

Clinical and Radiological Profile of Pulmonary Hydatid Cysts

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Abstract

Original Research Article

Introduction: Pulmonary hydatid cyst (KHP) is a major health problem, both in frequency and severity. **Methods:** To determine the epidemiological profile, the clinical and radiological aspects of KHP, we conducted a retrospective descriptive study of 184 cases identified in the respiratory diseases department of the Ibn Rochd Hospital in Casablanca between 2005 and 2019. **Results:** 93 women and 91 men. The average age was 38.6 years. The revealing signs were dominated by chest pain (84%). The diagnosis of pulmonary hydatidosis was based on imaging and hydatid serology. Thoracic CT was performed in 94% of cases. Bronchoscopy had visualized watery membranes in 22.3% of cases. Hydatid cyst was unique in 58% of cases, associated with liver damage in 19% of cases, heart damage and pulmonary artery in 1.6% of cases. Treatment was surgical in 70.6% of cases. The progression was good in 83% of cases. **Conclusion:** Hydatid disease is characterized by clinical polymorphism. Chest X-ray is often sufficient to make the diagnosis, while chest CT is still the choice for complicated cysts.

Keywords: Clinical aspects; epidemiology; pulmonary hydatid cyst; imaging.

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INTRODUCTION

Pulmonary hydatidosis is an anthroponosis due to the development of the larval form of *Taenia Echinococcus Granulosus* in humans. It is a cosmopolitan infection that plagues sheep and cattle farming areas, especially around the Mediterranean. Lung involvement represents 20-45% of all water locations [1] and is the second location after liver involvement. The positive diagnosis of pulmonary hydatid cysts (KHP) is based on epidemiological, clinical, biological and radiological arguments.

METHODS

This is a retrospective descriptive study of 184 cases of pulmonary hydatid cysts collected in the respiratory diseases department of the Ibn Rochd Hospital in Casablanca over a period of 14 years, from January 2005 to December 2019. The data were processed by Excel.

RESULTS

These were 93 women and 91 men, ranging in age from 14 to 75 with an average age of 38.6. The majority of our patients (65%) were under the age of 35. The majority of the patients were rural (129 cases or 70%). Fifty-eight patients or 31.5% had contact with

dogs. For the profession seven patients were butchers, nine were shepherds, and five were concrete sellers. A history of hydatid cyst surgery was found in eight cases including four pulmonary and four hepatic. The average time between onset of symptoms and hospitalization was two months. Clinical symptomatology consisted of chest pain in 155 cases (84%), cough in 147 cases (80%), hemoptysis in 67 cases (36%), hydatidoptysis in 26 cases (14.1%) and syncope in two cases (1%).

Five patients were asymptomatic and the discovery was accidental during a preoperative assessment in seven patients and during a pre-employment assessment in five cases. General condition was maintained in 81% of cases. The pleuro-pulmonary examination was normal in 129 cases (70%) and found submatity in 36 cases (19.5%), condensation syndrome in seven cases (3.8%) and fluid and mixed effusion syndrome in six cases each (3%). The abdominal examination had objectified hepatomegaly in ten cases (5.4%) and splenomegaly in six cases (3.2%). thoracic radiography had objectified variable aspects (opacity at regular edges in 53.2% of cases, hydroaeric waved level image in 20.6% of cases, balloon release appearance in 9.2% of cases fig.1, pleural opacity in 3% of cases, and rattle image in 1.6% of cases), these aspects are summarized in Table I.

Chest computed tomography was performed in 173 patients (94%) and had different scannographic aspects (a very limited fluid mass in 52% of cases, a hydroaeric image in 13.2% of cases, a balloon release aspect in 9.8% of cases fig 2, a hydropneumothorax in 2.8% of cases, and a rattle image in 2.3% of cases), these aspects are illustrated in Table II. It also made it possible to individualize hydatid cysts associated in particular hepatic in 19% of cases, splenic in 6.5% of cases, mediastinal in 3% of cases. Cardiac and pulmonary artery involvement were noted in 1.6% of cases and pericardial, pleural (fig. 3), diaphragmatic (fig. 4) and aortic involvement in 0.5% of cases each.

