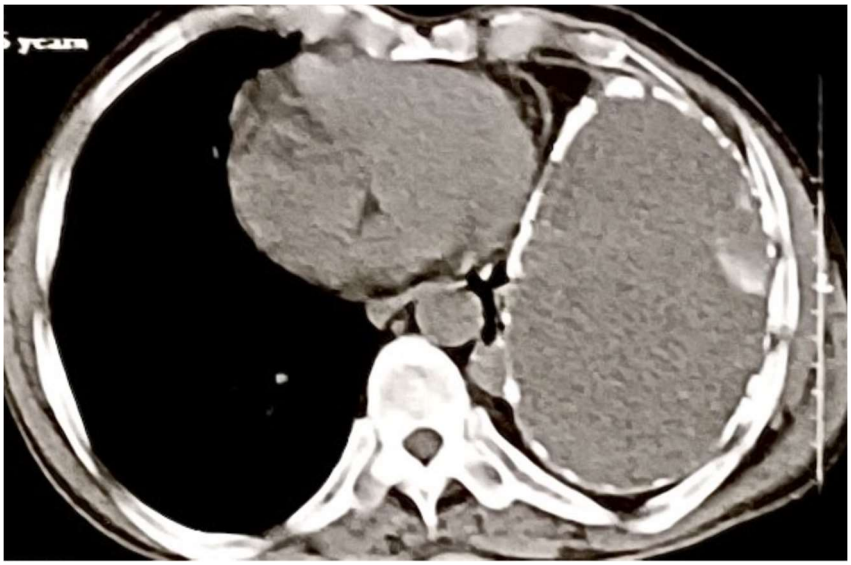

Figure 2 Mediastinal window of High-resolution Computed Tomography (HRCT) Thorax showing a large encapsulated pulmonary Hydatid Cyst

Based on the above diagnosis the patient was directed for thoracotomy (Figure *3)*. The surgery's goal was to completely excise the HC while conserving the lung tissue as much as feasible along with decortications of the left lung. Thoracotomy was performed under general anaesthesia in order to cause ipsilateral lung collapse with double-lumen endotracheal tubes. The adhesions were released after opening the chest wall and tissues in the periphery region were protected with towels moistened in 20% Hypertonic saline solution (HSS) and any alteration of the lung was avoided until the HC was evacuated followed by sterilization of the HC by aspiration of the fluid and replacing it with HSS, the HC was then aspirated at the site by a trocar and by suction, through this trocar, the components of the HC were evacuated. The cyst walls ruptured after the contents were removed, allowing the pericyst to be incised and opened. The contents that remained along with whitish pus fluid (100-200ml) were evacuated under direct vision and pus aspirated was sent for cytopathological examination and in addition to this, the pericystic area was partially resected (Figure 5). A small biopsy of lung tissue was obtained and was sent for histopathological examination. The remaining cavities were treated with HSS and the anaesthesiologist was instructed to ventilate the operated lung in order to precisely locate all bronchial openings by observing the air bubbles in the HSS and all bronchial leaks were sealed with absorbable sutures. With easily absorbed purse-string sutures, the cavity was completely sealed (capitonnage). After that, an intercostal drainage tube was inserted into the pleural cavity.

Figure 3 Thoracotomy while removing hydatid cyst

Figure 4 Intra-operative image showing various layers covering Hydatid Cyst

Pus aspirated during thoracotomy sent for cytology confirmed infected parasitic cyst – hydatid cyst (Figure 5). Lung biopsy specimen sent for Histo-pathological examination (HPE) showed calcified hydatid cyst (Figure 6).

