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(CASE REPORT)

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Vertebral and Spinal Cord Hydatidosis: A Case Series

Israe ALMAGHRIBI *, Yahya Farah, Mustapha EL BAYAD, Mustapha HEMAMA, Nizare EL FATEMI and Moulay Rachid EL MAAQILI

Department of Neurosurgery, Hospital IBN SINA-Rabat, Mohammed V University of Rabat, Morocco.

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Abstract

Vertebral and Spinal Cord Hydatidosis is an infectious pathology caused by the parasite Echinococcus granulosus. It most often affects the liver and lungs. Spinal hydatidosis represents only 1% of cases, and constitutes a public health problem, particularly in endemic areas, notably in Morocco. Despite its generally benign nature due to its rarity and local growth, vertebra medullary hydatid cyst is classified as a malignant disease due to its high potential for dissemination, which can lead to a notable incidence of recurrence and morbidity. The vertebro-medullary location affects the clinical presentation. Diagnosis has evolved through the magnetic resonance imaging and advances in the genetics, genomics and molecular epidemiology of parasites. Surgery is the treatment of choice associated with anti-parasitic medical treatment with albendazole is recommended to prevent recurrences. The prognosis is uncertain due to the risk of local recurrence and spinal cord compression. Here in, we present a retrospective study of a series of 9 cases of Vertebral and Spinal Cord Hydatidosis diagnosed at the IBN SINA RABAT hospital, during the period from 2009 to January of 2024. Data from radiological, clinical, therapeutic, and evolutionary studies were reviewed. As a conclusion, the objective of this work is to implement awareness-raising actions among the Moroccan population to establish early diagnosis and to involve means of prevention, and to emphasize the importance of therapeutic care to avoid possible recurrences.

Keywords: Hydatidosis; Cysts; Spinal cord; Posterior decompressive; Laminectomy; Albendazole

1. Introduction

Hydatid disease is caused by the tissue development of Echinococcus Granulosus, a parasite which is found in the adult state in the small intestine of canines. Intermediate hosts include herbivores, while humans can be affected following accidental ingestion of food contaminated with the parasite's eggs [1]. In the Mediterranean region, especially in Morocco, it is regarded as endemic [2]. Hydatid cysts are mainly seen in vascularized organs such as the liver, lungs and brain. Although bone involvement is rare, the spinal location is notable in 44% of cases, probably related to the vascular richness of the spine, often associated with spinal cord involvement, hence the term vertebro-medullary hydatidosis [3-4]. To date, the treatment of vertebromedullary hydatidosis remains complex, mainly surgical, and the effectiveness of an association with medical treatment remains to be proven. It is important to note that the problem of recidivism still exists [5]. The aim of our work is to carry out a descriptive analysis while evaluating the epidemiological and clinical characteristics, as well as the medical and surgical management, and to examine the evolution of cases of vertebromedullary hydatidosis diagnosed at the neurosurgery department of IBN SINA-RABAT hospital, extending from 2009 to 2024, while analyzing and discussing our results compared to those of the literature.

* Corresponding author: Israe ALMAGHRIBI

2. Clinical Presentation

This is a retrospective descriptive study of the epidemiological, clinical, paraclinical and therapeutic characteristics of a series of 9 cases of vertebro-medullary hydatidosis collected and analyzed according to a pre-established operating sheet over a period of 15 years (January 2009–January 2024) in the Department of Neurosurgery of the hospital IBN SINA in Rabat. A review of the clinical, radiological, therapeutic and evolutionary features was carried out.

2.1. Epidemiological features

- The average age of patients was 40 years, with a clear male predominance (77% of cases), while 67% of patients came from rural areas.
- The osseous form of hydatidosis is rare; it does not exceed 2% of all hydatid locations, while the most common location is the thoracic spine, representing approximately 50%, followed by the lumbosacral region 29%, and the lumbar spine, representing approximately 21%.

Table 1 Demographical features of patients

Cases	Age	Sex	location
Case N° 1	Male	40 Y	L5–S1
Case N° 2	Male	29 Y	T8-T10
Case N° 3	Male	45 Y	L4
Case N° 4	Male	26 Y	T8
Case N° 5	Male	36 Y	T12-L1
Case N° 6	Male	48 Y	L1-L3
Case N° 7	Female	45 Y	T1-T3
Case N° 8	Male	35 Y	L5–S2
Case N° 9	Female	61 Y	T8-T11

