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CASE REPORT / ПРИКАЗ БОЛЕСНИКА

Hydatid cyst of the thoracic spine – where can we make a mistake?

Uroš Novaković[1,2], Milan Savić[2,3], Miloš Vasić[1], Valerija Teodosić[1,2], Slaviša Zagorac[1,2]

[1]University Clinical Center of Serbia, Clinic for Orthopedic Surgery and Traumatology, Belgrade, Serbia;
[2]University of Belgrade, Faculty of Medicine, Belgrade, Serbia;
[3]University Clinical Center of Serbia, Clinic of Thoracic surgery, Belgrade, Serbia

SUMMARY

Introduction Spinal hydatid cyst disease, caused by *Echinococcus granulosus*, is a rare yet challenging condition often overlooked in differential diagnoses.

Case outline We present the case of a 41-year-old woman with a spinal hydatid cyst initially misdiagnosed despite persistent thoracic spine pain, night fevers, and a history of chiropractic and physical therapy treatments. Diagnostic confusion persisted despite various examinations, including X-rays, computed tomography scans, and magnetic resonance imaging, with radiological findings initially suggestive of osteomyelitis or neoplastic changes. Ultimately, surgical intervention revealed a hydatid cyst, emphasizing the diagnostic challenges posed by its nonspecific radiographic features. Treatment involved a corpectomy with spinal stabilization and postoperative albendazole therapy. Regular radiographic monitoring showed no recurrence or dissemination of the cyst.

Conclusion Despite the rarity of spinal hydatid cysts, they should be considered in cases of cystic lesions causing spinal compression. Surgery remains the cornerstone of treatment, often combined with long-term chemotherapy. This case underscores the importance of considering uncommon diagnoses in persistent spinal pathology and highlights the complexities of diagnosing and managing spinal hydatid cyst disease.

Keywords: spinal hydatid cyst; *Echinococcus granulosus*; misdiagnosis; surgical treatment; chemotherapy

INTRODUCTION

Infection of the spinal column are still one of the leading problems in diagnosis and treatment around the world. Most common is vertebral osteomyelitis and ranges from 2% to 16.7% of all cases. With an incidence peak from younger od 20 years of age and above 50 years of age and a predominance for male patients (male to female ratio 2:1 and 5:1 respectively). Mortality ranges from 2–4% and the dominant cause is monomicrobial bacterial infections (30–80%), but either a combination of bacterial agents or other microorganism can be the cause of rare spinal infections [1]. Among them, parasite *Echinococcus granulosus* is the cause of the hydatid cyst disease [2]. Humans can contract it as an intermediate host in the hydatid disease biological cycle. They become infected either directly from a dog bite or indirectly via consuming water or food that has been tainted with parasite eggs [3]. Cysts are typically seen in parenchymal organs, like the brain, liver, and lung, but it can be present in every part of the body, from the head to the toe, although the bone involvement is quite uncommon [4]. The incidence of osseous echinococcosis is low (approximately 0.5–4%). In osseous echinococcosis, spinal involvement is the most common form, though rare overall approximately 0.2–1% [5]. The most common spinal location is the thoracic spine (approximately 50%), followed by the lumbosacral region (approximately 29%) and the lumbar spine (approximately 21%) [6]. Spinal hydatid cysts have become a raising problem in recent years. Although vast array of laboratory tests and radiological diagnostic tools are available the spinal hydatid disease are generally overlooked in differential diagnosis possibly due to the rate of occurrence and to medical community lack of awareness [7]. In this case, we present a female patient with spinal hydatid cyst, which has been misdiagnosed.

CASE REPORT

A 41-year-old female presented with a several-month history of pain in her thoracic spine, accompanied by night pain and fever with no neurological impairment. She was physically very active and had no underlying comorbidities. She refused to undergo examination, and went to a chiropractor treatment who performed periarticular infiltrations, but there was no improvement. Subsequently, she initiated private physical therapy independently, without prior medical advice or referral. Despite long-term physical and drug therapy, the pain was still present, she underwent X-ray examination, which were with no pathological finding. Laboratory results revealed a slight elevation in the erythrocyte sedimentation rate. She also underwent psychiatric examination; a

