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Primary paravertebral hydatid cyst in children: unusual location and literature review.

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Abstract

Cystic Echinococcosis is a parasitic infection caused by Echinococcus granulosus, which is endemic in Algeria. The lungs and liver are primarily affected in children. Spinal location is uncommon and often characterised by insidious manifestations. Without timely surgical intervention supported by medical therapy, the condition can lead to significant morbidity and a high risk of recurrence. We report the case of a 12-year-old child with repeated hospitalisations for a psoas abscess. Further evaluation revealed a pre-spinal hydatid cyst, with MRI confirming a primary localisation in the lumbosacral area. The patient improved following surgical and medical treatment; however, the risk of recurrence remains significant. In endemic areas, cystic lesions can occur anywhere in the body. Highlighting the importance of considering echinococcosis. The current serological and imaging methods aid in diagnosis with high accuracy. To avoid complications and morbidity, cystic echinococcosis should be diagnosed paravertebrally as soon as possible. Treatment ranges from medical management to radical surgical excision.

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Background

Cystic Echinococcosis (CE), formerly known as hydatid disease, is secondary to a metacestode belonging to the Echinococcus granulosus sensu latu[1]. It is an infection spread worldwide, particularly in the Mediterranean rim, including Algeria. Uncommon primary locations can present with atypical features, delaying diagnosis owing to the insidious onset of symptoms and complicating therapeutic management[2].

This report describes an exceptional case of a primary paravertebral hydatid cyst of the lumbar spine, without involvement of any other sites, while emphasising the importance of medical imaging in the diagnosis and the success of treatment achieved in our department.

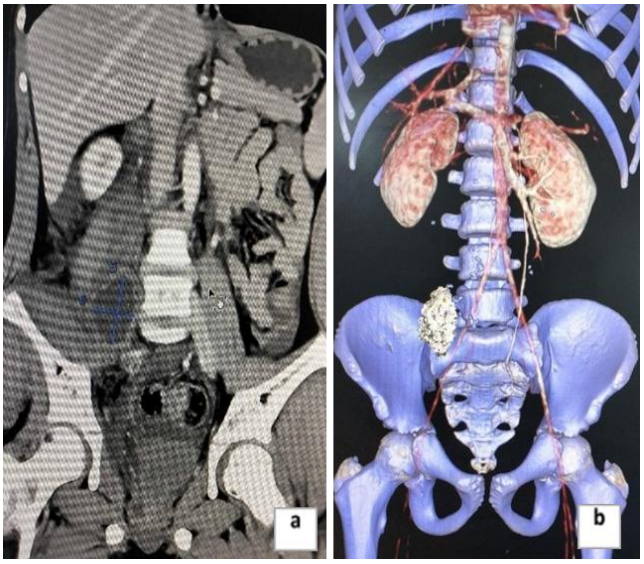
Case Presentation

A 12-year-old boy from rural origins was referred to our department after two prior hospitalisations for a psoas muscle abscess, for which he had received medical treatment.

Physical examination revealed that the child was in a good general condition with no signs of either fever or weight loss. The abdominal palpation reveals pain in the right lumbar region associated with a slightly palpable mass fixed to the deep abdominal plane. Both blood tests and tuberculosis explorations were negative. An indirect hemagglutination test for hydatid cyst was conducted and came back positive at 1/2560 UI. Abdominopelvic ultrasound revealed a hypoechoic image with an echogenic wall located in the distal third of the right psoas muscle. As for the abdominopelvic CT scan, it revealed a right

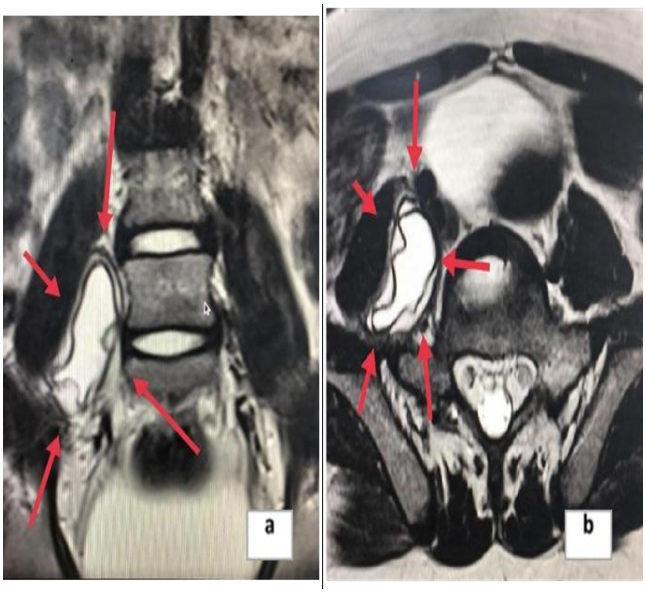
paravertebral cystic image without invasion of the medulla and other locations (Figure 1).

Figure 1: Abdominopelvic CT scan: (a) sagittal section: pre-spinal cystic formation; (b) a 3D reconstruction with a cyst located on L5-S1.



