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J Case Rep Images Infect Dis 2024;7(2):1–7.
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CASE REPORT PEER REVIEWED | OPEN ACCESS

Spinal hydatid disease: A case report

Ketevan Tsanava, Elene Shengelia, Lia Trapaidze, Lali Khurtsia, Ahmed Abdelkader, Natali Shulaia

ABSTRACT

Introduction: The parasitic tapeworm *Echinococcus granulosus* causes cystic echinococcosis, also known as hydatidosis. It is a serious medical condition that can show up in a lot of different ways. Although spinal involvement occurs in about 45% of bone echinococcosis cases, it represents only 0.5–4% of all echinococcosis cases. The disease can spread to the spine through hematogenous dissemination, direct invasion, or cerebrospinal fluid seeding from ruptured cysts. While the liver and lungs are the most commonly affected organs, spinal involvement can lead to severe complications, including radiculopathy, motor deficits, and paraparesis. Imaging, such as computed tomography (CT) and (MRI), typically makes the diagnosis by revealing cystic and osteolytic lesions.

Case Report: The patient is a 51-year-old male from the Kakheti region (a region in Georgia) who presented with progressive generalized weakness, nausea, vomiting, and lumbar pain. He had a history of spinal echinococcosis, diagnosed 15 years ago, but was unable to complete antiprotozoal treatment due to intolerance to albendazole. Over the years, his condition deteriorated, leading to lower paraplegia and urinary incontinence, necessitating a suprapubic cystostomy. Imaging revealed extensive septated cystic lesions in Th4-S1, which spread to the ribs and subcutaneous tissues. Despite neurosurgical intervention to reduce the cyst burden, the infestation persisted. Multiple hospitalizations were required for complications, such as urinary tract infections and pneumonia. Joint supervision managed the patient's condition, but he continued to experience spherocyte discharge from numerous fistulas.

Conclusion: This case underscores the severe and chronic nature of spinal echinococcal infection, particularly when complete antiprotozoal treatment is not feasible. The patient did not experience severe neurological complications despite the extensive disease burden and the development of multiple vertebral-cutaneous fistulas, possibly due to the continuous drainage of spherocytes. This unusual clinical course highlights the importance of comprehensive management and the potential impact of innovative approaches on managing complex parasitic infections.

Keywords: Hydatid spine disease, Spinal echinococcosis, Spinal hydatidosis, Vertebral-cutaneous fistula

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Received: 03 April 2024
Accepted: 17 June 2024
Published: 30 July 2024

How to cite this article

Tsanava K, Shengelia E, Trapaidze L, Khurtsia L, Abdelkader A, Shulaia N. Spinal hydatid disease: A case report. J Case Rep Images Infect Dis 2024;7(2):1–7.

Article ID: 100014Z16KT2024

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doi: 10.5348/100014Z16KT2024CR

INTRODUCTION

Due to the fact that they are frequently interrupted in the parasitic infection cycle, humans are inadvertent hosts.

Spinal hydatid disease can occur through three routes: hematogenous dissemination to spinal structures

(the most frequent route), direct invasion from extraspinal structures, or cerebrospinal fluid seeding from ruptured cysts of cerebral hydatid disease.

Seventy percent of cystic lesions identified in human subjects are located in the liver, whereas the remaining 20% occur in the lungs; affecting the central nervous system in only 3% of cases (Figure 1). Virtually any organ or tissue can be impacted. Due to the resulting effects, the vertebral column, which is implicated in 50% of the cases (0.5–4%), is particularly susceptible to complications [1–3].

Clinical manifestations may include radiculopathy, motor deficits/paraparesis, sensory disturbances, acute to chronic back pain, and sphincter involvement, contingent upon the site of the cysts. On vertebral body CT scans, multiple cysts, and osteolytic expansile lesions are frequently identified [4, 5].

