CASE REPORTS

UNUSUAL MULTIFOCAL LOCALIZATIONS OF HYDATIDOSI

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Abstract

Hydatid disease is a major public health problem in Mediterranean countries, North Africa (including Morocco), Asia, Australia, New Zealand, and certain South American countries, where agricultural and pastoral practices remain largely traditional. This parasitic disease results from infection with the larval stage of Echinococcus granulosus. Diaphragmatic involvement is rare, even in regions where the disease is endemic.

We present the case of a 31-year-old patient who had previously suffered from hepatic and renal hydatid infection and who developed multifocal disease affecting the lung parenchyma, diaphragmatic tissue, and pancreas two years after the initial diagnosis. The therapeutic management of the thoracic lesions consisted of surgery followed by prolonged administration of benzimidazole-based pharmacological treatment, specifically albendazole.

Keywords: *hydatid disease, pulmonary, diaphragmatic, pancreas, thoracic surgery, albendazole.*

INTRODUCTION

Human echinococcosis, also known as hydatid disease or hydatidosis, is an anthropozoonosis caused by a parasite, specifically the tapeworm Echinococcus. Cystic echinococcosis is caused by the Echinococcus granulosus species complex. This parasitic disease is endemic in livestock-raising regions, particularly in countries in the Mediterranean basin, North Africa, Asia, Australia, and New Zealand, where the agro-pastoral sector remains relatively unindustrialized. Hydatid disease is also endemic in certain South American countries such as Argentina, Brazil, Chile, Peru, and Uruguay, with an incidence rate ranging from 0.012 to 13 per 100,000 inhabitants[1].

Morocco, a country with a long tradition of livestock farming, is one of the countries affected in the Maghreb region, particularly among rural populations. The parasite's reproductive cycle is complex and can be summarized in two hosts: dogs and other canids are the definitive hosts, and herbivores are the intermediate hosts. Humans, accidental intermediate hosts, become infected by ingesting microscopic eggs present in the feces of infected dogs, mainly through hand-to-mouth contact or by ingesting water or food contaminated with dog feces. The most common visceral location is the liver (50 to 77%), followed by the lungs (15 to 47%). Diaphragmatic hydatid disease is rare, with an incidence of between 0.8 and 1.5% of all thoracic localizations[1]. Less common locations for hydatid cysts are the bones, kidneys, spleen, muscles, and central nervous system.

However, although rare and generally asymptomatic, hydatid cysts can develop in other organs: bones, brain, kidneys, pancreas, and heart. Multiple locations, with concomitant pulmonary, diaphragmatic, and pancreatic involvement, are even rarer in the literature. Due to their relatively symptomatic nature, pulmonary hydatid cysts are often diagnosed at the stage of complications. Clinical manifestations can therefore be misleading. Clinical symptoms are generally related to the progressive nature of the cystic lesion, its compressive effect on intrathoracic organs, secondary infection, or rupture, depending on whether it is located in the pleura or the respiratory tract.

Cough, the main symptom (52%-62%), can be associated with dyspnea (large hydatid cyst), hemoptysis, chest pain or fever. Immune-allergic symptoms may also be

