

Hydatid disease of the ribs: An exceptional location

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Background. Hydatid disease is a zoonosis caused by larval stages of cestodes belonging to the genus *Echinococcus*. The rib location is exceptional. It presents a real diagnostic and therapeutic challenge.

Objective. To describe the clinical, serological and radiological features and surgical management of rib hydatidosis.

Methods. This is a retrospective study conducted over 4 years, on five cases of rib hydatidosis. We analysed the clinical and radiological presentations and the adopted therapeutic procedure.

Results. The average age of our patients was 44 years, without gender predominance. The clinical signs were dominated by the presence of an immobile swelling of fluid consistency without inflammatory sign, accompanied by moderate and intermittent localised pain. The laboratory assessment was nonspecific. The radiological assessment, including chest X-ray and thoracic computed tomography, with and without contrast, was essential in order to assess the extent of the lesion. Thoracic magnetic resonance imaging was requested in one case because of suspicions of a spinal extension. All of our patients underwent a rib excision accompanied by medical treatment of albendazole 24 hours after the surgery. The follow-up ranged from 1 year to 4 years and did not show any recurrence.

Conclusion. Hydatidosis of the ribs is an exceptional location of hydatid disease. The diagnosis was based on radiology and intraoperative exploration. The treatment remained essentially surgical by rib excision with anthelmintic drugs to prevent recurrence.

Keywords. hydatid disease, genus *Echinococcus*, retrospective study.

Afr J Thoracic Crit Care Med 2022;28(3):115-118. <https://doi.org/10.7196/AJTCCM.2022.v28i3.193>

Hydatid disease is a zoonosis resulting from the growth of the larval form of *Echinococcus granulosus* (family Taeniidae).^[1] Involvement of the bones is rare, and the rib location is exceptional even in endemic countries like Morocco. It poses diagnostic and therapeutic challenges because of the clinical latency of hydatid disease and the severity of its prognosis linked to the risk of local recurrence and medullary compression, in the forms extended to the spine. The aim of our study was to present the clinical and radiological presentation and specify the surgical singularity in hydatid disease of the ribs.

Methods

This was a retrospective study conducted between 2016 and 2020, involving five patients operated on for rib hydatidosis. Cases with incomplete medical files were excluded from the study.

Different variables were taken into account: age, sex, geographic area, anatomical localisation of the cyst, dimensions, symptoms, signs, laboratory tests, imaging, and the surgical approach.

Results

The average age of our patients was 44 years, without gender predominance; 80% of the patients lived in rural areas. Previous history: One patient had been operated on for hepatopulmonary hydatidosis, one patient reported pathological fracture in the affected rib, and another had been operated on for pulmonary and vertebral hydatidosis (Table 1). The clinical symptoms were insidious in all cases, characterised by the presence of a swelling of fluid consistency (100% of cases), and moderate intermittent pain, localised (40% of



Fig. 1. Swelling without inflammatory sign localised at the level of the scar of the PLT.

cases) (Fig. 1). The laboratory assessment showed hypereosinophilia in 40% of cases and negative hydatid serology in all patients. Chest X-ray was requested first in 60% of our patients, showing well-defined rounded opacity associated with osteolysis (Fig. 2), and ultrasound of the soft tissues was requested first in only 40% of cases, showing hypoechoic multivesicular lesions. Thoracic computed tomography (CT), with and without contrast, was requested in 80% of our patients and confirmed the diagnosis in all cases (Figs 3 and 4). Thoracic magnetic resonance imaging (MRI) was requested for one patient for suspected spinal extension (Fig. 5).

Surgical treatment was instituted for all of our patients. The approach may be just an elective incision or thoracotomy. The choice of approach depends on the location of the lesion. The surgery consists of the removal of the cyst with rib excision through healthy areas.

Table 1. Clinical characteristics of patients and treatment

	Age	Sex	Previous history	Clinical	Symptoms delay	Location	Surgery	Medical treatment	Follow-up (mo.)
Case 1	63	F		Tumefaction + pain	15 yrs	Right 11th rib	RE	Albendazole	26
Case 2	29	F	Three episodes of pathological fracture of the rib	Tumefaction	8 mo.	Left 6th rib	PLT + RE	Albendazole	48
Case 3	52	M	Operated for hepatopulmonary hydatidosis	Tumefaction + pain	7 mo.	Right 8th and 9th rib	RE	Albendazole	34
Case 4	34	M		Tumefaction	6 mo.	Right 2nd rib	PLT Shaw-Paulson + RE	Albendazole	36
Case 5	41	M	Operated for pulmonary hydatidosis Operated for vertebral hydatidosis	Tumefaction	4 yrs	Right 7th and 8th rib	RE	Albendazole	36

F = female; M = male; RE = rib excision; PLT = posterolateral thoracotomy.



