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Case Report

Management of costovertebral hydatid cyst: A case report and review of the literature

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ABSTRACT

The hydatid bone cyst is very rare; it represents 2% for all locations combined. Vertebral involvement is often asymptomatic with an incidence of 40%; the diagnosis is made when complications arise related to the extension of the lesions inside towards the medullary canal or outside towards the rib grill. Surgery alone or in combination with medical treatment is essential whenever possible. Often emergency relief for neurological disorders; it must be carcinological in order to compensate for recurrences which are frequent. We report the observation of a 26-year-old young patient with a costovertebral hydatid cyst revealed by chest pain with paraparesis. The evolution was favorable after surgical excision associated with albendazol.

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1. Introduction

Hydatidosis is an anthrozoosis; due to the development of the larval form of *Echinococcus granulosus* in humans, the bone localization is very rare; the costovertebral location present 40% of bone hydatid, his evolution is very insidious with the risk of spinal cord compression and neurological disorders; this often imposes an emergency management associating the so-called carcinological surgical excision with laminectomy. We report in this case a young patient with a left costovertebral hydatid cyst.

2. Case Report

We received a 26-year-old man, Moroccan patient, coming from a rural area, he is being in contact with dogs; he has complained for 5 years of a left posterior basithoracic pain, with progressive aggravation without any respiratory signs. The clinical examination found painful palpation of the dorsal rachis, without palpable mass or associated

neurological deficit. The pleuro-pulmonary and all somatic examination was unremarkable. The chest X-ray (Figure 1) showed a well-limited homogeneous opacity; of posterior mediastinal appearance with a left paravertebral aspect.

The thoracic CT scan (Figure 2) showed a cystic formation with fluid content and multiple vesicles; occupying the left costovertebral angle with lysis of the posterior arch of the 7th, 8th and 9th rib. Given the context, a hydatid serology was requested and came back positive. Magnetic resonance imaging (MRI) was performed to look for endocanal invasion showing a foraminal extension D7-D8, with spinal cord compression (Figure 3 a,b).

We thus retained the diagnosis of costovertebral hydatid cyst with spinal cord compression, our patient benefited from an emergency surgical treatment in collaboration with the neurosurgery department, we carried out under posterolateral thoracotomy and protection of the operating site by compresses soaked in hypertonic serum, a complete resection and disinsertion of the posterior costal arches of K7, K8, K9 and of the affected tissues, resection of the affected vertebrae and laminectomy for decompression of

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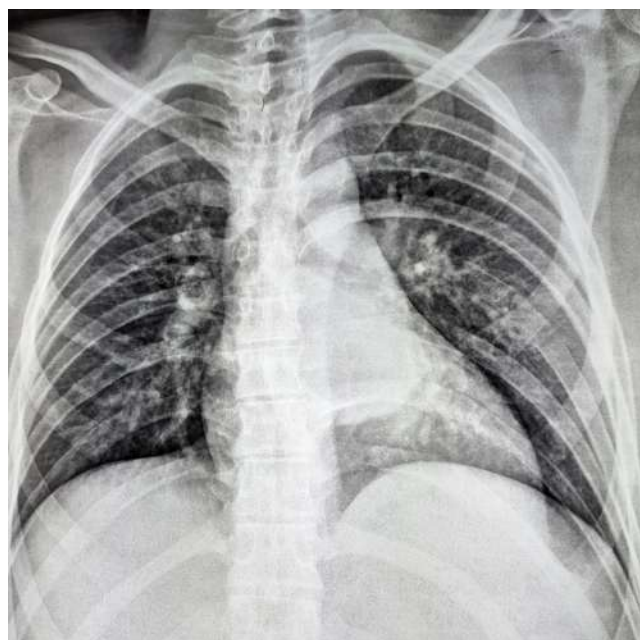


Fig. 1: Chest X-ray showing posterior mediastinal opacity in relation to costovertebral hydatid cyst

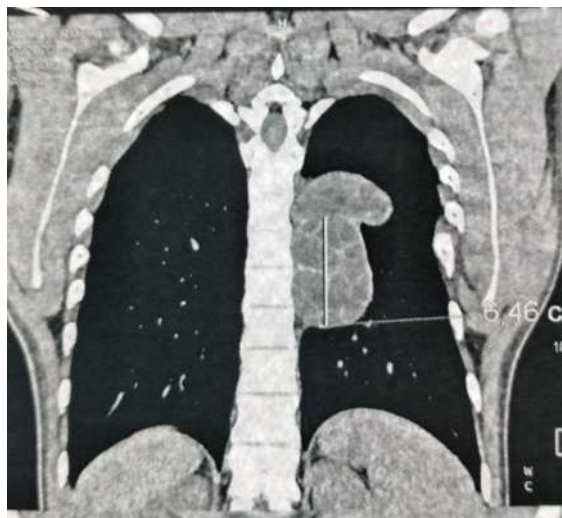


Fig. 2: Coronal thoracic CT section, mediastinal window showing a paravertebral multi-vesicular cystic formation

the spinal cord. (Figure 4). Stabilization of the spine by osteosynthesis with a screwed plate, and reconstruction of the chest wall with a vicryl plate. Placement of a thoracic drainage and closure.

