SAR Journal of Surgery

Abbreviated Key Title: *SAR J Surg* Home page: <u>https://sarpublication.com/journal/sarjs/home</u> DOI: 10.36346/sarjs.2024.v05i03.001



Original Research Article

Surgical Management for Complicated Hydatid Cyst of the Lung about 94 Cases

Khalil Ghebouli^{1*}, Abelnacer Haddam¹, Karim Meskouri¹, Mohammed Amine Kecir², Mehdi Belbekri², Sedik Merouani², Houda Amrane², Kenza Bendjoudi³

¹Department of Thoracic Cardiovascular Surgery and Organ Transplantation, University Hospital Mustapha Algiers, Algeria ²Department of Thoracic Surgery and Organ Transplantation, University Hospital Ibn Badis Constantine, Algeria ³Department of Anesthesia, University Hospital Mustapha, Algiers, Algeria

*Corresponding Author: Khalil Ghebouli

Department of Thoracic Cardiovascular Surgery and Organ Transplantation, University Hospital Mustapha Algiers, Algeria

Article History: | Received: 24.03.2024 | Accepted: 02.05.2024 | Published: 04.05.2024 |

Abstract: *Introduction*: Pulmonary hydatid disease is a parasitic infestation endemic in many sheep and cattle farming regions, particularly in Algeria. Complicated forms are a frequent occurrence and characterized by radio-clinical polymorphism and therapeutic difficulties. *Material and Methods*: This is a retrospective study (2017—2023) including 94 cases of complicated hydatid cyst of lung. *Results*: Complicated hydatid cyst of lung represented 22.76% of all hydatid cyst of lung treated surgically in our department. The average age was 27.8 years (4—56 years) with a slight male predominance 57.45%. The circumstances of discovery were dominated by cough in 54.26% and hemoptysis in 32.99%. The definitive diagnosis was made on the radio-clinical and biological findings. The treatment was surgical in all cases, conservative treatment was practiced in 87.03%, and parenchymal resection was performed in 16 patients. Postoperative hydatid recurrence was noted in only one patient. *Conclusion*: Hydatid cyst of the lung is a common zoonosis in Algeria. Its treatment is essentially surgical and must be initiated quickly once the diagnosis is established in order to avoid progression towards complications that are sources of diagnostic and therapeutic difficulties. *Keywords*: Hydatid Cyst of Lung, Complications, Surgery.

Copyright © 2024 The Author(s): This is an open-access article distributed under the terms of the Creative Commons Attribution 4.0 International License (CC BY-NC 4.0) which permits unrestricted use, distribution, and reproduction in any medium for non-commercial use provided the original author and source are credited.

INTRODUCTION

Pulmonary hydatidosis is occurs in a hyperendemic manner in countries where sheep and cattle are raised, particularly in Algeria [1].

This zoonosis represents a serious public health problem and has a significant socio-economic impact. Echinococcus granulosus infection constitutes a significant financial constraint derived from human health costs and livestock production losses [2].

It is a disease wrongly considered as benign, which can become serious because of its complications which can pose diagnostic difficulties given their radioclinical polymorphism, the therapeutic problems linked to the severity and the complexity of their lesions. In certain cases and which can endanger the patient's vital prognosis [3]. Complicated pulmonary hydatidosis is defined as "any ruptured cyst in the bronchi or in the pleural cavity, infected, and/or in multiple locations" [4].

PATIENTS AND METHODS

This is a retrospective observational study carried out over a period of 6 years from January 2017 to May 2023. We collected 94 (22.76%) cases of patients with complicated pulmonary hydatidosis among 413 patients hospitalized and treated surgically in our department for pulmonary hydatidosis.

The different variables studied were age, sex, clinical symptoms, radiological and biological data and the type of surgery performed.