Hydatid cyst was unique in 108 cases (58%), bifocal in 36 cases (19.5%), and multiple in 40 cases (21.7%). Localization in the right lung was most common in 126 cases (68%). Hydatid cyst was healthy in 89 cases (48.3%), cracked in 27 cases (14.6%), and broken in 68 cases (36.9%). Abdominal ultrasound performed systematically in all our patients, had visualized a hepatic hydatid cyst associated in 35 cases (19%) and splenic in 12 cases (6.5%). The cardiac ultrasound showed an objective biauricular cyst (left and right atrium) multi-vesicular, associated with pericardial and pulmonary artery cysts in one case, ventricular cyst and pulmonary artery in two cases. THE thoracic MRI performed in two patients confirmed lung, arterial and cardiac localization of cysts. Flexible

bronchoscopy was performed in all our patients, it objectified watery membranes in 41 cases (22.3%) and the presence of scolex in bronchial aspiration fluid in nine cases (4.8%). Hydatid serology was positive in 99 cases (55%). Treatment was surgical in 130 cases (70.6%), associated with medical treatment in 7 cases (5.3%). Medical treatment alone was indicated in 54 inoperable cases (40 multiple hydatidose cases, one pulmonary artery site case, and 13 inoperable patient cases). It was based on Albendazole orally at the rate of 10-15mg/kg/d in two doses with cycles of 28 days separated by two weeks interval with an average number of 7 therapeutic cycles. The surgical treatment was done by conservative or/and radical methods, the conservative methods being the kystectomy in 65 cases, the aspiration puncture and perikystectomy in 28 cases. Segmentectomy in 28 cases and lobectomy in nine cases. Video-assisted thoracic surgery (VATS) is performed through a mini-thoracotomy was the most commonly used method in 111 cases. Surgical removal of cardiac and arterial cysts was performed under extracorporeal circulation (ECC) in two cases and an aortic cyst was discovered intraoperative in one case. Prevention measures and education were systematic in all of our patients. The evolution was good in 154 cases (83.6%), a recidivism was noted in five cases (2.7%) and three deaths (1.6%) were reported as a result of a post PH chronic hydatid embolism. Twenty-two patients (11.9%) were lost.

Table-I: Radiological aspects of KHP

Radiological aspect	Number of patients	%
Homogeneous opacity with regular edges	98	53,2
Hydro-aeric image with irregular level	38	20,6
Hydro-aeric image with regular level	12	6,5
Balloon release aspect	17	9,2
Inhomogeneous opacity with blurred edges	9	4,8
Opacity of pleural type	6	3
Image in fungal ball	3	1,6
Cavity image	1	0,5

Table-II: Scan aspects of KHP

Scannographic aspects	Number of patients	%
Well limited fluid mass	90	52
Hydro-aeric image with corrugated level	23	13,2
Hydro-aeric image with regular level	7	4
Balloon release aspect	17	9,8
Multi vesicular appearance	12	6,9
Pseudotumoral appearance	6	3,4
Crimp aspect	5	2,8
Hydropneumothorax	5	2,8
Image in fungal ball	4	2,3
Cavity image with membrane retention	1	0,5

Table-III: Comparative table of clinical signs by authors

	Racil [23]. N %		Our series N %	
Cough	41	78,8	147	80
Chest pain	28	53,8	155	84
Hemoptysis	37	71,2	67	36
Hydatidoptysis	22	42,3	26	14,2
Syncope	-	-	2	0,1
Asymptomatic	-	-	5	2,7

