

Juvenile Primary Mediastinal Hydatid Cyst

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Abstract

Introduction: Echinococcosis is a zoonosis that is still endemic and represents a major public health problem in many parts of the world, including Tunisia. The primitive forms of the unusual locations (extra-pulmonary and extra-hepatic) of this parasitosis are exceptional. We report an original case of juvenile primary mediastinal hydatid cyst.

Case Report: A 12-year-old patient, with no notable pathological history, was explored for right lateral thoracic pain, progressive worsening for two months, with recent dyspnea.

Somatic examination, electrocardiogram, and basic biological assessment were without abnormalities. Chest X-ray showed a right, well-defined, homogenous para-cardiac opacity, which proved to be in the posterior mediastinum on the right profile view of chest radiograph. Further investigation concluded to a primary and isolated mediastinal hydatid cyst. The patient was operated through a posterolateral thoracotomy with simple operative follow-up, and a subsequent favorable evolution.

Conclusion: As exceptional as it is, this location of the hydatid disease deserve to be known and evoked in front of any respiratory signs that is not proven, especially in endemic countries, because its potential complications may be life-threatening.

Keywords: Hydatid Cyst; Mediastinum; Echinococcosis; Hydatidosis

Introduction

Echinococcosis is a zoonosis that is still endemic and represents a major public health problem in many parts of the world, including Tunisia [1-5]. The most frequent locations of human hydatid disease are the liver and lungs: 65 - 70% and 25% respectively [2,4]. In addition, all organs and/or tissues of the human body may be the seat of single or multiple hydatid cyst [6-8].

These extra-pulmonary and extra-hepatic sites are described as atypical or unusual by the majority of authors [6-8], or even “ectopic” and “aberrant” by some [9].

The primitive forms of these unusual locations are exceptional: only 0.9% in the series of Lianos GD., et al. collected over 33 years of experience [10].

Because of their scarcity and the lack of knowledge by the majority of medical practitioners, these unusual, but sometimes potentially fatal [11], localizations of human echinococcosis often represent a real diagnostic and therapeutic challenge, especially since clinical presentations are not stereotyped [6,10].

We report an exceptional case of juvenile primary mediastinal hydatid cyst, diagnosed in a child of twelve years, which is very uncommon; indeed, in the large series of 235 cases of intrathoracic echinococcosis arising in 222 juvenile patients, only two cases of mediastinal hydatid cyst were noted (0.85%) [12].

Case Report

A 12-year-old patient, with no notable pathological history, was explored at our department (Department of Internal Medicine. Military Hospital of Gabes, Tunisia) for right lateral thoracic pain, progressive worsening for two months, with recent dyspnea.

The somatic examination found an afebrile patient, eupneic at rest, and with a stable hemodynamic state. The electrocardiogram was without abnormalities as well as the basic biological assessment: Leucocytes: 8 200/mm³, hemoglobin: 12.3 g/dl, platelets: 198 000/mm³, erythrocyte sedimentation rate: 22 mm/H1, C-reactive protein: 3.2 mg/l, creatinine: 82 μmol/l, ionogram: sodium at 142 mmol/l and potassium at 4.3 mmol/l, fasting blood glucose: 5.02 mmol/l, troponin Ic <0.01 ng/ml, aspartate aminotransferase: 22 IU/l, alanine aminotransferase: 28 IU/l, creatine kinase: 80 IU/l, Lactate dehydrogenase: 132 IU/l, and normal profile of electrophoresis of serum proteins.

The chest X-ray showed a right, well-defined, homogenous para-cardiac opacity on posteroanterior view, which proved to be in the posterior mediastinum on the right-profile view (Figure 1).

