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Case Report

Primary Lumbar Extradural Hydatid Cyst: Case Report

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Abstract

Vertebral hydatid cysts are found in <1% of all cases of hydatidosis. This localization has an infiltrative malignant nature, affecting the vertebral body with possible extension in the epidural space. Primary extradural hydatid cyst of the spine without any other systemic involvement is extremely rare entity. We report a case in young a man and we review different aspect of this pathology.

Keywords: Hydatidosis, hydatid cyst, primary, Spine, extradural, Echinococcus granulosus, Bone involvement, Spinal cord compression, Albendazole.

INTRODUCTION

Primary epidural hydatid cyst of the spinal canal is very rare condition, characterized by the presence of extra-dural hydatid vesicles without associated bone lesions. Spinal involvement generally develops secondarily, with direct expansion from the lung or abdominal portal venous anastomoses or foci. However, primary spinal involvement without another focus can also be observed [1]. Carrea and Murphy reported the first case, in 1964 [2], then sporadic cases have been published [3, 4]. A rare case of primary lumbar extradural hydatid cyst, in 37 years old man, which causes cauda equina compression, is reported and clinical presentation, diagnosis and surgical treatment are discussed.

Case Report

A male shepherd of 37-year-old with history of lumbar radicular pain, numbness in both lower extremities for several years, treated in our department for cauda equina compression evolving for three months prior to admission. Neurological examination revealed incomplete paraplegia with 2/5 bilateral muscle strength with hypoesthesia below T10, perianal hypoesthesia. There was loss of patella and Achilles reflexes, as well as urinary and anal incontinence. The patient was classified as ASIA C. Magnetic resonance imaging (MRI) of the thoraco-lumbar region showed an extradural cystic lesions with a “bunch of grape” appearance and regular contour located in posterior. There was cerebrospinal fluid like signal intensity on T1- and T2-weighted images. The lesion had excessively compressed the dural sac and caudal roots, and extended from T12 to L4 with paraspinal extension through neural foramina widening (Figure-1), without any bone or disk involvement. Ultrasonography and thoracic and abdomio-pelvic CT scan was normal, and serological tests (specific ELISA/Western blot) proved negative. The patient underwent urgent laminectomy of 5 levels from T12 to L4; multiples size extradural vesicles were found in para and intraspinal with no adhesion to meninges or nervous structures. Total removal of the cysts and their foraminal extension was performed without rupture (Figure-2). The area was freely irrigated with hypertonic saline solution. Given the extent of the laminectomy the patient has undergone posterior transpedicular screw stabilization. Parasitological examination found cysts without hydatic scolex and the diagnosis of hydatid cyst was confirmed on histopathologic examination. The patient was taken in to the rehabilitation program immediately. And Albendazole treatment was initiated in the early postoperative stage: 400 mg twice a day during 6 months with 15-day free-drug intervals. The outcome was marked by recovery of the motor deficit and sphincter disorders. No recurrence has been found over 2-year follow-up.

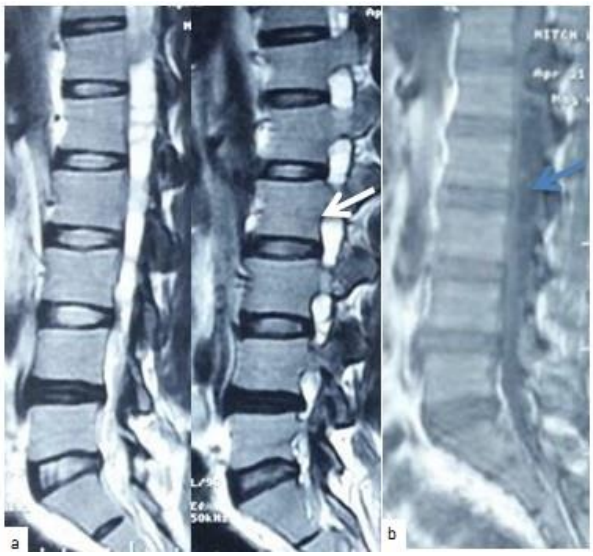

Figure 1: Preoperative sagittal T2 (a) and T1 (b) weighted spinal MRI images showing cystic lesions located in extradural space, extended from T12 to L4 (bleu arrow), with paraspinal extension through neural foramina widening (arrow)