2.2. Clinical Presentation

Table 2 Clinical Presentation

Case Number	Time to onset of symptoms	Functional signs	Clinical Examination	Frankel Score
1	1 year	Lumbosciatalgia + Genito-Sphincter Disorders (Urinary Incontinence)+ Lower limb deficit	left monoparesis 4/5 (lower limb)	Grade D
2	2 years	Back pain	Dorsal kyphosis	Grade E
3	4 months	Lumbosciatalgia + Genito-Sphincter Disorders+ Lower limb deficit+fever	Paraparesis 4/5	Grade D
4	1 month	lower back pain+ Urinary Incontinence	paraplegia	Grade B
5	20 days	Back pain+ Lower limb deficit	Spastic paraplegia	Grade B
6	6 months	Lumbosciatalgia + Genito-Sphincter Disorders (erectile dysfunction) + Lower limb deficit	right monoparesis 4/5 (lower limb)	Grade D
7	4 months	Upper back pain+ Urinary Retention + Deficit	Spastic paraplegia	Grade A
8	2 months	Lumbosciatalgia + Genito-Sphincter Disorders +Deficit	Spastic paraplegia	Grade B
9	15 days	Back pain + Urinary Retention + Constipation + deficit	Spastic paraplegia	Grade A

The circumstances of discovery revealed by clinical symptoms were: the predominance of low back pain representing 44% of cases, compared to back pain (25% of cases). In contrast, 87% of patients presented with deficit symptoms. Sensory disorders were present in only 43.75% of cases, with sphincter disorders in 68.75% of our patients.

The most common clinical picture is spinal cord compression, which is identified in 43% of clinical cases, with cauda equina syndrome in 50% of clinical presentations.

2.3. Diagnostic Workup

The hydatid serology using the ELISA technique is the most commonly used specific assessment with a sensitivity greater than 90%, while vertebro-medullary MRI is the most efficient.

The management is based on surgery, aimed at decompressing the spinal cord and resecting hydatid cysts without rupture to prevent recurrence.

2.4. Surgical Intervention

Surgical techniques are varied; posterior resection and laminectomy in 75% of our cases, while one case was treated with an anterior approach by corpectomy, while another required both anterior approaches for cyst removal and a posterior approach for laminectomy. Half of the surgical procedures, included a stabilization procedure using osteosynthesis.

The use of medical treatment with Albendazole (100% of cases) has demonstrated significant results in preventing recurrences.

Relapses are common, with approximately one-third of treated patients (30% of cases) experiencing multiple recurrences accompanied by lesion extension.

Table 3 Management

Case Number	First Surgery	Postsurgical Recovery	Postoperative complications/ recurrence
1	Laminectomy T2+ posterior decompression	Complete recovery	-
2	laminectomy T8, T9 and T10 with osteosynthesis + Evacuation of cysts	no preoperative deficit	-
3	laminectomy and arthrectomy L5 + posterior decompression	Sensory and partial motor	Recurrence
4	laminectomy T8-T9 +osteosynthesis T7-T8-T9 + anterior Evacuation of cysts	Sensory and partial motor	-
5	Laminectomy T12-L1 + posterior decompression	Complete recovery	-
6	Laminectomy L3 + + posterior decompression + osteosynthesis T11-L3	Complete recovery	Recurrence + Surgical site infection
7	Corpectomy T1-T2 with insertion of a pyrameche-type cage + excision of the remodeled cysts	Complete recovery	Anterior migration of the Pyrameche + Recurrence
8	laminectomy L5 + posterior decompression	Sensory and partial motor	-
9	Laminectomy T8, T9 and T10 + Osteosynthesis T7-T8 and T10-T11 +Excision Evacuation of cysts	Sensory and partial motor	-

Figure 1 Sagittal and axial sections of the dorsal CT scan of one of our patients showing a right costovertebral lesion process extending from T7 to T12 with compression fracture of T8 and reduction of the medullary canal at the level of T8-T9

