

Relapsing Primary Spinal Cord Hydatid Cyst: A Case Report and Review of Literature

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ABSTRACT

A 35-year-old man complaining of progressive paresis in his lower extremities presented to the neurosurgical ward. The patient had history of spinal hydatid cysts and had undergone surgical resection 5 years ago. Multiple lesions were observed at the regions of previous surgery on spine magnetic resonance imaging. Thus, this patient had recurrent spinal cord hydatid cysts. Patient was prescribed albendazole

400 mg twice a day and then underwent surgical resection of the hydatid cyst with T-12 to L-4 laminectomy. Following surgery, patient's symptoms improved gradually after the surgery and with regular physiotherapy sessions during the follow-up period. In addition to the limb weakness, the patient recovered control of urination and defecation with time.

Keywords: Hydatid Cyst; Spine; Magnetic Resonance Imaging; Surgery

INTRODUCTION

Hydatid cyst (hydatidosis) is referred to the infection of the human body by a tapeworm cestode (*Echinococcus granulosus*) and is endemic in countries with warm climate. The life cycle of the worm involves animals but may lead to human infection through the ingestion of tapeworm eggs embedded in the infective animal products. Although the cysts can develop anywhere in the human body, hydatid cyst occurs mainly in the liver (80-90%), lungs and the spleen [1-5]. The incidence of musculoskeletal hydatidosis has been reported to be less than 2.5%. Prevalent sites of musculoskeletal hydatid cyst based on decreasing frequency include pelvis, sacrum, long bone metaphyses, skull, spine and the ribs [6-9]. Spinal hydatid cyst is very rare (accounting for less than 1% of all the hydatidosis) [10] and when diagnosed radiologically by magnetic resonance imaging (MRI), it is usually an absolute indication for surgery. Between different types of spinal hydatid cysts, intradural lesions are the rarest [9]. Because of thin hydatid cyst wall and difficulty in removal of spinal hydatid cyst due to narrow spinal space, cyst ruptures are common often resulting in multiple cyst recurrence [11-12]. Therefore, spinal hydatidosis recurrence is very common and, based on literature report, varies

between 30 to 100%. Its mortality rate has been reported as between 3% and more than 50% on average of 5 years after starting of the disease symptoms [13-17]. We report a case of recurrent spinal hydatid cyst in a 35-year-old man with symptoms of paraparesis that recurred after the first surgical resection of the primary spinal hydatid cyst 5 years earlier.

CASE REPORT

A 35-year-old man from Malayer (a small town in Iran), with known history of surgically resected spinal cord hydatid cyst, presented to the neurosurgical ward complaining of weakness in the lower limbs. About 5 years ago, the patient had presented to another hospital for similar symptoms (lower limbs weakness), was diagnosed with spinal hydatosis, and the cyst was surgically removed.

On this presentation, his symptoms had begun about 40 days earlier with bilateral weakness of lower extremities leading to complete paresis and an atonic bladder. Patient also developed fecal incontinence. Past history was significant only for smoking over last 10 years. Being from a traditional family in Malayer, he had been a shepherd and a farmer throughout his life with history of contact with sheep and dogs. On neurological examination, muscles in the lower

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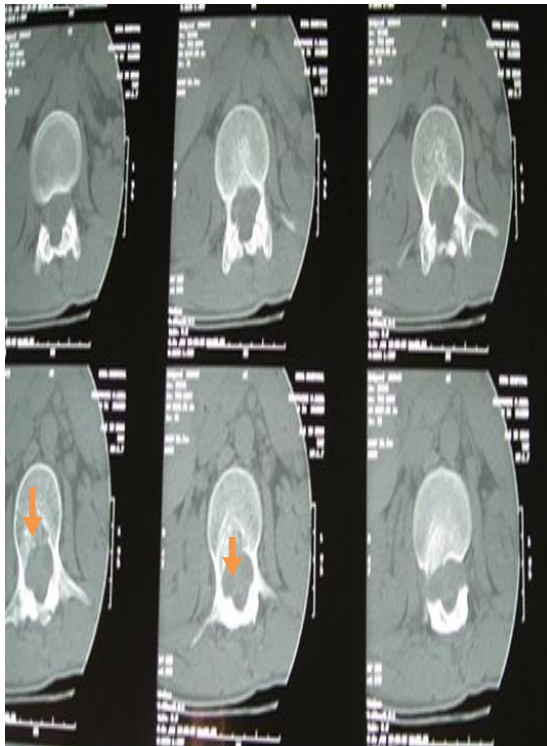
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Figure 1: T1W MRI showing multiple hypo-intense lesions in thoracolumbar spinal cord and vertebral column from T12 to L4



