

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.

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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 hydatid 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 (blue arrow), with paraspinous extension through neural foramina widening (arrow)



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



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

DISCUSSION

Hydatid disease or hydatidosis is a common anthroponosis 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 extraneural 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, extraneural, 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 extraneural 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 |
| Baybek 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 <i>et al.</i> , (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 Recovery |
| 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%–50%), followed by the lumbar (range, 20%–39%), 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 aneurysmal 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 scolical 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 Albendazole 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 scolical 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.

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

Disclosures: None

REFERENCES

1. Karadereler, S., Orakdögen, M., Kiliç, K., & Özdoğan, C. (2002). Primary spinal extradural hydatid cyst in a child: case report and review of the literature. *European Spine Journal*, 11(5), 500-503.

2. Carrera, R., & Murphy, G. (1964). Primary Hydatid Cyst of the Spinal Cord. *Acta Neurol Lat AM*, 10: 308-312.
3. İşlekel, S., Zileli, M., & Erşahin, Y. (1998). Intradural spinal hydatid cysts. *European Spine Journal*, 7(2), 162-164.
4. San Martin Sanchez, L., Lopez Zafra, J. J., de la Riva Aguilar, A., Oliva Alonso, J. M., & Donnay Brisa, G. (1980). Hydatidose sous-durale rachidienne. A propos d'une observation. *Neurochirurgie*, 26, 235-238.
5. Neumayr, A., Tamarozzi, F., Goblirsch, S., Blum, J., & Brunetti, E. (2013). Spinal cystic echinococcosis—a systematic analysis and review of the literature: part 1. Epidemiology and anatomy. *PLoS neglected tropical diseases*, 7(9), e2450.
6. Schnepfer, G. D., & Johnson, W. D. (2004). Recurrent spinal hydatidosis in North America: case report and review of the literature. *Neurosurgical focus*, 17(6), 1-6.
7. Pamir, M. N., Ozduman, K., & Elmaci, I. (2002). Spinal hydatid disease. *Spinal cord*, 40(4), 153-160.
8. Xin, L., Wang, Z., & Fan, S. (2009). Magnetic resonance imaging and computerised tomography findings in an intraspinal extradural hydatid cyst mimicking tuberculous spondylitis: a case report. *Cases journal*, 2(1), 7109.
9. Benzagmout, M., Kamaoui, I., Chakour, K., & Chaoui, M. E. (2009). Primary spinal epidural hydatid cyst with intrathoracic extension. *Neurosciences*, 14(1), 81-83.
10. Layadi, F., Boubrik, M., Ait, A. E. Q., & Ait, S. B. (2005). Primary sacral epidural hydatid cyst: A case report. *Journal de radiologie*, 86(9 Pt 1), 1040-1042.
11. Sener, R., Calli, C., Kitis, O., & Yalman, O. (2001). Multiple, primary spinal–paraspinal hydatid cysts. *European radiology*, 11(11), 2314-2316.
12. Gopal, N. N., Chauhan, S. P. S., & Yogesh, N. (2007). Primary spinal extradural hydatid cyst causing spinal cord compression. *Indian journal of orthopaedics*, 41(1), 76-78.
13. Bhojraj, S. Y., & Shetty, N. R. (1999). Primary hydatid disease of the spine: an unusual cause of progressive paraplegia: Case report and review of literature. *Journal of Neurosurgery: Spine*, 91(2), 216-218.
14. Kafaji, A., Al-Zain, T., Lemcke, J., & Al-Zain, F. (2013). Spinal manifestation of hydatid disease: a case series of 36 patients. *World neurosurgery*, 80(5), 620-626.
15. Gennari, A., Almairac, F., Litrico, S., Albert, C., Marty, P., & Paquis, P. (2016). Spinal cord compression due to a primary vertebral hydatid disease: a rare case report in metropolitan France and a literature review. *Neurochirurgie*, 62(4), 226-228.
16. Gezerkan, Y., Ökten, A. I., Çavuş, G., Açık, V., & Bilgin, E. (2017). Spinal hydatid cyst disease. *World neurosurgery*, 108, 407-417.
17. Trivedi, A., Shukla, S., Singh, K., & Sharma, V. (2007). Giant intracranial hydatid cyst. *Journal of Pediatric Neurosciences*, 2(2), 72-74.
18. Bilgic, S., Kose, O., Sehirlioglu, A., Safaz, I., & Ozkan, H. (2009). Primary paraspinal hydatid cyst treated with puncture, aspiration, injection and re-aspiration (PAIR) technique: a case report. *European Spine Journal*, 18(2), 165-167.
19. Sucu, K., & Zileli, M. (2002). Omurga Ve Omurilik Enfeksiyonları. In: Zileli, M., & Özer, A. F. (Eds). *Omurilik Ve Omurga Cerrahisi*. Cilt 2, Izmir, 1150-1154.
20. Mandell, D. B. (1995). Principles and Practice of Infectious Diseases, 4th Edn, Vol 2. Churchill-Liv- Ingstone, New York, 2550.
21. Pamir, M. N., Ozduman, K., & Elmaci, I. (2002). Spinal hydatid disease. *Spinal cord*, 40(4), 153-160.
22. Saimot, A. G., Cremieux, A. C., Hay, J. M., Meulemans, A., Giovanangeli, M. D., Delaitre, B., & Coulaud, J. P. (1983). Albendazole as a potential treatment for human hydatidosis. *The Lancet*, 322(8351), 652-656.
23. Stamatakis, M., Sargedi, C., Stefanaki, C., Safioleas, C., Matthaipoulou, I., & Safioleas, M. (2009). Anthelmintic treatment: an adjuvant therapeutic strategy against *Echinococcus granulosus*. *Parasitology international*, 58(2), 115-120.
24. Teggi, A., Lastilla, M. G., & De Rosa, F. (1993). Therapy of human hydatid disease with mebendazole and albendazole. *Antimicrobial agents and chemotherapy*, 37(8), 1679-1684.
25. Özek, M. (1994). Complications of central nervous system hydatid disease. *Pediatric neurosurgery*, 20(1), 84-91.
26. Sharma, N. K., Chitkara, N., Bakshi, N., & Gupta, P. (2003). Primary spinal extradural hydatid cyst. *Neurology India*, 51(1), 89-90.
27. Awasthy, N., & Chand, K. (2005). Primary hydatid disease of the spine: an unusual case. *British journal of neurosurgery*, 19(5), 425-427.
28. Karakasli, A., Yilmaz, M., Mucuoglu, A. O., & Yurt, A. (2015). A large primary dumbbell hydatid cyst causing neural foraminal widening of the thoracic spine: A case report and literature review. *International journal of surgery case reports*, 8, 55-58.
29. Jaiswal, S., Jaiswal, A. K., Jain, M., Behari, S., & Pandey, R. (2009). Primary spinal extradural hydatid cyst causing paraplegia. *Indian Journal of Pathology and Microbiology*, 52(3), 432-433.
30. Eloqayli, H., Matalka, I., & Daoud, S. (2010). Primary spinal extradural hydatid cyst in a 4-year-

