Primary spinal hydatid presenting as Cauda Equina Syndrome: A rare case report

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Abstract

Primary hydatid of spinal cord is a very rare presentation of relatively common disease. We present the case of 23-year-old female with cauda equina syndrome diagnosed to have a primary spinal hydatid which mimicked Pott's spine. Initial diagnosis was made with the help of history taking, clinical examination and investigations like MRI and later confirmed with the help of biopsy. High suspicion is required for diagnosis, as it is a rare disease. Complete resection along with Albendazole therapy is the current choice of treatment. Further studies are required for the same as there is a lack of guidelines for management.

Keywords: Hydatid cyst, Spinal cord, Echinococcus, Albendazole.

Introduction

Hydatid disease (echinococcosis) is a zoonosis caused by Echinococcus tapeworm (Dog tape worm). The parasite affects the humans, and other animals like sheep, horse, mice[1]. Two main species which affects the men are Echinococcus granulosus and Echinococcus multilocularis[2]. It commonly involves liver (in 75% cases) then lungs (15% cases). Bone involvement is very rare (0.5-2%) and the most common bone involved is vertebra[3].

Spinal hydatid disease can be classified into- (1) Primary intramedullary hydatid cyst, (2) Intradural extramedullary hydatid cyst, (3) Extradural intraspinal hydatid cyst, (4) Hydatid disease of the vertebrae and (5) Para vertebral hydatid disease, according to the sites it involves- Braithwaite and Lees classification[4,5]. In this case report we present the primary hydatid disease of the spinal cord crossing across the inter vertebral foramina (extramedullary type) and presenting as Cauda Equina Syndrome.

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Case Report

A 23-year-old female, manual laborer by occupation hailing from Kota, Hadoti region of Rajasthan, presented with chief complaints of Back pain & weakness of both the lower limbs for 1 year, loss of sensation over foot for 3 months & dribbling of urine for 1 month.

Back pain was insidious in onset initially boring type; later progressed to sharp shooting type, radiating to the right lower limb, often aggravated on sleeping supine; which makes her sit up with support at night times. Pain was associated with low grade continuous type of fever.

Weakness was insidious in onset, asymmetrical, started in right lower limb first, progressing from distal to proximal parts. Later involved the left lower limb in the same fashion over 5-6 months, making the patient bed ridden for 3 months.

Patient also complains of involuntary dribbling of urine and decreased sensation of filling of bladder for 1 month.

On general physical examination her pulse rate was 82 beats per minute, Blood pressure was 120/70 mm of mercury, she was afebrile. There was no pallor, icterus, cyanosis, clubbing, generalized lymphadenopathy and oedema. On CNS examination power was grade 1 in ankle (all movements), grade 3 in right knee joint and grade 4 in right hip adduction. Left knee & hip abduction was normal. Sensory loss was also noted in the region of L3, L4, L5. Deep tendon reflexes were diminished

in right ankle & knee, DTRs were normal on left side. Plantar reflex was mute on both sides. There was a swelling of around 10×5 cm in lumbosacral, para spinal region, soft in consistency with well-defined borders. On contracting the muscle, size of the swelling decreased. Pain & tenderness were absent & skin over it was normal. Spinal tenderness was present in lower spine. Bladder was full. Straight Leg Raising test, Lasegue's test, crossed leg test was positive.

Immediately ultra-sonography of the swelling was done & it showed right psoas muscle collections extending into para vertebral region. ESR & CRP were normal. MRI lumbosacral spine suggested multiple cystic lesions in right pre and para vertebral regions extending to spinal canal through neuronal foramina L4- L5 & L5-S1. Provisional diagnosis of ?Infective etiology (hydatid) ?Neurogenic tumor. We did CECT abdomen to rule out the presence of any other primary lesions. The lesion was surgically removed and biopsy confirmed the diagnosis of hydatid cyst. After surgical decompression patient improved such that, she could stand & walk without support within 1 month. Patient was treated with Albendazole 400 mg BD peri operatively for 28 days.

