

Case Report

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Spinal Hydatid Disease Mimics Malignancy: Lack of Correct Diagnosis of Spinal Infection for Several Years

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Abstract

Summary: Hydatid disease or Cystic Echinococcosis (CE) is a zoonosis caused by *Echinococcus granulosus* to which human can become an incidental intermediate host. Almost any organ can be affected, however spinal CE is relatively rare. We report a case of spinal CE mimicking malignancy with delay of diagnosis for several years. The patient had corpectomy and spondylodesis surgery for major destructive processes and biopsies came back negative for malignancy and infection. After two years without symptoms, the disease relapsed rapidly with major paresis. On suspicion, serologic analysis confirmed CE diagnosis. Anthelmintic treatment was instituted and a secondary surgery was performed by 360-degree approach aimed at hydatid cyst-removal, spinal canal decompression and prevention of further progression. Two-and-a-half years later the disease remained stable. CE and other infestations can cause severe disease-patterns even in non-endemic areas. Suspicion must be raised when histopathological and microbiological findings are not in accordance with the clinical presentation and the general condition of the patient.

Keywords: *Echinococcus granulosus*; Hydatid cyst; Spinal echinococcosis; Spinal infestation

Introduction

Hydatid cyst disease in human also known as cystic echinococcosis (CE) is well described through history [1-3]. CE is a zoonosis usually caused by the larval stage of the tapeworm *Echinococcus granulosus*. Domestic dogs and other canine are the definitive host, while man can replace the usual intermediate host, the sheep, goat and cattle [3,4]. The ova are shed in the feces of the host animal and ingested by the intermediate host. After the intestinal passage, they develop into the larva stage and eventually enter the portal vein circulation and get trapped. The predominant sites for the infestation in human are liver (up to 70%) and lungs (up to 20%) where cysts are formed. However, any part of the body can be affected [5]. The tapeworm is widespread, most common in tempered zones where sheep are raised, mainly South America,

Middle East, Southeast Asia, China, north and east Africa, Russia and the Mediterranean areas which include Greece and the former Yugoslavia [1,4,5]. The aim of this article is to report a case of disseminated CE mimicking spinal malignancy in which correct diagnosis was delayed several years.

Case Presentation

A 64-years-old male native from the rural countryside of the Balkans. When he was in his 20s, he was operated at a local hospital in the former Yugoslavia for lumbar radiculopathy and an acquired drop-foot. The right sided drop-foot persisted after surgery. He immigrated to Denmark in the early 1990s. In 2013, at the age of 60 years, he sustained a minor fall. Severe back pain and radicular pain in both legs progressed rapidly over a few days. An MRI revealed spondylodiscitis L4-L5 with abscess in the vertebral bodies and in the surrounding soft tissue (Figure 1).

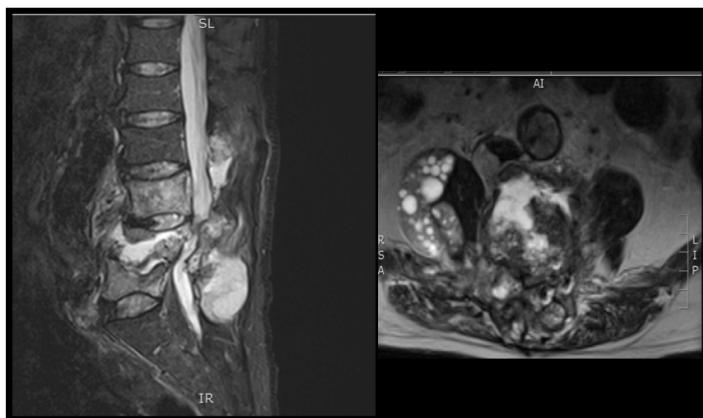


Figure 1: Pre-operative MRI of the sagittal plane (left) and axial plane (right) of the lumbar region showing destruction of the L4 vertebra.

A slight elevation of C-Reactive Protein (CRP) level and eosinophil leucocyte count were the only biochemical markers of infection, and he presented no other clinical signs of an ongoing infection. L3-L5 Corpectomy and L1-S1 spondylodesis was performed for major destructive processes. Surprisingly, the intraoperative biopsies were found negative for both malignancy and infection including TB and mycosis.

After being asymptomatic for two years he experienced a relapse of low back pain and radicular pain. A rapid progressing gait impairment with major paresis of the right leg developed to an extend that he had to manually aid limb motion. His general condition was rapidly deteriorating. The physical examination also revealed a large fluctuating migrating hump at the surgical site (Figure 2).

