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### RESEARCH ARTICLE

#### CARDIAC HYDATID CYST: A RARE CASE REPORT

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#### Abstract

**Introduction:** Hydatidosis, an anthroozoonosis prevalent in many sheep-farming countries, continues to pose a significant health concern. Cardiac localization represents a rare and distinctive manifestation, accounting for less than 2% of total cases. This report details a case where surgical intervention, coupled with antiparasitic chemotherapy, was used for effective treatment.

**Patients and Methods:** A 36-years-old female, residing in a rural area renowned for agricultural and livestock activities, was under cardiology supervision due to rheumatic polyvalvulopathy. Having undergone hepatic hydatid cyst surgery one year earlier, The reason for her consultation in cardiology was exertional dyspnea, left thoracic pain, palpitations, afebrility, and asthenia. The disease diagnosis relied on imaging findings. Treatment involved the urgent surgical excision of the cyst, followed by the oral administration of albendazole at 400mg x 2 per day, initiated 48 hours postoperatively. With no contraindications, the patient underwent continuous biological monitoring during hospitalization, reporting no adverse effects. The antiparasitic regimen persisted for 4 months.

**Conclusion:** Albendazole, whether used as a standalone therapy or in combination, remains a highly effective treatment. It can either substitute for or complement surgical approaches, offering the flexibility of extended treatment durations. Vigilant monitoring of hepatic function is imperative in these instances to identify potential adverse effects. Two-dimensional transthoracic echocardiography plays a pivotal role in tracking patient progress and evaluating treatment efficacy. Prevention stands out as the most effective strategy against hydatid cysts, emphasizing collective awareness and proactive patient management to avert cyst dissemination.

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#### Introduction:-

Hydatidosis, an anthroozoonosis caused by the larva of Echinococcus granulosus, specifically manifests as cystic echinococcosis (CE) according to the World Health Organization (WHO). This parasitic disease remains prevalent in various sheep-farming regions, including South America, Central Asia, Australia, Eastern Europe, and North

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America [1,2,3]. In Africa, it notably impacts North Africa and East Africa, with Kenya recording the highest incidence globally at 220 cases per 100,000 inhabitants. In Morocco, a country characterized by sheep farming, the conducive humidity for larval development, coupled with suboptimal hygiene practices and interactions between humans and the canine species, contributes to the transmission. In 2006, the Ministry of Health and Social Protection reported an incidence of 4.5 cases per 100,000 inhabitants [4,5].

The life cycle of the parasite involves the sheep as the intermediate host and the dog as the definitive host. Humans inadvertently become intermediate hosts via the ingestion of eggs present in dog feces, either directly or indirectly. Following hepatic transit, the *Echinococcus granulosus* larva reaches the right cardiac cavities through the venous return (IVC) and follows the cardiac circulation (RA>RV>LUNG>LA>LV) before entering the general circulation [6,7,8]. Subsequently, it may migrate to various organs, with the liver being the most affected (50 to 70% of cases), followed by the lung (25 to 40%), and less frequently the spleen (2 to 5%), kidney (2 to 5%), bones, brain, and other rare locations [9,10]. Notably, cardiac involvement is considered rare, accounting for less than 2% compared to other hydatid locations [11, 12, 13].

The primary aim of our study is to present a compelling case of cardiac hydatid cyst (HC) in a young woman admitted to the Cardiac Surgery Department at the Mohamed V Military Teaching Hospital in Rabat. We meticulously outline the circumstances surrounding its discovery and diagnosis, the chosen treatment approach, the subsequent evolution of the condition, and provide a comprehensive review of existing literature on this rare manifestation.

### **Patients and Methods:-**

The patient, a 36-year-old mother of two residing in the rural region of Khemisset, known for its agricultural activity and livestock farming, was under cardiology follow-up for rheumatic polyvalvulopathy. She had previously undergone surgery for a hepatic hydatid cyst one year ago.

Admitted to the cardiac surgery department following a consultation due to progressively worsening exertional dyspnea, left thoracic pain, and palpitations, all occurring in the context of afebrility and asthenia, the patient's cardiopulmonary examination revealed no significant abnormalities. The electrocardiogram indicated a regular sinus rhythm at 119 beats per minute, with a well-placed heart axis, a constant PR interval of 120 ms, and negative T waves inferiorly and laterally (the postoperative ECG remained unchanged).

Chest X-ray displayed a normal cardiac silhouette with good pleuro-parenchymal transparency. Transthoracic echocardiography revealed the presence of a 74x51mm extracardiac mass with a non-vascularized tissue appearance, heterogeneous echostructure, and some empty echoic areas. It was located at the cardiac apex, exerting a mass effect on the apical region of both ventricles.

