

## CASE REPORT

# A Rare Presentation of Primary Hydatid Cyst in an Adolescent – A Case Report

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### ABSTRACT

The aim of this case study was to report a very uncommon and rarely seen, but serious condition, involving spine. Hydatid cyst is caused by *Echinococcus granulosus* mainly. It is rarely found in spine, lumbar spine is least commonly involved in spine. A 14 years old boy with lower back pain radiating to both lower limbs observed in the hospital. Associated with progressive weakness and sensory deficit in both lower limbs investigations showed a space occupying lytic lesion extending from L3-S1 with cystic nature, showing nerve root compression at L3, L4 and L5. Managed with excision of cyst through posterior approach to lumbar spine and stabilized with posterior spinal instrumentation L2-L5 and S2Al screw. Hydatid cyst in lumbar spine should be considered as differential diagnosis of cystic lesions of spine. Treatment of hydatid cyst in spine is primarily removal of lesion. Postoperatively the patient improved, mobilized on first post-operative day and has been complication free in one-week follow up.

**Keywords:** Hydatid Cyst; Spine; *Echinococcus granulosus*; Lumbosacral Spine.

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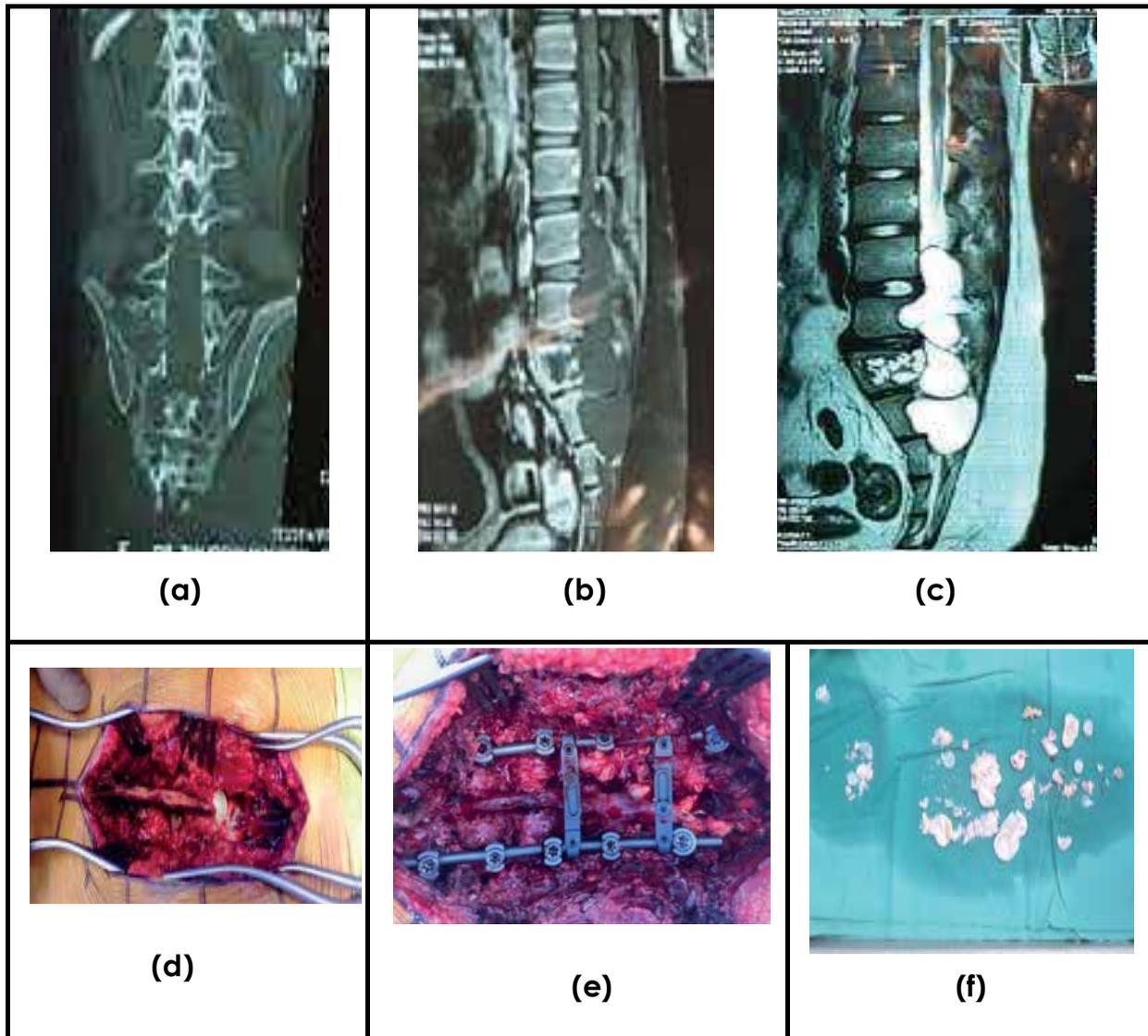
### INTRODUCTION

Hydatid means "watery cyst" in a Greek language. This disease is caused by the *Echinococcus granulosus* and *Echinococcus multilocularis*<sup>1</sup>. It is not very common as humans are intermediary host. Usually found in lungs and liver commonly sites involved. Spine is least involved site comprising of 1% of all hydatid cyst cases<sup>2-5</sup>. It is commonly found in young patients those are in contact with dogs<sup>3</sup>.