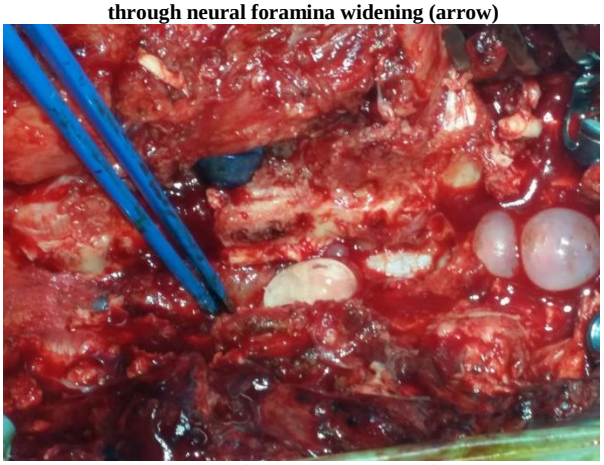

Figure 2: multiples size of extradural vesicles found in para and intraspinal through widening neural foramina, with no adhesion to

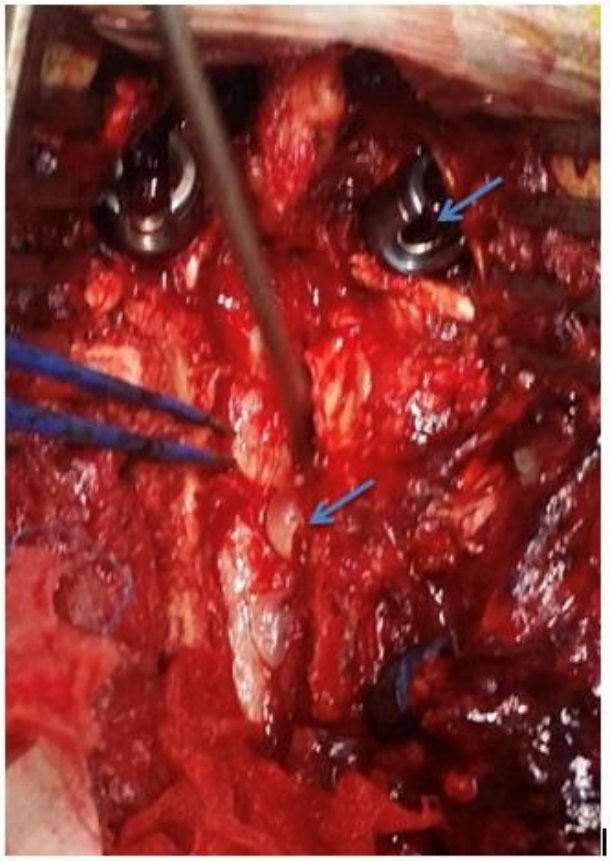

Figure 3: Laminectomy, extradural cysts, (arrow) Total Removal, And Posterior Transpedicular Screw Stabilization (arrow)

DISCUSSION

Hydatid disease or hydatidosis is a common anthropozoonosis in the endemic countries of North Africa, Central Asia and the Middle East [6, 7]. Human especially young people can be accidental intermediate hosts by ingesting food contaminated by Echinococcus cestode or by direct contact when petting an infested dog, which is the main host of the parasite. Fifty to 75% of these patients are children and young adults, with a male predominance in most series reported in the literature [17]. The most affected organs are the liver (65%e70%) and the lung (20%e27%). The frequency of osseous involvement in hydatid disease is 1% [36, 37]. It most commonly occurs in the spine (50%) and pelvis [5, 9, 12]. Cestodes usually spread to the spine by direct extension from pulmonary, abdominal or pelvic. However, primary extradural hydatid cyst without vertebral involvement is very rare [8, 9]. An analysis of the literature using the following key words: Hydatidosis hydatid cyst, primary, Spine, extradural, MRI, Surgery, Echinococcus granulosus, Bone involvement, Spinal cord compression, Albendazole. Allowed us to identify 24 cases, 3 published in Morocco including present paper [47, 31]. All available cases of primary spinal extradural hydatid cysts are presented in Table-1.