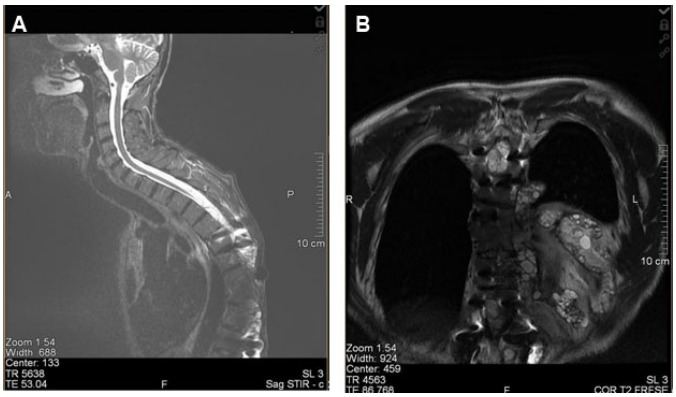

Figure 1: (A and B) MRI images from 15.12.2021. Septated cystic lesions can be observed starting from Th4 level continuing throughout the thoracic spine. Visualization is limited due to metallic implants. Extensive perifocal infiltration can be observed to the left of the spine at Th7 level on coronal plane infiltrating paravertebral soft tissue.

CASE REPORT

A 51-year-old male patient from the Kakheti region was admitted to the hospital with progressive, generalized weakness, nausea-vomiting, and pain in the lumbar and back areas.

According to the patient, the weakness and the above-mentioned symptoms have been progressing for about the last fifteen years. The conventional treatment of radiculopathy and polyneuropathy was not successful.

His social history was significant, as he had been a stock farmer since childhood. Approximately fifteen years ago, he was diagnosed with echinococcosis of the spinal cord and hydatid disease. According to the patient, for the past 15 years, he has been under the supervision of a parasitologist, but the clinical presentation and current state of the condition proved otherwise. Approximately 15 years ago, after the diagnosis was confirmed, the course of treatment with albendazole was initiated; however, due to the patient's intolerance of the drug, the treatment course could not be completed on multiple occasions. Hence, a full course of antiprotozoal treatment has never been accomplished with this patient.

Due to inadequate treatment during the last seven years, the disease progressed, and the patient developed lower paraplegia and urinary incontinence, which led to suprapubic cystostomy insertion.

Later in 2021, the patient was admitted to our hospital due to progressive generalized weakness and constant nausea and vomiting. Both clinical and radiological evaluations in our department concluded that the disease has not only spread to the spinal cord but also to the ribs (Figure 2).

It is important to note that the patient developed multiple fistulas that connected vertebrae to skin in the spinal region over several years of illness.

To alleviate the symptoms, the patient underwent neurosurgical intervention, which aimed to clear the bones from echinococcus. However, the advanced state of the infestation interfered with the procedure. As a result, this intervention somewhat reduced the number of larvae in the spinal cord but could not clear the area entirely.

The presence of *E. granulosus* was confirmed by the parasitological examination of the lesions. IgG tests for *E. granulosus* were continued to be positive >124.

It was evident that solely relying on the invasive procedure without combining it with antiprotozoal medications was insufficient to achieve the goal. However, as already stated, the patient had previously expressed intolerance against antiprotozoal medications. With the consent of the patient, we attempted to once again start antiprotozoal treatment with albendazole at a dose of 10–15 mg/kg/day, but after two to three days of administration, nausea, vomiting, severe abdominal

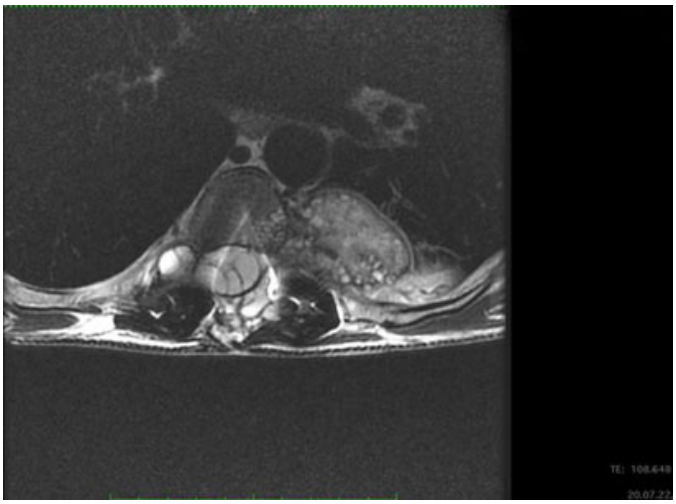

Figure 2: At the level of Th7-L1 in the left paravertebral soft tissues, spreading to the ribs in the subcutaneous soft tissues, the dimensions of the existing masses are increased by 5–10 mm, the structure is very solid, contrast involvement of the substance is peripheral, of moderate intensity, the degree of perifocal infiltration is increased. At the level of the 11th and 12th thoracic vertebrae, on the left paravertebral and ring line, a subcutaneous cyst-tissue mass increased (transverse size 3,771.8 cm).

pain, and dizziness manifested. As a result, the patient refused to take the medicine further.