On admission, all patients underwent a complete clinical examination, radiological examinations (chest x-ray, chest scan and abdominal

Citation: Khalil Ghebouli, Abelnacer Haddam, Karim Meskouri, Mohammed Amine Kecir, Mehdi Belbekri, Sedik Merouani, Houda Amrane, Kenza Bendjoudi (2024). Surgical Management for Complicated Hydatid Cyst of the Lung about 94 Cases. *SAR J Surg*, *5*(3), 22-28.

ultrasound looking for secondary locations), a standard biological assessment and hydatid serology.

For bilateral forms we opted for a two-stage surgery one month apart. Those with an abdominal location were secondarily referred to visceral surgery.

All patients benefited from additional treatment with albendazol which was continued for three months.

Our study aims to present the therapeutic modalities used in the surgical management of complicated hydatid cyst as well as their results in terms of morbidity and mortality. The average duration of follow-up is 4.1 years.

RESULTS

Of the 413 patients treated surgically for pulmonary hydatidosis, 94 patients (22.76%) presented a complicated hydatid cyst. Fifty-four patients were male (57.45%) and forty were female (42.55%). Their ages vary between 4 and 56 years, with the majority being between the 2nd and 4th decades.

The notion of hydatid infection was found in 65 patients (69.15%). Five patients were operated on for hydatid cyst of the liver several years previously and one patient operated on for a hydatid cyst of the contralateral lung. The majority of patients (87.23%) lived in average or poor socio-economic conditions.

The clinical picture was dominated by cough in 51 cases, hemoptysis in 21 cases, chest pain in 16 cases, salty vomit in 30 cases and biliptysis in a single patient. The general signs were dominated by fever observed in 8 patients. In 4 patients the discovery of the cyst was fortuitous during a systematic radiography.

The hydatid cyst of the lung was solitary in 90.43% of cases, multiple in 9 patients (9.57%). including 5 cases of bilateral location: 2 in 6 patients, 3 in 2 patients and 5 in a single patient. The hydatid cyst of the lung was located on the right in 50 patients and on the left in 40 patients.

The radiological presentation of complicated form was variable, different aspects were found, an excavated image with or without hydro-aeric level in 58 cases (Figure 1, 2, 3), followed by an image of poorly limited heterogeneous opacity in 19 cases. An appearance of hydro-pneumothorax in 6 cases and pleurisy in 2 cases. Multiple opacities were noted in 9 patients.



Figure 1: Fissured hydatid cyst Gas crescent image



Figure 2: Ruptured hydatid cyst Image of a regular hydroaeric level



Figure 3: Ruptured hydatid cyst Bell image

Easier access to chest CT has made this examination systematic before any surgical procedure. The main CT aspects encountered were a hydro-aerial image with regular level in 24.47%, followed by a round opacity with irregular edges in 20.21%, the appearance

of a floating membrane in 17.02% and the bell image in 12.77% cases.

CT revealed associated pleurisy in 8 cases (8.51%), liver HK in 14 cases (14.89%) (Figure 4).



Figure 4: CT scan appearance of a ruptured hydatid cyst in the pleura with pleural effusion

Bronchial endoscopy was performed on 30 patients, showing hydatid membranes in 9 of them. Hydatid serology was positive in 61 patients. Systematic abdominal ultrasound showed hepatic locations in 14 cases. Cardiac ultrasound was performed systematically in multiple forms and did not reveal cardiac hydatid localization.

Therapeutically, all patients benefited from surgical treatment. 93 thoracotomies (93.94%) and 6 video-assisted thoracic surgery (VATS) procedures (6.06%) were performed. One hundred and eight operative procedures were performed: The procedures performed were: 14 decortications (14.14%), a conservative procedure by a simple cystotomy in 8 cases and a cystectomy-pericystectomies-blinding of the fistulas in 94 cases (87.03%), resection parenchymal by segmentectomy or lobectomy (Figure 5) in 16 cases including one patient who presented with a biliobronchial fistula having previously benefited from drainage of the bile duct by an endoscopic sphincterotomy before performing a right lower lobectomy, no pneumonectomy been practiced (Table I).