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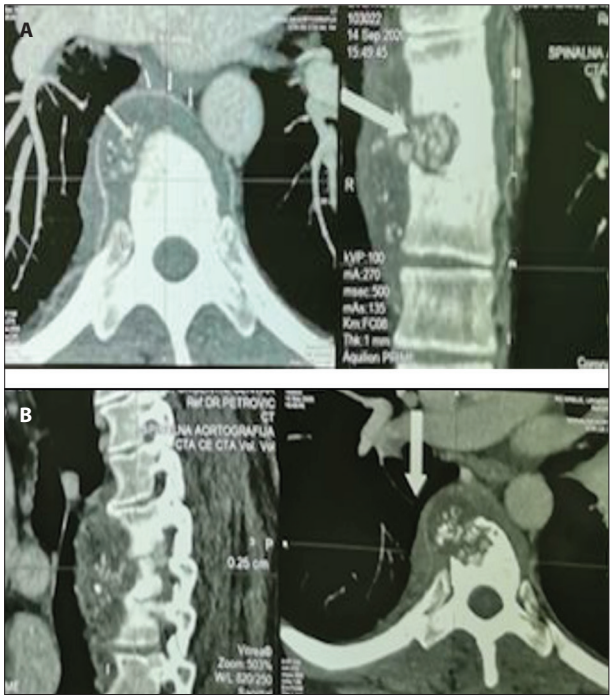
Correspondence to:
Valerija TEODOSIĆ
University Clinical Center of Serbia
Clinic for Orthopedic Surgery and Traumatology
Pasterova 2
11000 Belgrade
Serbia
valerija.teodosic996@gmail.com



Figures 1A and 1B. Preoperative computed tomography scan of thoracic spine: axial and sagittal section of multiple levels of thoracic spine which showed paravertebral and prevertebral change from Th5–Th9 with infiltration and osteolysis of vertebral bodies, disc space and spinal canal, with connection to S6 lung segment

diagnosis of anxiety had been established. Despite drugs and physical therapy, the pain was worsening. Finally, three months after the initial onset of symptoms, the patient underwent a computed tomography (CT) scan, which showed paravertebral and prevertebral change from Th5–Th9 with infiltration and osteolysis of vertebral bodies, disc space and spinal canal, with connection to S6 lung segment. Radiological conclusion was osteomyelitis or lung tumor with lymphadenopathy (T3N1M1a) (Figures 1A and 1B). A nuclear magnetic resonance imaging (MRI) of the thoracic spine was conducted which revealed changes from Th5 to Th8, posterior ribs VI and VII, prevertebral and paravertebral, and in the lung, spread epidural with foramina infiltration and compression on roots Th6 and Th7. The radiological conclusion: tuberculous (TBC) spondylitis or neoplastic change (Figure

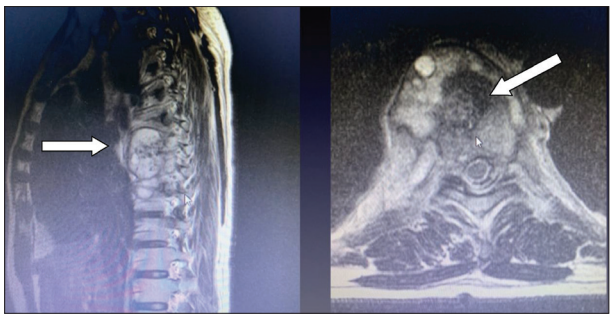


Figure 2. Preoperative nuclear magnetic resonance of thoracic spine: axial and sagittal section of multiple levels of thoracic spine which showed a visible cyst that invades most of several vertebral bodies and the spinal canal, also S6 lung segment

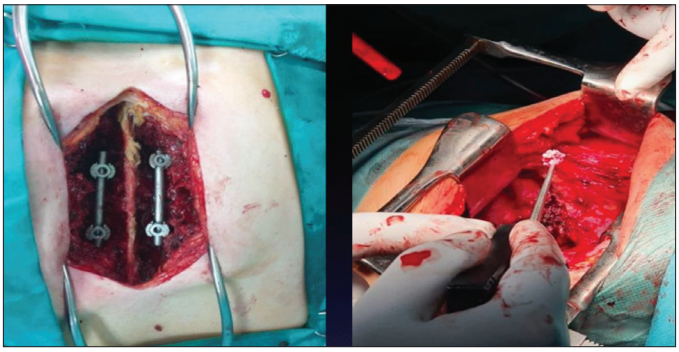


Figure 3. Intraoperative findings: transpedicular stabilization with screws

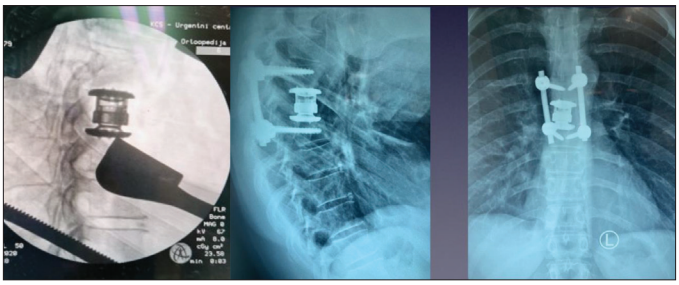


Figure 4. Postoperative findings: X-ray – anteroposterior and lateral view: properly positioned osteosynthesis material

