

## Case Report: Cystic Pulmonary Hydatidosis, a Forgotten Disease

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### Abstract

Human echinococcosis is a zoonotic parasitic disease caused by the larval stage of the tapeworm of the genus *Echinococcus* [1]. Cystic echinococcosis or hydatidosis, results from an infection with *Echinococcus granulosus*, the prevalent form in Uruguay. It continues to be an important cause of morbidity and mortality in tropical and subtropical countries. The World Health Organization (WHO), reports a global prevalence of more than 1 million people, causing 19.300 deaths in 2015 [2].

**Keywords:** Pulmonary Disease; Zoonotic; Cystic; Hydatidosis

### Introduction

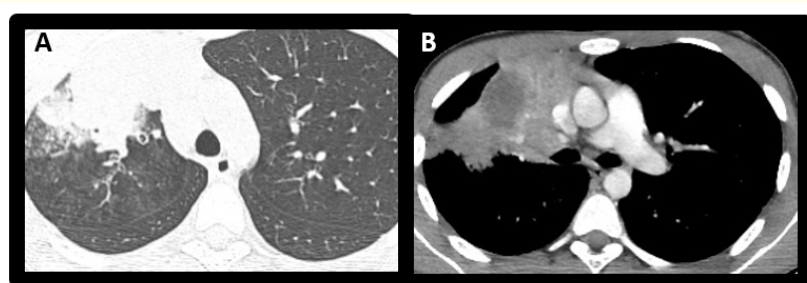
It has a global distribution, with presence in every continent. The Pan American Health Organization (PAHO) states it is highly endemic in rural areas of South America, especially in Uruguay, Argentina, Chile, Perú and Brazil, with a prevalence between 2% to 6% or even more [3].

In Uruguay, the only data registered comes from a specific rural village named La Paloma in Durazno department, which observed a prevalence of 5.6% [4]. Although it is mandatory to notify its diagnosis, it is still considered subdiagnosed.

We report a case of a young male who presented pulmonary hydatidosis with diagnostic and treatment difficulties.

### Case Report

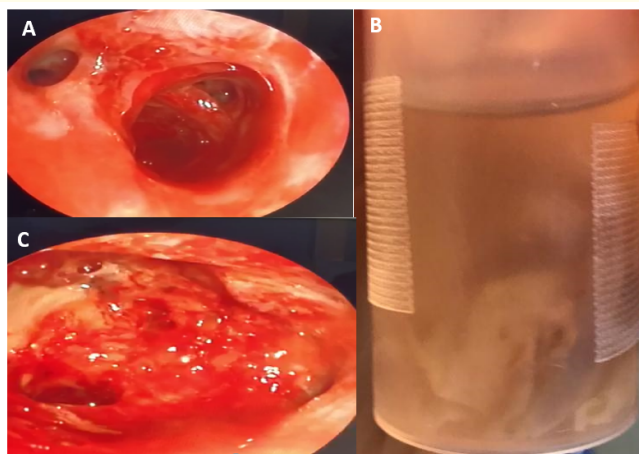
15-years-old male, born and raised in a rural area of Uruguay. His grandmother had a pulmonary hydatid disease during infancy. The patient presented an acute tracheobronchitis that was treated with antibiotics, followed 30 days after by a nonmassive hemoptysis. Chest radiology showed an image interpreted as a pulmonary abscess (Figure 1), therefore, antibiotics were administered for 6 weeks. During treatment, he developed a maculopapular rash with fever (Figure 2). One month later, the fever was back, and a new chest radiology showed no changes. A bronchoscopy evidenced a complete obstruction of the anterior bronchial segment of the right superior lobe, due to the presence of a white membrane that was subsequently removed and analyzed, leaving a residual bronchial cavity (Figure 3). A bacteriologic test was performed which revealed the presence of *Klebsiella pneumoniae* and *Streptococcus aureus*. Histopathologic analysis showed a hyaline membrane, pathognomonic finding of pulmonary hydatidosis (Figure 4). The patient was prescribed Albendazole 15 mg/kg/day before surgical treatment.



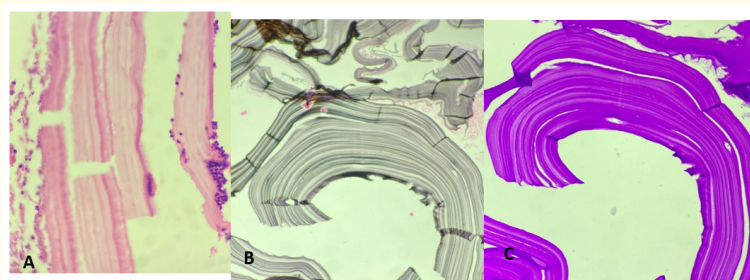
**Figure 1:** Radiograph of the chest, nonmassive hemoptysis. (A) Pulmonary window with consolidation of the anterior segment of the right upper lobe. (B) Mediastinal window inhomogeneous consolidation showing hydatid cyst drawn in pencil.