The advanced state of the spinal cord-cutaneous fistulas resulted in the outflow of spherocytes on the skin surface. It was estimated that within an hour, an average of 10–15 spherocytes appeared on the surface of the skin, and at least 300 echinococcal spherocytes would be removed from the body per 24 hours (Figure 1A and B).

The patient was discharged under the joint outpatient supervision of internists, parasitologists, and neurosurgeons.

In the past three years, the patient has been hospitalized several times in our clinic due to severe urinary tract infection, acute pneumonia, post-renal obstruction of the urinary tract, and acute kidney failure.

The last hospitalization took place three months ago due to the infiltration of soft tissues surrounding the fistula in the spinal cord, which obstructed the spinal cord-cutaneous fistula. Opening and drainage of the fistulas resolved the issue.

It is important to acknowledge that, up to the present time, the discharge of spherocytes persists from the numerous fistulas. The rate of discharge is observed to be intermittent, characterized by periodic fluctuations in velocity.

DISCUSSION

Hydatid disease represents a significant concern in the field of public health. While bone involvement is infrequent in human cystic echinococcosis (CE), the spine is affected in roughly 50% of cases. Regardless of progress in pharmacological therapy, surgery, and diagnostic imaging, spinal echinococcosis continues to be associated with significant morbidity, mortality, and disability.

The medical literature indicates that bone echinococcosis is not frequently reported, possibly due to a lack of awareness within the medical community. This lack of awareness often leads to high rates of misdiagnosis, which can have significant financial consequences, negative impacts on treatment outcomes, and complications. According to an Italian study, the annual national financial burden averages around four million euros [6].

The disease does not happen very often because the liver and lungs catch most of the larvae. Skeletal hydatidosis happens when the larval forms that get filtered out of the liver and lungs settle in bone tissue [2]. The features of bony disease are very similar to those of vertebral osteomyelitis. However, bony disease does not typically show signs of osteoporosis or sclerosis, and there is no damage to the vertebral discs. The cysts themselves have a density similar to that of cerebrospinal fluid and very rarely show calcification [7].

Bone hydatid disease is characterized by the absence of identifiable radiological findings. The main objective of CT is to detect vertebral structural damage. It is clear that hydatid cyst imaging is very flexible because these lesions can look like giant cell tumors, metastases of tuberculosis, or they may not enhance contrast, even though they still have the characteristic pattern of cystic lesions. Magnetic resonance imaging is regarded as the foremost imaging modality. The hydatid cyst is mostly made up of a single, thin wall, and the signal intensity of its contents is similar to that of cerebrospinal fluid when seen on an MRI. Diffusion-weighted imaging can provide additional information regarding the lesion, enabling the distinction between abscesses and complicated infected hydatidosis (Figure 3A–C).

The EC diagnosis is challenging due to the lack of typical clinical appearance and image characteristics. Epidemiological risk factors have a fundamental role in facilitating early diagnosis. The course of symptomatic disease might range from acute onset to prolonged clinical courses. Pain is the most typical clinical presentation, followed by pathological fractures and symptoms of spinal cord compression, such as medullary syndrome. This was reflected in our case, where the patient presented no specific symptoms and there was no alteration in the laboratory except for polymerase chain reaction (PCR) increases.