Table-1: All primary spinal extradural hydatid cysts published in the literature (24 cases including this paper)

	No of patient	Age	Genre	Location	Serology	Neurologic status	Radiology	Treatment	Outcome
Carrea R, Murphy G	-	-	-	-	-	-	-	-	-
Pluchino and Lodrini (1981) [38]	1	56	Male	T10–L2	Casoni(–)	Paraparesis	Myelography MRI	Posterior	Complete recovery
Wani and al (1989) [39]	1	14	Male	T9–10		Paraparesis	CT, MRI	Surgery	Complete recovery
Kars and al (1990) [40]	1	40	Male	C5-6	Casoni(–)	Tetraparesis	Myelography	Posterior	Normal
Bavbek and al (1992) [2]	1	40	Male	T5-9		Normal	Myelography MRI	Posterior	No change
Tekkök and Benli (1993) [42]	1	54	Male	L2-5		Cauda equina synd.	MRI	Posterior	No change
Baysefer (1996) [43]	2	21 22	Male	T5-T6		Paraparesis	CT,MRI	Posterior	Improved Improved
Pandey and Chaudhari (1997) [44]	1	15	Male	S1-2		Paraparesis	MRI	Posterior Posterior	Improved
Bayar and al (1997) [45]	1	30	Female	L5-S1		Cauda equina synd.	Myelography,MRI	Posterior	Improved
Berk *et al*., (1998) [46]	1	17	Male	T7-9		S1 radicular findings	CT, MRI	Posterior Posterior	complete recovery
Bouklata and al (2000) [47]	1	08	Male	T8-11		Paraparesis	Myelography	Posterior	Complete recovery
Karadereler (2002) [1]	1	08	Male	L2-5	Elisa (–)	Cauda equina synd.	Myelography MRI	Posterior	Normal
Sharma NK (2003) [26]	1	40	FEMALE	L1-L2		paraparesis	MRI	Posterior	Complete recovery
Awasthy (2005) [27]	1	15	Male	T4-T5		Paraparesis	MRI	Posterior	Complete recovery
NN Gopal (2007) [12]	1	38	Man	T2-T3	Negative		MRI	Posterior	Complete Recovry
Sushila and al (2009) [29]	1	21	Female	T10		Paraplegia	MRI	posterior	Improved
Eloqayli and al (2010) [30]	1	04	Male	T6-T9		Paraplegia	MRI	Posterior	No change
Boulahroud and al [31] (2012)	1	44	Female	T11-L4	negative.	paraplegia	MRI	posterior	Improved
Karakasli, al (2015) [28]	1	17	Male	T3-T4	ELISA(-)	paraparesis	MRI	Posterior	Complete recovery
I.Dogan and al (2015) [32]	1	09	female	T12		paraparesis	MRI	Posterior	Complete recovery
Gennari A And Al (2016) [33]	1	25	Female	T8, T10		Paraparesi	MR	Posterior	Complete Recovery
Mnari W (2016) [34]	1	42	Female	T11-L3	positive	paraparesis	MRI	Posterior	Complete recovery
Sridharan S (2017) [35]	1	64	Female	D8-D12		Paraplegia	MRI	Posterior	Improved
N Raouzi 2019 present paper	1	37	Male	T12 -L4	Negatif	Paraparesis	MRI	Posterior	Complete recovery

The involvement of the spine without any other systemic localization can be explained through the direct porto-vertebral venous shunt theory: in rare instances, the disease begins from the extradural area, suggesting that the parasite's embryo is possibly being carried through the porto-vertebral venous shunts. Then, the parasite reaches the retroperitoneum, spinal and paraspinal structures via lumbar epidural venous plexuses [10, 11]. In a large series of patients with spinal HC disease, the most prevalent location was the

thoracic area (range, 45%e50%), followed by the lumbar (range, 20%e39%), sacral (20%), and cervical (10%) areas [16]. This emphasis on the spine, especially the thoracic and lumbar regions in spinal HC, is attributed to heavy local vascularization and the rich blood circulation of the spongy vertebral bones. Spinal cord compression is a frequent presentation but neurological symptoms are various and non-specific [13]. The different series seem to underline an important rate of paraparesis at presentation (61% to 73%), associated or not with back pain (27.8% to 43%), bladder dysfunction (11.1% to 32%), sensory loss (24%), and radicular pain (27% to 60%) [14, 15]. In our case the hydatid cyst was extradural and located in the lumbar