**Figure 2:** Maculopapular rash.



**Figure 3:** Fibrobronchoscopy: (A) Dilation of the anterior segment of the right upper lobar bronchus. (B) Material extracted. (C) Residual endobronchial cavity in segment 3 right upper lobe, biopsy.



**Figure 4:** Histopathology of extracted membranous material: hydatid cyst cuticular membrane. Hyaline, anhistic and striated. Compatible with the cuticular membrane of hydatid cysts. (A) Hematoxylin-eosin; (B) Methenamine argenticum; (C) PAS (Periodic Acid/Schiff) positive.

## Discussion

Hydatidosis is an infection of herbivorous animals and humans caused by parasites of the genus *Echinococcus*. There are four species, the prevalent form in Uruguay is *Echinococcus granulosus*. It may affect different organs, but most commonly targets the liver, followed by the lungs [5]. The biological state of the hydatid determines its capacity to affect organs. This state also affects the clinical presentation of the disease, laboratory results and therapeutic plan [6].

The life cycle of *E. granulosus* involves multiple participants and stages. Once humans ingest the eggs from canine feces, they release oncospheres that penetrate the intestinal wall and migrate through the portal system into various organs, affecting the liver initially. If

they keep traveling through the inferior vena cava, they reach the heart and potentially the lungs. That stage is known as the primitive hydatidosis, giving rise to the next form, the hydatid cyst, which includes the adventitial layer (from a host inflammatory reaction). Once in the lungs, it generally resides in the lower lobules or the right hemithorax [6]. The cyst may break (bronchial and/or pleural rupture) or may become infected with bacteria [6].

In this particular case, we have two principal problem as: the diagnosis and the treatment.

About the diagnosis, the primitive hydatidosis infection was asymptomatic. During this stage, the diagnosis is usually made as an incidental finding upon chest radiology [6]. The patient’s first clinical sign was hemoptysis, pointing out the rupture of the cyst, that was facilitated by the acute tracheobronchitis. But that diagnostic was not so easy at first time, and it has been interpreted as a pulmonary abscess. Additionally, the patient manifested a systemic reaction to the disease, seen clinically as a skin rash (Figure 2), that may be explained as a hypersensitivity reaction triggered by the release of antigens from the ruptured cyst [6,7]. That was also not interpreted correctly in first moment, and it was thought an allergic reaction to the antibiotic.

The rupture of the cyst allows the colonization of bacteria (*S. aureus* and *K. pneumoniae*) and is what usually makes the patient seek for medical evaluation. The bronchoscopy results in a bronchial dissemination of the parasite and the removal of the cyst leaves a cavitory lung lesion and bronchiectasis (Figure 5).

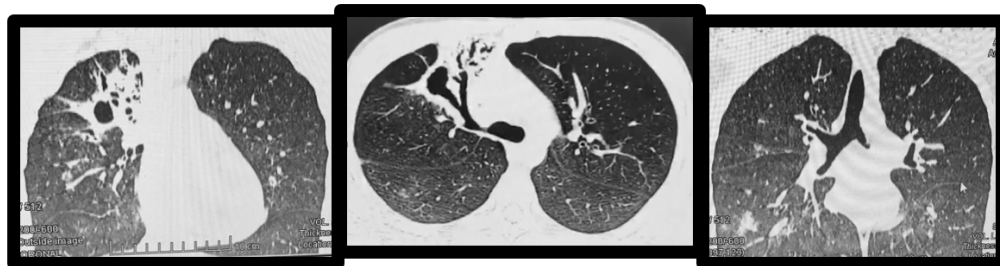


Figure 5: Sequel cavity with bronchial dilation.

The importance of considering the different stages of the disease lies in the fact that the death of the parasite does not put an end to the disease but creates an even more challenging and serious situation. This being said, the goal is to reach an early diagnosis in hope of preventing rupture of the cyst and posterior systemic involvement in multiple locations [7].

The diagnosis of cystic echinococcosis is obtained by analyzing its epidemiology, clinical presentation, imaging, serology and, in certain cases, a pathological anatomy evaluation.

