

CASE REPORT

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Hydatid disease causing paraplegia: A case report with literature review

Hewa M. Mustafa, Nuraddin H. Muhammad, Fahmi H. Kakamad, Rawand Abdulrahman Essa, Hawbash M. Rahem

ABSTRACT

Introduction: Hydatid disease is a parasitic infection, commonly affecting liver and lung. Musculoskeletal system is rarely affected. The aim of this study is to report a case of paraplegia caused by hydatid cyst with a brief literature review. **Case Report:** A 22-year-old male presented with complete paralysis of both lower limbs for five days. Magnetic resonance imaging showed a big hydatid cyst in the mid dorsal spine and posterior part of the chest. Under general anesthesia, left posterolateral thoracotomy was performed, there was 3x4 cm mass consisting of multiple hydatid cysts that attached to thoracic wall and invaded adjacent vertebral bodies. The cysts were removed in toto. The patient was normal after three weeks of surgery. **Conclusion:** Hydatid disease although extremely rare, might cause reversible paraplegia. Operation is the management of choice.

Keywords: Hydatid cyst, Paraplegia, Spinal column

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INTRODUCTION

Hydatid disease (watery cysts, HD) is a Greek term, refers to infestation by tapeworm *Echinococcus*. Although there are several types of *Echinococcus*, humans are mainly infected by *Echinococcus granulosus* through fecal–oral route. Affection by *Echinococcus multilocularis* is rare in humans but it causes more severe and malignant form of alveolar HD [1]. Encystation in various organs occurs after infection with worm larval stage [2]. Liver and lungs are the two most commonly affected organs accounting for 90% of all human hydatidosis, (75% for liver and 15% for lungs). Other organs are affected by 10% [3]. Bone and muscle involvement is extremely rare [4, 5]. Primary vertebral HD is a very rare condition (less than 1% of all cases of HD), in which dorsal spinal column is most commonly affected [2]. Compression of the cords and roots by mass effect is the main etiology of clinical findings. The diagnosis of HD is confirmed by histopathological examination which shows eosinophilic laminated membrane (stained by Eosin and Hematoxylin) invaded by inflammatory cells. The cyst is surrounded by

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granulomatous inflammation induced by seepage of the content [2]. Presentation of HD by paraplegia resulting from spinal cord compression by the cyst is very rare with only 21 reported cases in literature [1–3, 6–20]. The aim of this study is to report a case of hydatid disease presenting with complete paralysis of both lower limbs with a brief review of literature.

CASE REPORT

A 22-year-old male presented with complete paralysis of both lower limbs associated with constipation and bladder incontinence for five days. Babinski sign was up-going. Hematological tests were within the normal range. Magnetic resonance imaging (MRI) scan showed a big hydatid cyst in the mid dorsal spine and posterior part of the chest (posterior mediastinum) (Figure 1). Under general anesthesia, left posterolateral thoracotomy (6th intercostal space) was performed, there was 3x4 cm mass consisting of multiple hydatid cysts that attached to thoracic wall and invaded adjacent vertebral bodies causing pressure to the spinal cord (Figure 2). The cysts were removed in toto. At first postoperative day, the patient regained some flickering leg movements ranging from grade 1–2. At third postoperative day, he was discharged from hospital and complete recovery with return of grade 4 was seen after three weeks of surgery.

DISCUSSION

Hydatid disease which is a parasitic infection is caused by *Echinococcus granulosus* through a larval stage. Man is an accidental intermediate host for the worm which may affect any body part especially liver and lung [9]. Hydatid disease of the spine is a slowly progressing, silent disease, developing into neurological deficit after long period of latency [6]. When it affects the lumbar spine, the onset is even more delayed in comparison to the thoracic involvement [8]. In the current case, in which dorsal spine was involved, the patient developed complete paraplegia within five days. Spine hydatid disease is a dangerous disease with mortality rate reaching 14–58% [8]. It has been categorized by Braithwaite and Lees into five major subtypes [21]:

- Hydatid disease of vertebrae
- Paravertebral hydatid disease
- Primary intramedullary hydatid disease
- Intradural extramedullary hydatid disease
- Extradural intraspinal hydatid disease

The current case was extradural intraspinal hydatid disease.

Spinal involvement by hydatid disease has been reported in various age groups. Parot et al. reported a 50-year-old male presented with chest pain and cough followed by progressive paraplegia [13]. Hydatid disease causing paraplegia in a four-year-old child was reported

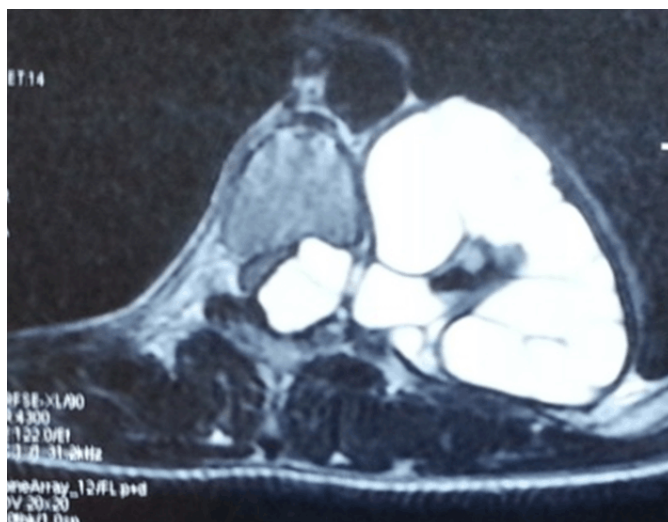


Figure 1: Magnetic resonance imaging scan of the dorsal spine showing a hyperdense lobulated mass compressing and invading the D10 and D11.