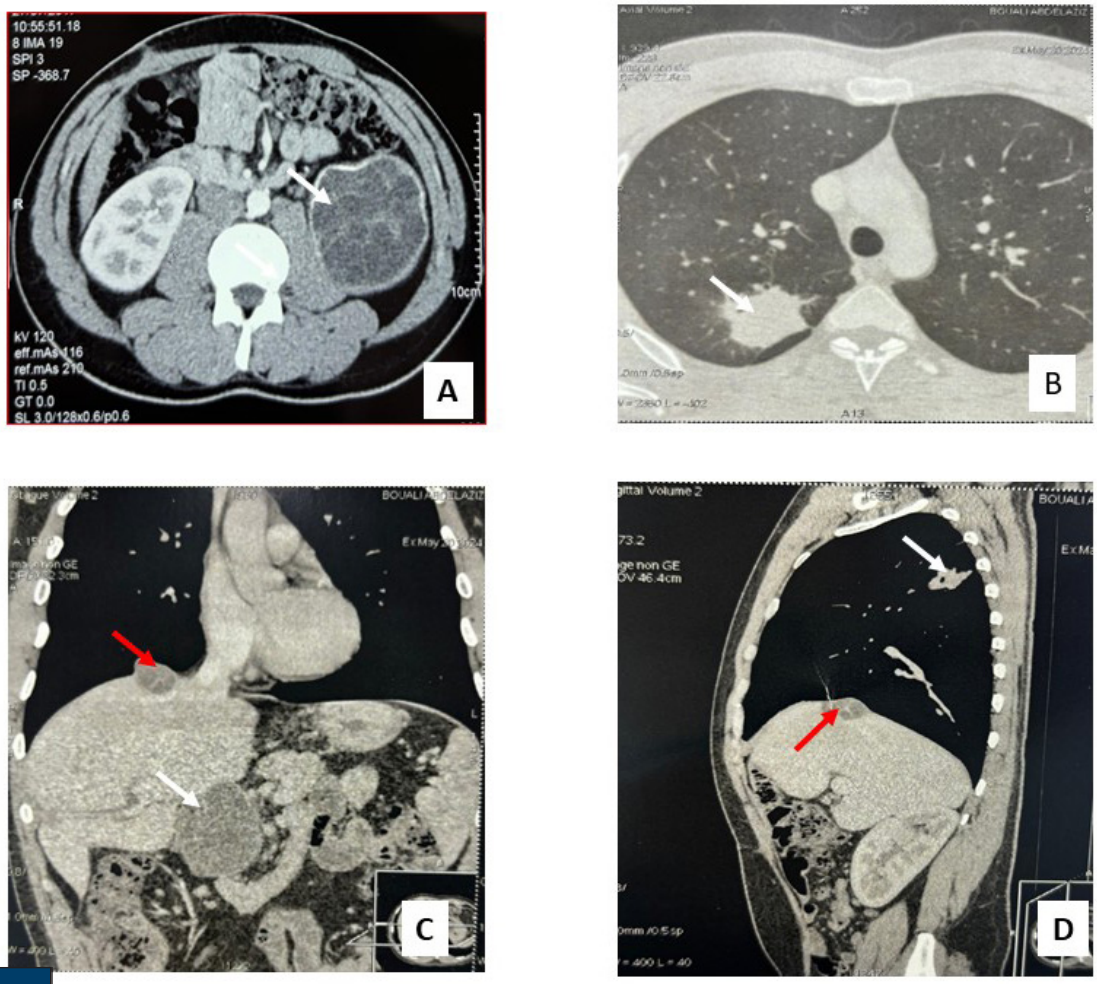

Figure 1

(A) Abdominal CT scan: left renal hydatid cyst before previous surgeries (white arrow). (B) Thoracic CT: mass of the upper right lobe (white arrow). (C) Thoraco-abdominal CT: diaphragmatic (Red arrow) and pancreatic (white arrow) hydatid cyst. (D) Thoraco-abdominal CT scan showing a diaphragmatic (Red arrow) and pulmonary hydatid cyst (white arrow)

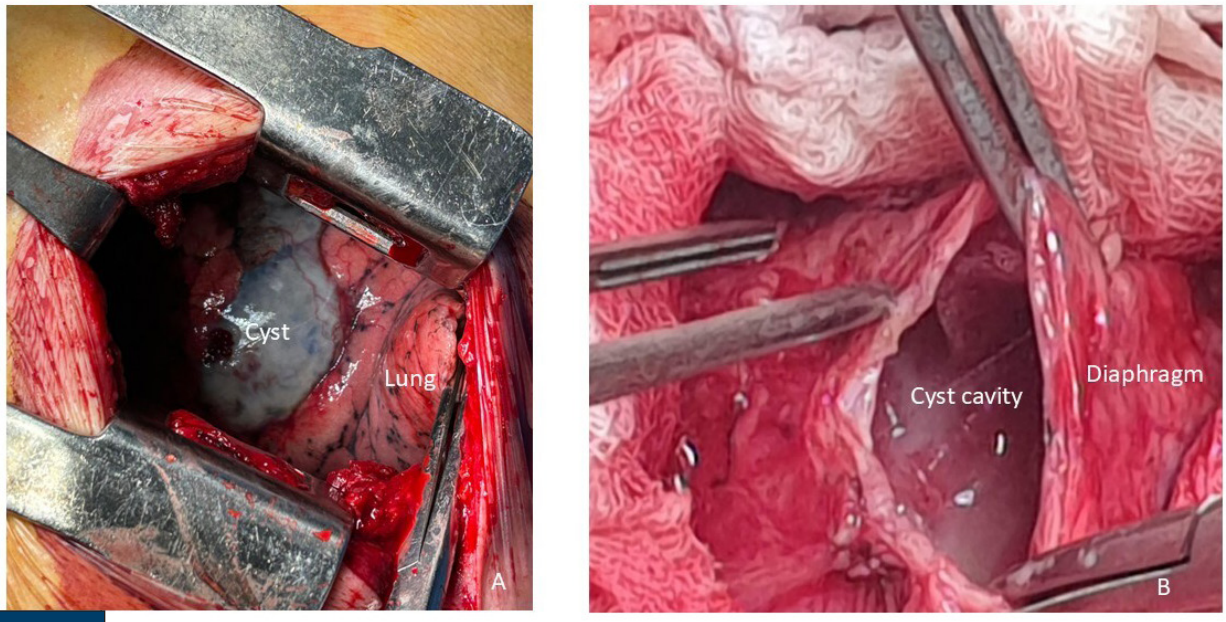

Figure 2

Figure 2: Operative views (A) lung hydatid cyst. (B) diaphragmatic hydatid cyst after extraction of the hydatid membrane

present and develop in a low-level manner. Hydatid vomiting, also called hydatidoptysis, is a pathognomonic phenomenon of intrabronchial rupture of a hydatid cyst[1].