Figure 5: Right lower lobectomy on lobe destroyed by hydatid cyst of the lung

	8	
Surgical procedure	Nomber	Percentage
cys +Pcys+ BF	83	76.85%
Decortication + cys +Pcys+ BF	11	10.19%
simple cys	8	7.40%
Lobectomiy without decortication	10	9.26%
Decortication + lobectomy	3	2.78%
Segmentectomy	3	2.78%

Table I: Distribution of surgical procedures

Cys: cystectomie Pcys: pericystectomie BF: Blinding of the fistulas Mortality was nil, however we deplored three cases of postoperative pyothorax and one case of haemothorax which benefited from surgical drainage.

A lack of pulmonary re-expansion was observed in a one case, clearly improved by active respiratory physiotherapy.

Py

Haemothorax

Lack of pulmonary re-expansion

Secondary pleural hydatidosis

The average duration of drainage varied from 4 to 18 days with an average of 5.3 days.

Two patients experienced hepatic toxicity from albendazol requiring treatment to be stopped.

The long-term outcome was favorable for all patients except one case who presented with secondary pleural hydatidosis which was surgically treated (Table II).

۰.

0.93%

0.93%

0.93%

Table II: Distribution according the complications			
omplication	Nombre de patient	Pourcentage	
othorax	3	2.78%	

1

1

1

DISCUSSION

Pulmonary hydatid infections represent 20 to 40% of all hydatid cysts and more than 90% of intrathoracic hydatid cysts. They generally affect young adults aged 20 to 30, with no gender predominance. In our series the average age is 27.8 years with a slight male predominance. The average age of subjects with hydatid cysts has varied according to global studies between 24 years in the Thameur study [5], covering 1619 with a male-female sex ratio of 1.1 and 33 in the Ghoshal study [6]. Pulmonary hydatid cysts are characterized by a diversity of anatomoradio-clinical forms. They are associated, in 17 to 50% of cases, with another extrapulmonary localization including 2 to 5% intra-thoracic and 6 to 30% hepatic [7], 14.89% in our series the lung localization was associated with liver hydatid cyst. The complicated form is not a rare occurrence and represents 22.76% of cases in our series.

The symptomatology of the hydatid cyst of the lung, apart from any complications, remains relatively latent and not very noisy, with a phase of growth of the cyst which can vary from a few weeks to a few years. Indeed, and generally speaking, the cyst only begins to really symptomatic when it becomes complicated [8].

The earliest sign manifested was cough (54.26%) in our study. This can have several aspects, it can be tenacious, brief or quintuous, dry at the beginning then productive later, accompanied by mucous or mucopurulent expectoration reflecting the rupture and superinfection of the cyst [9, 10].

Chest pain, found in 17.02% of cases, reflects the development of the cyst towards the pleura or the chest wall rich in nervous elements [10]. Its location depends on the cyst, its intensity is variable, most often discrete, localized or diffuse, it can be a side point type, sometimes a stab [11, 12], or very rarely, a radicular type having mentioned a Pancoast and Tobias syndrome [13, 14]. Dyspnea was also observed in 12.77% of cases, its intensity depends on the volume of the cyst which impairs respiratory function by compressing more or less extensive pulmonary parenchymal territories [15].

Hemoptysis, present in 32.99% of cases in our series, is the main functional symptom [12-16], it encourages the patient to consult early. According to Alaoui *et al.*, [16], hemoptysis is often minimal in cases of hydatid cyst of the lung. Hemoptysis signals the beginnings of rupture of the cyst in the bronchi or pleura [8].

According to Yéna *et al.*, [17], the only pathognomonic clinical manifestation of pulmonary hydatidosis is the rare externalization of hydatid materials through vomit. It can cause allergic accidents, bronchopulmonary suppuration and bronchogenic dissemination [18]. In our study, 28 patients (29.79%) presented with vomiting.

Intrapleural rupture of the hydatid cyst of the lung, whether acute or insidious, remains a rare complication (0.1 to 6% of cases) [19], in our series it is of the order of 8.51%. It can occur spontaneously without apparent cause, favored by the size of the cyst and its peripheral localiation, or be caused by a disturbance in the balance of intra-thoracic pressures during a coughing fit, chest trauma or accidental puncture of the cyst [19].