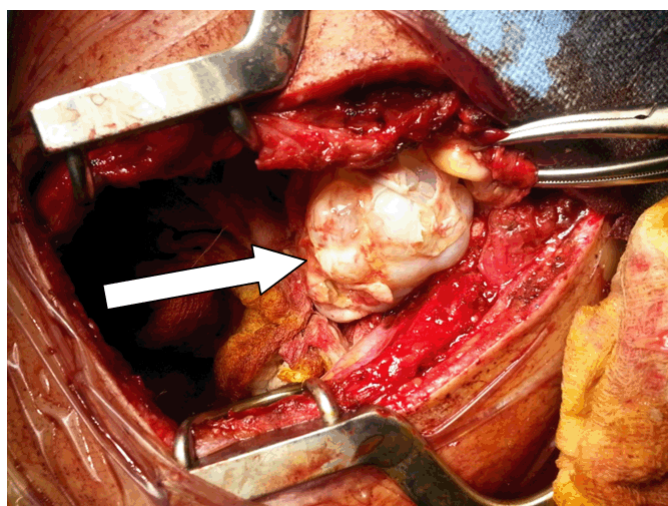


Figure 2: Intraoperative findings of lobulated laminated mass in left costovertebral junction (white arrow).

by Eloqayl et al. [9]. The age of the current case is 22 years which is among the similar age group reported by other studies [2, 12, 18]. Presentation of HD varies according to the organ involved. Paraplegia caused by HD may cause diagnostic dilemma. Baram et al. reported a 40-year-old lady presenting with paraplegia for four month duration undergoing lumbar laminectomy as she was diagnosed as L4,L5 disc prolapse [6]. Hassan et al. presented a 40-year-old male with paraplegia for two years. Provisional diagnosis of lymphoma was done. Fine-needle aspiration was performed under radiological guidance which showed laminated eosinophilic membrane with few scolices which confirmed the diagnosis of HD [20]. Sharma et al. reported a 14-year-old child presented with progressive paraplegia. They put him on antituberculosis therapy for six months without response. Later, operation for laminectomy (D5 to D8) revealed spinal HD [17].

Diagnosis of hydatid disease of spine is considered in endemic areas and classically, spinal hydatid cyst is diagnosed by magnetic resonance imaging (MRI) scan, on both T1 and T2, the wall appears as hypodense ring and on T2, the content is hyperdense [6, 9]. The differential diagnoses include pyogenic infection, vertebral tuberculosis, fibrous dysplasia, malignancies, enchondroma, multiple myeloma, hyperparathyroidism, hematoma, abscess and giant cell tumors [7, 11].

The current treatment standard for hydatid disease is total surgical removal of the cysts and spinal cord decompression before irreversible damage occurs, followed by medical therapy for about 3–9 months [7, 11].

It is not well known, to which degree spinal cord injuries induced by HD, cure. Dongel et al. state that rate of recovery from spinal cord injury by HD is inversely proportional to the duration of the symptoms. According to them, prolonged compression causes nerve ischemia with subsequent irreversible spinal cord injury [8, 11]. This idea might be challenged by what are reported in literature. Baram et al. reported complete recovery after four months paraplegia in a 40-year-old lady who presented with posterior mediastinal mass invading dorsal vertebrae [6]. On the other hand, Eloqayli et al. did not observe any improvement in a four-year-old male child with history of four weeks paraplegia after complete surgical evacuation of the hydatid cyst involving D6 to D9 [9]. Also, Fiennes and associates reported a 39-year-old female presented with sudden onset of paraplegia for one week duration caused by HD affecting D3 and D4 with only mild improvement after surgical cystectomy [10]. The current case with history of five days paraplegia retained full power of both lower limbs and normal continence four months after surgical evacuation.

Primary prevention and control of the disease are necessary which include good sanitation, standard deworming of domestic animals and clean water supply [11].

CONCLUSION

Hydatid disease of the spine is a very rare disease, causing paraplegia is even rarer. Magnetic resonance imaging scan is the diagnostic tool of choice. Surgical evacuation is the standard management strategy. Outcome is variable and unexpected.

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Hewa M. Mustafa – Substantial contributions to conception and design, Acquisition of data, Revising it critically for important intellectual content, Final approval of the version to be published

Nuraddin H. Muhammad – Substantial contributions to conception and design, Acquisition of data, Revising it critically for important intellectual content, Final approval of the version to be published

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Rawand Abdulrahman Essa – Substantial contributions to conception and design, Drafting the article, Final approval of the version to be published

Hawbash M. Rahem – Substantial contributions to conception and design, Drafting the article, Final approval of the version to be published

Guarantor of Submission

The corresponding author is the guarantor of submission.

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Conflict of Interest

Authors declare no conflict of interest.

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