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An Unusual Presentation Of A Spinal Hydatid Cyst – An Operative Surprise

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ABSTRACT

Isolated primary spinal hydatid disease without any visceral involvement is a rare finding. A middle aged male presented to our institute with a history of localized swelling over the back since 7 years and difficulty in walking since 3 months. On examination, there was a 20x15cm diffuse swelling visible over the back. MRI scan was suggestive of a plexiform neurofibroma, hence surgical excision was planned. After doing a *D*11 to L2 level hemi-laminectomy , the lesion was dissected in the muscular plane and into the foraminal part. While performing internal debulking , the lesion was incised which then revealed a bunch of pearly white cysts within, much to our surprise. Once these cysts were identified, utmost care was taken to avoid rupture of cysts. On follow up after 3 months, patient was symptom free, ambulatory without support and the MRI showed no remnant or residual of the hydatid disease.

Keywords: *Spinal hydatid disease, Isolated echinococcosis, Hemi-laminectomy, Pearly white cysts, Misdiagnosed neurofibroma, Surgical excision*

1. INTRODUCTION

Hydatid is a Greek word which means ‘watery cyst’. Echinococcus granulosus also known as ‘dog tapeworm’ which is a helminth of the Cestoda class is a major causative factor of this zoonotic disease. (1) E. granulosus and E. multilocularis are the two main species identified which are endemic to the temperate climate. (2) Echinococcus granulosus is one of 20 neglected tropical diseases as identified by the World Health Organization. (1) In primary hydatid disease most commonly involved organs are the liver (60-70%) and lungs (10-15%). Bone involvement is uncommon and a rare complication and occurs by hematogenous route in about 0.5-4% of patients. (3) The bones affected in decreasing order of incidence are pelvis, sacrum, metaphysis of long bones, skull, spine and ribs. (1) Isolated primary spinal hydatid disease without any visceral involvement is a rare finding. (4) With consideration to spinal involvement, thoracic spine is the most affected site followed by lumbar, sacral and cervical spine. (5) The only definitive treatment for spinal osseous hydatidosis is surgery. Multiple options for treatment have been mentioned in literature including simple Dr.ainage or debridement, curettage and resection of infected bone, anterior and posterior decompression and stabilization. (6)

We present a case of an isolated spinal hydatid cyst of the thoraco-lumbar spine who presented with neurological deficits.

2. CASE REPORT

A middle aged male, farmer by occupation presented to our institute with a history of localized swelling over the back since 7 years and difficulty in walking since 3 months. There was associated history of back pain predominantly over the swelling with no radicular pain in both lower limbs. The swelling over the back was neglected, was initially small to begin with when noticed 7 years ago and gradually increased in size. He complained of heaviness in both the lower limbs which led to reduced pace while walking as compared to others around him over a period of 3 months. No bowel and bladder complaints and no history of significant weight loss. Being a farmer by occupation he would be in regular contact with cattle, however he did not have a pet and no history of any underlying diseases. No positive family history for hydatid disease.

On examination, there was a 20x15cm diffuse swelling visible over the back approximately extending from the lower thoracic to lumbo-sacral region and into the right flank. *(Fig 1)* The swelling was firm in consistency, mobile with no reducibility and pulsatality. There was hypertonia of both lower limbs with an exaggerated knee and ankle reflex. Plantars were extensor. Power remained 4/5 in both lower limbs and had to be ambulated on a wheelchair.

Figure 1 – Patient in prone position. Diffuse swelling visible over the back approximately extending from the lower thoracic to lumbo-sacral region and into the right flank.

Investigations

Contrast MRI scan was suggestive of a T1 hypo intense and T2 hyper intense, large multiloculated, multiseptate lesion with cystic and solid component in the right posterior abdominal wall in muscular plane, extending into the intraspinal region through widening of right neural foramina from D11 to L4 level. No enhancement seen on post contrast images. Lesion appeared to cause varying severity of compression over the spinal cord, conus medullaris and cauda equine nerve roots, probable radiological diagnosis of plexiform neurofibroma. *(Fig 2 a b)*

Figure 2a – Pre-operative MRI contrast images (sagittal view)

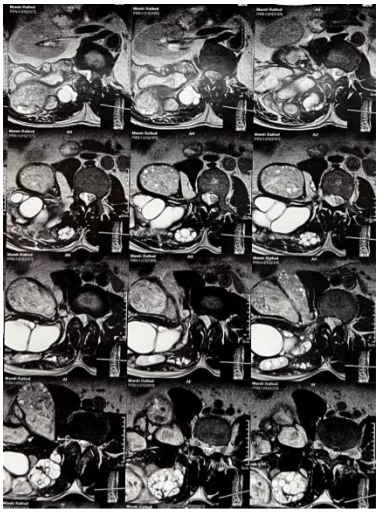

Fig 2b – Pre-operative MRI contrast images (axial view)