Spine is involved secondary to spread from pulmonary, abdominal or pelvic infection, most commonly involving the thoracic spine, in decreasing order of frequency to lumbar, sacral and cervical spine<sup>4,6</sup>. We presented a case of boy with hydatid cyst in lumbosacral spine, with progressive weakness and sensory deficit bilateral lower limbs that was a very rare site for involvement of hydatid cyst.

### CASE PRESENTATION

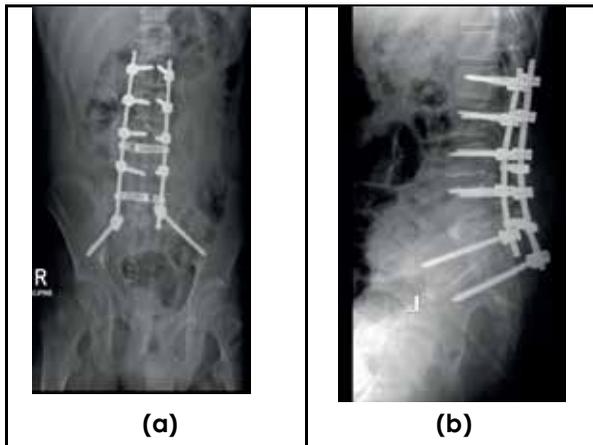
A 14-years old, male patient no known comorbid presented to outpatient department with complaint of pain in lower back since 1 year. Pain was gradual in onset and progressed with time, does not give any history of trauma. Pain was radiating to bilateral lower limbs associated with sensory deficit and progressive weakness bilateral lower limbs. Since last few months mobility was restricted to home, because of pain and progressive weakness patient was hardly able to walk for more than thirty minutes. On examination healthy boy weighing 69kg, walks with waddling gait. Motor examination was unremarkable in bilateral lower limbs. There was sensory deficit in right foot at distribution of Lumbar 5-Sacral 1, ankle jerk was absent. MRI (Magnetic Resonance Imaging) and CT (Computerized Tomography) was performed (Figure 1a-f) which showed space occupying lytic lesion extending from L3-S1 with cystic nature, with nerve root compression at L3, L4 and L5.



**Figure 1: (a) Coronal cuts of CT scan showing involvement and extent of cyst in lumbosacral spine, also involving the right side sacrum (b) Sagittal cut of MRI T-1 image – showing dark signals around the involved area, as CSF and fluid appears dark. Sagittal cut of MRI (c) T-2 image – showing high uptake along posterior border of L3-S1 also involving vertebral body of S1 (d, e) Intraoperative images of cyst and fusion surgery (f) Postoperative removed cyst.**

Patient was planned for excision of cystic lesion and spinal instrumentation through posterior approach. After taking consent from family patient was given general anesthesia and was kept in prone position, skin incision was given following midline approach to lumbo-sacral spine. While doing soft tissue dissection he was found to have cystic lesion extending from spine superficially on right side with clear fluid and lytic sacral bone. Cyst was excised completely and sent for frozen section per-operatively and it came out to be hydatid cyst. Excision was done followed by posterior spinal instrumentation from

L2-L5 and S2A1 screw (Sacrum 2 Alar iliac) connected with rods and connector (Figure 2a, b). Later decompression was completed and curettage of all necrotic bone was performed and irrigation done with hypertonic saline. Patient remained stable on immediate post-operative day, with intact lower limbs neurology. Patient was ambulated full weight bearing on first post operation day without any brace, with relieved pre-operative symptoms; hospital course was uneventful and discharged home on fourth post-operative day without any walking aid.