CLINICAL CASE

A 31-year-old male patient complains of a persistent cough for one month, with an episode of blood-streaked sputum. The cough was predominantly nocturnal, refractory to antitussive treatment and evolving in a state of apyrexia. The patient denied any contact with dogs or other domestic animals. No personal history or relatives suffering from tuberculosis was mentioned. In 2016 and 2023, the patient underwent surgical and medical treatment for hepatic and renal hydatid cysts (total left nephrectomy) respectively. The patient received long-term medical treatment with albendazole after the two surgical procedures, the duration of which remains unclear. On admission, clinical examination was unremarkable, with except for scars from previous surgeries.

Taking into account the previous medical history, the patient underwent a chest X-ray and a thoracoabdominal CT scan with intravenous injection of contrast. Imaging revealed a lung parenchymal condensation zone (01), originating from the right upper lobe (RUL) (Fig.1:B-D), suggesting a probable infectious origin. In addition, there were two (02) masses suggestive of cystic lesions on the diaphragm and the head of the pancreas(Fig.1:C-D)

The overall laboratory work-up showed a mild hepatic cytolysis without associated cholestasis. The other biological assessment tests were unremarkable. Bronchial secretions tested negative for scolex and acid-fast bacilli (AFB) after fibroscopy. Hydatid serology was positive. These results, combined with the patient's medical history, suggested a multifocal hydatid disease.

Treatment, after patient consent, consisted of surgical management followed by medical treatment using albendazole therapy. The patient underwent a conservative right posterolateral thoracotomy. Exploration of the pleural cavity revealed a generally good-quality lung, with fine adhesions at the level of the pulmonary cystic lesion (Fig. 2:A). After protecting the operating field using soakeds compresses in scolicide solution (in our case hydrogen peroxide), we performed a cystectomy using the Barrett technique. This technique is based on puncture-aspiration of the cystic contents followed by cystectomy, extraction of the membrane, and padding of the residual cavity.

For the diaphragmatic lesion, we performed a resection of the protruding dome (Fig. 2:B), followed by a diaphragmatic reconstruction using separate stitches with non-absorbable sutures. Although, no obvious contamination of the pleural cavity and intrathoracic organs during the intervention, and due to the recurrent nature of hydatid disease, we proceeded at the end of the operation to an abundant lavage of the pleural cavity with serum mixed with oxygenated water. No intraoperative complications were observed. Postoperative management was uneventful, with early walking and removal of the chest tube 72 hours after surgery. The patient was initiated on Albendazole therapy 48 hours post-operatively at a dosage of 400 milligrams twice daily, administered in 28-day cycles with 14-day intervals, for a total duration of 6 months. Post-surgical follow-up protocol consisted of monthly clinical evaluations with concurrent complete blood count and liver function tests from hospital discharge

DISCUSSION

Hydatid disease is a major health problem in Morocco. It can develop in any organ, with a strong predilection for the liver (50 to 77%) and lungs (15 to 47%)[1]. Cases of hydatid disease affecting multiple organs are increasingly being described in the literature. However, simultaneous multifocal involvement of the lungs, diaphragm, and pancreas is exceptional. Diaphragmatic hydatid disease accounts for only 0.8% to 1.5% of intrathoracic hydatid locations, while pancreatic involvement, which is even rarer, is reported in 0.3% to 2% of cases[2].

In 70% of cases, pulmonary hydatid disease is solitary and preferentially affects the lower lobes, mainly on the right side[1]. Several anatomical, physiological, and circulatory reasons explain this preferential localization. From a physiological point of view, the western part of the lung could explain the preferential localization of Echinococcus granulosus at the base. Cardiac output distribution is uneven depending on the patient's position, with the bases receiving more perfusion when the patient is sitting or standing[1]. In addition, blood flow is often greater in the right lung, which promotes the capture of parasitic embryos. Secondary hydatid disease results from the dissemination of fertile hydatid elements (scolex or daughter vesicles) released from a primary cyst. This can occur through direct rupture of the serosa or an organ, through secondary hydatid embolism due to vessel rupture, or through secondary bronchial hydatid disease due to endobronchial dissemination[1]. In our case, the presence of hydatid cystic lesions above and below the diaphragm could be due to secondary embolic disease, particularly given the patient's history.