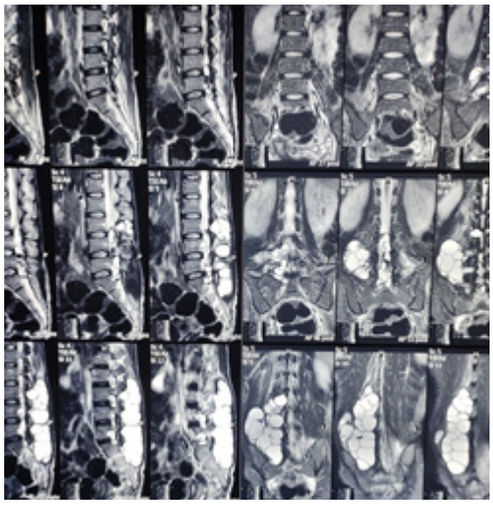


Figure1. MRI spine showing multiple cystic lesions in lumbosacral region

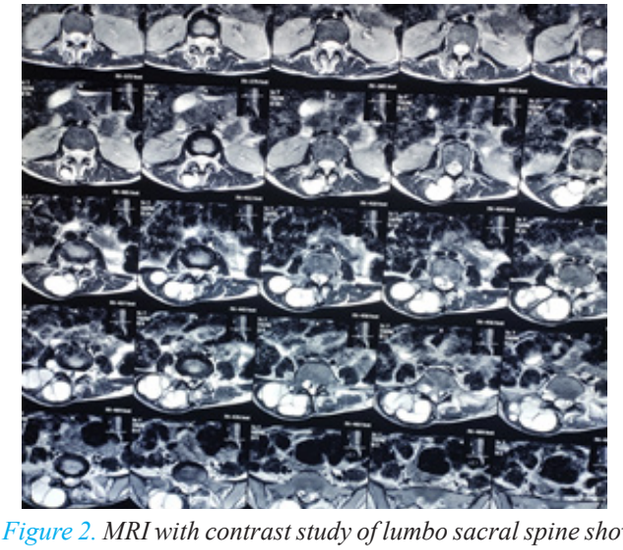
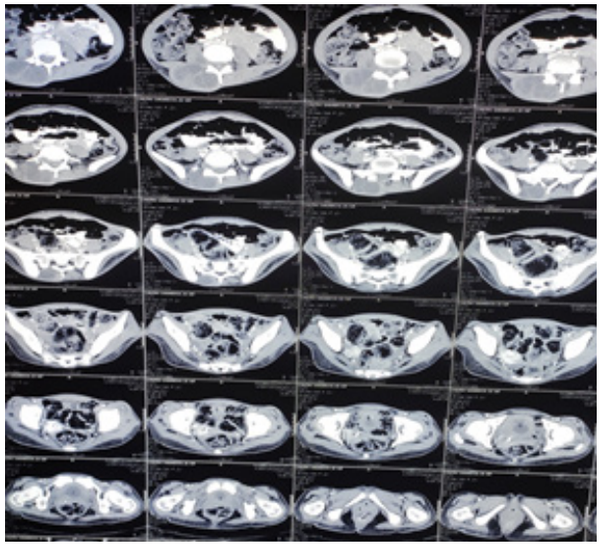


Figure 2. MRI with contrast study of lumbo sacral spine showing multiple cystic lesion



Fiureg 3. Contrast enhanced CT Abdomen and pelvis- showing cysts only in paraspinal region suggesting it as a primary.

Fiureg 4. Patient after treatment able to stand and walk with support.

Discussion

Spinal hydatid is a rare disease caused by Echinococcus granulosus and occasionally by echinococcus multilocularis. Source of infection is livestock, mainly dogs. The former is the more benign form which is characterized by cyst formation, whereas the latter is not encapsulated and presents as a ramifying, porous and necrotic mass with has mortality nearly 100%[6].

Among the bone hydatid diseases spinal hydatids are more common (50%). Since the venous plexus around spine have no valves and slow blood flow in this area. In the spinal cord thoracic vertebrae involvement is the most common[7]. It can be originated in the vertebral canal wall, or it can invade into the spinal canal through the inter vertebral foramen and compress the

spinal cord. Proliferation and spread can be outward along the Haversian osteon lamella system. When the cyst breaks through the cortical bone into the spinal canal, compression of the spinal cord or nerve root may occur, leading to symptoms. Symptoms at presentation may include para paresis at presentation (61–73%), associated or not with back pain (27.8–43%), bladder dysfunction (11.1–32%), sensory loss (24%), and radicular pain (27–60%)[11].

