Case Report

An Unusual Case of Hydatid Cyst

S.A. Mirhoseini MD*, J. Ahmadi MD**

ABSTRACT

Hydatidosis is a parasitic disease in all over the world which is caused by a Cestode (tape worm). Liver, lung, and brain are the most Common involved organs and involvement of muscles and bones is unusual. We report a 32years old man who had low back and radicular leg pain. He had a paravertebral mass with involvement of Spinal column. This patient was operated with differential diagnosis of tumor or hydatid cyst. The final diagnosis was hydatidosis of paravertebral muscles and vertebral column.

Keywords: hydatidosis, p aravertebral muscle hydatid cyst, spinal column hydatid cyst

The patient was a 32 years old man with low back pain and radicular pain in both legs for one month. His past history for any other illness was negative. He didn't have any history of coughing and respiratory symptoms. He had good general condition. In examination we found low back and paravertebral tenderness in lumbar area. Radicular pain was more severe in left leg. Straight leg raising test (SLR) was negative in both legs. He didn't have any motor and Sensory deficit, sphincter dysfunction, mascular atrophy or gate disturbance. Deep tendon reflex (DTR) in right leg was normal and a littel exaggerated in left leg. Other examinations was normal.

In lumbar spine MRI there was large cystidbb lesions in left paravertebral muscles and in spinal canal, which was hypointense in T1 weighted and hyperintense in T2 weighted images. The lesion extended into spinal canal but had no obvious comperesion on techal sac (Figures 1 and 2). Chest x Ray was normal.

We operated the patient. Paravertebral muscles were explored meticulously and cysts were dissected from muscles. One of the cysts was ruptured during operation and a clear and colorless liquid came out. All of cysts were removed. Spinous process and lamina of L3 vertebra were involved which were removed and cyst extension in the spinal canal was removed too. Cysts had not involved dura and were

completely extradural with mild comperesion on techal sac. After that all the operation field was irrigated with hypertonic salin. The pathologic diagnosis was hydatid cyst.

Discussion

Hydatic cyst is caused by two kind of tape worms (cestodes); Echinococcus granulosus and echinococcus multilocularis. This disease is a worldwide problem ¹. Iran is one of the endemic environments for this disease 2. Liver, Lungs, and brain are the most common involved organs and involvement of bone, and muscles is uncommon ³. Bones and mucsles can be involved both primary or secondary to more common involved parts. The prevalence of muscle involvement is 3% and for bore is 1.1 to 4% from nonendemic to endemic areas ¹. Spinal column is involved in 50% of bone involvement cases. Men are involved more than women. The peak age of disease is 21-40 years old ¹. Patients with spinal column hydatid cyst have: Lumbar pain, paresthesia, paraparesia and even paraplegia, or sphincter dysfunction. Neurological symptoms of this disease are due to spinal involvement and compression effects on cord, or cord direct involvement ¹. Our patient didn't had neurological deficit. He had low back pain with feeling of pain in both tighs which could be due to mild compresion on techal sac but there was no compresion on nerve roots, and so SLR test was negative in both legs.

Correspondence to: Dr. Jalal Ahmadi, Department of Neurosurgery, Isfahan University of Medical Science, Isfahan, Iran.

E-mail: J-ahmadi@resident.mui.ac.ir

^{*}Assistant professor, Department of Neurosurgery, Isfahan University of Medical Science, Isfahan, Iran.

^{**}Resident, Department of Neurosrgery, Isfahan University of Medical Science, Isfanhan, Iran.

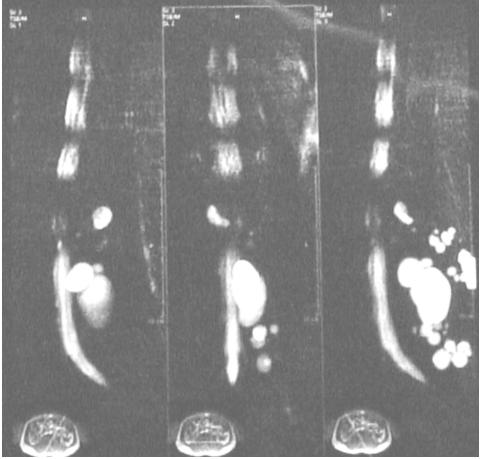


Figure 1. MRI myelogram of patient demonstrating multiple cysts in paraspinal space.