The epidemiology of the disease is of the utmost importance to prevent the use of invasive procedures that risk rupturing the cyst, as happened with this patient. It is also key to bear in mind that the disease develops in an agricultural environment and that the most frequent route of transmission to humans is by accidental consumption of infected food, soil or water [3].

Clinical presentation depends on the size of the hydatid cyst, the hydatid itself, and also on its integrity and risk of rupture that can cause dissemination.

Ultrasound is the Gold Standard in the diagnosis of liver involvement. The WHO classifies the cysts based on ultrasonographic images, categorizing them by their type and size, which determine the severity and further treatment [8].

Serological tests are useful in detecting the disease, however, a negative result does not rule it out [9,10]. There are numerous diagnostic serological techniques, recommending: indirect hemagglutination (IHA), indirect immunofluorescent (IFI) and ELISA. In this case will be useful for the diagnostic differential with the abscess at first time. The sensitivity of any of these tests depend on the location of the cyst and its physiological state. Moreover, immunodiagnostic tests are more sensitive in hepatic hydatidosis, rather than in pulmonary hydatidosis.

Although histologic diagnosis is usually not necessary, it could be found accidentally or in post surgery. That is what happened with this case, and it was one of the reasons of sharing it.

### Treatment, control and prevention

Surgery is recommended in every stage of the disease, as it involves a complete removal of every component of the cyst (hydatid, adventitial layer and the cavitory lung lesion) [11]. The most convenient should be to extract it in early stages; diagnostic delays (like in this case) carries into difficulties in the surgical technique. Medical treatment, pre and post-surgery, with Albendazole in a dose of 10 - 15 mg/kg/day, has gained relevance, due to its efficacy and effectiveness both as a protoscolicide, as well as reducing the size and intracystic tension [12].

This particular case was studied by a multidisciplinary team, mainly due to its diagnostic difficulties. It was decided that a medical approach would be the best option, followed by surgical intervention.

### Conclusion

Lastly, it is important to state that this zoonotic disease must be controlled by sanitary measures in every affected country. In Uruguay the Zoonotic Committee is responsible of establishing guidelines in this regard. Measures adopted include educating rural population about this disease and its control, insisting on not feeding dogs with raw viscera, controlling dog population by veterinary services, restricting home slaughter of livestock and sheep and surveilling human hydatidosis, followed by a mandatory notification to the sanitary authorities [13,14]. These measures tend to stimulate the control of the disease by local communities and prevent its transmission. As it is considered a major public health problem, an urgent control is key to alleviate its effects on the health system as it entails high costs along with a lengthy hospital stay for the patient [7].

### Bibliography

1. Eckert J., *et al.* WHO/OIE Manual on Echinococcosis in Humans and Animals: A Public Health Problem of Global Concern 104 (2002).
2. Pavletic CF., *et al.* "Cystic echinococcosis in South America: a call for action". *Pan American Journal of Public Health* 41 (2017): 1.
3. Organization W health. WHO (2020).
4. Cohen H., *et al.* "Human cystic echinococcosis in a Uruguayan community: A sonographic, serologic, and epidemiologic study". *American Journal of Tropical Medicine and Hygiene* 59.4 (1998): 620-627.
5. Brunetti E., *et al.* "Expert consensus for the diagnosis and treatment of cystic and alveolar echinococcosis in humans". *Acta Tropica* 114.1 (2010): 1-16.
6. Atias A. "Parasitología Médica". In: *Parasitología Medica*. 1998th edititon. Mediterráneo (1998): 339-354.

7. Souza-Santos R. "Zoonosis y enfermedades transmisibles comunes al hombre y a los animales". *Cadernos de Saude Publica* 21.3 (2005): 988-989.
8. Macpherson CNL, *et al.* "International classification of ultrasound images in cystic echinococcosis for application in clinical and field epidemiological settings". *Acta Tropica* 85.2 (2003): 253-261.
9. Biava M., *et al.* "Laboratory Diagnosis of Cystic Hydatid Disease (2001): 10-14.
10. Ortona E., *et al.* "An update on immunodiagnosis of cystic echinococcosis". *Clinical and Developmental Immunology* (2012): 165-171.
11. Agudelo Higueta NI and Brunetti E MCC. "Cystic echinococcosis". *Journal of Clinical Microbiology* 48.5 (2016): 42-43.
12. Becerril MA. "Parasitología Médica". In: *Parasitología Medica*. 4<sup>th</sup> Edition (2014): 187-194.
13. Uruguay CZ. *Zoonosis Uruguay*.
14. Dora JF, *et al.* *Hidatidosis - 3er Reunión del Proyecto Subregional Cono Sur de Control y Vigilancia de la Hidatidosis* (2006).

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