Figure 2: Lumbar CT scan shows hyper-intense lesions in thoracolumbar spinal cord and vertebral column from T12 to L4



limbs were atrophied, power in both lower limbs was decreased to 1/5 with loss of position sense in his left foot, but not the right foot. Patellar and plantar reflexes were absent in both feet. All laboratory tests were negative including tests for hepatitis, human immunodeficiency virus (HIV), and other infectious diseases. Serological tests were also negative for anti-Echinococcus granulosus antibody. Chest X-ray, abdomen and pelvic sonography and computed tomography (CT) scan revealed no lesions or abnormality. Spinal region MRI showed multiple low intense lesions in T-1 (Figure 1) and high intense lesions in T-2 within the vertebral column and spinal cord with involvement of the intradural and extradural portions. Furthermore, lumbar CT scan also shows hyper-intense lesions in thoracolumbar spinal cord (Figure 2). The lesions were distributed from T-12 to L-4 segment and appeared to be a recurrence of the earlier hydatid cyst resected 5 years ago.

Albendazole 400 mg twice a day was prescribed following consultation with infectious disease service. Patient underwent surgical resection of the hydatid cyst through T-12 to L-4 laminectomy. Performing the surgery of hydatidosis, dura matter was opened and all the floating cysts were removed.

Physiotherapy was started after the first postoperative day, the patient's symptoms improved gradually after the surgery and with regular physiotherapy sessions during the follow-up period. In addition to the limb weakness, the patient regained control of urination and defecation with time. Other family members were also screened for the disease with serological evaluation for anti-Echinococcus antibodies.

DISCUSSION

Hydatid cyst is a disease caused by two forms of Echinococcus; the common type is Echinococcus granulosus and the other parasite is Echinococcus multilocularis [18]. In 1708, Bildoo reported hydatosis of bone affecting the humerus [19]. The first case of spinal hydatidosis was reported in 1807 by Chaussier [20] and Reydellet reported the first surgical excision of the spinal hydatid cysts in 1819 [21].

Initially, hydatidosis involves the soft tissue and then spreads to the bones. Although it is very rare that the cyst develop within the spinal cord, the growth of the cyst is responsible for the neurological deficit and other symptoms [22]. Braithwaite and Lees classified these lesions in 5

types: 1) primary intramedullary hydatid cyst; 2) intradural extramedullary hydatid cyst; 3) extradural intraspinal hydatid cyst; 4) hydatid disease of the vertebrae; and 5) paravertebral hydatid disease; of these the first three types are uncommon [23]. The deficit may be a result of direct invasion of the spinal cord or pressure on the nerve roots or even can occur due to the collapse of the fractured vertebral bodies which had been damaged by hydatidosis [6, 24-25]. Recurrence is the major problem of surgical management of spinal canal hydatidosis with some studies reporting the recurrence rate of 30-100% [13]. However, with improved imaging modalities and removal of cysts and use of anti helminthic drugs, the recurrence rate has decreased with a consequent increase in life expectancy [18]. The best method for follow up of hydatidosis and recurrence diagnosis is through MRI [26].

The growth and dissemination of hydatid cyst in thoracolumbar spinal cord within 5 years after the first excision was responsible for the clinical presentations in our patient.

The diagnosis of hydatid cyst was made based on clinical presentation, history of exposure to a potential source of infection, personal history of hydatid cysts, and radiologic imaging, with final confirmation by histopathological studies. Hydatid cyst has no special features on MRI. In endemic countries, some MRI features may suggest an infection; these features consist of: dense calcification rim around a lytic lesion, a water like signal for daughter cysts, a muscle like one for the parent cyst in T1W with multiloculation, and hyper intensity of parent and daughter cysts on a T2W with the appearance of rose or wheel [27].

Albendazole 400 mg twice a day for 28 days has been recommended pre-operatively to reduce the internal pressure of the cyst and to avoid hydatidosis recurrence; albendazole may be repeated after the surgery for one year to assure prevention of the relapse [28].

The initial treatment of choice is surgical decompression by laminectomy and debridement of paravertebral lesions and removal of the entire cysts. Although irrigation of the surgical site with hypertonic saline or cetrinide may have a scolical effect, inadvertent dural opening may lead to the recurrence of hydatid cyst [3, 29]. The treatment for recurrent cyst is repeated extensive excision if applicable. Although recurrence of spinal hydatid cysts occurs in some cases, it seems that extensive surgical resection and proper medical treatment, with effective agents is

efficacious to reduce the chance of recurrence.

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