All of the body muscles can be involved by hydatid cyst. Involvements of limb muscles ^{4, 5, 6}, gluteal mucles ⁷, Sternocleidomastoid muscle ⁸, temporalis muscles ⁹, paravertebral muscles ¹⁰ and even extraocular muscles ¹¹ have been reported. Muscle involvement produces nonspecific symptoms with local edema, thus in all cases with muscular mass, hydatid cyst should be one of differential diagnosis ¹².

Several reports of pravertebral muscles involvement are peresent which non specific symptoms like local edema, pain, tenderness, and back pain are reported in all of them ^{10,13,14}. In our patient's paravertebral muscles, posterior spinal elements and extradural space were involved and it is difficult to say that which of them was the primary site of infection. Spine might be the first place of infection and muscular and canal involvement were secondary to it. In this kind of involvement, recurrence rate is high and despite of surgical resection, 30 to 60 percent of

recurrence has been reported with poor prognosis ¹. The most common infected parts in spinal column are thoracic and lumbar areas. Generally, the involvement begins in vertebral body and extends to other parts of the spine. Disk space is usually spared ¹⁶. In our patient, involvement was in L3 vertebra and lamina and spinous process were infected but L3 body was intact.

The MRI scan characteristically shows an image resembeling a bunch of grapes. Cyst wall is hypointense in both T1 and T2 weighted images. Cyst walls are thin, regular, not septated, and do not show enhancement ¹⁶. MRI images were not completely typical in our patient. He had multiple cysts with different sizes in left paravertebral muscle.

Cysts intensity was homogenous (hypointens in T1 and hyperintens in T2 images) and we coulden't see dougheter cysts in them. They had a hypointens wall. Vertebral involvement wasn't obvious in MRI

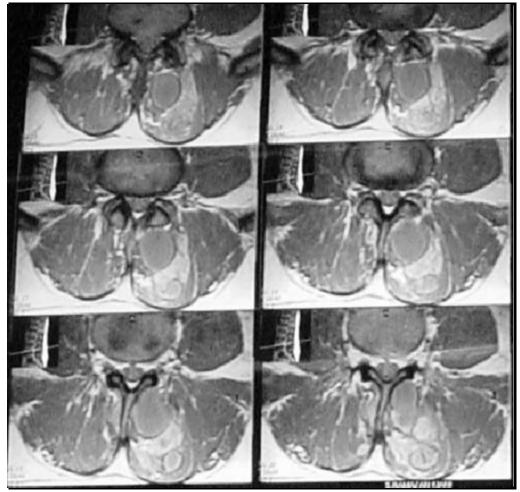


Figure 2. T1 lateral axial MRI of patient demonstrating multiple cysts in left para spinal space.

images, but extension of lesion into spinal canal was seen.MRI study with contrast media wasnot done(Figures 1,2).

Chest X ray and sonography of abdomen and pelvic cavity was normal, so that involvement of muscles and spinal column in our patient was primary which is rare and unusual.

Surgical resection is the treatment of choice in hydatid cyst. Theraputic aim in muscular involvement is pericystectomy and resection of cysts without rupture ¹⁷, and, in vertebral involvement is resection of all infected parts ¹⁶.Decompression of spinal column without resection of anterior infected parts of vertebra have been reported with 50 percent recurrence ¹⁷. In our patient, we resected all the bony involved parts. Because of Severe adhesion and large extent of infected museles, some of intramuscular cysts were

ruptured during the resection (Figure.3). Cyst rupture has two potentially dangerous results: ¹ anaphylaxis and ² local recurrence ¹⁵. We irrigated operation field with 30% hypertonic salin in order to prevention of recurrence.

Antihelminthic drugs should be used in all patients whom we coulden't do a radical surgery for them and received just a palliative surgery ¹⁶. Albendazol is the drug of choice ³, but because of low bone penenterance of albendazol, spinal cyst is resistant to it. Combination of albendazol and praziquantel has been successfully used in treatment of spinal hydatid cyst and is recommended in extensive involvement of spinal column in which the surgery has high risk of morbidity and mortality ¹. We refered our patient to an infectious disease specialist for chemotherapy and albendazol was prescribed for him.