CT and MRI show the anatomical position of the lesion, the osseous portion of the lesion, extension, and neuronal involvement. MRI is the exam of choice in case of suspicion of Spinal Hydatid Cyst Disease. The typical appearance is that of well-circumscribed, cystic lesions, as a multiloculated mass with CSF-like signal intensities. Hypointense T1-weighted images, hyperintense on T2-weighted images with sharply defined, hypointense cyst wall without enhancement following intravenous gadolinium, in some cases an enhancement reflect the vascularity of the pericyst in case of muscle hydatid cyst [18]. But there is no contrast enhancement either in extradural or intradural hydatid cysts [12]. Extradural spread of hydatid cysts through widened neural foramina into muscles may result in a “bunch of grapes” appearance [10, 18]. The other differential diagnosis of cystic lesion of sacrum includes developmental cysts (epidermoid and dermoid cyst, teratoma, neurenteric and retrorectal cystic) anterior sacral meningocele, necrotic sacral chordoma, schwannoma, arachnoid cyst, and anevrysmal bone cyst. In our case, the MRI and CT scan of the spine revealed no vertebral involvement.

Serologic enzyme-linked immunosorbent assay, Western blot, indirect hemagglutination assay, and polymerase chain reaction are 80–100% sensitive and 88–96% specific for liver cyst, and 50–56% for lung but less sensitive for the other organs (25–56%) [20]. Generally the treatment is successful when the immunologic test becomes negative [19]. In this present case, the serological tests were negative.

Surgery remains the optimal treatment for spinal HC. The objectives are, total removal of the cyst without rupture then establishing diagnosis, decompression of nervous structures and stabilization of the affected spine. Procedure and surgical approach is depending on the localization and extent of the disease. The stability of the vertebral column is frequently affected in hydatid cyst of the anterior column that requires systematic stabilization using different systems. In our case there was no bone involvement except neural foraminal widening we judged that the extended laminectomy on 5 levels required stabilization, transpedicular screw systems was used. To reduce risk of recurrence and sterilize the cysts systematic preoperative irrigation of cysts and soft tissues is classically recommended, the solutions used most often are scolicidal solutions of 95% ethanol and 20% hypertonic saline [21]. The hydatid cyst is a parasitic disease. Adjuvant anthelmintic chemotherapy is essential to control the disease and prevent recurrence [22]. Both albendazole and mebendazole (10 mg/kg/day) can be used continuously or periodically (with washout periods) from three months to one year [23]. Albendazole is preferred because of its specific pharmacologic features, such as better oral absorption and higher intracystic penetration [24]. However, no consensus exists regarding the length of the anthelmintic treatment period. Our patient was administered Albendazol treatment (400mg, twice a day) for six months postoperatively.

The published case review concludes, that the primary epidural hydatid cyst without a bone involvement has good long-term outcome, posterior approach is the gold standard. The recurrences are rare which depends on the complete resection of all parasitic lesions, intraoperative use of scolicidal agents and the preoperative and postoperative use of albendazole. However, systemic hydatid cyst, spine involvement and mid-thoracic localization have a poorer neurologic outcome according to decreased blood supply of the spinal cord in this level, And to the infiltrative nature of the disease [25].

CONCLUSION

Primary extradural hydatid cyst has only occasionally been reported in the literature, however it should be considered in the differential diagnosis of spinal cystic lesion. Radiological diagnosis and determination of hydatid cyst extension are usually provided via MRI. The final diagnosis is made; via surgical exploration which is the main treatment. Postoperative antihelminthic chemotherapy might reduce the recurrence rate. However, conducting preventive programs can reduce the incidence of this serious disease.

Conflits of Interest: The authors declare that there are no conflicts of interest regarding the publication of this article.

Disclosures: None

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