According to the work of Halezeroglu *et al.*, [20], the frequency of signs is proportional to the size of the cyst (p=0.004). The size of the cyst varies, from 2 to 10 cm [21], sometimes even larger and can occupy an entire lobe, or even an entire lung [22]. Some authors describe the cyst as voluminous when it exceeds two thirds of the lung. ipsilateral hemithorax, while others agree on the figure of 10 cm in diameter [8-20].

The radiological presentation of the complicated hydatid cyst is very polymorphic:

The pneumocyst with floating membrane giving the sign of the floating membrane of

Belot or the sign of the "water lily" of camalote described by Lagos Garcia and Segers in 1924, popularized by the work of Dévé in 1925. It is the most characteristic sign and the most frequent which follows the sign of the "double airy crescent" and which corresponds to the secondary collapse of the hydatid membrane and its floating on the surface of the hydatid fluid [23].

The image of a perfectly horizontal hydro-aerial level, a rarer aspect which is seen in the event of complete evacuation of the membrane or its total immersion in the hydatid liquid.

The cracked hydatid cyst results in a gaseous crescent at the upper pole (the Argentinian sign "del bandete claro"), or "gaseous meniscus" described by Morquio [8-24], or also called a "gaseous crescent".

This crescent is sometimes encountered in a sloping position, producing the sign of the "inverted airy crescent" linked to the existence of a localized hydatid pericystic synechia [15].

Dry membrane retention or membrane incarceration, studied by Del Campo in 1947, and by Curtillet in 1949. It presents itself in the form of flaming, tapered, irregular images, with blurred boundaries, directed towards the hilum, or "cirrus-cumulus" by Liaras and Houël.

The intra-pleural rupture of the cyst is reflected radiologically by a gaseous, liquid or mixed effusion which obscures the radiological image. The diagnosis can only be confirmed by puncture except in a known patient with a pulmonary hydatid cyst or in the radiological presence of an image of a floating membrane supernatant in the pleural fluid.

Multiple pulmonary hydatidosis presents differently depending on whether it is primary or secondary:

Primary multiple pulmonary hydatidosis manifests radiologically as round opacities of varying dimensions and number, but true "balloon release" remains rare [18-23].

Secondary multiple pulmonary hydatid disease is suspected given the small size of the cysts, the multiple, bilateral and symmetrical nature of the lesions and their peripheral and basal location along the arterial axes [25].

The managment of pulmonary hydatid cyst is essentially surgical.

Thoracotomy constitutes the classic surgical approach practiced in 93.94% of our patients. Video thoracoscopy constitutes an alternative to thoracotomy and seems particularly attractive in children [26], and in

bilateral lesions but poses the problem of management of bronchopleural fistulas at the end of the procedure. We performed 6 video thoracoscopies for bilateral forms to better preserve respiratory function.

For the surgical treatment of complicated hydatid cyst we opted for conservative treatment (87.03%). Pericystectomy and blinding of bronchial fistulas are essential to guarantee simple surgical outcomes, especially in cases of ruptured and infected cysts and/or in the presence of a thick pericyst.

Treatment of the residual cavity can be done by padding or ideally by the flattening technique provided there is a perfect seal.

In the event of a large cyst exceeding 50% of the lobe, lobectomy is the rule, subject to pulmonary function allowing an uneconomical procedure.

Regular excisions are indicated in cases of cyst associated with irreversible lesions leading to the destruction of the surrounding pulmonary parenchyma.

Multiple cysts treated according to the same principles depending on the condition of each cyst taken separately and the condition of the surrounding parenchyma.

In case of unilateral polycystic disease, conservative treatment is preferred given the risk of recurrence, especially in endemic areas, however padding should be avoided so as not to tie up the lung.

Bilateral cysts are usually treated in two stages, the attitude is to first operate on the lung which contains the most healthy cysts. If all the cysts are healthy, you should operate on the side of the largest cyst.