Owing to compression of sacral root below L3, our patient presented with early asymmetrical radicular pain in lumbosacral nerve root and then symptoms progressed to involve motor weakness and sensory symptoms typically suggestive of cauda equina syndrome. Involvement of bladder is "autonomous bladder" suggestive of sacral root involvement. Since sensory roots for bladder is involved, sensation of fullness of bladder will not be appreciated, leading to overflow incontinence associated with large bladder. Absence of ankle reflex suggestive of S1-S2 involvement & preserved knee reflex on left side suggests non involvement of L4/L3 roots on left side & involvement of the same on the right side. Sensory involvement of same nerve distribution adds to diagnosis. Pain typically increases in sleeping supine position and spinal tenderness is suggestive of destructive pathology since the para spinal muscles relax while sleeping & increase the laxity of vertebrae. Positive crossed leg test and bladder involvement suggests need of surgical decompression. Since patient was having low grade fever, our primary diagnosis was Pott's spine with para spinal cold abscess as it is more common. Lack of suspicion of spinal hydatid leads to primary diagnosis of Pott's spine. But normal ESR & CRP were 1st contradictions to our diagnosis, followed by imaging studies. Later, biopsy confirmed hydatid disease. CECT abdomen ruled out the involvement of other common sites like liver. Only spinal cord involvement alone (primary) made the presentation of the disease rare. For this case, primary hydatid infestation of the spine without any other systemic involvement can only 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[8–10].

Due to lack of suspicion and rarity of the presentation, early diagnosis is difficult. Literatures agrees that even serology does not add much to the diagnosis. The sensitivity rates of serological diagnosis of the liver, lungs, and other organs are 80–100%, 50–56%, and 25–56%, respectively[6]. The differential lesions in the Imaging includes aneurysmal bone cyst, giant cell tumor of bone, isolated bone cysts, arachnoid cysts, fibro cystic diseases, chondrosarcomas, and tuberculosis, etc[10,12]. Surgical decompression is the mainstay of treatment. Avoiding spillage of content during surgery to prevent allergic reactions and recurrence is most important. Perioperative use of Albendazole is useful along with other symptomatic support according to literature and we followed the same. Patient improved and could stand and walk without support. More studies are needed for the same.

Conclusion

Hydatid cyst across the inter vertebral foramen has been reported only sporadically in the literature, and the primary cyst is extremely rare. For diagnosis, hydatid cyst disease should be considered when imaging findings indicate cystic changes in the spinal region and the patient having the suspicious history, even if the serology test is negative. For treatment, the main method is still surgical treatment, and great attention should be paid to completely remove the cyst to avoid extravasation of the cystic fluid which results in spread of infection. As the incidence of this disease is extremely low and rarely encountered in clinical practice, this case is reported in the hope of improving the understanding, diagnosing it and treating it in time.

References

1. Eckert J, Deplazes P. Biological, epidemiological, and clinical aspects of echinococcosis, a zoonosis of increasing concern. Clin Microbiol Rev.2004.17:107–35.
2. Ben Ayed, M., Kamoun, N., Makni, K., Ben Romdhane, K.: Kystehydatique, 281 cas à localisation inhabituelle, observés au coursd'unepériode de dix ans. 1972–1981. Tunis. Med. 64:389, 1986.
3. Senol N, Kerman M, Ozturk A, Ucmakli E. A rare parasitic infection: primary intradural extramedullary hydatid cyst. Turkish Neurosurg.2016.26:460– 2. doi: 10.5137/1019-5149. Jtn.12209-14.1
4. Braithwaite PA, Lees RF. Vertebral hydatid disease: radiological assessment. Radiology. 1981. 140:763–6.
5. Zhang Z, Fan J, Dang Y, Xu R, Shen C. Primary intramedullary hydatid cyst: a case report and literature review. Eur Spine J. 2017. 26(Suppl. 1):107– 10.
6. Karakasli A, Yilmaz M, Mucuoglu AO, Yurt A. A large primary dumbbell hydatid cyst causing neural foraminal widening of the thoracic spine: a case report and literature review. Int J Surg Case Rep. 2015. 8c:55– 8.
7. Lam KS, Faraj A, Mulholland RC, Finch RGJS. Medical decompression of vertebral hydatidosis. Spine. 1997. 22:2050–5.
8. Karadereler S, Orakdögen M, Kiliç K, Ozdogan C. Primary spinal extradural hydatid cyst in a child: case report and review of the literature. Eur Spine J. 2002. 11:500–3.
9. Dighe MP, Balasubramaniam S, Nadkarni TD. Transthoracic approach for ventrally situated paraspinal extradural hydatid cyst of the dorsal spine. J Craniov Junction Spine. 2018. 9:271–3.
10. Joshi KC, Jagannath AT, Varma RG. Dumbbell shaped spinal hydatidosis. Neurology India. 2014. 62:113–4.
11. Gennari A, Almairac F, Litrico S, Albert C, Marty P, Paquis P. Spinal cord compression due to a primary vertebral hydatid disease: a rare case report in metropolitan France and a literature review. Neuro-Chirurgie. 2016. 62:226– 8.
12. Berk C, Ciftçi E, Erdogan A. MRI in primary intraspinal extradural hydatid disease: case report. Neuroradiology. 1998. 40:390–2.