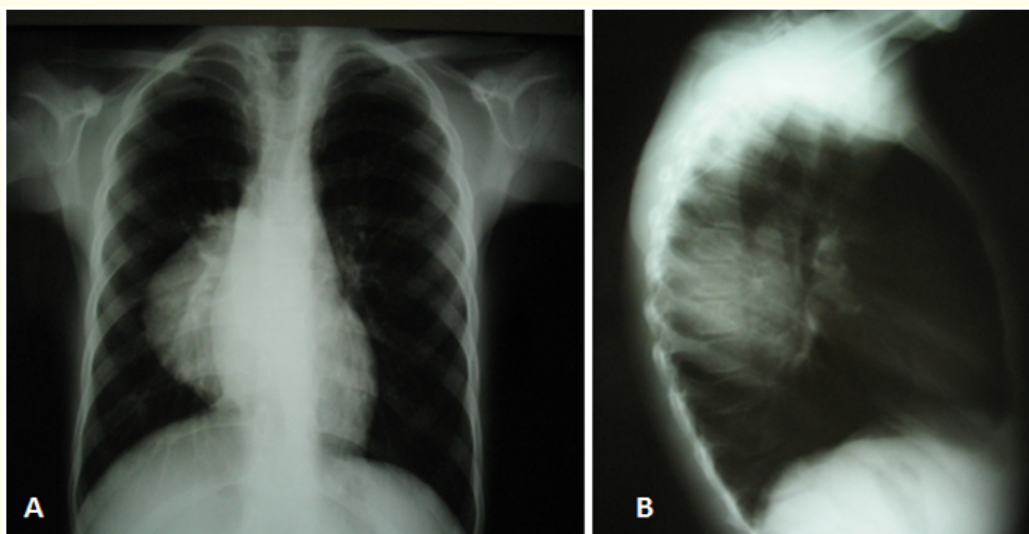


Figure 1: Chest X-ray: homogeneous and regular right paracardial opacity on posteroanterior view (A), in the right posterior mediastinum on the right profile view (B).

Thoracic computed tomography showed a homogeneous and well-defined cystic mass of the right posterior mediastinum, displacing the parenchyma of the lungs and taking the contrast in a peripheral and homogeneous way compatible with a simple hydatid cyst (Figure 2).

The hydatid serodiagnosis was weakly positive. The abdominopelvic computed tomography showed no other cystic lesions.

The child was from a rural area of southeastern Tunisia, farmer’s son, with a notion of close contact with sheep and dogs, explaining how he had contracted this infection. It was transferred to the Pneumologic hospital of Ariana (Tunis, Tunisia) to be operated.

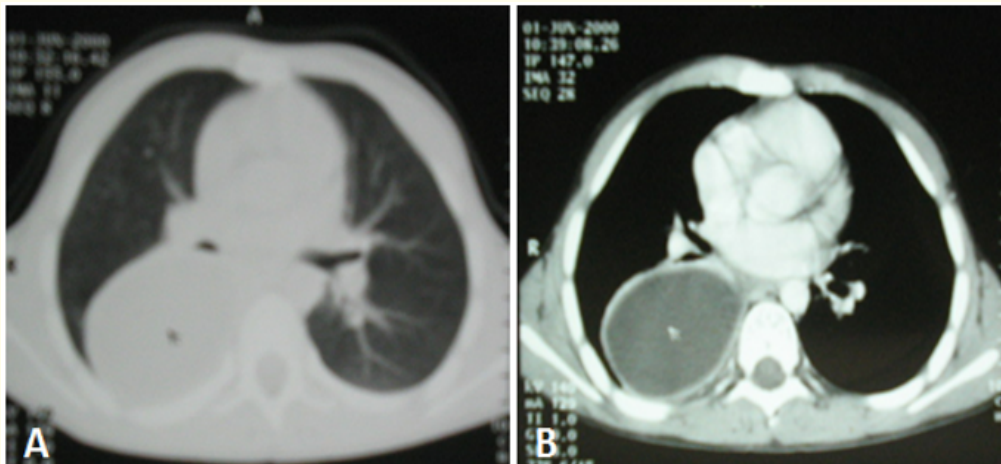


Figure 2: Thoracic CT in axial section without (A) and with (B) contrast: right posterior mediastinal cyst with regular peripheral contrast enhancement.

The patient was operated through a posterolateral thoracotomy. Total excision of the cyst and subtotal excision of the residual cavity was performed with simple operative follow-up, and a subsequent favorable evolution with return to the wall of the pulmonary parenchyma. Histological examination was performed confirming the diagnosis of hydatidosis.

Additional medical treatment with Albendazole 400 mg/day perioperative was prescribed (two weeks before surgery and six weeks postoperatively). No recurrence was noted at two years of regular survey.