- old child. *British journal of neurosurgery*, 24(5), 602-603.
31. Boulahroud, O., Dao, I., El Asri, C. A., & Boucetta, M. (2012). Primary extradural hydatid cyst extended to paraspinal muscles. *Journal of neurosciences in rural practice*, 3(3), 358-360.
32. Dogan, I., Kahilogullari, G., Guner, E., & Unlu, A. (2015). A rare and unexpected clinical progress and location on a primary extradural spinal hydatid cyst in a pediatric patient: a case report. *Child's Nervous System*, 31(8), 1407-1411.
33. Gennari, A., Almairac, F., Litrico, S., Albert, C., Marty, P., & Paquis, P. (2016). Spinal cord compression due to a primary vertebral hydatid disease: a rare case report in metropolitan France and a literature review. *Neurochirurgie*, 62(4), 226-228.
34. Mnari, W., Maatouk, M., Kilani, M., & Golli, M. (2016). A rare etiology of failed epidural anesthesia and paraparesis: Primary intracanal hydatid cysts. *Joint, bone, spine: revue du rhumatisme*, 83(2), 239-240.
35. Sridharan, S., Narayana, G. J., Chidambaram, K., & Jayachandiran, A. P. (2017). Primary paraspinal hydatidosis causing acute paraplegia. *Journal of neurosciences in rural practice*, 8(3), 472-474.
36. Schnepfer, G. D., & Johnson, W. D. (2004). Recurrent spinal hydatidosis in North America: case report and review of the literature. *Neurosurgical focus*, 17(6), 1-6.
37. Pedrosa, I., Saiz, A., Arrazola, J., Ferreirós, J., & Pedrosa, C. S. (2000). Hydatid Disease: Radiologic and Pathologic Features and Complications 1: (CME available in print version and on RSNA Link). *Radiographics*, 20(3), 795-817.
38. Pluchino, F., & Lodrini, S. (1981). Multiple primitive epidural spinal hydatid cysts. *Acta neurochirurgica*, 59(3-4), 257-262.
39. Wani, M. A., Taheri, S. A., Babu, M. L., Ahangar, G. A., & Wani, H. (1989). Primary spinal extradural hydatid cyst. *Neurosurgery*, 24(4), 631-632.
40. Kars, H. Z., Hekimoglu, B., & Cepoglu, C. (1990). Spinal epidural hydatid cyst: radiological and ultrasonographical workup of a case. *European journal of radiology*, 11(3), 212-214.
41. Bavbek, M., Inci, S., Tahta, K., & Bertan, V. (1992). Primary multiple spinal extradural hydatid cysts of the literature: case report and review. *Spinal Cord*, 30(7), 517-519.
42. Tekkök, I. H., & Benli, K. (1993). Primary spinal extradural hydatid disease: report of a case with magnetic resonance characteristics and pathological correlation. *Neurosurgery*, 33(2), 320-323.
43. Baysefer, A., Gönül, E., Canakci, Z., Erdoğan, E., Aydoğan, N., & Kayali, H. (1996). Hydatid disease of the spine. *Spinal Cord*, 34(5), 297-300.
44. Pandey, M., & Chaudhari, M. P. (1997). Primary hydatid cyst of sacral spinal canal: case report. *Neurosurgery*, 40(2), 407-409.
45. Bayar, M. A., Erdem, Y., & Habip, N. (1997). Primary Intraspinal Extradural Hydatid Disease Causing Radicular Compression. *Turkish Neurosurgery*, 7(1-2): 33-35.
46. Berk, C., Ciftci, E., & Erdoğan, A. (1998). MRI in primary intraspinal extradural hydatid disease: case report. *Neuroradiology*, 40(6), 390-392.
47. Bouklata, S., El-Mahi, M., Karmouni, W., El-Hassani, M. R., Chakir, N., Jiddane, M., & Boukhrissi, N. (2000). Isolated dorsal extradural hydatid cyst. A case report. *Journal of Neuroradiology*, 27(4), 285-286.