**Figure 2a, b: Post operative images of posterior spinal instrumentation L2-L5 and S2AI screws.**

### DISCUSSION

Hydatid disease is usually formed in liver (60-70%) and lungs (10-15%). Bones are involved rarely which is 0.5 to 2%<sup>7,8</sup>. Neurological deficit have been reported to be found in patients with hydatid cyst of spine in 25-84% of cases<sup>9</sup>. It is more common in males' 11:1 female with an age range of 7 years to 56 years average age of 27 years. The parasite extends the paravertebral tissue or the epidural space through the cortex and pore front of the vertebral body. Cyst can progress from body of vertebrae to anterior space, on the sides to extradural space and paravertebral tissues, and to spinal canal putting pressure on the cord, causing radiculopathy symptoms and neurological deficit. Cyst is usually located in posterior element of spine or posterior aspect of cord<sup>10</sup>. It is more common in sheep raising areas of world, it affects human by ingesting the eggs excreted by dogs.

Hydatid cyst has two layers, inner germinal layer and outer fibrous layer. It has two types unilocular and multilocular, commonest among them is unilocular. Three layers of hydatid cyst are further classified histopathologically, peripheral adventitial layer consist of eosinophils, intermediate cuticular layer and inner germinal layer<sup>11</sup>. Other conditions can be expected such as syrinx, hematoma, arachnoid cyst, epidermoid cyst, hemangioblastoma, myelomalacia and astrocytoma like cystic neoplasms.

Braithwaite and Lees<sup>7</sup> classified the spinal disease in five types: 1) primary intramedullary hydatid cyst, 2) intradural extramedullary hydatid cyst, 3) extradural intraspinal hydatid cyst, 4) spinal hydatid cyst disease of the spine and 5) paravertebral hydatid cyst disease. In our case, it was type 4 spinal hydatid cyst diseases of spine.

Clinical manifestation of hydatid cyst usually shows no sign or symptoms other than symptoms related

to cord compression<sup>12</sup>. Patient usually presents with chronic continuous lower back pain with radicular pain. Because of pressure over the cord or nerve roots in lower spine, patient may presents with neurological deficit, it can cause paraplegia in 25% to 84% of cases. Symptoms found in our patient were chronic lower back pain since 1 year, with radiculopathy in both lower limbs and now progressing to neurological deficit.

To confirm the diagnosis CT and MRI is necessary to see extant of disease. CT scan is done to see bone involvement and damage to vertebral structures. As hydatid cyst contents has same density as CSF MRI is considered as best imaging tool. The density of the cyst fluid is the same as the CSF in T1-and T2-weighted imaging<sup>13</sup>. Irregular vertebral body margins with non-enhancing flattened sausage shaped lesions, non-septated walls are all signs of vertebral hydatid disease. Cyst lying in extradural area are always multiple involving the bone<sup>14</sup>.

Lesion should be completely excised as a block and instrumentation must be performed for stabilization. Location of cyst and surgical area should be washed off with hypertonic saline. Goal of surgery is too remove the cyst without rupturing it, and it should be excised as much as possible without damaging any adjacent structure. However, it is not possible to do complete radical excision of lesion<sup>15</sup>. certain solution has not been proved yet, how potent they are, but most of the studies recommend use of (3%,10%,20% hypertonic saline, 0.5% betadine, 0.5% silver nitrate and 2% formalin<sup>10</sup>.

Surgical procedure varies from only excision of cyst to complete decompression depending on extant of disease; MRI (magnetic resonance imaging) is performed on regular follow up in post-operative period to ensure that recurrence is picked up, as re-operation is usually required. 30%-40% recurrence rate is found in previous studies<sup>16</sup>. Surgery is choice of treatment for spinal hydatid cyst as most of patients present with signs of cord compression, which requires immediate intervention. Surgeons usually act promptly in these cases.

Cyst has a tendency to be burst during the surgery, and its recurrence will exceed to 40% as reported before<sup>17</sup>. If there is, a single cyst or solid cyst wall recovery is possible post-surgical excision. It is often been misdiagnosed, a review from one study found that error among radiologists averages from 3% to 5%, in the same study it was noted that 26 % of cases diagnosis is missed clinically<sup>18</sup>. For the primary diagnosis of hydatid cyst and for follow up in outpatient department immunodiagnostic is useful. For serodiagnosis of hydatid cyst complement fixation test was the primary immunologic method. Most reliable test for diagnosis of hydatid cyst is Casoni test, which is positive in 90% of cases<sup>19</sup>.

## CONCLUSION

Hydatid cyst is rare disease involving spine, but it is related to very severe morbidity. Patient with hydatid cyst of spine may end up in neurological deficit. Although it is a rare case but possibility should be suspected in patients with chronic back pain. Cystic lesion in spine diagnosed with help of MRI and CT scan should be considered as hydatid cyst under the differentials until proven.

## ACKNOWLEDGEMENTS

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## CONFLICT OF INTEREST

The authors declare no conflict of interest.

## PATIENT CONSENT

Patient had been informed regarding the study and written consent was taken.

## AUTHOR'S CONTRIBUTION

All authors contributed equally in this case study.

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