Early View Article: Online published version of an accepted article before publication in the final form.

Journal Name: Edorium Journal of Infectious Diseases

doi: To be assigned

Early view version published: November 11, 2017

How to cite the article: Mustafa HM, Muhammad NH, Kakamad FH, Essa RA, Rahem HM. Hydatid disease causing paraplegia; a case report with review of literature. Edorium Journal of Infectious Diseases. Forthcoming 2017.

TYPE OF ARTICLE: Case Report

TITLE: Hydatid disease causing paraplegia; a case report with review of literature

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Short Running Title: Paraplegia caused by Hydatid cyst.

Guarantor of Submission: The corresponding author is the guarantor of submission.

ABSTRACT

Introduction

Hydatid disease is a parasitic infection, commonly affecting liver and lung. Muscular-skeletal system is rarely affected. The aim of this study is to report a case of paraplegia caused by Hydatid cyst with brief literature review.

Case Report

A 22-year-old male presented with complete paralysis of both lower limbs for 5 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 3 x 4 centimeter 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.

INTRODUCTION

Hydatid disease (watery cysts, HD) is a Greek term, refers to infestation by tapeworm Echinococcus. Although there are several types of Echinococus, humans are mainly infected by Echinococcus granulosus through faeco-oral root. 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]. Livers and lungs are the two most commonly affected organs accounting for 90% of all human hydatidosis, (75% for liver, 15% for lungs). Other organs are affected by ten percent [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 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 HD presenting with complete paralysis of both lower limbs with brief review of the literature.

CASE REPORT

A 22-year-old male presented with complete paralysis of both lower limbs associated with constipation and bladder incontinence for 5 days. Babinski sign was up-going. Hematological tests were within normal range. Magnetic resonance imaging (MRI) 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 3 x 4 centimeter 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

HD which is a parasitic infection is caused by *Echinococcus granulosis* through a larval stage. Man is an accidental intermediate host for the worm which may affect any body part especially liver and lung [9]. HD of the spine is a slowly progressing, silent disease, developing into nerulogoical deficit after long period of latency [6]. When it affects the lumber 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 HD is a dangerous disease with mortality rate reaching 14-58% [8]. It has been categorized by Braithwaite and Lees into five major subtypes [21]:

1. HD of vertebrae
2. Paravertebral HD
3. Primary intramedullary HD
4. Intradural extramedullary HD
5. Extradural intraspinal HD

The current case was extradural intraspinal HD (subtype 5).

Spinal involvement by HD 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]. HD causing paraplegia in a 4 year old child was reported by Eloqayl and his colleagues [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 and associates reported a 40-year-old lady presenting with paraplegia for four month duration undergoing lumber laminectomy as she was diagnosed as L4,L5 disc prolapse [6]. Hassan et al presented a 40-year-old man with paraplegia for 2 years. Provisional diagnosis of lymphoma was done. Fine needle aspiration was performed under radiological guidance which showed laminated esinophilic membrane with few scolices which confirmed the diagnosis of HD [20]. Sharma and his colleagues reported a 14-year-

old child presented with progressive paraplegia. They put him on antituberculosis therapy for 6 months without response. Later, operation for laminectomy (D5 to D8) revealed spinal HD [17].

Diagnosis of HD of spine is considered in endemic areas and classically, spinal Hydatid cyst is diagnosed by magnetic resonance imaging (MRI), 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, vertebtal tuberculosis, fibrous dysplasia, malignancies, enchondroma, multiple myeloma, hyperparathyroidism, hematoma, abscess and giant cell tumors [7][11].

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

It is not well known, to which degree spinal cord injuries induced by HD, cure [11]. Dongel and colleagues 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 [11]. This idea might be challenged by what are reported in literature. Baram et al reported complete recovery after 4 month 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 4-year-male child with history of 4 week paraplegia after complete surgical evacuation of the Hydatid cyst involving D6 to D9 [9]. Also, Fiennes and associates reported a 39-year-old lady 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 5 day paraplegia retained full power of both lower limbs and normal continence 4 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

HD of the spine is a very rare disease, causing paraplegia is even rarer. MRI is the diagnostic tool of choice. Surgical evacuation is the standard management strategy. Outcome is variable and unexpected.