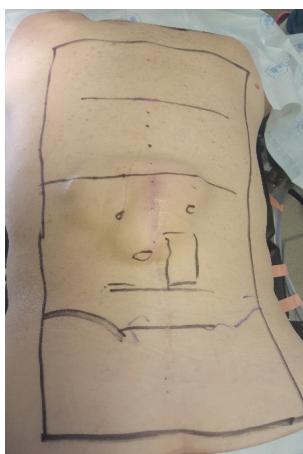


Figure 2: Clinical presentation of the large migrating fluctuating hump before the second operation.

Laboratory blood analysis were largely normal, again except for slight elevations of CRP level and eosinophil leucocyte count. Repeated MRI and a PET-CT was conducted, showing multiple cystic tumors in the area of the lumbar spine. The cystic tumors had increased in both size and numbers and spread to lung, liver, retro-peritoneum and the abdominal cavity (Figure 3). Especially the tissue surrounding the abdominal aorta was affected, causing an aortic aneurism. Open biopsy in the lumbar region on suspicion of malignancy or infection was again found to be negative. On suspicion of CE, serologic analysis confirmed *Echinococcus granulosus* helminth infestation.

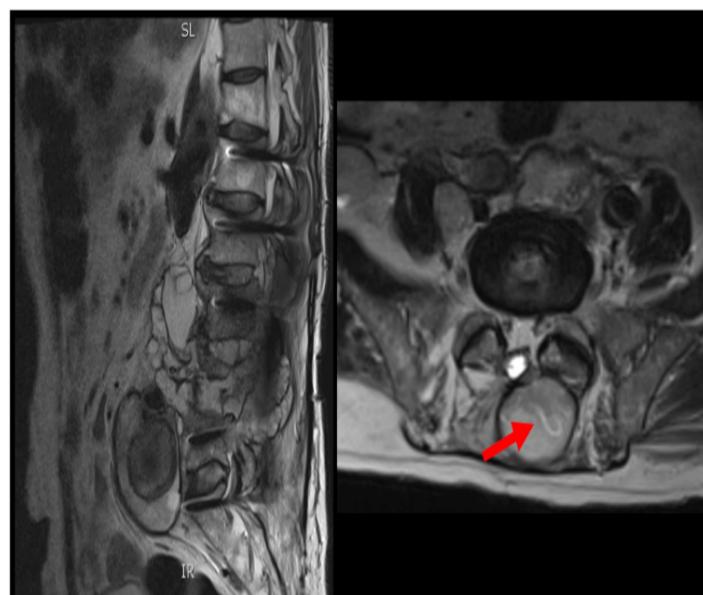


Figure 3: Repeated MRI before open biopsy and second surgery, sagittal plane (left) and axial plane (right), and a serpentine structure (arrow) inside a cyst presumably representing a collapsed cyst membrane.

Anthelmintic albendazole treatment was instituted and by combined anterior and posterior approach a secondary surgery aimed at removing hydatid cysts surrounding the lumbar spine and to prevent further progression of lower limb symptoms was performed Figures 4,5. Albendazole treatment was continued (perhaps life-long treatment) and Praziquantel was added to the anthelmintic chemotherapy for one year. Despite preventive measures, the patient experienced severe complications during the first months that followed: pulmonary embolism and cardiac arrest but he was successfully resuscitated without sequelae. Later, his aortic aneurism ruptured which caused intestinal ischemia requiring resection and ileostomy. Two-and-a-half years after CE diagnosis and institution of anthelmintic treatment the disease remained stable.

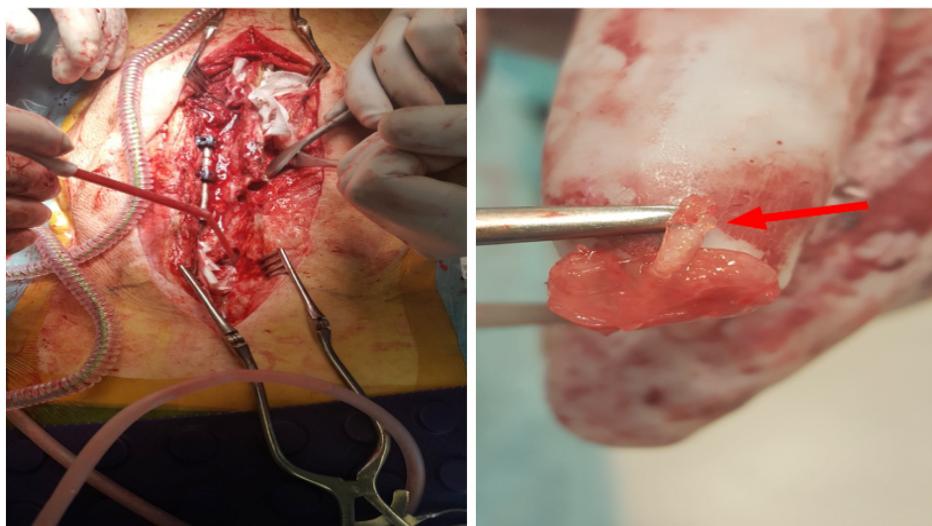


Figure 4: Intraoperative images of posterior procedure for instrumented stabilization of the spine and cyst removal (left). Excised hydatid cyst cut open (right), presenting a serpentine-like structure (arrow).

