



Spinal Hydatid Disease: A Series of 4 Cases

KEYWORDS

Dr Naresh Krishnani

HOD Neurosurgery Sumandeep Vidyapeeth

Dr Deepak Bandlish

(Assistant Prof) Neurosurgery , Sumandeep Vidyapeeth

Dr Bhagwati Salgotra

(Associate prof) Neurosurgery Sumandeep Vidyapeeth

***Dr Kunal Kishore**

(Resident) Neurosurgery , Sumandeep Vidyapeeth
*Corresponding Author

Dr Mehul Solanki

(Resident) Neurosurgery , Sumandeep Vidyapeeth

Introduction:

Hydatid disease in humans is caused by the cystic (larval) stage of the tapeworm *Echinococcus granulosus*, which is endemic to the temperate climate. Canines are the primary host. The life cycle of *E granulosus* may also involve sheep, cattle, goats, and humans. This infection is transmitted orally via eggs shed in the feces of infected animals. Hydatid disease is not uncommon in Gujarat state, where cattle rearing is a common occupation. Primary hydatidosis is common in the liver, spleen, and lungs (1-3). Theoretically, it can occur at any site except teeth, hair, and nails (4). Musculo-skeletal involvement is secondary and uncommon, with an incidence of less than 2.5%. It affects the pelvis and sacrum, metaphyses of the long bones, skull, spine, and ribs in decreasing order of incidence. Spinal involvement is rare, with an incidence of less than 1% (5-8).

The aim of this presentation is to share our experiences of the pitfalls and challenges in the diagnosis and management of spinal hydatid disease and to provide a perspective through review of the literature.

Clinical Presentation

CASE	AGE/SEX	COMPLAINTS	EXAMINATION
1	21/M	LOWER LIMB PARESIS	GRADE II POWER NO SWELLING
2	34/M	LOWER LIMB WEAKNESS	GRADE III POWER NO SWELLING
3	26/M	LOWER LIMB PARESIS	GRADE II POWER NO SWELLING
4	28/M	LOWER LIMB PARESIS	GRADE I POWER NO SWELLING

Diagnostic Workup

Patients were investigated with routine hematologic investigations including blood counts, plain radiographs of the spine in anteroposterior and lateral views, and underwent ultrasonographic examination of the abdomen to rule out visceral hydatid disease. Magnetic resonance imaging (MRI) was done in all patients which showed hyperintense lesion probably extradural and extramedullary.

Management

Surgical Intervention

All patients underwent surgery to excise the cysts and had

a posterior laminectomy performed through the posterior approach for neurologic decompression at the level of spinal involvement. Due care was taken to prevent rupture while removing the cysts, which contain hundreds, even thousands, of protoscolices, each of which can form a new hydatid cyst. The surrounding surgical field was packed with mops to prevent local spillage. However, scolical solutions, such as hypertonic saline and cetrimide, were not used during surgery for fear of chemical damage to the cord.

Follow-up

In all patients, histopathologic confirmation of the diagnosis was obtained and antihelminthic therapy with 400 mg of albendazole 3 times daily was prescribed for 1 year. The patients were followed every 6 weeks initially, in the form of neurologic reassessment and imaging investigations.

DISCUSSION

Hydatid disease usually affects the soft tissues first and bones are involved later. Out of our 4 patients, none had detectable primary lesions on ultrasonography of the abdomen. In bone, hydatid growth continues in an outward direction in surrounding cancellous bone, with destruction by pressure necrosis and resorption leading to exogenous cyst formation. Thus, pericyst does not form in the bone hydatid (4). There is no pus formation or sequestration of bone.

The neurologic complications are the result of invasive intradural and extradural growth of the cysts causing direct compression. Destruction of the bones causes mechanical instability and secondary neurologic damage. This mixed picture poses a confusing picture for diagnosis and treatment planning (3,5,6). These 4 patients were referred to our center because of incorrect diagnosis and inappropriate treatment. Because of lack of clinical suspicion of hydatid disease, imaging investigations were inadequate and biopsy reports inconclusive.

On plain radiographs, multiple cystic lesions in multiple contiguous vertebral bodies and appendages are seen, usually without vertebral collapse and with sparing of intervertebral disks. Involvement of contiguous ribs and paravertebral masses are suggestive of hydatid disease (2). There is absence of the classical paradiscal sclerosis and sequestration that is suggestive of tuberculous infection.

CT scan with IV contrast does not show any enhancement. Myelogram demonstrates multi-level cord compression providing precise anatomic localization of the lesions (8). Postmyelogram CT scan is useful when MRI is not available.