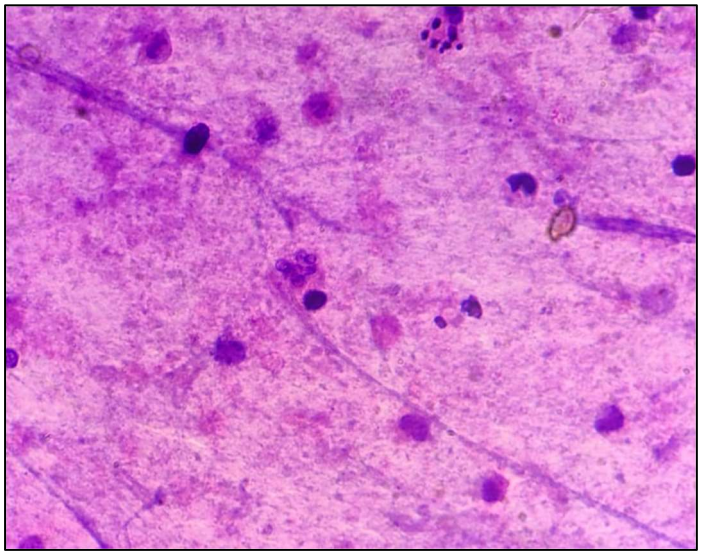

Figure 5 Pus cytology -hydatid cyst showing foamy macrophages

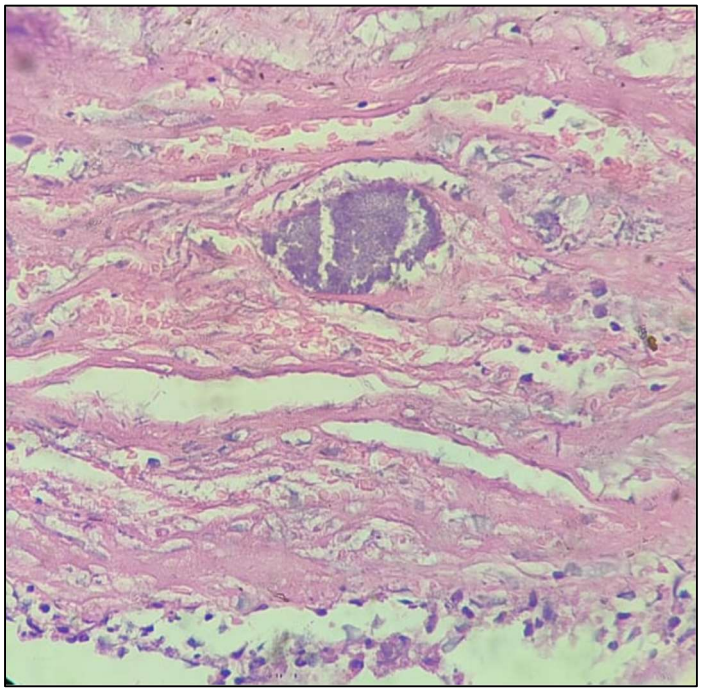

Figure 6 Lung tissue Histopathological features suggestive of calcified Hydatid cyst

The patient post operatively was kept in observation with medical intervention consisting of intravenous (IV) antibiotics Meropenem and Metronidazole and albendazole (antihelminthic). Albendazole is the drug of choice in this condition and plan is to continue albendazole for minimum 3 months with regular monitoring of Liver function test. Patient was discharged with albendazole 400mg twice daily. On follow up after 21 days, Chest X-ray (Figure 7) done showed resolution compared to the previous X-ray, liver function test was normal and albendazole was continued. The patient is adhering well to the above-described treatment