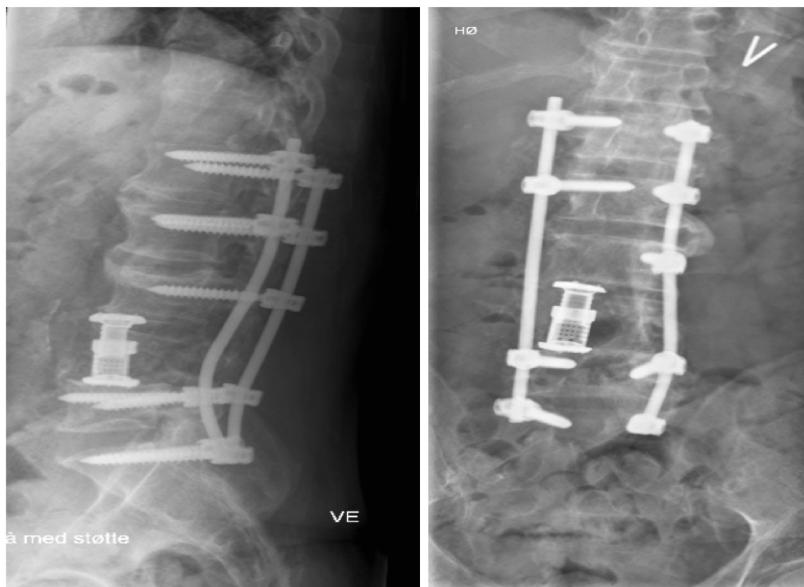


Figure 5: Radiographic status of instrumentation after the second surgery, in sagittal plane (left) and frontal plane (right).

Discussion

CE is uncommon in Scandinavia and diagnosed cases usually originates from high-incidence countries. Skeletal involvement including spinal CE is rare (0.5-4% of CE cases) [1,2]. Spinal CE can be classified into 5 radiological types (Dew/ Braithwaite & Lees classification [6]): intramedullary, intradural, extradural, cyst of the vertebra and paravertebral lesion [6]. This case of spinal CE is classified as a combination of the later three subtypes and was aggravated by concurrent cysts to lung and liver.

CE can be fatal, Belhasan-Garcia, et al. followed 567 mixed CE patients in Spain and found 11 deaths directly related CE [7]. Spinal CE with paraplegia can be a truly lethal disease: Karray, et al. reported a mortality of 6 out of 13 patients (46%) but other reported mortality rates ranging from 14-56% in direct relation to CE [8] and Mills et al reported an average survival length of 5 years after onset of paraplegia in spinal CE [2]. Combined radical surgery and anthelmintic systemic drug treatment with benzimidazole compounds (albendazole for osseous involvement) is the recognized standard [9-11]. Applying intra-operative local

chemotherapy seems logical, but the agent has to be left in contact with the cyst for at least 15 minutes to be effective [11] which may prove difficult in some cases. Risk-effectiveness assessments have deemed formalin obsolete for that purpose, but 90% ethanol, 20% hypertonic saline or 0.5% cetrimide solution can be used. PAIR procedure can be applied as an alternative: first ultrasound guided percutaneous puncture and aspiration from the cyst, then injection of a protoscolicidal compound (e.g. 90% ethanol or 20% hypertonic saline) for a minimum duration of 15 minutes followed by re-aspiration [11]. Complete cyst excision is often difficult in the spine due to invasiveness and risk of cyst rupture. Cyst rupture can cause spreading of the disease, recurrence and risk of an anaphylactic reaction [9]. In this case radical excision of all the paravertebral cysts was impossible. We applied no local anthelmintic treatment, but a successful spinal canal decompression was archived together with improved mobility, reduced radiculopathy and improved general health status post-op and in follow-up. After the surgery the patient regained ambulation and the radicular symptoms were reduced.

Conclusion

This case is a rare presentation of hydatid disease or cystic echinococcosis in which correct diagnosis was delayed for several years. Parasite-infections can be the cause of severe disease patterns even in non-endemic areas. This case report highlights the importance of anamnestic exposures in immigrants and travelers alike. An increased suspicion must be raised when the histopathological and microbiological findings are not in accordance with the clinical presentation and the general condition of the patient. Appropriate paraclinical tests (e.g. specific Ig or PCR analysis) should be performed on suspicion to obtain correct diagnosis to ensure timely diagnosis and proper treatment.

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