Table-IV: Different scan aspects by authors

	Racil [23]. (n : 53) N %		Notre série (n : 173)N%	
Round or oval image, sharp boundaries	-	-	90	-
Hydro-aeric image regular water level	8	12,5	7	-
Hydro-aeric image water level corrugated	15	23,4	23	-
Image in fungal ball	6	9,4	4	-
Pseudotumoral appearance	19	27,7	6	-
Balloon release aspect	3	4,7	17	-
Hydropneumoyhorax	3	10,9	5	-
Cavitory Image with membrane retention	4	6,2	1	-

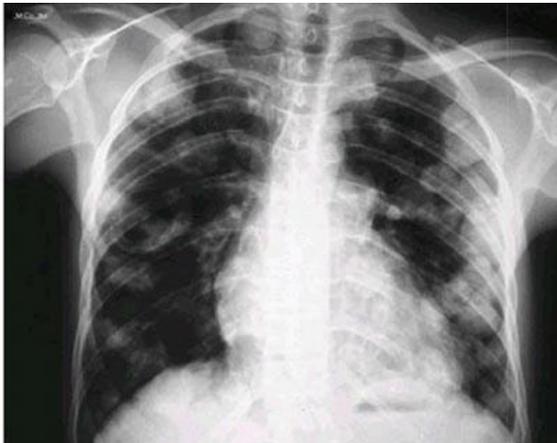


Fig-1: Chest Radio Showing United Nations Balloon Drop Appearance



Fig-3: Thoracic CT with right pleural and pulmonary fluid cyst



Fig-2: Thoracic CT of multiple pulmonary hydatidosis

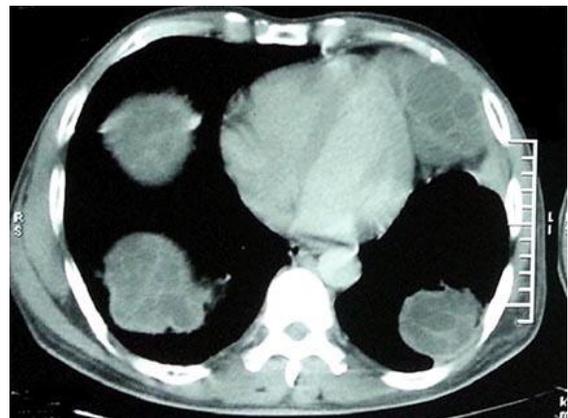


Fig-4: Thoracic CT with pulmonary and diaphragmatic hydatid cysts

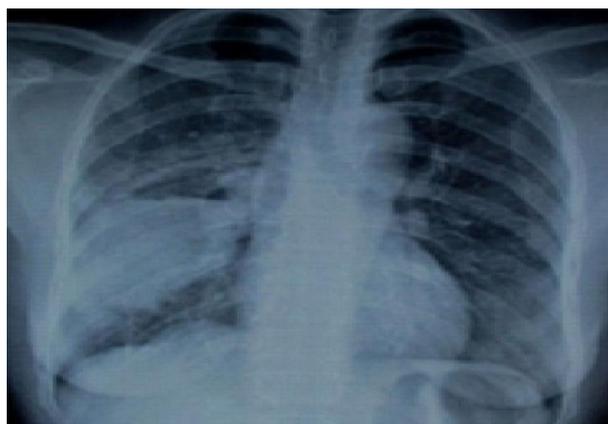


Fig-5: Radiothorax objectifying a hydro-aeric image with irregular level

DISCUSSION

Hydatidosis, or hydatid disease, is a cosmopolitan parasitosis that plagues the state endemic. It is responsible for accidental contamination of humans by a parasite metacestode of which the final host is the dog and the intermediate host is a herbivore. In Europe, hydatidosis is much rarer; it usually affects emigrants from endemic areas [2]. The results of the studies indicate that its prevalence in the areas between 1% and 10% [28]. The highest prevalence is found in the temperate zone countries including the countries of the Mediterranean region, the centre and the countries of Eastern Europe, central Asia, China, Australia, and some Sub-Saharan Africa and South America [3,4]. In Morocco, it represents a real public health problem due to its chronic nature and complications According to the Interdepartmental Counter-Terrorism Committee Hydatidosis/Echinococcosis from 2007 in Morocco [5], the frequency of liver damage is 50-70% of cases followed by lung damage, which remains the second location of predilection, which is 25-40% of cases. Bone localization is 0.5-2.5% and the heart level is 0.02 to 2%. Multiple contributing factors, environmental, socio-cultural and socio-economic, are at the origin of this hyper-endemicity. In 2011, a prevalence of 7.76 per 100,000 inhabitants was recorded in Morocco [6] with high frequency in some areas of the country, especially the West, the Middle Atlas and Eastern Morocco [7]. The rural origin of the patients is predominant in our context, found in 70% of patients. This result was confirmed by other authors [8]. A male predominance was noted in most body studies[9], while there was had no sex difference in our series. Hydatidosis is the prerogative of the adult, it is rare in children and the elderly. The average age of our patients was 38.6 years, this joins the AMOUIAN series [10]. The pulmonary localization of the hydatid cyst 2nd rank after liver localization. Indeed After intestinal passage, the larva reaches liver then lung with possibility of passage through the systemic circulation giving localizations sometimes rare and unusual throughout the body [11]. During its evolution in the lung, the fluid cyst gradually increases in volume and between in contact with the bronchi causing erosion of their walls and preening to

