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## Bilateral pulmonary hydatidosis associated with uncommon muscular localization

Houssef Messaoudi<sup>a</sup>, Bochra Zayène<sup>a,\*</sup>, Imen Ben Ismail<sup>b</sup>, Mokhles Lajmi<sup>a</sup>, Hatem Lahdhili<sup>a</sup>, Saber Hachicha<sup>a</sup>, Slim Chenik<sup>a</sup><sup>a</sup> Department of Cardiac and Thoracic Surgery, The Military Hospital of Instruction of Tunis, Tunisia<sup>b</sup> Department of General Surgery, Traumatology and Severe Burns Center Ben Arous, Tunisia

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## ABSTRACT

**INTRODUCTION:** The lung is the second most commonly affected organ by hydatidosis, and the bilateral involvement is rare even in endemic regions.**CASE PRESENTATION:** We report the case of a 27-year-old patient who presented with right basithoracic pain and cough evolving for three months. Thoracic CT scan revealed two homogeneous, rounded cystic formations enhancing after injection of the contrast media, located in the lateral basal segments of the lower lobe. An abdominal CT scan was performed to rule out a hepatic localization of the hydatid cyst, revealed a cystic formation of the left psoas muscle. The diagnosis of bilateral hydatid lung cyst associated with hydatid psoas muscle location was then made. The patient underwent a two-stage thoracic surgery. The second step involved partial cystectomy of the psoas muscle hydatid cyst via a left iliac incision and using an extraperitoneal approach. The postoperative course was uneventful.**DISCUSSION:** Management of bilateral pulmonary hydatid cyst is controversial. Some authors recommend operating bilateral cysts in two-stage surgery, with an interval of three to four weeks between procedures. The involvement of the psoas muscle is rare and is generally secondary to the rupture of splenic, hepatic or renal hydatid cysts. Generally, its diagnosis is delayed as the latter is most of the time asymptomatic. **CONCLUSION:** Bilateral pulmonary hydatidosis associated with hydatid cyst of the psoas muscle is a rare entity. Radiological investigations and especially CT scan are the mainstay of diagnosis. Surgery remains to be the treatment modality of choice.© 2020 Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

## 1. Introduction

Hydatidosis is an anthroponosis due to the development of the larval form of *taenia Echinococcus granulosus* in humans [1]. Even though the most commonly affected organs are the liver (55–70%) and lungs (18–35%), hydatid cyst can affect all organs in the human body. The incidence of unusual sites is about 8–10% [2]. The primary retroperitoneal location in the psoas muscle remains exceptional, representing 3% of muscle locations [3]. It is an endemic disease in many regions and presents a public health problem in Tunisia with an incidence of 12.6/100,000 inhabitants [4].

We report the case of an unusual presentation of bilateral pulmonary hydatid cyst associated with a psoas muscle location.

The work has been reported in line with the SCARE criteria [5].

## 2. Case presentation

A 27-year-old patient with no past medical history, presented with right basithoracic pain and cough evolving for three months without fever. Physical examination revealed slight basithoracic tenderness. The patient had no history of hydatid vomiting or close contact with dogs or other animals. A chest X-ray showed two bilateral basal opacities with very limited water tones (Fig. 1). A further thoracic CT scan revealed the presence of two homogeneous, rounded fluid formations with regular, thin-walled, bilobed contours enhancing after injection of the contrast media. They were located in the lateral basal segments of the lower lobe measuring 58 × 78 mm on the right, 80 × 96 × 72 mm on the left, without any complications (Fig. 2). An abdominal CT scan was performed in order to rule out a hepatic localization of the hydatid cyst, revealed a homogeneous cystic formation of the left psoas muscle. The latter had an oval shape with regular contours and thin wall, enhancing after injection of the contrast media. It was measuring

\* Corresponding author.

E-mail address: [bochra.zayene@gmail.com](mailto:bochra.zayene@gmail.com) (B. Zayène).