2). She underwent bronchoscopy (with normal findings), QuantiFERON-TB test (the findings were negative) and a Tru-cut biopsy of the lungs which have shown monocyte/histocyte proliferation, possible inflammatory myofibroblastic tumor or pneumonitis. The patient was referred to an oncologist who indicated radiotherapy of the infiltrated segment. However, the patient declined this modality of treatment, and was referred to a spinal surgeon. After a clinical examination and a thorough reviewing existent medical documentation and available radiographic sources, we suggested surgical treatment, open transthoracic approach with a biopsy of the affected region and a corpectomy with an implantation of an expandable cage and subsequently a posterior transpedicular stabilization (Figures 3 and 4). Postoperatively there was no neurological impairment. Biopsy of the lesion has shown a hydatid cyst caused by *Echinococcus granulosus*. Postoperatively, the patient was treated with albendazole for six months and routinely monitored by both a spinal surgeon and an infectious diseases specialist. Follow-up imaging was performed at three, six, 12, and 24 months, with no evidence of cyst recurrence or dissemination.

Ethics: The study was approved by the Ethics Committee of the University of Belgrade Faculty of Medicine (Decision number 1051/3), and carried out in accordance with the Helsinki Declaration and its amendments. This case report analyzes data collected in the period preceding the writing of this article, in accordance with the provisions of the Law on the Protection of Personal Data of the Republic of Serbia. Insight into the patient records in this case report was obtained in accordance with Article 3 of the above-mentioned Law, which necessitates a precise definition of how this data may be used. Informed consent was obtained

from the patient. In keeping with the University Clinical Center of Serbia regulations, the patient consented to participate in any retrospective studies by signing her consent to hospitalization and treatment, as well as to the use of her medical data in this case report.

DISCUSSION

Spinal hydatid cyst illness is a rare and challenging condition to manage. An endemic condition, spinal hydatid cyst disease is typically observed in nations along the Mediterranean and in the Middle East [8]. The most common spinal location is the thoracic spine (approximately 50%) and most patients with thoracic spine echinococcosis had a history of extraspinal cystic echinococcosis, most commonly of the lungs, liver, kidneys, and soft tissues [9]. Our patient had no comorbidities. Hydatid cysts typically extend beyond the vertebral bodies, frequently affecting the intervertebral discs, spinal cord, and posterior elements, with potential growth into the spinal canal [10]. Radiographic diagnosis is challenging because there are no specific findings consistent with spinal echinococcosis; misdiagnosis is common with radiographs only and the lesions can be confused with tumors such as metastases and chondroblastoma, or other infections such as tuberculosis, spinal or paraspinal abscess [11]. In our case, the X-ray were without any radiological features which are pathognomonic for hydatid cyst (zones of multilocular osteolysis) [12]. Typical CT finding is multilocal, round cyst, with erosion of vertebral body and posterior elements. Further, the cyst density can be measured in order to confirm diagnosis, which was not done in our case. MRI is more sensitive than CT and revealed multiple cystic, fluid-filled lesions with internal septations, forming a "bunch of grapes" appearance at multiple spinal levels [13]. Regarding laboratory findings, polymerase chain reactions, Western blots, indirect hemagglutination tests, and serological enzyme-linked immunosorbent assays are utilized for diagnosis [14]. In our case, CT and MRI scans revealed changes in anterior parts of vertebral body, including disc space. Our case was challenging regarding proper diagnosis and surgical approach. Our patient had a paravertebral and prevertebral change from Th5–Th9 with infiltration and osteolysis of vertebral bodies, disc space, and spinal canal, with connection to S6 lung segment. What was confusing for radiologist is involvement of disc space, which is not characteristic for hydatid cyst [15].