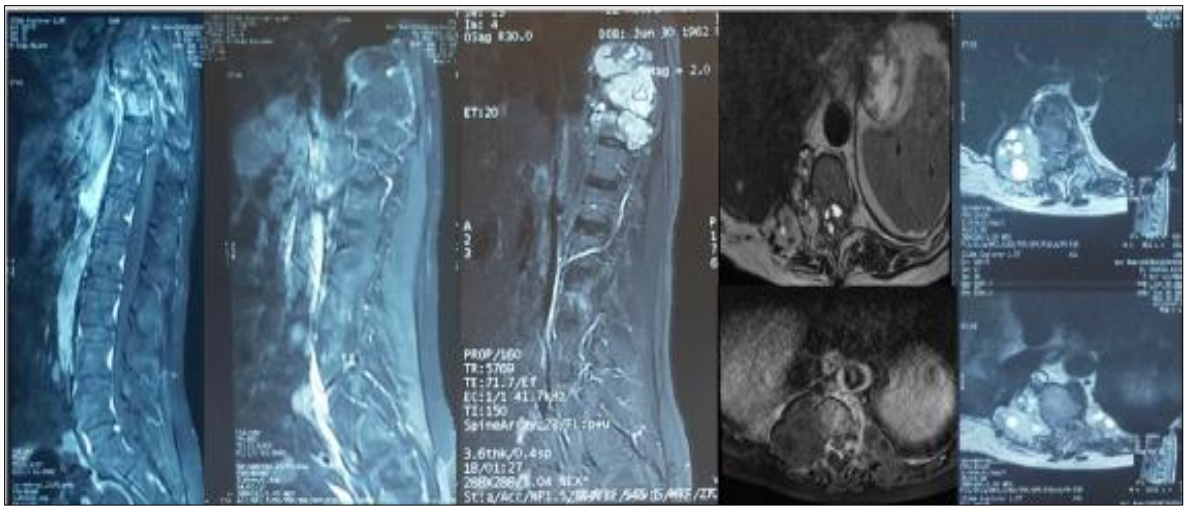

Figure 2 Axial and sagittal T2 and T1 weighted Magnetic resonance imaging (MRI) with contrast enhancement showing a right costo-vertebral process extended from T7 to T12, with polylobed tissue contours not enhanced after injection of godalinium containing rounded cystic formations related to hydatid vesicles, extending to the medullary canal

Figure 3 Intraoperative images of one of our patients showing the presence of multivesicular lesions of the costovertebral groove with a macroscopic appearance composed of these multiple daughter vesicles with a hydatid cyst membrane after cyst excision followed by lavage with hypertonic saline

Figure 4 Intraoperative and radiological images of the same patient showing the osteosynthesis procedure by pedicle screwing at D7-D8 and D10-D11, with laminectomy at D8, D9 and D10

3. Discussion

Hydatid disease of the spine usually manifests insidiously over years after the initial cyst infestation and is characterized by a long incubation period [6]. .and by the slow progression of cysts from the surrounding bone. The average time between the onset of symptoms and diagnosis was 3 months [4].

The presenting signs are generally nonspecific, frequently manifesting after local neurological complications resulting from spinal infiltration by hydatid vesicles that extend to intracanal structures, causing pain and neurological symptoms, or by forming peri-spinal paravertebral swellings [7]. These manifestations are recorded in 57% of cases, according to a study by Zlitni (2012) [8]. Morocco is endemic, with a prevalence of 7.76 per 100,000 inhabitants, the incidence of operability of 4.55 cases per 100,000 inhabitants places Morocco in third place in the Maghreb countries after Tunisia and Algeria [9]. The rural living environment is the most affected (65%) according to a study carried out in the region of Gharb Cherarda. This could be explained by the fact that rural populations are in permanent contact with the main host, the dog [9]. The proportion of patients in contact with dogs in our population reaching 78% underlines the significant role of contact with dogs, particularly in rural contexts.

Vertebro-medullary involvement of hydatidosis shows no age preference, affecting all age groups. However, the incidence is higher in young adults aged 30 to 36 years, with an overall mean age of 36.6 ± 1.2 years, which is attributable to diagnostic delay [10-11]. In our series, the mean age at diagnosis is 40.4 years, which is consistent with the literature where the most represented age group is between 30 and 40 years and 40 and 50 years. Men seem to be the most affected by this disease [12]. In our study population, the M/F sex ratio is 3.5. Other studies describe a lower M/F ratio such as Kafaji (2013) where this ratio was 1.35 M/F and the study by Luan (2022) demonstrated a ratio of 1.2 M/F [13-14].

Hepatic hydatidosis is the most common form, representing a frequency of 75% [17]. Although the lungs show a frequency of 15%, these two organs remain the most predominant, totaling 90% of cases. It is also possible for hydatidosis to develop in any organ of the body [9]. The osseous form of hydatidosis is rare despite its endemic nature in the Maghreb countries, not exceeding 2% of all hydatid locations [15]. It is associated with a visceral cyst in 12 to 25% of cases [16]. Among the osseous forms, spinal locations are the most frequent, representing 44% of cases. This is explained by the vascular richness of the spine and the venous plexuses which favor this manifestation, making it also particularly serious [16] [18]. In fact, this form constitutes 34% of cases of hydatidosis of the entire central nervous system [19]. The most common location in the vertebral segment is the thoracic spine, representing about 50% [18]. This is followed by the lumbosacral region, which constitutes about 29%, and the lumbar spine, which represents about 21% [10] [20]. In our context, three cases with dorsal hydatid location have a history of pulmonary location in two cases and mediastinal location in one case [21]. Hydatid cysts are not usually limited to the vertebral bodies, they affect the spinal cord and the posterior elements of the spine, being able to develop in the spinal canal [22].