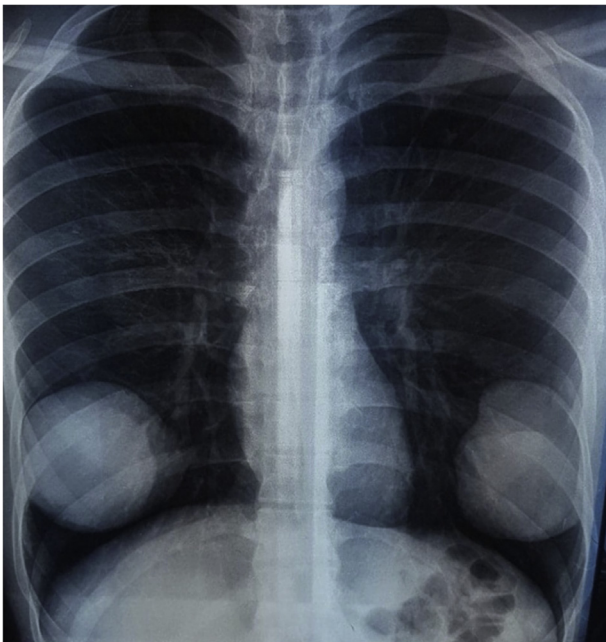


Fig. 1. Chest x-ray showing two laterobasal opacities.

42 × 33 × 57 mm and presents a close contact with the left external iliac vein with loss of the fatty safety line (Fig. 3). The diagnosis of bilateral hydatid lung cyst associated with hydatid psoas muscle location was made.

Surgical management was planned as follows: start with the pulmonary stage, and particularly with the right side which is the site of the largest cyst, then the left side with an interval of one month. The abdominal localization was treated secondarily. The patient underwent two-stage surgery. The first step of the surgical procedure was right lateral thoracotomy in the sixth intercostal space with cystectomy, peri-cystectomy and padding, followed one month later by the same procedure on the left side via left lateral thoracotomy. The second step involved partial cystectomy of the psoas muscle hydatid cyst, leaving a part of the



Fig. 3. Abdominal CT scan showing a cystic mass at the expense of the left psoas muscle.

pericyst against neurovascular structures, via a left iliac incision, and using an extraperitoneal approach. The patient was discharged well on oral albendazole with a posology of 400 mg per day for six months.

### 3. Discussion

Hydatidosis has a worldwide distribution and represents a serious health problem in endemic regions such as Mediterranean countries. It is caused by the larval stage of *Echinococcus granulosus*, whose infestation is secondary to the ingestion of taenia eggs eliminated by dogs [1]. After then, larvae penetrate the mucosa of the proximal portion of jejunum and may reach through the venous and lymphatic channels any region of the body where they transform into cysts [6].

It's well known that the lung is the second most affected organ after the liver [2]. Pulmonary hydatidosis has a unilateral localisation commonly. The bilateral involvement is unfrequent with a prevalence ranging from 4% to 26.7% in endemic regions [7].

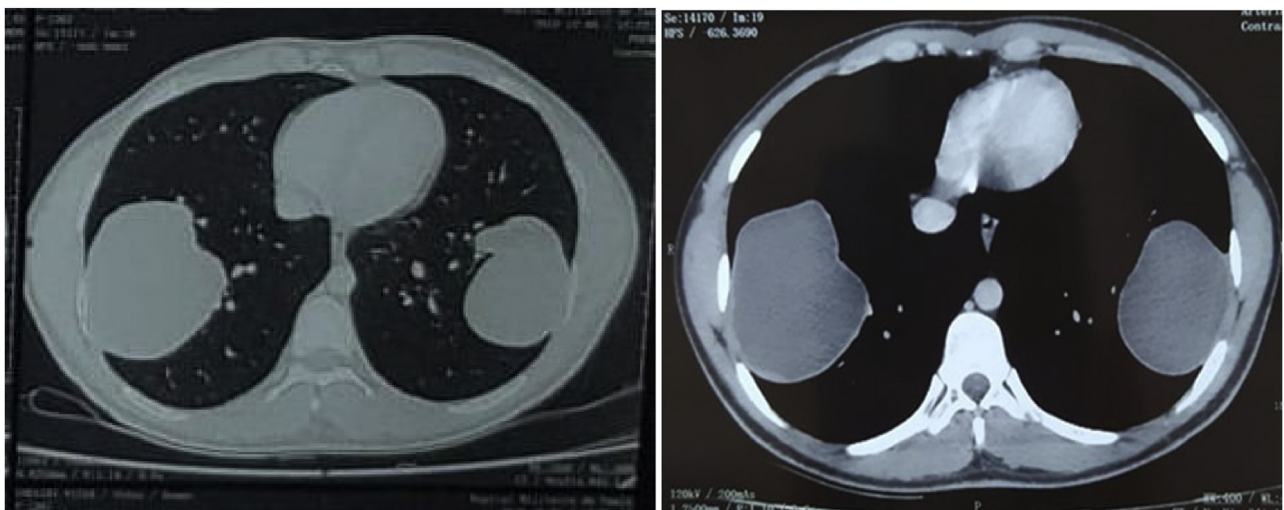


Fig. 2. Chest CT scan showing two laterobasal cystic formations.