CONFLICT OF INTEREST

None to be declared

AUTHOR'S CONTRIBUTIONS

Hewa M. Mustafa and

Group 1-Substantial contribution to the concept and design, supervising the operation

Group 2-Revising the manuscript

Group 3-Final approval of the manuscript

Nuraddin H. Muhammad

Group 1-Substantial contribution to the concept and design

Group 2-Revising the manuscript

Group 3-Final approval of the manuscript

Fahmi H. Kakamad

Group 1- Substantial contribution to the concept and design, acquisition the data

Group 2-Drafting the manuscript, Revising the manuscript

Group 3-Final approval of the manuscript

Rawand Abdulrahman Essa

Group 1-Substantial contribution to the concept and design, acquisition the data

Group 2-Drafting the manuscript

Group 3-Final approval of the manuscript

Hawbash M.Rahem

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FIGURE LEGENDS

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

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

FIGURES

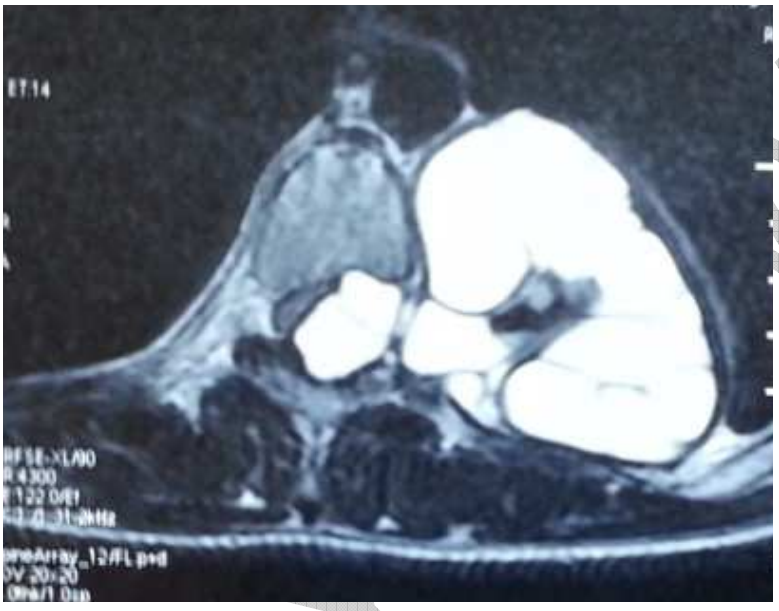

Figure 1: Magnetic resonance imaging of the dorsal spine showing a hyperdense obulated mass compressing and invading the D10 and D11.

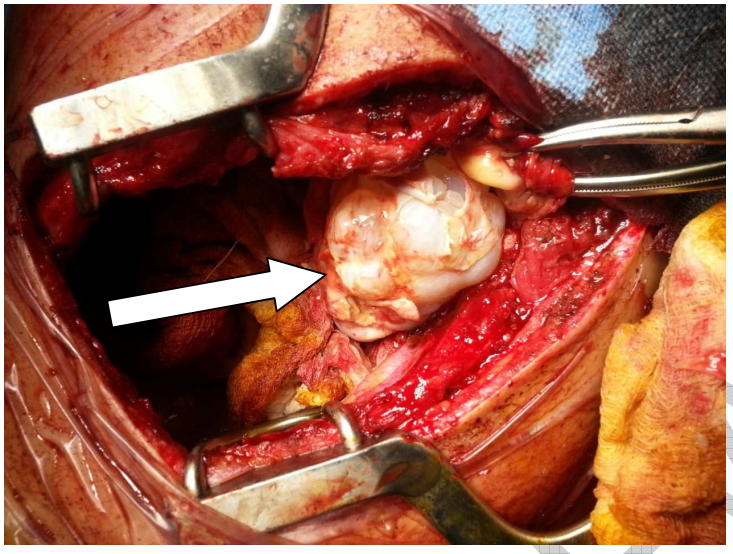

Figure 2: intraoperative findings of lobulated laminated mass in left costovertebral junction (white arrow).