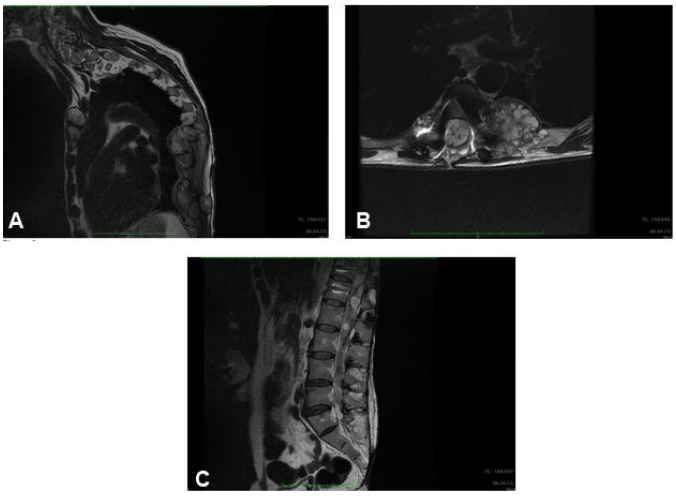

Figure 3: (A–C) Date 06.04.2023 compared to the previous study on 27.10.22. The intraspinal parasitic septated-cystic lesions at the Th4-S1 level are elevated and reflected at the Th3-S2 level. At the level of Th7-L1 in the left paravertebral soft tissues, spreading to the ribs in the subcutaneous soft tissues, as well as on the projection of the costal pleura, the sizes of existing masses increased, the maximum sizes 4.4/3.5 cm (4.1/2.4 cm with previous research), 6.7/2.9 cm (4.8/2.2 cm with previous research), 6.8/2.7 cm (4.9/2.1 cm), the structure is solid, and the degree of perifocal infiltration is increased. Also, at the level of the 11th and 12th thoracic vertebrae, the subcutaneous cystic-tissue mass increased on the left paravertebral and ring line. Compared to the previous study, all mass dimensions increased—negative dynamics.

The efficacy of benzimidazole derivatives (albendazole and mebendazole) as an antihelminthic therapy for visceral CE has been demonstrated. However, recent evidence suggests that treatment response is highly dependent on disease stage and cyst size. So, the data on how well benzimidazoles work to treat bone CE are still not clear, and the question of whether they can stop vertebral CE from happening again is still being discussed.

Albendazole medical therapy has been associated with favorable outcomes for patients diagnosed with inoperable spinal CE or disseminated disease, according to some authors. In a study involving 40 patients with inoperable vertebral hydatidosis who were treated with albendazole, El-Mufti et al. documented a 53% recovery rate at the two-year follow-up. On long-term follow-up, they concluded that additional medical therapy appears to delay the recurrence rate in surgically treated patients with spinal CE [8, 9].

There are reports in the literature of spinal CE lesions that were treated using a percutaneous approach [1]. These reports describe two patients who underwent percutaneous CT-guided treatment using the PAIR approach, which involves puncturing the cyst, aspirating the cyst fluid, injecting a scolicidal agent, and aspirating the cyst content again.

Complications

Cystic echinococcosis, also known as hydatid disease, is a parasitic disease caused by the larval form of the tapeworm (*E. granulosus*). The World Health Organization (WHO) lists it as one of the most neglected and geographically widespread parasitic diseases because it occurs on every continent except Antarctica [10].

The lifecycle of *E. granulosus* involves two hosts: one intermediate and one final. Dogs are commonly the final hosts, as the adult larvae stick to their small intestinal mucosa and the eggs are excreted with their feces. The intermediate hosts are humans and herbivores, such as cattle, sheep, goats, camels, horses, and pigs. The eggs hatch inside the intermediate host's body and can reach various sites through the circulatory system [11]. Musculoskeletal involvement is a rare occurrence, with an incidence rate of 0.5–4% in all cases of CE [4]. Half of all cases of CE occur in the spine, although it can parasitize almost any bone in the body [4]. The incidence in other bones is lower [12].

Common complications of hydatid cysts include cyst rupture, superinfection, and secondary bacterial infection [13]. Hydatid cyst disease affecting the central nervous system can present in various ways, depending on whether it affects the brain or the spinal cord. The condition is mostly found in children and usually affects the white matter of the brain. Treatment usually involves surgery, and complications depend on factors such as the location, size, and number of cysts present, as well as the level of contamination. The most common complication is the rupture of the cysts, which can lead to severe inflammation or anaphylactic response.