Thoracic angioscanner highlighted a lesion associated with the left ventricle (LV) apex, measuring 6.27 cm in the exoventricular development without intracystic nodulations but with small calcifications at its periphery. This suggested a hydatid cyst originating from the LV apex, with no other anomalies detected in cardiac cavities or walls. Abdominal CT scan revealed no other visceral locations.

Biological assessment showed negative troponin, normocytic normochromic anemia, and negative hydatid serology. Regarding treatment, the foundation was surgical, involving extracorporeal circulation (ECC) for the excision of the cyst, which had taken the form of a left ventricular intra-pericardial lateral mass, with a cleavage plane between the mass and the left ventricle.

In addition to surgery, medical treatment included the prescription of Albendazole orally at 400mg x 2 per day, initiated 48 hours postoperatively. Hepatic function remained normal throughout the hospitalization, and this dosage was maintained for 4 months post-discharge.

### **Discussion:-**

Cardiac hydatidosis is a rare condition, accounting for 0.2% to 2% of reported cases of hydatid cysts, typically associated with anterior or contemporary visceral involvement (pulmonary, hepatic, or mediastinal) [49]. Approximately 60% of cardiac CE cases are asymptomatic, especially when located in the left ventricle [14,15].

Discovery often occurs incidentally during systematic examinations (chest X-ray, echocardiography) or is prompted by interview elements (rural context, contact with dogs, and sheep). Functional symptoms are nonspecific, varying based on cardiac localization, cyst size, and number, commonly presenting as precordial pain, palpitations, chest angina, syncope, exertional dyspnea, in addition to hypersensitivity, skin rashes, and pruritus [49]. In our case, the patient had several risk factors: coming from a rural environment, a history of contact with dogs, and a previous hepatic hydatidosis. Clinically, our patient was symptomatic (dyspnea, thoracic pain, and palpitations).

Paraclinical examination techniques play a crucial role in diagnosing cardiac CE in 80% of cases [36]. Chest X-ray reveals cysts as round or oval opacities [37][38]. Two-dimensional transthoracic echocardiography is the preferred technique, demonstrating them as highly suggestive echolucent formations with thin walls [16][17]. It provides precise localization, lesion cystic relationships, and assessment of its impact on adjacent structures. The latest generation of multi-slice thoracic CT confirms a positive diagnosis and establishes an extension assessment to adjacent structures [39]. Nuclear Magnetic Resonance Imaging (MRI) has a major role in confirming the diagnosis in cases of uncertainty or discrepancies between ultrasound and CT results. ECG must be systematically performed, potentially revealing conduction disorders such as complete atrioventricular block, reversible after surgery [18]. It may also show rhythm disturbances [19] depending on the cyst's location, evolutionary stage, and potential complications, although these signs are nonspecific, and ECG disturbances occur in 40% of cases [40]. The extracardiac mass detected by transthoracic echocardiography at the cardiac apex raised the possibility of hydatid recurrence, prompting further examinations with thoracic angioscanner and CT for confirmation and to define the stage and different locations.

Biology plays a crucial role in confirming the disease diagnosis, although direct examination (search for parasitic material: protoscolices or membranous fragments) is rarely relied upon in practice, limited to pulmonary cyst cases (broncho-pulmonary lavage or pulmonary vomit). Biopsies and punctures constitute an absolute contraindication due to the almost inevitable risk of cyst dissemination to other locations [41]. Some nonspecific tests, such as elevated eosinophilia, increased total IgE, or inflammatory syndrome (elevated ESR and neutrophils), can be observed. Other specific examinations, usually immunological diagnostic tests such as the search for specific IgE or antiparasitic serology using various techniques, should consider that some cardiac CE cases may be serologically silent, either due to calcification or small size, or in patients with immune deficiencies, resulting in a falsely negative serology [42]. False positives may also occur in the presence of other helminthiasis (alveolar echinococcosis and cysticercosis) or in cases of neoplastic pathology or immune disorders [43]. Interpretation of results should rely on at least two techniques, one quantitative and the other qualitative [20]. The French National Authority for Health (HAS) recommends using two techniques for screening [indirect hemagglutination (IHA) and enzyme-linked immunosorbent assay (ELISA or EIA)], in addition to confirmation by immunoblot and quantitative technique for patient monitoring [21]. Hydatid serology is positive in only 50% of cardiac hydatidosis cases [22].

In our patient's case, the negative High-Sensitivity Troponin assay helped rule out myocardial infarction risk. Subsequent examinations revealed normocytic normochromic anemia. Serology was systematically requested due to the patient's hydatid cyst history, resulting in a negative outcome but not excluding the diagnostic hypothesis. However, parasitological examination was not requested.

The treatment of CE is primarily surgical, aiming to prevent complications related to compression or the potentially fatal risk of cyst rupture. It involves excision through pericystectomy or cystectomy [23,24], with encouraging postoperative results [25].