Intrapleural rupture of the hydatid cyst often requires decortication (14 cases in our study) of the localized pleural pockets associated with the treatment of the pulmonary or even hepatic cyst(s) in question. In the event of pleural seeding, the cysts become fixed in the subpleural region and it is then appropriate to perform extensive parietal and mediastinal pleurectomies associated with prolonged cleaning of the pleural cavity using scolicidal solutions.

In the case of biliobronchial fistulas which is becoming less and less frequent than previously (1.06%) in our series, the cure of liver lesions and drainage of the common bile duct in our opinion must be carried out first.

Surgery still remains the radical treatment for hydatid cysts, however, a considerable improvement in management can be provided by medical treatment to secure a surgical procedure. We recommend systematically a antihelminthic treatment for 3 months subject to a correct liver and hematological assessment. Hydatid recurrences can occur within variable periods, linked to incomplete excisions, fistulization of cysts with systemic passage and hydatid emboli. The recurrence rate varies from 0.5 to 11.3% in the literature [24-28]. In our series, it was 0.93%.

CONCLUSION

Hydatid cyst of the lung is a benign but serious pathology due to its complications, often occurring in young and active subjects.

Complicated forms of hydatidosis are not rare and are characterized by their radioclinical polymorphism. The management of hydatid cyst of the lung remains surgical, it is essentially conservative. Medical treatment takes more space in complicated forms to optimize surgical management.

DECLARATIONS

Acknowledgement: None to declare.

Ethical Statement: All procedures performed in this case study involving human participants, were in accordance with the ethical standards of the Faculty of Medicine of Algiers and with the 1964 Helsinki declaration and its later amendments. Written and informed consent for publication was obtained from the patient.

Funding: We attest that we have not received any financial support or grants from any institution during the realization of this report.

Conflict of Interest: The authors have no conflict of interests to declare.

REFERENCE

- 1. Schantz, P. M. (2006). Progress in diagnosis, treatment and elimination of echinococcosis and cysticercosis. *Parasitology International*, *55*, S7-S13.
- 2. Otero-Abad, B., & Torgerson, P. R. (2013). A systematic review of the epidemiology of echinococcosis in domestic and wild animals. *PLoS neglected tropical diseases*, 7(6), e2249.
- Racil, H. (2009). Coll. Kystes hydatiques compliqués du poumon. *Rev Mal Respir*, 26, 727-34. Doi : 10.1019/20094056.
- Kervancioĝlu, R., Bayram, M., & Elbeyli, L. (1999). CT findings in pulmonary hydatid disease. *Acta Radiologica*, 40(5), 510-514.
- Thameur, H., Chenik, S., Abdelmoulah, S., Bey, M., Hachicha, S., Chemingui, M., ... & Chaouch, H. (2000). Thoracic hydatidosis. A review of 1619 cases. *Revue de pneumologie clinique*, 56(1), 7-15.
- Ghoshal, A. G., Sarkar, S., Saha, K., Sarkar, U., Kundu, S., & Chatterjee, S. (2012). Hydatid lung disease: an analysis of five years cumulative data from Kolkata. *The Journal of the Association of Physicians of India*, 60, 12-16.