some complications such as suppuration, endobronchial rupture, inflammation and pulmonary calcification. The clinical manifestations of the pulmonary hydatid cyst are diverse and depend on its size, location and especially it's complicated or not. Small healthy pulmonary cysts are often asymptomatic from radiological discovery often incidental [12]. Unlike the voluminous cyst, which is accompanied by non-specific clinical manifestations such as chest pain, dry cough and hemoptysis. Ruptured pulmonary hydatid cysts in the bronchi are most often manifested by hydatidoptysis, which is the only specific sign of KHP, or by hemoptysis or repetitive bronchial suppurations [13,14]. In case of ruptured cyst in the pleura, brutal pain and dyspnea are the revealing symptoms. Although exceptional, some allergic manifestations may exist, such as a state of anaphylactic shock noted in a case in our series, in a patient who had a ruptured KHP in the pleura.

Clinical examination of the rib cage may be normal or reveal bronchial rails, condensation syndrome, cavity syndrome or fluid or mixed effusion syndrome when the cyst is broken in the pleura or enlarged. All these clinical aspects were noted in our series. Hydatidoptysis, the only specific sign which corresponds to the discharge through the mouth and/or the nose of a clear liquid with a salty taste in which membrane debris is found, was objectified in 14.1% of cases, while the discovery of the hydatid cyst was fortuitous in five cases. According to the authors, Table III summarizes the different functional signs represented mainly by cough, chest pain, haemoptysis and hydatidoptysis. Other associated locations, especially hepatic, must be systematically searched. Khps most often sit at the level of the right lung, preferably at the level of the lower lobes and can be bilateral in 20% of cases and multiple in 30% of cases [15]. In our series the KHP was in right seat in 68% of the cases, they were multiple in 21.7% of the cases, so our results join those of the literature. Imaging plays an important role in the diagnosis of KHP. Different radiological aspects may exist and vary depending on the evolutionary stage of KHP and complications. On thoracic radiography, a healthy hydatid cyst is manifested by a round or oval opacity with clear contours, of water density, realizing an aspect «cannonball», this is the most frequent aspect, found in 52.2 to 78% in the literature [16, 17]. In our study, this aspect is noted in 52% of cases. When cracked, it is manifested by a round image surmounted by the classic gaseous meniscus realizing the pneumokyst image. In case of ruptured cyst in the bronchi, the aspect found is a hydro-aeric image with regular or irregular level (fig. 5) corresponding to the typical floating membrane image performing the sign of Nenufar which is a pathognomonic sign noted in 16-30% of complicated KHP cases [18,19]. It was found in 17.2% of cases in our series. More rarely we can observe the double arc image of Ivassinevitch or bridge opacity which

corresponds to the endocyst partially detached above the liquid level. In case of vomited cyst, the radiological aspect varies according to the degree of evacuation of the cyst; one can find a cockade image that presents itself as a round opacity with blurred boundaries, surrounded by fine ring clarity or an image of a paddled membrane or dry membrane retention [19].