The majority of patients present with a spinal syndrome characterized by progressive onset of low back pain accompanied by radicular pain [23]. Our study significantly highlights the predominance of pain located in the lumbar region, representing 44.4% of cases, compared to dorsal localization, which constitutes 33.3% of back pain. Muscle

weakness of the limbs manifests at a later stage. Other symptoms observed include radiculopathy, myelopathy and pathological fractures [10-11]. Pathological fracture was observed in one patient in our population. Consequently, paraparesis, sensory disturbances and sphincter disorders are frequently observed [24]. Using the Fränkel score to compare our results with the literature, we observe that 22.2% of cases present a Grade A according to the Fränkel classification.

Vertebro-medullary MRI is emerging as the imaging tool of choice for diagnosing vertebro-medullary hydatidosis, accurately highlighting its exact location and relationship to anatomical structures such as the spinal cord, as well as its extension into the surrounding soft tissues and to the spinal cord [25-26]. Compared to computed tomography (CT), it is recognized as a superior imaging modality, particularly in the assessment of recurrences after surgical treatment [8]. In 90% of cases of spinal hydatidosis, the lesion is limited to the bone and the epidural space [27]. All segments of the spine can be affected, with variable frequency, manifesting mainly as involvement of the thoracic and lumbar spine [7] [28]. Initial involvement of the cervical site is particularly rare, and synchronous multifocal association of thoracic and cervical involvement is extremely rare [29-30].

Usually, the diagnosis is based on the use of well-established techniques such as enzyme-linked immunosorbent assay, Western blot, indirect hemagglutination tests [31]. These methods are commonly used to detect the presence of anti-echinococcal antibodies (immunoglobulin G), as well as polymerase chain reaction [4] [32]. In our context, 55.6% of the population benefited from a specific test by ELISA technique, with a positivity rate reaching 100% of the tests. Hydatid serology by ELISA technique is the most commonly used specific assessment with a sensitivity greater than 90% [33]. The anatomopathological examination is essential to confirm the diagnosis, cystic vitality and changes especially during the first occurrence of hydatidosis and is done after surgical resection [19]. The diagnosis of vertebro-medullary hydatidosis is complex and must be considered in the presence of a combination of epidemiological elements, clinical course, radiological findings, including MRI, and serological data [21]. However, diagnostic confirmation is based on the per-operative observation of multivesicular lesions consisting of daughter vesicles with their whitish membrane, in association with an anatomo-pathological or parasitological study of the material resected during surgery [6] [30].Specific antiparasitic medical treatment plays a crucial role in the management of spinal hydatid disease, constituting an essential adjunct to surgery to prevent recurrences and reduce complications when used postoperatively, particularly due to the difficulty in achieving total resection without rupture of the cystic vesicles. This treatment is based on two benzimidazole anthelmintics, Mebendazole and Albendazole [4].

The treatment of hydatid disease is primarily surgical. All experts recommend this surgical approach to eliminate the disease at the spinal level. The objective of surgery is to completely resect the hydatid cyst and its intact scolex, without causing rupture, in order to prevent recurrences through precautionary measures. Another major objective of surgery is spinal cord decompression, aimed at relieving the patient and recovering from any neurological deficits [30]. In addition to spinal cord decompression, another essential objective of surgery includes stabilization and strengthening of the affected spine as well as correction of spinal deformities resulting from infiltration of vesicles into the vertebral bodies, or pathological fracture following the disease [14]. However, complete resection of cysts often proves impossible due to their infiltrative nature and severe and extensive bone destruction, undefined anatomical features and existing nerve radicles [29]. For this purpose, several techniques have been adopted to stop the extension of hydatid vesicles [4].

According to the literature, the treatment of vertebro-medullary hydatidosis is based on various surgical options ranging from less invasive procedures to more serious surgical interventions. These options include simple drainage with debridement or curettage and resection of the infected bone, as well as posterior or anterior decompression and stabilization [34] [35].

Scolicidal treatment was applied by all authors, using mainly hypertonic saline solution in all invasive surgical procedures [14][29]. Due to the significant risk of relapse, it is imperative to ensure regular follow-up of patients. The prognosis of the disease is closely related to the frequency of recurrences reaching 40 to 60% and to major surgical interventions, attributable to the malignant tumor nature of the disease due to its capacity for infiltration and bone destruction (white cancer) [14] [27] [36].

4. Conclusion

Vertebro-medullary hydatidosis is very rare. Diagnosis in radiology is based mainly on magnetic resonance imaging. Surgery is the preferred treatment option. It is considered a very aggressive lesion due to the extent of the lesions and the constant recurrence and iIts diagnosis is generally late, constituting a public health problem in Morocco.

Compliance with ethical standards

Disclosure of conflict of interest

No conflict of interest to be disclosed.

Statement of informed consent

Informed consent was obtained from all individual participants included in the study.

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