Fig. 2. Chest X-ray showing well-limited basithoracic parietal opacity.

We used a scolical agent (H_2O_2) to prevent parasite dissemination. Pathological examination confirmed the diagnosis by visualising the hydatid membrane and clarifying its multivesicular character. After 24 hours post-surgery, and for 6 months, medical treatment with albendazole was provided for all the patients. The follow-up for all patients was uneventful and no recurrence was noted.

Discussion

Hydatid cyst is defined by the development of a larval form of *Echinococcus granulosus* transmitted to humans either by direct contact with dogs or through ingestion of contaminated food. It can infest several locations in the body, such as the liver, lung or kidney.^[1]

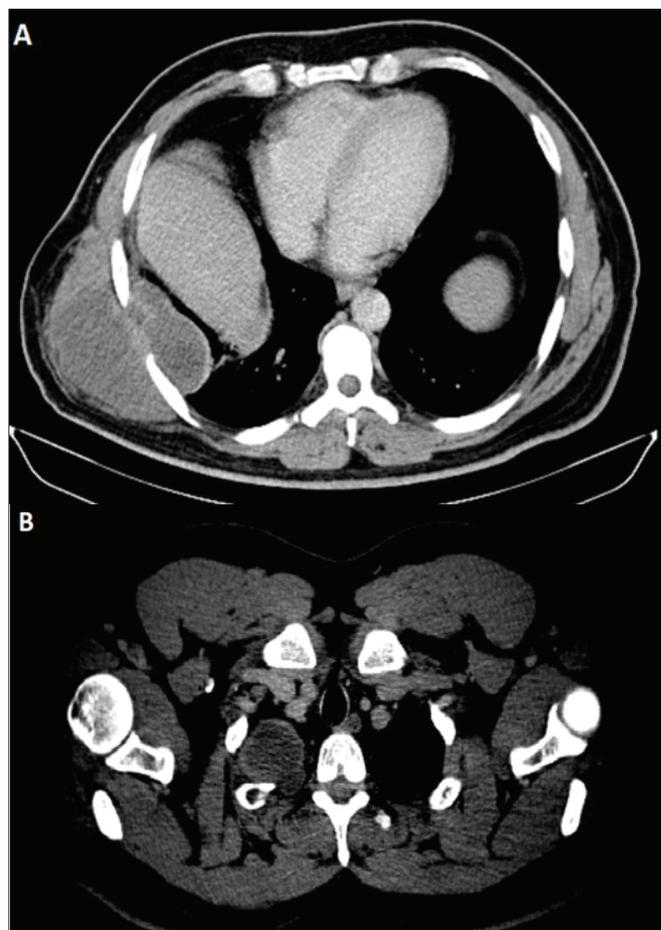


Fig. 3. Thoracic CT showing a parietal cystic mass with costal lysis of: A. the right 8th and 9th ribs; B. the right 2nd rib.

Bone location is rare and represents only 0.5 - 2% of all general hydatid localisations;^[2] rib involvement is exceptional and its frequency is estimated at 0.18 - 1.21%.^[3] Hydatid osteopathy is infiltrative,

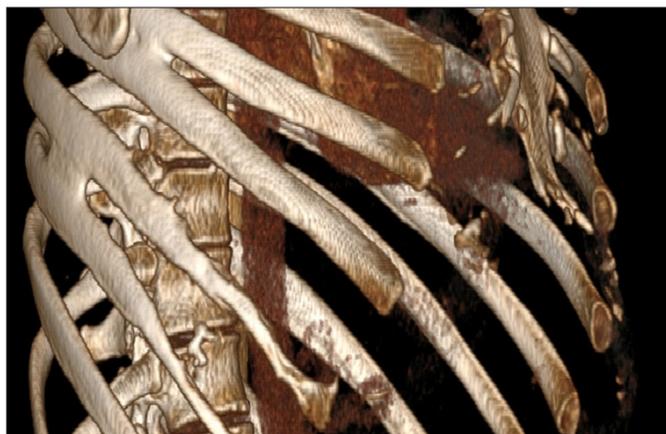


Fig. 4. Thoracic CT scan with bone reconstruction demonstrating costal lysis of the right 8th and 9th ribs.