Thoracic ultrasound is currently considered a complement to thoracic radiography in the exploration of pulmonary hydatidosis [18]. In addition, the latter lack sensitivity and remains relatively limited in employment. The limits of thoracic ultrasound are good indications for thoracic computed tomography in hydatid pathology, although it is reserved only for complicated and atypical forms. It confirms the cystic nature of pulmonary opacity and can count lesions and look for other possible chest locations that may go unnoticed by standard radiography. Different scannologic aspects Table IV, can exist and depend on the state of the cyst. A healthy cyst is most often presented as a single fluid mass that is sometimes voluminous, homogeneous, well-limited, surrounded by a smooth and even wall. When the cysts are multiple, they achieve a «balloon release» aspect noted in 9.8% of cases.

In case of a ruptured cyst in the bronchi, DB Lewall and SJ Mc Corkell [20], allowed to individualize 6 CT stages according to the evolution of the cyst:

Stage I or "chestnut ring" appearance: is due to the entry of a minimal amount of air between the endocyst and the pericyst, without detachment of the endocyst.

Stage II or aspect of "crescent" or "inverted crescent": corresponds to partial detachment of the endocyst.

Stage III or appearance of "trapped sightings" and "honeycomb": the result of a total detachment of the endocyst without evacuation of the liquid contents. The appearance of 'trapped light' is defined by the presence of small air bubbles trapped between the folds of the endocyst detached within the fluid content. When the bubbles are very numerous, they realize the «honeycomb» aspect.

Stage IV: presents as hydro-aeric images with regular or irregular level in connection with a detachment of the endocyst and partial evacuation of the fluid content of the cyst realizing either a «double arc» aspect where the endocyst is partially collared, either an aspect of the "water lily" or the endocyst is totally sticky and floats on the liquid content, or the "piggyback" aspect where the endocyst is totally detached and remains immersed in the declive hydatid

fluid. This aspect is noted in 2.8% of cases in our study. The latter three aspects are pathognomonic of KHP

Stage V: aspect of dry retention, it is the result of a complete evacuation of the hydatid fluid and the retraction of the endocyst realizing an aspect of «ball of wool» or «bell» or even a pseudotumoral aspect. The latter two aspects were noted in 2.3% and 3.4% respectively in our series.

Stage VI or sequellary aspect, it is the result of a complete elimination of the contents of the KHP leaving a thin-walled or thick-walled aerated cavity or a nonseptal linear dense image. The biological diagnosis of hydatidosis is based on hydatid serology which maintains an interest in post-therapeutic monitoring and whose sensitivity increases in case of complicated and hepatic hydatid cysts [21,22]. Qualitative hydatid serology (immunoelectrophoresis, electrosyneresis), and quantitative serology (direct hemagglutination, indirect immunofluorescence, ELISA), makes it possible to guide the diagnosis in 40 to 65% of lung locations [29]. In our series, the hydatid serology was positive and guided the diagnosis in 55% of cases while it was negative in 45% of cases. This can be explained by the frequency of healthy and intact cysts in our patients.

Flexible bronchoscopy has a diagnostic interest when it visualizes pearly white membranes in bronchial light and individualizes scolex in bronchial aspiration fluid. These indications remain limited and are carried out especially in case of diagnostic doubt, in case of hemoptysis or to eliminate neoplasia [23]. In our context, it was required by surgeons before any surgery and was performed in all our patients. It objectified watery membranes in 41 cases (22.3%) and scolex in bronchial aspiration fluid in nine cases (4.8%).