The diagnostic procedures that were conducted were bioscopy, QuantiFERON-TB test, and a Tru-cut biopsy of the lungs, as well as a CT scan and a nuclear MRI scan. Biopsy is indicated when other radiological findings are not clear [16]. However, in hydatid cyst, biopsy can lead to the rupture and systematic reaction, hence biopsy should be avoided. After Tru-cut biopsy in our case, the diagnosis of inflammatory pseudo tumor has been established. We had no experience with this kind of tumor and in the literature, we found many names related to this lesion like fibrous xanthoma, plasma cell granuloma, pseudosarcoma, lymphoid hamartoma, myxoid hamartoma, inflammatory myofibrohistiocytic proliferation, benign myofibroblastoma, and most recently, inflammatory myofibroblastic tumor [17]. The treatment is based on two pillars: chemotherapy and surgical treatment. Surgical treatment is the treatment of choice. Among diverse surgical approaches and techniques, posterior laminectomy and fusion is the treatment of choice. However, it can be combined with thoracotomy [18]. According to literature, the main drawbacks regarding the surgical treatment are: systematic allergic reaction, recurrence, and general contraindications [19]. Pharmacologic treatment consists of administration of anti-parasitic drugs such as benzimidazoles (albendazole and mebendazole) [20]. According to the Turgut [21], patients that have a low rate of recurrence are those with combined pharmacological therapy (5% in patients treated with combined modalities, and 32% in those patients who only underwent surgery). We were against radiation therapy and chemotherapy, which was suggested by radiologist and we offered surgical treatment in order to confirm diagnosis, to make complete resection of the lesion and to stabilize the spinal column, which was expected after complete removal of the cyst. The aim of spinal surgery, depending on where the condition is located, is to decompress the spinal cord and, if necessary, to stabilize the spinal column to make up for the stability that was lost during cyst excision. This postulate is related to all spinal pathologies. We performed corpectomy with an implantation of an expandable cage, and a posterior transpedicular stabilization and cyst removal, as we could not perform anterior fixation due to lack of availability of implants. Albendazole can be helpful in preventing or delaying recurrence as well as preventing intraoperative dissemination of the cyst, even though it is well known that it cannot guarantee recovery or prevent recurrence when used alone [3]. This is especially true when used as an auxiliary application in patients who are ineligible for surgery or in conjunction with surgical treatment. Our patient was treated with albendazole (intravenously administered) for six months after the initial surgery. A cysticidal substance (hypertonic 30% saline, cetrimide, or 70–95% ethanol) can be used. During the procedure our patient underwent, we did not use any type of cysticidal substance, only hypertonic saline solution was used as an irrigation method as well as an iodine solution [22]. The risk of recurrence has varied between 30% and 100% [23, 24]. Our patient underwent regular radiological follow-up at three, six, 12, and 24 months post-surgery, with no evidence of cyst recurrence or dissemination. The limitation was that the patient was lost to follow up after two years of initial operation. We are aware that spinal hydatid cyst has been already described in literature and the diagnosis and treatment options are well known and evidence-based. However, our opinion was that this case is interesting due to several peculiarities: wrong patient behavior, wrong working diagnosis, histopathological finding of the biopsy, as well as the type of surgical treatment.

Our case showed that hydatid cyst is still a challenging medical issue, despite available diagnostic tools, due to the various and unspecific clinical and radiological features.

The clinical course leading to diagnosis can be long and difficult. The spinal hydatid cyst should be considered in the differential diagnosis when there are cystic lesions that cause osteolysis and spinal compression. The therapy of choice should always be surgery and long-term chemotherapy.

Conflict of interest: None declared.

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Хидатидна циста торакалне кичме – где можемо погрешити?

Урош Новаковић[1,2], Милан Савић[2,3], Милош Васић[1], Валерија Теодосић[1,2], Славиша Загорац[1,2]

[1]Универзитетски клинички центар Србије, Клиника за ортопедску хирургију и трауматологију, Београд, Србија;
[2]Универзитет у Београду, Медицински факултет, Београд, Србија;
[3]Универзитетски клинички центар Србије, Клиника за торакалну хирургију, Београд, Србија

САЖЕТАК

Увод Цистична ехинококоза (или хидатидна циста) локализована на кичменом стубу, узрокована цестодом *Echinococcus granulosus*, представља ретко али изазовно стање, које се често занемарује у диференцијалној дијагностици.

Приказ случаја Представљамо случај 41-годишње жене са цистичном ехинококозом на кичменом стубу, која је првобитно погрешно дијагностикована упркос упорним боловима у торакалној кичми, ноћној грозници и историји киропрактичне и физикалне терапије. Неизвесност у дијагностиковању болести трајала је упркос разним прегледима, укључујући рендгенске снимке, скенере и магнетну резонанцу, са радиолошким налазима који су првобитно указивали на остеомијелитис или неопластичне промене. На крају, хируршка интервенција је открила хидатидну цисту, наглашавајући дијагностичке изазове које представљају њене неспецифичне радиографске карактеристике. Лечење је било оперативно, урађена је корпектомија са стабилизацијом кичме, а укључивало је и постоперативну терапију албендазолом. Редовно радиографско праћење није показало рецидив или дисеминацију цисте.

Закључак Иако су хидатидне цисте на кичменом стубу ретке, треба их узети у обзир у случајевима цистичних лезија које изазивају компресију кичмене мождине. Хирургија остаје основни начин лечења, често у комбинацији са дуготрајном хемиотерапијом. Овај случај наглашава важност разматрања неуобичајених дијагноза у трајној патологији кичменог стуба, као и сложеност дијагностиковања и лечења болести хидатидне цисте локализоване на кичменом стубу.

Кључне речи: хидатидна циста; кичмени стуб; *Echinococcus granulosus*; погрешна дијагноза; хируршко лечење; хемиотерапија