MRI shows soft tissue masses in paraspinal muscles, which are spherical thin-walled, fluid-filled parent cysts. The formation of internal daughter cysts forms a grape-bunch-like appearance. Similarly, continuous intraspinal extension appears as vertebral and rib lesions, representing intradural and extradural multiple cysts compressing the cord.

MRI is superior to CT for demonstrating neural involvement (9,10,11). All of these studies helped establish a preoperative diagnosis in our cases. The diagnosis can be definitely confirmed histopathologically after decompressive surgeries by observing the allergic-type tissue reaction in the surrounding tissue and demonstration of the cyst wall histology and scolices.

Efficacy of albendazole for primary bony hydatid involvement is questionable; postoperative albendazole therapy seems only to retard recurrence (3,12,13). All of our patients received 400 mg of albendazole 3 times daily for 1 year, but recurrence rate was still 100%.

The aim of surgery is removal of all of the cysts early in the course of the disease (3). Usually, posterior spinal decompression through laminectomy and debridement of paravertebral lesions is the initial surgery (1,3), but complete clearance is difficult because of invasive diffuse spread within the bone and canal (13). Often, spillage of fluid caused by cyst rupture leads to subsequent recurrence (2,13). According to the guidelines for excisional surgery, the surgical area needs to be irrigated with hypertonic saline (3). We avoided this irrigation to prevent chemical damage to the spinal cord; we took enough care not to rupture the cysts; and we attempted the intact removal of all of the visible cysts by wide laminectomy. Ideally, these lesions should be treated by radical operation with circumferential approach and extensive removal of all cysts and affected bone and soft tissues (3,13).

CONCLUSION:

Hydatid disease is not uncommon in rural areas of Gujarat state. It is acquired in childhood, and usually presents in adulthood. Musculoskeletal involvement is a less common presentation, of which, spinal involvement is infrequent. The infection may be misdiagnosed initially as tuberculosis of the spine, which delays proper diagnosis and intervention. In addition to the many difficulties in the diagnosis, the management of the disease is even more challenging because of a high recurrence rate, requiring extensive and repeated spinal surgeries with high rates of complications and significant long-term morbidity and mortality. Results are seldom satisfactory and prognosis is usually poor. Control of conditions favorable to tapeworm transmission is key to preventing hydatidosis in humans.

REFERENCES:

1. Fares Y, Khazim R, El Zaatari MM, Haddad GF, Bannes PR. Spinal hydatid disease and its neurological complications. *Scand J Infect Dis.* 2003;35(6-7):394-396.
2. Islekel S, Zileli M, Ersahin Y. Intradural spinal hydatid cyst. *Eur Spine J.* 1998;7(2):162-164.
3. Schneppenheim M, Jerosch J. Echinococcosis granulosa/cysticulus of the tibia. *Arch Orthop Trauma Surg.* 2003;123(2-3):107-111. Epub. April 2003.

4. Islekel S, Ersahin T, Zileli M, et al. Spinal hydatid disease. *Spinal Cord.* 1998;36(3):166-170.
5. Beggs I. The radiology of hydatid disease. *Am J Roentgenol.* 1985;145:639-648.
6. Sapkar GS, Stathakopoulos DP, Babis GC, Tsarouchas JK. Hydatid disease of bone and joints 8 cases followed for 4-16 years. *Acta Orthop Scand.* 1998;69(1):89-94.
7. Turgut M. Hydatid disease of spine: a survey study from Turkey. *Infection.* 1997;25:221-226.
8. Claudon M, Bracard S, Plenat F, Regent D, Bernadac P, Picard L. Spine involvement in alveolar echinococcosis assessment of 2 cases. *Radiology.* 1987;162:571-572.
9. Braithwaite PA, Lees RF. Vertebral hydatid disease: radiological assessment. *Radiology.* 1981;140:763-766.
10. Taourel P, Marty-Ane B, Charasset S, Mattei M, Devred PH, Bruel JM. Hydatid cyst of the liver Comparison of CT and MRI. *J Comput Assist Tomogr.* 1993;17:80-85.
11. Severino A, Marani D, Canossa GC, et al. Hydatid disease: MR imaging study. *Radiology.* 1990;175:701-706.
12. Pedrosa I, Saiz A, Arrazola J, Ferraires J, Pedroza CS. Hydatid disease: radiological and pathological features and complications. *Radiographics.* 2000;20(3):795-817.
13. Mazyad MA, Morsy TA, Habib KS. Vertebral unilocular hydatidosis in a shepherd and his wife. *J Egypt Soc Parasitol.* 1999;29(2):547-550.