Drug treatment is provided by the Benzimidazole class, with Albendazole being the treatment of choice. It reduces cyst size, parasite viability, and recurrence frequency [44]. Its efficacy varies but is insufficient for a complete cure, making it an alternative to surgery when contraindicated. It is also used in preventing the risk of cyst dissemination when numerous, calcified, or small, as well as in preventing local recurrences. Some authors recommend it systematically postoperatively [45].

Regarding dosage, remarkable results have been reported at a dose of 15 mg/kg twice daily for a 5-month course [26]. Albendazole's cysticidal activity occurs during the first 2 or 3 months of treatment; if no cyst size reduction is observed after 2 months, its continuation becomes unproductive [46]. Prophylactically, Albendazole is prescribed in addition to surgery, preoperatively (2 courses) inducing cyst sterilization to minimize the risk of dissemination during surgery, and postoperatively (at least 2 courses) in cases where live cysts are discovered or in cases of cyst

rupture or hydatid fluid release during the procedure. For curative chemotherapy, the average treatment duration ranges from 6 to 12 months, with a daily dose of 800 mg in 2 doses (>60 kg) and 15 mg/kg in 2 doses (<60 kg) in 28-day courses separated by 14-day intervals to reduce toxicity. Continuous treatment beyond 9 months has shown improved effectiveness [47]. Several other cases of cardiac hydatid cysts have been reported associated with other locations such as the brain, lungs, liver, and kidneys. In each case, the effective protocol involved Albendazole 400mg, once or twice daily for 28 days, in 3 to 5 courses, spaced 14 days apart. In some instances, it was prescribed at 200mg twice daily for 5 to 24 successive months, administered in combination with Praziquantel at 40mg/kg daily.

Regarding treatment-related intolerances and adverse effects, elevated transaminases have been reported in 15% of cases, with a potential risk of neutropenia and abdominal pain, which may also signal possible abdominal cyst rupture in cases of disseminated cysts. Pharmacokinetically, Albendazole induces enzymatic activity in the cytochrome P450 group, CYP1A1, and CYP1A2 at the transcriptional level [27]. This induction should be considered for its potential clinical impact on the metabolism of other drugs, necessitating dose adjustments. It has been reported that the combination with St. John's Wort, a widely used plant extract as an antidepressant, activates Albendazole metabolism [28]. Concurrent use of cimetidine and praziquantel with Albendazole also induces enzymatic activity, resulting in decreased plasma Albendazole levels due to increased metabolism [29]. This effect has also been described when combined with grapefruit juice [30].

While Albendazole does not have specific contraindications for prescription, its use in pregnant women is not recommended (demonstrated teratogenicity in rats and rabbits) [48]. Treatment monitoring is mandatory throughout the course, including blood tests such as complete blood count (CBC), transaminases (bi-monthly during the first month, then monthly and quarterly after the third month of treatment), serology (IgG search by ELISA technique, with no added value), and radiology (the most reliable means of monitoring): changes in cyst size, density, shape, or intracystic echogenicity. The risk of recurrence remains possible after 2 or 3 years of recovery, warranting extended follow-up possibly for life, as recommended by the WHO, to detect potential morphological changes of the parasite and ensure better management in case of relapse, which remains sensitive to Albendazole [31, 32, 33, 34, 35].

The patient's management began with surgical excision of the cystic mass, followed by antiparasitic chemotherapy initiated 48 hours after the intervention to complete the treatment and prevent recurrences. Hepatic function remained normal throughout the hospitalization (15 days). The choice of Albendazole was justified by its rapid digestive absorption and good diffusion to cysts, and our patient had no contraindications or drug interactions. The patient's evolution was favorable.

During the follow-up examination after one-month, transthoracic echography was prescribed to assess treatment and search for potential recurrences. Additionally, biochemical and complete blood count examinations showed no notable anomalies. Our patient was reviewed after 3 months with the same examinations (echographic and biological) for the complete cessation of medical treatment.

### **Conclusion:-**

In conclusion, hydatidosis continues to prevail in endemic areas, with its cardiac manifestations being rare but the most feared due to their nonspecific clinical presentation. Prevention remains the most effective means to combat the disease, relying on collective awareness to interrupt the biological cycle of the parasite through early diagnosis and patient management before any metastatic dissemination of cysts.

Two-dimensional transthoracic echocardiography plays a crucial role in detecting cysts and monitoring patient progress, while MRI and CT scans are also significant, especially in ruling out alternative diagnoses for cystic masses.

Albendazole, whether used as monotherapy or in combination, stands out as the most effective treatment, serving as a replacement or complement to surgical intervention, with variable treatment durations. We should notice the importance of patient follow-up during the treatment to assess efficacy and monitor potential side effects.

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