Fig. 5. T2-weighted MRI image showing a hyperintense cystic lesion with a hypointense rim located within the right 7th rib.

progressive, with numerous microvesicles and without encystment of the parasite.^[1]

Rib hydatidosis can be primary or secondary owing to spontaneous rupture of a pulmonary or mediastinal cyst or by puncture of a subpleural location.^[4] Two of our patients had a history of pulmonary hydatid cyst. The clinical signs of rib hydatidosis are nonspecific; it is characterised by a slow evolution with moderate intermittent chest pain at the site of the lesion, while clinical examination reveals a local tumefaction without inflammatory signs, which generally reflects an extension to the soft parts of the parasitic lesions, but in some cases can be asymptomatic.^[4-6] The laboratory assessment may show hypereosinophilia but the rest is unremarkable. The hydatid serology (total and specific immunoglobulin E (IgE), enzyme-linked immunosorbent assay (ELISA), Western blot, immunoprecipitation, indirect haemagglutination test (IHT)) may be positive, but remains unspecific^[4,5] In our cases, hydatid serology was performed for all

patients, all of which came back negative, confirming the non-sensitivity of this examination. The radiological assessment plays a dual role: first, it allows the lesion to be located, and second, it provides the opportunity to find other locations. The chest X-ray remains the first-line examination, as it allows the lesion to be located. Rib hydatidosis manifests as an opacity of anterior or posterior parietal appearance, parallel to the axis of the bone without modification of its shape.^[7] Soft-tissue ultrasound can also be requested as a first step; it will show a hypoechoic multivesicular image with a posterior shadow.^[4] These two examinations, mentioned above, remain nonspecific.

The thoracic CT, with and without contrast, remains the gold standard for diagnosis of rib hydatidosis. It highlights multilocular lesions of fluid density with fine partitions, not enhanced after injection with contrast, and also makes it possible to show bone lysis and to specify the condition of the cortex which can be either blown, rolled or broken with endo- or exothoracic extension. At the level of the soft parts, the diffusion of the process gives a characteristic appearance of a cystic mass with a thin wall or an image of an abscess with a thick wall, or even a pseudotumour heterogeneous appearance.^[4,5] A thoracic MRI is requested especially for costovertebral locations where it is mandatory for preoperative assessment. It shows images with variable shape, rounded or oval, with fine walls, low signal on T1-weighted sequences and high signal on T2-weighted sequences, not taking the contrast except sometimes at the periphery; though a T1 hypersignal may be observed in relation to content rich in proteins.^[5,8,9] In our series, the chest X-ray and the soft-tissue ultrasound were the first examinations requested but they did not confirm the diagnosis. The CT, on the other hand, allowed the diagnosis to be confirmed in 80% of the cases. The MRI was requested in only one case because of a suspected spinal extension.

The differential diagnosis of rib hydatidosis includes malignant bone tumours (plasmacytoma or metastases), benign bone tumour (aneurysmal cysts, neurofibromas or giant cell tumours) and osteomyelitis caused by *Mycobacterium tuberculosis* or common pathogens.^[4]

The treatment of rib hydatidosis is based firstly on surgery, which consists of a large rib excision, passing through a healthy area; and secondly on medical treatment, based on albendazole pre and post surgery at a dose of 10 or 15 mg/kg (for 6 - 9 months) to minimise the risk of recurrence.^[4-6,10] In our series, rib excision through an elective incision was performed in most cases, but in one case the excision was partial through a posterolateral thoracotomy (PLT Shaw-Paulson), given the location of the lesion with placement of a chest tube. All our patients benefited from albendazole treatment 24 hours after surgery.

Conclusion

Hydatidosis of the ribs is an exceptional location of hydatid disease. The diagnosis is based on radiology and intraoperative exploration. The treatment remains essentially surgical by rib excision and anthelmintic treatment to prevent recurrence.

Declaration. None.

Acknowledgements. None.

Author contributions. All authors contributed to the study conception and design. All authors read and approved the final version.

Funding. The authors received no financial support for the research, authorship, and/or publication of this article.

Conflicts of interest. None.

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Accepted 8 July 2022.