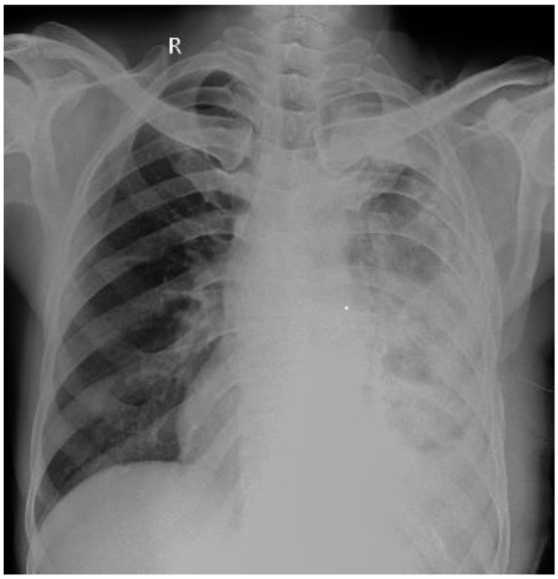

Figure 7 Chest X-ray Postero-anterior (PA) view showed resolution of the lesion

3. DISCUSSION

EC parasite causes HC which is a zoonosis. *E. granulosus* and *E. multilocularis* are the two species of EC among which *E. granulosus* is most frequently encountered in humans. In underdeveloped regions where animal husbandry is widespread and there is no veterinary control, HC disease is a serious public health issue. The definitive hosts are dogs and other predators whereas; the intermediate hosts are sheep and other ruminant animals. Humans are these parasite's secondary hosts and they become infected by consuming food or water that has been infected with these parasites' eggs (Kumar et al., 2012). In the present case, the patient had a history of contact with sheep and goats which acts as definitive hosts for the parasite whereas, humans act as an intermediate host in which infection occurs by ingestion of EC eggs that are found in faeces of dogs and several other wilds (Baruah et al., 2020).

A combination of serological tests and radiological imaging can be used to diagnose HC (Almutairi and Al-Rajhi, 2018; Bakir et al., 2004). Cystic lesions should be diagnosed by Ultrasonography (USG). When the area is inaccessible, the preferred choice of imaging should be Computed tomography (CT) and Magnetic resonance imaging (MRI) with strongly T2-weighted series (Darwish et al., 2020; Stojkovic et al., 2012). In this case, the diagnosis was confirmed using chest x-ray and CT along with serological tests with ELISA that confirmed *E. granulosus*. Since the liver is the most typical site for HC, a USG of the pelvis and abdomen was performed, but no abnormalities were noted.

Treatment parameters for hydatid cysts involve both surgical and medical approaches for which it requires a multidisciplinary team of consultant haematologist, anaesthesiologist, pulmonologist and surgeons. The procedures used to treat this uncommon ailment are not conventional, are underreported and should be customized according to the site, size and compliance of the HC (Almutairi and Al-Rajhi, 2018). Complex cysts are best treated surgically, which typically involves thoracotomy and ranges from puncture and aspiration of the cyst's contents to partial excision of the entire organ (Junghanss et al., 2008). In the present case, the treatment involved both surgical approach through thoracotomy and post-operative medical approach involving intravenous antibiotics and Albendazole antihelminthic as it is considered to be the main medical treatment for HC (Almutairi and Al-Rajhi, 2018; Kumar et al., 2012; Tercan et al., 2005).

However, consideration should be given to preventive interventions such as community information campaigns and adequate hand washing after interaction with animals like dogs along with patient education focussing on informing the population at risk about the illness, its carriers, its transmission and the best ways to prevent it (Ben-Abdallah et al., 2019). Hence, this case report concluded that the patient adhered well to both the interventions and to prevent such incidences prophylactic measures such as proper hand sanitation practices after contact with animals like dogs, and awareness campaigns in communities, are essential.

4. CONCLUSION

HC is a public health issue that is frequently seen in developing and underdeveloped nations. Humans contract HC by consuming vegetables and water contaminated by infected dogs. The use of imaging techniques like USG and CT as part of the patient's work-up has become essential. However, in rural health centres with no availability of imaging techniques like USG and CT result in

delay in diagnosis. HC are uncommon infectious condition of the lungs that needs proper and effective management. Postponements in treatment or delay in diagnosis create a cascade of problems that, if left unattended may become life-threatening and hence it is very important for primary care physicians to have awareness about this condition. Surgery is considered as the preferred course of treatment for HCs, regardless of size and whether it is intact or ruptured; also, it is safe to perform, has low morbidity and has a very low fatality rate. Hence, this case report highlights an effective surgical and adherence to medications in an older adult with hydatid cyst resulting in the cure of the disease.

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Author Contribution

All authors equally contributed in this case report.

Informed Consent

Written consent was obtained from the patient.

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Conflict of interest

The authors declare that there is no conflict of interests.

Data and materials availability

All data sets collected during this study are available upon reasonable request from the corresponding author.

