

Hydatid Cyst of Thoracic Spine: A Rare Cause of Paraplegia

Rahul Soangra¹, Mool Chand Soangra²

¹Virginia Tech-Wake Forest University, School of Biomedical Engineering and Sciences, Blacksburg 24060, United States

²Department of Orthopedics and Spinal Surgery, Dr (Col) Ali Omar Askar Hospital and Neurocenter, Sbia, Bengasheer, Tripoli, Libya

Abstract: Primary spinal hydatid cysts are uncommon and extradural involvement is rare. We report an unusual case of Paraplegia due to hydatid disease of primary site of infection extra spinal muscles in thoracic region from T5 to T8 level extending into a daughter cyst at T9 level, is main cause of acute Paraplegia. Magnetic resonance imaging (MRI) showed an extradural round cystic lesion in the spinal canal filling the posterior and left postero-lateral aspect at T9 level, markedly compressing the cord. The cyst was removed after laminectomy and opening of the dural sac, with excision of extraspinal mass in left paravertebral region from T5 to T8 level. Histopathological examination confirmed a diagnosis of hydatid cyst. Early decompression surgery of the spine with chemotherapy is the treatment of choice for the disease.

Keywords: Hydatid cyst, paraplegia, extradural, intraosseous, extraosseous

1. Introduction

Hydatid cyst is a parasitic disease caused by echinococcus granulosus occurring in humans as a result of faeco-oral contamination. Hydatid cysts are primarily located in the liver or lung and are uncommon in the spine [1]. Spinal manifestation of the disease was defined first by Churrier in 1807. Osseous involvement is seen in 0.5 to 2% and approximately half of these are located in the vertebrae out of which in 50% involvement is seen in thoracic area [2].

2. Case Report

A 40 year old male was admitted in male ortho- spinal unit with history of paraplegia with acute onset since three weeks. There was no history of fever or trauma. His symptoms started with gradual loss of motor functions in both lower limbs which led to complete paraplegia with the sensory deficit below the umbilicus.

MRI showed an extradural round cystic lesion with low signal intensity on T1 and high signal intensity on T2 weighted images at thoracic 9 level markedly compressing the cord which was flattened and displaced. Marginal enhancement after gadolinium contrast was noted. Another extra spinal large soft tissue mass lesion located in Left Paravertebral Thoracic Region was seen infiltrating the left posterior spinal muscles at level of D5 to D8 in contact with posterior neural elements of these vertebra without any evidence of bone erosion. This mass was hypointense on T1 weighted images, hyper intense on FFE. There was no pathology in cranial, cervical and lumbar spines. Tomography of the abdomen and lungs and abdomen ultrasonography showed no abnormality, blood parameter investigations are within normal limits except high erythrocyte sedimentation rate.

Laminectomy decompression was done with a favorable outcome. During surgery excision of capsulated cystic mass

in left paravertebral thoracic region was carried out tracing down to T9 level. The mass consisted of creamy white gelatinous material which was slippery and difficult to catch by tissue forceps. It was excised and laminectomy at T9 level done. While exposing with gentle manipulation by dissector the cyst ruptured like a balloon and the area was irrigated with hydrogen peroxide and saline.



Figure 1: T1WI image shows extradural extramedullary CSF signal intensity cystic lesion hydatid cyst.

Post operatively on next day the patient showed significant improvement. He started feeling sensation and movement of lower limbs started gradually increasing. He was discharged a month after initial hospitalization and has been followed up for 9 years after surgery and has only minimal weakness in right lower limb. The latest MRI of dorso-lumbar was fine and shows no recurrence of hydatid cyst.

3. Discussion

Many studies have reported paraplegia as the presenting sign of spinal hydatidosis [3,4,5,6]. Our patient also presented

with symptoms and signs related to spinal cord compression such as low back pain, radicular leg pain and paraparesis for few weeks which culminated in acute paraplegia due to rapid compression. Paraplegia results either from the cysts invading the spinal canal and causing compression or ischemic changes of the spinal cord or cauda equine [4,6]. Spinal hydatid cysts may be classified as intraosseous or extraosseous. The intraosseous cysts may be further divided into solitary or osteohydatidosis form. The solitary form represents an area of multiloculated destruction without periosteal reaction or soft tissue swelling. This lesion is not self-limiting but gradually grows in all directions.



Figure 2: T1WI postoperative image shows complete removal of hydatid cyst.

In many cases the disease involves the spine primarily and the disease extends into the neighbouring structures [7]. This form is frequently associated with paraspinal soft tissue swelling as was seen in our case. The extraosseous form is secondary, which involves the ribs by contiguity; the mechanism of involvement is by pressure erosion.

The extraosseous form is most commonly seen due to ruptured primary cyst either spontaneous or during surgery. Periosteal reaction if present is suggestive of secondary infection. Computerized tomography (CT) scan provides the precise anatomical location and details of the lesion along with bone destruction and definition of paraspinal and intraspinal extension which generally do not enhance following contrast administration [8, 9].

Delineation of intraspinal extension is also possible with computer assisted myelography. MRI is helpful in verifying the extent of the disease, texture of cyst, degree of medullary involvement and viability of cyst [10, 11, 12]. On T1 weighted images, there is a mixed morphological appearance. High signal intensity content of the cyst may correlate with high cell or protein content which is suggestive of extension of parasite host reaction. Daughter cysts are more hypointense than the parent cyst on T1 weighted images. The cyst wall or capsule is seen as a low intensity rim, which shows mild enhancement following intravenous gadolinium. On T2 weighted imaging the daughter cysts are of slightly higher signal intensity than parent cyst.