- Bouhaouala, M. H., Hendaoui, L., Charfi, M. R., Drissi, C., Tlili-Graies, K., Mechmèche, R., & Kilani, T. (2007). Hydatidose thoracique. EMC-Radiodiagnostic- Coeur-Poumon (Elsevier Masson SAS, Paris), 1-17.
- Haddam, A. N. (2015). Echinococcose kystique pulmonaire. Chirurgie de la cavité adventielle. Thèse N°283.
- 9. Ghalim, A. (1995). *Le kyste hydatique pulmonaire* : à propos de 294 cas. Thèse Méd.; Casablanca; n°240.
- Gharbi, H. A., Ben Chehida, F., Bardi, I., Horchani, H., & Ben Romdhane, H. (1993). Radiodiagnosticcoeur-poumon. *Parasitoses pulmonaires. Paris: Éditions techniques-Encycl Med Chir.*
- 11. Pene, P., NOSNY, E. P., & Nosny, E. P. (1986). Parasitose à développement pulmonaire. Hydatidose pulmonaire. EMC. *Poumon*, 6003-L10.
- Salih, O. K., Topcuoğlu, M. Ş., Çelik, Ş. K., Ulus, T., & Tokcan, A. (1998). Surgical treatment of hydatid cysts of the lung: analysis of 405 patients. *Canadian journal of surgery*, 41(2), 131.
- Alaoui-Yazidi, A., Sibaï, H., Kafih, A., El Meziane, A., & Bartal, M. (1995). Syndrome de Pancoast et Tobias d'étiologie hydatique. *Revue des maladies respiratoires*, 12(1), 49-52.
- 14. Sabir, L., Afif, H., Berrada, Z., Safieddine, S., Aichane, A., & Bouayad, Z. (2007). Kyste hydatique pulmonaire : à propos de 181 cas Revue des Maladies Respiratoires, 24(4), 112-116.
- 15. Batache, A. Kyste hydatique du poumon (à propos de 178 cas). Thèse méd.; Casablanca; 1994 n°71.
- Alaoui, Y. A., Bartal, M., & El Boutahiri, A. (1991). Coll. Profil étiologique des hémoptysies dans un service de pneumologie (291 cas). Rev. *Mar. Méd. Santé, 13*(2), 27-34.
- Yéna, S., Sanogo, ZZ, Kéïta, A., Sangaré, D., Sidibé, S., Delaye, A., ... & Soumaré, S. (2002, May). Pulmonary hydatid cyst surgery in Mali. In *Annals of Surgery* (Vol. 127, No. 5, pp. 350-355). Elsevier Masson.
- 18. Rifki, S. (2001). Le traitement chirurgical du kyste hydatique du poumon. *Maghreb Médical*, 21, 192-5.
- Kilani, T., Ben Safta, Z., Jamoussi, M., Ben Driss, M., Thameur, H., & Abid, A. (1988). Les complications pleurales du kyste hydatique du poumon (à propos de seize cas). In *Annales de chirurgie (Paris)* (Vol. 42, No. 2, pp. 145-148).
- Halezeroglu, S., Muharrdm, C., Uysal, A., Senol, C., Keles, M., & Arman, B. (1997). Giant hydatid cysts of the lung. *J Thorac Cardiovasc Surg*, *113*, 712-7.
- Bouhaouala, M. H., Ladeb, M. F., & Ben Hamouda, M. (1989). Coll. Radiologie de la maladie hydatique. Feuil. *Radiol*, 29(2), 141-2.
- 22. Grenier, P. H. (1988). coll. Imagerie thoracique de l'adulte. Médecine Sciences Flammarion; Paris.
- 23. Kheder, A. B., El Mekki, L., & Hamza, M. F. Coll. Les aspects épidémiologiques, cliniques, radiologiques et immunologiques du kyste hydatique pulmonaire en Tunisie.

- 24. Larbaoui, D. (1989). Le kyste hydatique du poumon. *Revue de pneumologie clinique (Paris)*, 45(2), 49-63.
- Ramos, G., Md, P., Antonio Orduña, M., & Mariano García-yuste, M. (2001). Hydatid cyst of the lung: diagnosis and treatment. *World journal of surgery*, 25(1), 46.
- Afif, H., Aichane, A., Trombati, N., Bahlaoui, A., Bouayad, Z., & Boumzebra, M. (2000). Hydatidose pulmonaire multiple en lâcher de ballons avec

localisation cardiaque. *Revue des maladies respiratoires*, 17(3), 697-699.

- 27. Jidejian, Y. D. (1979). *Hydatid disease*. Ed. Dar el-Mashreq, Beirut, 202p.
- Thameur, H., Chenik, S., Abdelmoulah, S., Bey, M., Hachicha, S., & Chemingui, M. (2000). Les localisations thoraciques de l'hydatidose: A partir de 1 619 observations. *Revue de pneumologie clinique* (*Paris*), 56(1), 7-15.