Radiological imaging plays an important role in the diagnosis and surgical management of hydatid disease. Chest CT scans can be used to determine whether surgery should be conservative or radical with regard to the lung parenchyma[1]. Abdominal and hepatic ultrasound, followed by magnetic resonance imaging (MRI), are the most sensitive diagnostic tools for pancreatic hydatid disease, and several blood tests are available to detect echinococcosis-specific antigens and serum antibodies. The ELISA test, which is generally positive, has a combined mean specificity and sensitivity of 90%, with a diagnostic accuracy of 92.3%[4].

Surgery is always the first-line treatment for hydatid disease, provided the patient is operable. Preservation of the lung parenchyma is the cornerstone of surgical management. The aim is to remove the hydatid cyst, treat the residual cavity, manage complications, and prevent recurrence[1]. The surgical

approach depends on the location and size of the lesions.

In our case, the approach chosen was a conservative right posterolateral thoracotomy via the 5TH intercostal space. This approach allowed for better assessment of the lesions located in the posterior segment of the RUL and at the diaphragm, thus limiting the risk of intraoperative dissemination.

The thoracoabdominal CT scan revealed a well-defined peripheral cystic formation with heterogeneous contents measuring 70 mm in the major axis and 42 mm, located in the posterior segment of the right lobe (Fig. 1: B, D). At the level of the diaphragm, a multivesicular cystic lesion was observed, measuring 40 mm x 30 mm and located in its tendinous center, with no underlying hepatic involvement (Fig. 1: D). This rare and difficult-to-diagnose location could be attributed to lactic acid produced by muscle contractility. This lactic acid prevents the parasite from settling and developing in muscle tissue[2]. Treatment is mainly surgical, although percutaneous aspiration remains an option for patients who cannot undergo surgery. Depending on whether the cyst has developed on the upper or lower surface of the diaphragm, it will be approached by thoracotomy or by median or subcostal laparotomy. However, a combined approach may be considered in certain specific cases.

In our case, the cyst developed on the thoracic surface of the diaphragm, which, in addition to the cystectomy performed on the right upper lobe (RUL), allowed us to use only the thoracic approach. The procedure to be followed depends largely on the size of the cyst.

For large cysts, resection of the prominent dome seems to be the most appropriate solution, as it minimizes the risk of posterior diaphragmatic hernia or rupture, even after muscle or prosthetic repair. Cystectomy is performed for small diaphragmatic hydatid cysts. As there is less tissue loss, simple repair with non-absorbable sutures and, in exceptional cases, diaphragmatic reconstruction can be performed. Our patient underwent cystectomy with diaphragmatic suturing using non-absorbable sutures. This surgical procedure allowed complete resection of the cystic lesions without contamination of the pleural cavity.

Among extra-thoracic locations, pancreatic cysts remain rare. As pancreatic cysts are isolated in most cases, diagnosis is generally very difficult. As in our case, most cystic lesions are located in the cephalic position (57%). Other locations, involving the body (24%) and tail (19%) of the pancreas, are also observed [3,4].

In our case, after multidisciplinary discussion with the visceral surgery department, pancreatic surgery was scheduled within 30 days of the operation, subject to favorable progress and prior agreement from the anesthesiologist.

Medical treatment of echinococcosis has many advantages, particularly as an alternative or complement to surgery. It reduces the parasite load, prepares patients for surgery, allows for the management of inoperable or high-risk cases, prevents recurrence, and improves patients' quality of life. Benzimidazoles are the most effective molecules, particularly albendazole at a dose of 10 mg/kg/day, preferably 2 to 4 weeks before surgery, then continued after the operation. In our case, the size, peripheral nature, and resectability of the cystic lesions, as well as the initial hepatic cytolysis, were all arguments in favor of postponing preoperative medical treatment.

Some authors recommend that medical treatment be administered systematically after surgery. As Kabiri et al. have established, the duration of treatment is not clearly defined in the literature[1,5]. In our case, the patient received an intermittent protocol based on albendazole, at a dose of 400 mg every 12 hours for 28 days with 14-day breaks for 6 months. The results must be evaluated in the long term, generally 12 to 18 months after the start of treatment[5].

CONCLUSION

Multifocal hydatid disease affecting the lungs, pancreas, and diaphragm is a rare form of echinococcosis. The combination of these three organs is even rarer, if not exceptional. Diagnosis is based on epidemiological, clinical, biological, and, above all, radiological data, combined with serum antibody testing. Computed tomography (CT) is the examination of choice for determining the location and size of a hydatid cyst. Treatment is mainly surgical, with or without medical treatment. Medical treatment with albendazole is essential in cases of multiple hydatid disease or intraoperative cyst rupture[5]. Long-term medical follow-up is necessary due to the high risk of recurrence.

Conflicts of interest: The authors declare no conflict of interest.

Authors' contributions: All the authors contributed to the conduct of this work. All authors also declare that they have read and approved the final version of the manuscript.

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