The postoperative course was complicated at 5 days by the appearance of a reinit collection in front of the thoracotomy with serum content, which led to the diagnosis of a hydromatics that required percutaneous drainage. The postoperative radiography was satisfactory. The patient was treated with albendazol pre and postoperatively at a

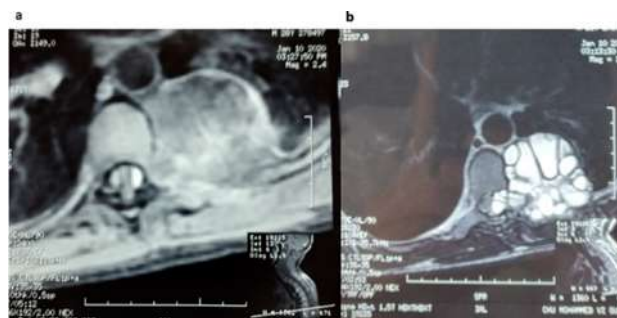


Fig. 3: a): T1-weighted axial slice of thoracic MRI shows a hypo intense cystic formation related to the hydatid cyst, b): T2-weighted axial slice of thoracic MRI shows a hyper intense cystic formation with endocanal extension of the vesicles and compression of the marrow



Fig. 4: Intra-operative view showing costovertebral resection and osteosynthesis of the spine

dose of 400 mg/day 6 days/7 for 6 months, at long term evolution (30 months) the patient had no recurrence of the disease, and it has no notable functional repercussions, notably paralysis or paresthesia, apart from a chronic neurological pain associated with a deformation of the thoracic wall having required a care in physical medicine and rehabilitation.(Figure 5)

3. Discussion

Hydatid cyst is a parasitic zoonosis present throughout the world, particularly in countries around the Mediterranean. It is caused by the larval form of the dog tapeworm (*Echinococcus Granulosus*), which constitutes the definitive host of the parasite. Man is an accidental intermediate host, infestation requires the proximity of man to breeding areas, this disease is favored by poor hygiene conditions and promiscuity in contact with stray dogs. The infestation in humans is done only by ingestion of eggs, which can remain



Fig. 5: Control chest x-ray after 2 years of evolution

viable for months in the environment.¹

The main location is the liver (60.8-80%) followed by the lungs (10-22.4%), the bone location is rare, present only 0.2 to 3% of all cases of hydatid cyst.² The common sites are the ribs, vertebrae (40%) and the upper extremity of the long bones. The costovertebral localization of hydatid disease constitutes a serious form due to the complexity of its therapeutic management, which requires the so-called carcinological surgery.^{3,4} Vertebral hydatidosis is often the initial lesion and the costal involvement is due to the contiguity with the posterior arch of the rib; this localization is explained by an arterio-venous system around the vertebral column favoring the deposition of hydatid material around the vertebro-medullary capillaries.^{3,5}

Clinical symptoms are generally insidious, as in our patient's case, and are limited to thoracic or spinal pain, which can be attributed to back pain or lumbosacral pain. The emergency is the appearance of neurological signs such as paresis or paralysis, often of the lower limbs, and thus constitutes a turning point in the evolution of the disease, that means spinal cord compression.⁶ Sometimes, the appearance of a parietal mass can be revealing, especially if the invasion is more costal than vertebral, often extending to the peri-cystic musculoaponeurotic soft tissue.^{3,5,7,8} The diagnosis is often presumptive based on a set of epidemiological, clinical and radiological arguments; confirmation is often only possible at the cost of surgery to bring back hydatid material for histopathological study.

The thoracic radiography; often performed in first intention in front of the presence of thoracic pain, it shows a well limited opacity of paravertebral seat with poly-lobed contours realizing the aspect of a posterior mediastinal opacity;³ as it is the case of our patient. At this stage, the differential diagnosis with other pathologies of the posterior mediastinum, in particular neurogenic tumors, requires a

thoracic CT scan and the use of magnetic resonance imaging in case of suspected spinal cord injury.

Computed tomography (CT) has an important role in the diagnosis of this pathology. The scannographic aspect is a cystic lesion containing multiple vesicles. It also allows a locoregional extension work-up; search for an associated pulmonary hydatid cyst. It allows an analysis of bony cortices degree of involvement (osteolysis; swelling; condensation...) and of the posterior wall, which will lead to the suspicion of an endocanal extension.⁹

Magnetic resonance imaging (MRI) has an essential place in this localization, it shows characteristic hydatid images in hyposignal on T1-weighted sequences and in hypersignal on T2-weighted sequences, unchanged after injection of gadolinium. It studies the relationship of the cyst with the spinal cord and soft tissues. MRI can also be used to judge the viability of the cyst by showing a T2 hypersignal if the cyst is viable; the absence of this hypersignal often indicates softening, which is a sign that the cyst has died.¹⁰

In our case, MRI confirmed the foraminal extension and the compression of the medulla by the presence of endocanal hydatid daughter vesicles. The onset of this compression may be acute and result in definitive paraplegia. Imaging therefore plays an important role in confirming spinal cord compression, requiring urgent spinal decompression surgery. This surgery must be associated with a monobloc carcinological resection of the costovertebral lesion as much as possible (costo vertebral disinsertion; vertebrectomy...), with stabilization of the spine by osteosynthesis. Recurrence is frequent after intraoperative effraction of the hydatid process; or incomplete resection. This recurrence varies between 30% and 100% according to the series.¹¹⁻¹⁶

Medical treatment is still necessary before and after surgery in order to prevent recurrence, for which repeat surgery is often impossible and the results are disastrous. This treatment is based on albendazole at a dose of 400 mg per day for 6 to 18 months;¹⁷ this treatment can be recommended as the only therapy in inoperable forms.

4. Conclusion

The costovertebral hydatid cyst is a rare localization, but serious because of the difficulty of its therapeutic management. The vertebral instability and the recurrence after surgery constitute a major handicap of the disease. The combination of surgical treatment with the most complete excision possible, the intraoperative use of scolicide to compensate for the almost inevitable invasion of the vesicles, and medical treatment with albendazol is the only way to guarantee the management.

5. Source of Funding

None.

6. Conflict of Interest

The authors declare that they have no conflicts of interest.

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