REFERENCES AND NOTES

1. Alloubi I. Thoracic Hydatid Cyst: Clinical Presentation, Radiological Features and Surgical Treatment. Principles and Practice of Cardiothoracic Surgery. Firstenberg MS (editor): Intech Open, London 2013. doi: 10.5772/53533
2. Almutairi A, Al-Rajhi S. Case Report of Hydatid Cyst in the Pulmonary Artery Uncommon Presentation: CT and MRI Findings. Case Rep Radiol 2018; 2018:1301072. doi: 10.1155/2018/1301072
3. Aqqad A, Hamdi B, Louhaichi S, Khalfallah I, Attia M, Zairi S, Ammar J, Hamzaoui A. Giant pulmonary hydatid cyst in children. Arch Pédiatrie 2021; 28:273-7. doi: 10.1016/j.arcped.2021.02.017
4. Arroud M, Afifi MA, El-Ghazi K, Nejjari C, Bouabdallah Y. Lung hydatid cysts in children: Comparison study between giant and non-giant cysts. Pediatr Surg Int 2009; 25:37-40. doi: 10.1007/s00383-008-2256-z
5. Bakir I, Enc Y, Cicek S. Hydatid Cyst in the Pulmonary Artery: An Uncommon Localization. Heart Surg Forum 2004; 7:13-5. doi: 10.1532/hsf.954
6. Baruah A, Sarma K, Barman B, Phukan P, Nath C, Boruah P, Rajkhowa P, Baruah M, Dutta A, Naku N. Clinical and Laboratory Presentation of Hydatid Disease: A Study from Northeast India. Cureus 2020; 12:e10260. doi: 10.7759/cureus.10260
7. Ben-Abdallah AK, Zouari M, Haj-Mansour M, Abid I, Ben-Dhaou M, Jallouli M, Mhiri R. Hydatid Cyst of the Lung in Children: A Diagnosis Not to Be Missed. Iran J Public Health 2019; 48:767-769.
8. Darwish B, Alsabek MB, Kakaje A. A complicated pulmonary hydatid cyst resembling a tumour in an adult on PET scan: A case report. J Surg Case Rep 2020; 2020(10):rjaa448. doi: 10.1093/jscr/rjaa448
9. Emlik D, Ödev K, Poyraz N, Kaya HE. Radiological Characteristics of Pulmonary Hydatid Cysts. Current Topics in Echinococcosis. Rodriguez-Morales AJ (editor): Intech Open, London 2015. doi: 10.5772/60884
10. Gattani R, Malhotra G, Deshpande SG, Ramteke H, Nayak K, Salwan A, Bhattacharjee A. Asymptomatic incidental primary pelvic hydatid cyst in a post-menopausal woman: A case report. Medical Science 2023; 27: e12ms2609. doi: 10.54905/disssi/v27i131/e12ms2609
11. Junghanss T, Silva A, Horton J, Chiodini P, Brunetti E. Clinical Management of Cystic Echinococcosis: State of the Art, Problems, and Perspectives. Am J Trop Med Hyg 2008; 79:301-11. doi: 10.4269/ajtmh.2008.79.301
12. Kumar S, Satija B, Mittal MK, Thukral BB. Unusual Mediastinal Dumbbell Tumor Mimicking an Aggressive

Malignancy. J Clin Imaging Sci 2012; 2:67. doi: 10.4103/2156-7514.103057
13. Onal O, Demir OF. The relation between the location and the perforation rate of lung hydatid cysts in children. Asian J Surg 2018; 41:422-426. doi: 10.1016/j.asjsur.2017.04.001
14. Pakala T, Molina M, Wu GY. Hepatic Echinococcal Cysts: A Review. J Clin Transl Hepatol 2016; 4:39-46. doi: 10.14218/JCTH.2015.00036
15. Stojkovic M, Rosenberger K, Kauczor HU, Junghanss T, Hosch W. Diagnosing and staging of cystic echinococcosis: How do CT and MRI perform in comparison to ultrasound? PLoS Negl Trop Dis 2012; 6:e1880. doi: 10.1371/journal.pntd.0001880
16. Tercan F, Kacar N, Kilic D, Oguzkurt L, Turkoz R, Habesoglu MA. Hydatid Cysts of the Bilateral Pulmonary Arteries and Left Ventricle Wall: Computed Tomography and Magnetic Resonance Imaging Findings. J Comput Assist Tomogr 2005; 29:31-33. doi: 10.1097/01.rct.0000153405.09602.c5
17. Yekeler E, Karaarslan K, Yazicioğlu A, Kaya S, Karaoğlanoğlu N. Lobectomy for pulmonary hydatid cyst. Turk J Med Sci 2013; 43(6):1024-1029. doi: 10.3906/sag-1302-16