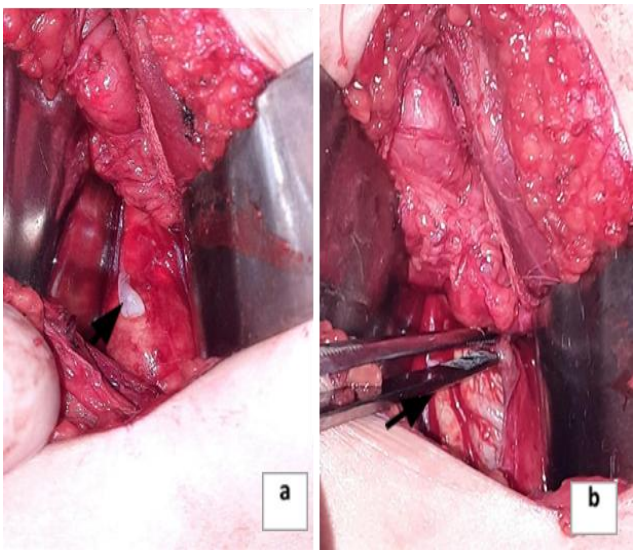
MRI demonstrated an oval cystic formation with low signal on T1 sequences, associated with pre-sacral membrane detachment, and no intracanal extension was demonstrated (Figure 2). The patient had undergone a surgical intervention, first by laparoscopic exploration, which did not conclude or visualize the cyst, and then a right para-rectal approach was conducted.

Figure 2: Abdominopelvic MRI: (a) sagittal section revealing cystic image extending from L5-S1 (red arrows); (b) axial section.



The latter visualised an intact right psoas muscle after its repression, and the opening of its aponeurosis, and a cystic mass of approximately 5 cm appeared extending in intimate contact with L5 and S1. The abdominal cavity was protected by hypertonic saline solution, partial cystectomy was done, and the germinal layer was removed. We proceeded with the sterilisation of the residual cavity with hypertonic saline solution and povidone-iodine for 20 minutes, then we proceeded with resection of the protruding dome (Figure 3), followed by drainage of the residual cavity.

Figure 3: Intraoperative image: (a) black arrow: germinal layer; (b) residual cavity.



The absence of protoscolex in the hydatid fluid was indicated in the parasitological examination; moreover, the histology confirmed the diagnosis of hydatid cyst.

The follow-up after the surgical treatment was marked by a positive evolution. An antiparasitic treatment based on albendazole was prescribed for 6 months to prevent recurrence. After 12 months, ultrasound imaging was requested, and no anomalies were observed.

Discussion

CE is still endemic around the Mediterranean rim, particularly in Algeria; it represents a significant public health problem, with the liver and lungs being the most commonly affected organs in children[3,4]. Vertebral involvement accounts for approximately 1% of all localisations. Spinal localisation generally

occurs by direct extension of an abdominal, pulmonary, or pelvic localisation and most often affects the dorsal region of the spine[5]. Vertebral cystic echinococcosis is most often located at the thoracic level (46-50%), followed by the lumbar spine (20-29%), then the sacral (20-23%), which is the least frequently affected. The localisation of the cervical region would be the least common. As for the primary intradural vertebral localisation, it is extremely rare [6, 7]. The frequency of vertebral localisation can be explained retrospectively: during a sudden increase in intra-abdominal pressure, the blood from the venules of the portal system would drain into the perispinal venous plexus at low pressure. This causes the passage of the hexacanth embryo to the vertebral or costal spongy tissue, thereby avoiding the hepatic and pulmonary filter. Unlike visceral localisations, the rigid structure of the bone prevents the formation of adventitia, hence allowing parasitic dissemination by microvesicular infiltration of the bone tissue. The progression procedure is based on a dual mechanism: diverticular budding and exogenous vesiculation[8].

Local hypervascularisation and the abundance of blood circulation are the reasons behind the thoracic and lumbar involvement[9]. Braithwaite and Lees have classified spinal involvement into 5 types: 1. Primary intramedullary, 2. Extramedullary intradural, 3. Extra-dural intraspinal, 4. Vertebral, 5. Paravertebral involvement[10] the latter corresponding to our case.

Due to this clinical latency, the diagnosis of the paravertebral form is often made during a neurological complication related to extensive bone infiltration. Thus, the rarity of the diagnosis in children may be explained by this evolution[8].

CT or MRI can easily diagnose pre-spinal cystic localisation as well as the follow-up. CT allows us to measure vertebral, intracanal, and soft parts of hydatid vesicle extension; however, MRI is currently the best medical imaging technique in terms of how it details the characteristics of the hydatid cyst. It also has an advantage over CT when it comes to assessing the connections of cysts with adjacent structures, in particular with soft tissues, especially in the spine and the spinal cord.

The differential diagnosis can be essentially made with spinal tuberculosis, an extramedullary neuroenteric cyst, or, more rarely, with certain tumors[6]. Surgery remains the "gold standard" in the choice of a therapeutic arsenal; it consists of a radical resection of the cyst. Unfortunately, this excision cannot be done because of the location; therefore, recurrences are frequent. Protecting the surgical field with hypertonic saline solution is recommended by several authors to avoid its dissemination. Antiparasitic medical treatment with albendazole is recommended in association with surgery in the pre- and postoperative period to prevent recurrences for a period of between 4 and 6 months[7]. The prognosis is unpredictable because of the high frequency of recurrences[9].

Conclusion

The paravertebral localisation of CE remains very rare, even rarer when it is primary; it represents a real diagnostic and therapeutic challenge due to its clinical latency, the difficult radiological approach, and its management protocol because of the unpredictable evolution.

This diagnosis should be kept in mind, especially in endemic countries; it must be made at an early stage to prevent complications and morbidity related to this parasitosis.

Surgical excision alongside anti-parasitic medications is the best treatment, irrespective of location; prevention remains the optimal way to reduce its frequency. Location: prevention remains the optimal way to reduce its frequency.

Conflict of Interest Statement: The authors declare that there is no conflict of interest regarding the publication of this article.

Consent for publication: 'Written informed consent for publication of their clinical details and/or clinical images was obtained from the parent or guardian of the patient

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