Clinically, uncomplicated lung hydatid cysts, are usually asymptomatic and incidentally diagnosed. In some cases, as in our case, the patient may present with cough dyspnea and chest pain.

The diagnosis of pulmonary hydatid cysts depends basically on radiological investigations. Chest radiograph and Computed tomography (CT) are the imaging modalities of choice. The chest x-ray is considered to be the initial diagnostic tool in pulmonary hydatid cyst with a sensibility neighbouring 90%. Computed tomography is useful for evaluating hydatid, especially complicated cyst and eliminating other differential diagnoses [8].

Surgery is still the unique therapeutic modality for pulmonary hydatid cyst. The aim is to remove the entire cyst with preservation of the maximum of lung parenchyma. Management of bilateral pulmonary hydatid cyst is controversial. Some authors recommend operating bilateral cysts in two-stage surgery, with an interval of three to four weeks between procedures [9]. The largest and uncomplicated hydatid cyst should be operated on the first [10]. Other authors demonstrated that one-stage surgery is superior to a classic two-stage operation as it decreases the morbidity, hospital stay, and cost [11].

The one-step surgery can be performed either through double thoracotomy or median sternotomy to reduce the cost of the intervention and avoid second general anaesthesia. It is mainly indicated for young, uncomplicated and peripheral cysts [10].

As we mentioned above, the larvae are able to pass through the liver filter and reach any organ of the body. Soft tissue involvement is unusual, and it is reported in 0.5–4.7% of cases and mainly affects the skeletal muscles (2–3%) [3]. The infrequency of muscle localisation could be explained by the muscle contractions and lactic acid production, making difficult the implantation and development of the embryo [12]. The involvement of the psoas muscle is rare and is generally secondary to the rupture of splenic, hepatic or renal hydatid cysts.

Generally, the diagnosis of hydatid cyst of the psoas muscle is delayed as the latter is most of the time asymptomatic, but enlarged cysts may compress the adjacent organs like ureter, kidney, or vertebra; then it becomes symptomatic [13]. In our case, the patient was asymptomatic.

Mainly, the diagnosis is made based on radiologic findings. Many imaging modalities (US, CT and MRI) are valuable, but MRI is the imaging method of choices since it enables accurate visualisation of structures and their relation to adjacent tissues [14].

Surgery is the only curative treatment modality for hydatid psoas cyst. It can be performed either through the intraperitoneal or extraperitoneal approach. Otherwise, the extraperitoneal approach is preferred in order to avoid intraperitoneal dissemination. As for the pulmonary localisation, the goal of surgery in case of psoas localisation is to remove the entire cyst, but total cystectomy is not possible most of the time because of the adhesions between the cyst and its neighbouring vessels and nerves [15].

Medical treatment has limited indications such as disseminated form, contra-indications to surgery, or as a postoperative preventive measure if there is hydatid fluid spillage preoperatively [16].

#### 4. Conclusion

Bilateral pulmonary hydatidosis associated with hydatid cyst of the psoas muscle is a rare entity. Radiological investigations and especially CT scan are the mainstay of diagnosis. Surgery remains to be the treatment modality of choice.

#### Declaration of Competing Interest

Authors declare no conflict of interest.

#### Funding

This study was not supported by any institution and company.

#### Ethical approval

Ethical approval was not required and patient identifying knowledge was not presented in the report.

#### Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

#### Author's contribution

Study concepts: Dr Messaoudi Houssem Manuscript writing: Dr Ben Ismail Imen, Dr Bochra Zayene. Helped in data interpretation and manuscript evaluation: Dr Mokhles Lajmi, Dr Lahdhili Hatem Data acquisition: Dr Saber Hachicha. Critical revision: Dr Chenik Slim.

Dr Hachicha Saber, Dr Messaoudi Houssem are the thoracic surgeons who performed the procedure.

#### Registration of research studies

It is not the case.

#### Guarantor

Dr Chenik Slim.

#### Provenance and peer review

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