Multiple hydatidosis is rare and its frequency varies between 11 and 30% [23]. It can be primitive following a massive and iterative infestation, or secondary to metastatic dissemination following an opening of a primitive KH in the venous system with migration of cysts to the right heart and pulmonary artery leading to hydatid emboli [23]. Bronchogenic spread is possible but exceptional. In case of multiple pulmonary hydatidosis, the search for other cardiac locations is necessary, hence the interest of a systematic echocardiography. Cardiac localization is rare [30, 31] even in endemic countries. It can be sub-endocardial, sub-epicardial, epicardopericardial, or intracardial (VD, VG, OD, OG), it is characterized by the severity of its spontaneous evolution and the frequency of its complications which are very often revealing. In our series, the echoheart objectified a biauricular cyst associated with pericardial and pulmonary artery cysts in one case and a ventricular cyst associated with a pulmonary artery cyst in two cases. Abdominal ultrasound is a systematic practice in the face of any

suspicion of pulmonary hydatidosis, it makes it possible to search for possible intra-abdominal hydatid locations that may pose therapeutic problems of chronological order [24]. In our series this allowed to visualize a hepatic hydatid cyst associated in 35 cases (19%) and splenic in 12 cases (6.5%). Thoracic magnetic resonance imaging, on the other hand, has very limited indications in thoracic hydatidosis. It is especially useful in cases of mediastinal, spinal or thoracic parietal localization associated with lung involvement [24]. It remains efficient and allows analyzing vascular structures and detecting cardiac damage. It confirmed the lung, arterial and cardiac localization of the cysts in two patients. KHP can be complicated by cracking which is the first complication during the evolution of a cyst, which can lead to further dissemination, or rupture that follows cracking and can be done at the bronchial level, translating into hydatidoptysis, or in the pleural cavity performing a secondary pleural hydatidosis and its frequency varies from 2.4% to 10.4% according to the literature [25,26]. It was noted in 5.6% of the cases in our series, or at the pericardium performing a hydatid pericarditis. It can persist a residual sequellary cavity with risk of aspergillar grafting. Pachypleuritis can occur following a chronic inflammatory reaction of the pleura. Finally the recurrence is possible following an incomplete surgical treatment, a leak of liquid after rupture, or a reinfestation.

The treatment of choice for KHP is surgery. Medical treatment with Albendazole at a rate of 10-14 mg/kg per day for a period of 3-6 months or in the form of 28-day cycles spaced 2 weeks apart [27] is indicated in multiple inoperable forms, in case of refusal of surgery or perioperative to reduce the risk of dissemination or relapses. In our series surgery was recommended in the majority of our patients, in 130 cases (70.6%), while medical treatment was recommended in 54 patients (29.3%).

The recidivism rate in the literature ranges from 0.5% to 11.3% [23]. In our series, it was 2.7%, this can be explained by incomplete surgical removal or by the persistence of hydatid cysts of small sizes gone unnoticed. The prevention of this pathology called "Dirty Hand Disease" focuses on vermifugation of final hosts (dog and sheep), strengthening food control, improving slaughterhouse hygiene and public awareness campaigns.

CONCLUSION

Through our study, it turns out that hydatid disease remains a common pathology in our context, characterized by variable and aspecific clinical signs outside of hydatidoptysis. Thoracic X-ray and abdominal ultrasound are the first-line examinations for the diagnosis of hydatidosis, however the contribution of thoracic TDM remains essential to better specify the number, location and characteristics of these cysts.

Thoracic MRI and echoheart, on the other hand, are not systematic and their indications remain limited. The treatment of choice is surgery, and prevention remains the cornerstone in management.

CONFLICTS OF INTEREST

The authors do not declare any conflict of interest.

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