Treatment

With suspicion of a neurofibroma, surgical excision was planned. A midline incision was taken extending from D10 to L5 level. A soft cystic swelling was visualized in the muscular plane in the right paraspinal region. *(Fig 3) D*11 to L2 level hemi-laminectomy was done. The lesion was causing significant dural tube compression. After performing a hemilaminectomy , the lesion was dissected in the muscular plane and into the foraminal part. With an aim to perform internal debulking of the lesion, the lesion was incised which then revealed a bunch of pearly white cysts within, much to our surprise. Once these cysts were identified, utmost care was taken to avoid rupture of cysts. *(Fig 4) Hypertonic* saline soaked gauze was placed in the surrounding region and gross total resection of the lesion was done. *(Fig 5)* There was no intra-operative rupture of the cysts or dural injury. The lesion was separated slowly from the underlying exiting nerve roots and none of them were sacrificed. The surgical region was washed with 20% hypertonic saline after excision of the lesion. As only hemilaminectomy was performed and there was no bony instability, we did not plan a posterior spinal stabilization.

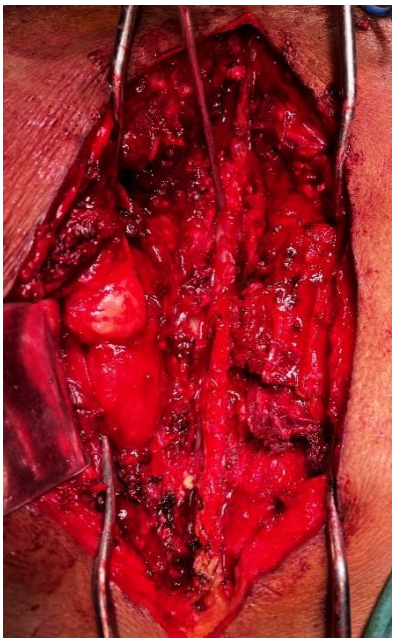

Fig 3 – Intra-operative image on initial exposure of the spine. Lesion seen in the right paraspinal region.

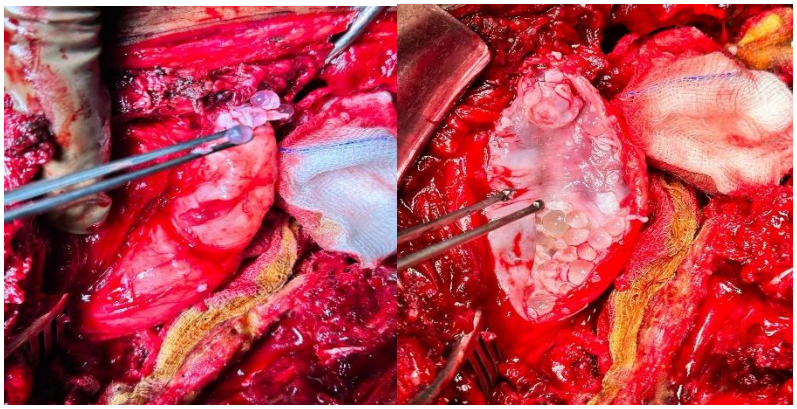

Fig 4 – Internal debulking of the lesion and visualization of the hydatid cysts

Fig 5 – Specimen of excised lesion

Outcome and Follow up

As a result of the operation, patient's neurological symptoms improved and he was discharged on antihelmintic therapy of albendazole 400mg BD for the next six months.

At follow up after 3 months, patient was symptom free, ambulatory without support and the neuraxis MRI showed no remnant or residual of the hydatid disease. *(Fig 6)*

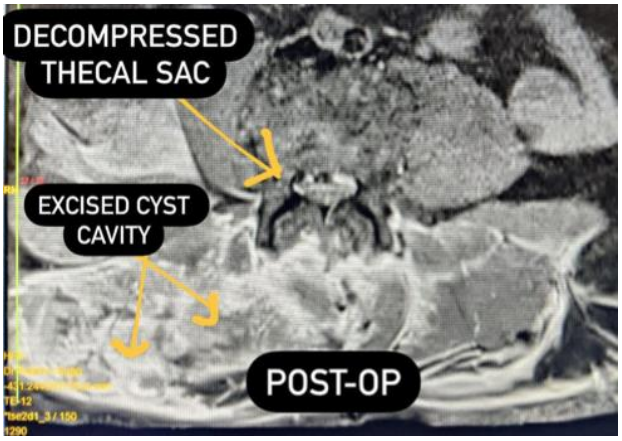

Fig 6 – Post operative MRI scan (axial view)

3. DISCUSSION

The overall incidence of bony hydatidosis remains rare, with vertebral location seen in half of these cases and generally affecting a single vertebra. (7) Hydatidosis is a major cause of disease burden worldwide and affects all age groups. The estimated post-operative mortality rate is 2.2% and recurrence rate is 6.5%. (8)