Discussion

Extra-pulmonary intra-thoracic hydatidosis, affecting all organs of the thorax except the lungs, remains rare even in areas of high endemicity; it represents only 5 to 7.5% of all thoracic hydatid cysts. The cyst is often secondary, and can be multiple [1-8].

The mediastinal localization remains exceptional. Its frequency varies between 0.1% and 4% of human hydatidosis according to series [5,9,10]. The primitive forms of this localization, such as our case, are among the rarest forms of human hydatid disease, accounting for 0.1% of all localizations and less than 1% of intrathoracic locations [2,11].

In large series, 45% to 55% of mediastinal hydatid cysts occur in the posterior mediastinum, 36% in the anterior mediastinum, whereas the medial mediastinum is affected in only 8 to 18% of cases [5,12].

In the mediastinum, the clinical symptomatology of the hydatid cyst is nonspecific, and can vary from a simple respiratory discomfort to an acute picture of massive pulmonary embolism. The clinical spectrum is dominated by signs of mediastinal compression and signs of neighboring intra-thoracic structures involvement [2,5,9].

The hydatid cyst of the posterior mediastinum can cause pains, dorsal para-vertebral masses without signs of malignancy or neurological signs, and possibility of erosion of the ribs and vertebrae. Cases of prolonged fever, dyspnoea and haemoptysis have been described [5,9,12]. Sometimes their discovery is fortuitous on the occasion of a chest X-ray [2,5,9].

The diagnosis must be made early to avoid complications that are rare but not exceptional: superinfection, fistulization, mediastinitis, sepsis, compression of neighboring vital structures (trachea, esophagus, cellular veins, recurrent nerve ...), pulmonary embolism, and rupture in the aorta, or in the pleural or cardiac cavities with risk of recurrence and development of secondary hydatidosis [2,5-7,13].

The chest X-ray directs the diagnosis by showing a mediastinal opacity, quite often rounded or oval, which can be partially or completely calcified. It can also show a mediastinal widening. Calcifications of the wall are rare but represent a significant diagnostic argument and are mainly observed during primary hydatid cysts of the mediastinum [5,9,11].

Thoracic ultrasonography confirms the diagnosis when the lesion is accessible; the abdominal ultrasound performed at the same time looks for another hydatid localization, particularly in the liver [9,11].

Thoracic computed tomography is of great benefit, particularly in the presence of daughter vesicles (multi-vesicular hydatid cyst). It classically targets a roughly rounded mediastinal mass, often well-defined, hypodense, homogeneous, compartmentalized or heterogeneous, and calcified or not depending on the cyst's evolutionary stage, while specifying its exact location, its relationship with the intra thoracic organs, and the presence of any associated lesions, including pleuropulmonary cysts [2,9,11].

In the absence of a multi-Burette CT, thoracic MRI is needed to specify the size of the cyst, its exact location, and the invasion of adjacent structures. MRI is interesting in case of posterior mediastinal cysts, in search of medullary involvement or in the presence of neurological signs [2,9,11].

The hydatid serology is not very contributive to the diagnosis of these localizations because remains often negative, and its negativity does not have to eliminate the diagnosis. It contributes mainly to postoperative follow-up [2,5,9,14].

Early management is necessary to avoid the occurrence of potentially fatal complications. Surgery is the treatment of choice. It obeys the general rules of hydatid surgery: eradicate the parasite and treat the residual cavity. The surgical approach depends on the exact seat and size of the cyst. Transcutaneous therapy is possible and increasingly practiced according to the PAIR method: puncture-aspiration-injection of scolicide agent-reaspiration. The scolicide agent used is often the hypertonic saline solution [2,5,9,14-16].

Medical treatment based on oral benzimidazole derivatives is reserved for complicated forms, inoperable forms, and patients with associated multiple hepatic or pulmonary hydatid cysts. It is also recommended in combination with transcutaneous or surgical treatment to prevent secondary hydatidosis and recurrence [2,5,9,14-16].

Conclusion

The mediastinal localization of the hydatid cyst is exceptional and very little known by clinicians.

This entity deserves to be known and suspected in front of any respiratory signs that is not proven, especially in endemic countries. Radiological investigations are of a great diagnostic contribution, especially that the hydatid serology remains most often negative. Potential complications may be life-threatening, hence the need for early diagnosis and timely and appropriate management.

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