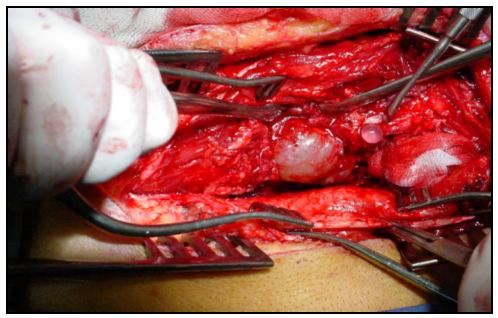


Figure 3. Image of cyst and a daughter cyst in para spinal muscles.

On the whole, we report a rare and unusual case of hydatidosis.this patient had primary muscle involvement with multiple cysts which is unusual. hydatidosis usually involves muscles secondary to involvement of other organs ¹². As muscles were more involved than bone, we think that involvement of

L3 vertebra was secondary to muscles, but if vertebral involvemnet was primary, that we are not sure, it is another unusual and interesting aspect of our patient; because primary vertebral involvement usually begins from vertebral body not posterior parts.

References

- 1. Lam KS, Faraj A, Mulholland R, Finch RG. Medical decompression of Vertebral Hydatidosis. Spine 1997 Sep;22(17):2050-5.
- 2. Saebi E. Parasitic disease in Iran. Vol 2. Iran: Islamic Republic publication & education; 1370.p.154-8.
- 3. Mandell GL, Bennett JE, Dolin R. Priciples and practice of Infections Diseases. Fifth edition. New York: Churchill Livingstone; 2000.
- 4. Ben M, Rad S, Mathlouthi A, Merai S, Ghrairi H, Mestiri I et all. Multiple Hydatid cysts of the thigh: the role of Magnetic resonance imaging. J Radiol 1998 Sep;79(9):877-9.
- 5. Meddeb N, Bachrouch N, Ellevch M, Sahli H, Cheour E, Labib S. Hydatid cyst of the adductor muscles. MR aspect, Apropos of 1 case. Bull Soc Pathol Exot 2001 May;94(2):106-8.
- 6. Bayram M, Sirikci A. Hydatic cyst located intermuscular area of the forearm: MR imaging findings. Eur J Radial 2000 Dec;36(3):130-2.
- 7. Chia ttoni MK, Jorge V, Jannke HA, Telo GH, Segala NC. Intramuscular hydatid cyst: a case report. Rev Soc Bras Med Trop 2003;36(4):527-9.
- 8. Gluncic I, Roje Z, Bradaric N, Petricevic A, Pisac VP, Gluncic V. Primary echinococcosis of the sternocleidomastoid muscle. Croat Med J 2001 Apr;42(2):196-8.
- 9. Sennaroglu L, Onercij M, Turan E, Sungur A. Infratemporal hydatid cyst -unusual location of echinococcosis. J Laryngol Otol 1994 Jul;108(7):601-3.
- 10. Tatari H, Baran O, Sanlidag T, Gore O, Ak D, Manisali M et all. Primary intramuscular hydatidosis of supraspinatus muscle. Arch Orthop Trauma Surg 2001;121(1-2):93-4.
- 11. Kiratli H, Bilgic S, Ozturkmen C, Aydin O. Intramuscular hydatid cyst of the medial rectus muscle. Am J Ophthalmol 2003 Jan;135(1):98-9.
- 12. Luhr T, Junginger T. Muscular echinococcal cysts. A rare differential diagnosis of a soft tissue swelling. Chirurg 1995 Dec;66(12):1275-6.

- 13. Calderoni PP, Galleti S. Echinococcosis of paravertebral space. Report of a case. Radiol Med (Torino) 1995 Dec;90(6):820-1.
- 14. Latino R, Costa S, Barbagallo E, Virzi A, Vagnoni G. Primary localization of a hydatid cyst in the major dorsal muscle: report of a case. Ann Ital Chir 1999 Jan-Feb;70(1):123-6.
- 15. Alexiadis G, Lambaropoulov M, Deftereos S, Papadopoulos N, Manavis J. Primary Muscular Hydatidosis. Acta Radiol 2002 Jul;43(4):428-30.
- 16. Govender TS, Aslam M, Parbhoo A, Corr P. Hydatid disease of the spine: A long-term Follow up After surgical treatment. Clin Orthop Relat Res 2000 Sep;(378):143-7.
- 17. Manes E, Santucci A. Echinococcosis: intramuscular localiyation. Cir Organi Mov 1990 Apr-Jan;75(2):189-96.