With regards to the life cycle of Echinococcus granulosus, it accidentally uses humans as intermediate host. Adult worms develop in the intestine of the permanent hosts mainly dogs and wolves and their eggs are shed in their feces. These eggs get consumed by herbivores like sheep and cattle. Humans in turn get infected by eating food contaminated with these parasite eggs or due to contact with these permanent hosts or their feces. After being consumed, the real larva hatches from the egg and burrows into the intestinal submucosa and then travels to the internal organs such as liver by veins or lymphatic vessels. Ninety percent of larva are eliminated by host reaction, however if they survive the hydatid cyst develops in the affected organ over a course of years. (1)

Primary infestation of the spine mainly occurs through the porto-vertebral shunts. Centre of the vertebral body is the first to get involved. The larva penetrates the cancellous bone and causes multivesicular and diffuse infiltration. There is aggressive proliferation along the line of least resistance, particularly the bony canals. Eventually, the daughter vesicles replace the medullary cavity and reaches the cortex and destroys it to spread to surrounding structures. (2)

Braithwaite and Lees classified spinal hydatid cyst based on location into five types – intramedullary, extramedullary intradural, extradural intraspinal, vertebral and paravertebral. (9) Our case was of extradural intraspinal type. Spinal hydatid cyst normally does not present with any pathognomic signs except those of direct compression of the cord. Weakness of limbs and paraplegia has been reported in about 25% of the cases. (6)

It is difficult to accurately come to a diagnosis of spinal hydatidosis radiologically. Some typical imaging features include sclerosis of involved bone, lack of intervertebral disc space involvement along with paraspinal extension of disease. CT may show honeycomb like appearance of spine along with rim calcification. It is commonly hypo intense on T1 weighted and hyper intense on T2 weighted imaging with multilocularity being a key feature. Viable cysts will demonstrate hyper intensity on T2 weighted images whereas hypo intensity on both T1 and T2 weighted images represents dead or dying cysts. (2) There are a number of conditions which resemble spinal echinococcosis radiologically such as Potts disease, brucellosis, osteomyelitis, aneurysmal bone cysts, spinal abscess, cancer and vertebral metastasis. (1) In our case, on initial radiological diagnosis we suspected it to be an extradural tumour most probably a neurofibroma and were caught by surprise on performing a laminectomy and visualization of the lesion.

The gold standard treatment for hydatid cysts is surgical excision of the whole cyst. Biopsy and aspiration of the cyst should be avoided as there is diffusion risk and anaphylaxis.(10) Thoracic spinal hydatid cysts are reported to have high possibility of malignant transformation and recurrence, hence radical excision should be the first line of management.(6) Type of operative procedure and extent of resection depends of various factors like location of cysts, extent of bony involvement and presence of bony instability.(1) The basic technique of surgery involves exposure of lesion and protection of adjacent normal tissue with gauze soaked in hypertonic saline. Care should be taken to avoid damage to the dura, as dural damage has been reported as a plausible mechanism of spread. Another important step is to avert rupture of cysts and spillage of contents as this can lead to an anaphylactic reaction and subsequent recurrence. (6) Recurrence is about 30-40% which is primarily due to rupture of the cysts intra-operatively. (4) However, even with all precautions taken rupture of cysts is a known complication. Various solutions are used to wash the operative field in case of a rupture of the cyst and this post-operative adjuvant antiparasitic chemotherapy mainly consists of solutions like hypertonic saline (3%,10%,20%), 0.5% betadine, 2% formalin and 0.5% silver nitrate. (10) World Health Organization recommends albendazole pharmacotherapy, with dosage of 10-15mg/kg/day for 6 months to 1 year after surgery to prevent recurrence of the cyst. (1)

4. CONCLUSION

- Our case represents a rare variety of isolated spinal hydatid disease involving the thoraco-lumbar spine without visceral involvement and presenting with neurological deficits. Bone involvement is uncommon and a rare complication and occurs by hematogenous route in about 0.5-4% of patients. The bones affected in decreasing order of incidence are pelvis, sacrum, metaphysis of long bones, skull, spine and ribs.
- Treatment of spinal hydatidosis is difficult and the treating team should be aware regarding the pre-operative evaluation and diagnosis of spinal hydatidosis and surgical technique.
- It is important to have a high degree of suspicion for spinal hydatid disease and an individualized surgical strategy should be carefully decided for each patient in their first intervention. Strict follow up and regular MRI scans are necessary to detect recurrence at an early stage.
- Type of operative procedure and extent of resection depends of various factors like location of cysts, extent of bony involvement and presence of bony instability. Care should be taken to avoid damage to the dura, as dural damage has been reported as a plausible mechanism of spread. Another important step is to avert rupture of cysts and spillage of contents as this can lead to an anaphylactic reaction and subsequent recurrence.

Conflict of Interest: None to declare

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