On the other hand, vertebral lesions can be invasive and cause neurological symptoms due to compression. Common symptoms include radicular pain and motor deficits, with up to half of patients experiencing paraparesis. This paper focuses on the natural history and complications that can arise when treating cerebral and vertebral hydatid cysts and discusses their clinical management. Treatment usually involves surgical excision and decompression, along with oral albendazole. After three years of follow-up, symptoms usually resolve, and the patient remains stable [14].

All patients in this study had either partial or complete lesions in their spinal cord, leading to recurrences in all cases and death in 18% of the cases. Infections in the spine can cause severe disability or even death [15]. Patients with non-spinal bone CE often present with nonspecific symptoms, such as pain and pathological fractures [16].

A patient's clinical history, along with laboratory, imaging, and serological tests, are essential in diagnosing the disease. Currently, the preferred clinical treatment involves radical surgical resection combined with chemotherapy. However, there is a high postsurgical recurrence rate of up to 40% [17].

Helminths or protozoa can get into the central nervous system (CNS) as adults or as larvae. This can cause neurological problems like meningitis, encephalitis, ventriculitis, myelitis, ischemic stroke, cerebral bleeding, venous thrombosis, or cerebral abscess. These conditions can manifest in the form of headaches, epilepsy, cognitive decline, weakness, confusion, impaired consciousness, coma, or focal neurological deficits [18].

The infiltrative nature of certain diseases can make it challenging to fully eliminate all parasitic lesions, which is necessary for achieving a long-term positive outcome. In cases where there are cystic lesions that erode bone and exert pressure on the spine, a spinal hydatid cyst should be considered as a potential diagnosis. By identifying this condition, patients can receive the best possible care, recover quickly, and resume their daily activities with confidence.

CONCLUSION

We believe that the case we are presenting highlights the severity of the spinal echinococcal infection. This echinococcal infection, which forms transcutaneous fistulas, is an extremely rare phenomenon. Even though the patient has virtually never undergone a complete course of treatment, during 15 years, no complications such as meningitis, encephalitis, ventriculitis, myelitis, ischemic stroke, hemorrhage, venous thrombosis or cerebral abscess, epilepsy, cognitive impairment, decreased consciousness, confusion, or coma, have been detected.

The patient was admitted to our clinic multiple times, and on each occasion, echinococcal, bacterial, and/or viral infectious diseases of the nervous system were never the reason for hospitalization.

Multiple vertebral-cutaneous fistulas on the body allow spherocytes (echinococcal larvae) to drain continuously onto the dorsal skin. We believe that this might be the reason why life-threatening neurological complications were not observed in this patient.

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Author Contributions

Ketevan Tsanava – Acquisition of data, Drafting the work, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Elene Shengelia – Conception of the work, Design of the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Lia Trapaidze – Acquisition of data, Drafting the work, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Lali Khurtsia – Conception of the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Ahmed Abdelkader – Conception of the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Natali Shulaia – Design of the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

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

Source of Support
None.

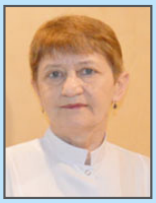
Consent Statement
Written informed consent was obtained from the patient for publication of this article.

Conflict of Interest
Authors declare no conflict of interest.

Data Availability
All relevant data are within the paper and its Supporting Information files.

ABOUT THE AUTHORS

Article citation: Tsanava K, Shengelia E, Trapaidze L, Khurtsia L, Abdelkader A, Shulaia N. Spinal hydatid disease: A case report. J Case Rep Images Infect Dis 2024;7(2):1–7.



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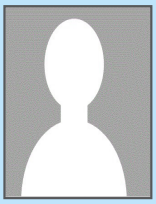

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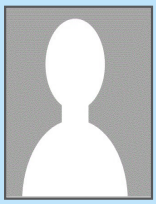
Ahmed Saleh Ahmed Abdelkader New Vision University in 2024 and is eager to contribute to the field of medicine as both a clinician and researcher. His interests lie in neurosurgery and oncology, and he is particularly motivated by the potential to bridge the gap between these disciplines to improve patient care. Dr. Ahmed has already begun his research journey by publishing papers in both neurosurgery and internal medicine, demonstrating his commitment to a well-rounded approach to healthcare.



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