Figure 3: Histopathology slide hydatid cyst.

Signal intensities may change with co-existing infection, calcification or haemorrhage. Extradural spread of the hydatid cyst through a widened neural foramen into the muscle planes, may result in a “bunch of grapes” appearance. The T2 weighted sequence also indicates whether a cyst is viable or not. A decrease in hyperintensity and an increase in hypointensity from collapse cyst wall is suggestive of succumbed cyst. Both CT and MRI may show endovesicular daughter cysts, which are frequently observed in hepatic diseases but are rare in musculoskeletal manifestation of this disease [8, 10].

The differential diagnosis of such a radiographic picture includes giant cell tumour, osteolytic metastases, plasmacytomas, aneurismal bone cyst and cystic neurofibromas [11]. Aspiration cytology is the procedure of choice in suspected cases of skeletal hydatid cyst, but biopsy is contraindicated. The standard of choice of therapy of the disease is radical resection of lesion in combination with antihelminthic drugs like mebendazol or albendazol [5, 7]. In isolated cases of infestation of a musculoskeletal region as seen in the present case, extensive curative surgical resection along with albendazol for a period of 3 months should be considered.

It is possible to evaluate successful surgery with removal of cyst post operatively using serological testing for the parasite. Furthermore, serological test can be used as a screening method, offering a diagnostic tool and can also indicate re-occurrence of the disease [6].

4 Conclusion

Rapidly developing neurological complications in the form of paraplegia due to spinal cord compression is presented in this case spinal hydatid disease which was treated by surgery with a favorable outcome. The aim in the treatment of hydatid cyst disease should be in accordance with curative surgery and radical removal of parasites. The WHO suggests an adjuvant chemotherapy with albendazol for at least 2 years after surgery.

We conclude that primary vertebral echinococcosis, although rare should be considered in destructive lesions of the vertebrae and spine in regions that the disease is endemic. Advanced imaging studies should be performed to diagnose

the disease. Early decompressive surgery of the spine, with chemotherapy, is the treatment of choice for these patients.



Dr. Mool Chand Soangra received his M.B.B.S and M.S. in Orthopedics from S.P. Medical College Bikaner. He has 35 years' experience in Orthopedic and spine surgeries.

References

- [1] Baysefer, A., Gonul, E., Canakci, Z., Erdogan, E., Aydogan, N., & Kayali, H. "Hydatid disease of the spine." *Spinal Cord*, 34(5), 297-300, 1996.
- [2] Bahloul, K., Ghorbel, M., Boudouara, M. Z., & Mansour, H. B. "Primary vertebral echinococcosis: four case reports and review of literature." *British Journal of Neurosurgery*, 20(5), 320-323, 2006.
- [3] Adilay, U., Tugcu, B., Gunaldi, O., Gunal, M., & Eseoglu, M. "Cauda equina syndrome caused by primary lumbosacral and pelvic hydatid cyst: A case report." *Minimally Invasive Neurosurgery*, 50(5), 292-295, 2007.
- [4] Fares, Y., Khazim, R., El Zaatari, M. M., Haddad, G. F., & Barnes, P. R. "Spinal hydatid disease and its neurological complications." *Scandinavian Journal of Infectious Diseases*, 35(6-7), 394-396, 2003.
- [5] Prabhakar, M. M., Acharya, A. J., Modi, D. R., & Jadav, B. "Spinal hydatid disease: A case series." *Journal of Spinal Cord Medicine*, 28(5), 426-431, 2005.
- [6] Taghipour, M., Zamanizadeh, B., & Zare, Z. "Primary extradural hydatid cyst of spine causing acute paraplegia." *Neurosurgery Quarterly*, 16(1), 50-51, 2006.
- [7] Kalva, S. P., Cherian, M., & Ramasamy, K. C. "Spinal hydatid cyst with dissection into posterior paravertebral muscles - A case report." *Rivista Di Neuroradiologia*, 14(1), 103-106, 2001.
- [8] Tsitouridis, I., & Dimitriadis, A. S. "CT and MRI in vertebral hydatid disease." *European Radiology*, 7(8), 1207-1210, 1997.
- [9] Tuzun, M., & Hekimoglu, B. "CT findings in skeletal cystic echinococcosis." *Acta Radiologica*, 43(5), 533-538, 2002.
- [10] Singh, S., Korah, I. P., Gibikote, S. V., Shyam, N. K., Nair, A., & Korula, A. "Sacral hydatidosis: value of MRI in the diagnosis." *Skeletal Radiology*, 27(9), 518-521, 1998.
- [11] Turgut, A. T., Altin, L., Topcu, S., Kilicoglu, B., Altinok, T., Kaptanoglu, E., et al. Unusual imaging characteristics of complicated hydatid disease. *European Journal of Radiology*, 63(1), 84-93, 2007.
- [12] Fahl, M., Haddad, F. S., Huballah, M., Kanaan, S., Husheimi, I., & Azizi, T. Magnetic-Resonance-Imaging in Intradural and Extradural Spinal Echinococcosis. *Clinical Imaging*, 18(3), 179-183, 1994.

Author Profile



Dr. Rahul Soangra received the B.E. and M.Tech. degrees in Chemical Engineering from Jai Narain Vyas University and Indian Institute of Technology Roorkee. He further received his M.S. in Industrial & Systems engineering and Ph.D in Biomedical engineering from Virginia Tech, USA.