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Case Report

Spinal cysticercosis: A case report ☆

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ABSTRACT

Spinal cysticercosis is a rare and severe cysticercosis complication. The pathology is from the inflammatory reaction and granuloma formation around the eggs, which cause focal neurological deficits. Because of the rarity of this condition, diagnosis may be delayed and confused with other myelopathies such as neoplasms or myelitis. We report a 42-year-old woman with low back pain and paraplegia. Magnetic resonance imaging showed a lesion in the spinal canal at the level of L4/L5 that was toward the diagnosis of myelitis. The patient underwent an open biopsy, and the result was granulomatosis caused by cysticercosis. The patient was then given an anticysticercosis medication and gradually recovered.

Introduction

Neurocysticercosis is caused by the larval stage of the tapeworm (*Taenia solium*). The disease is common in people who live in developing countries, such as in Latin America, Africa, and Asia, or who migrate from those countries to developed countries such as in Europe and North America [1].

The most common cause of parasitic infections in the central nervous system is cysticercosis [2–4]. On the other hand, cystic lesions of the spinal cord are uncommon. Various studies show that spinal cysticercosis accounts for only 1%-5% of neurocysticercosis [2,3,5,6]. Symptoms of spinal cysticercosis usually progress slowly, but when the cyst degenerates, it causes a granulomatous inflammatory response that can result in clinical manifestations of acute spinal cord compression [2,3].

We present the clinical report of a very rare case of spinal cysticercosis lesion that was diagnosed and treated surgically. We describe this clinical case that was operated on at Viet Duc Hospital in August 2022 and review the literature on the di-

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agnosis and treatment of spinal cord lesions caused by tapeworm larvae.

Case presentation

Medical history

A 42-year-old female patient of Thai ethnicity, working in agriculture, living in Vietnam's northwestern mountains area had experienced low back pain for around 1 year. The pain became more severe around 10 days before, accompanied by weakness in both legs and difficulty in defecation and urination.

Clinical examination

On clinical examination, the patient was found to have motor paresis of the lower extremities (muscle strength 4/5). The sensory disturbances comprised numbness on the back of the leg on both sides along the S1 root, decreased quadriceps tendon and heel tendons reflexes on both sides, and sphincter disturbance.

Imaging examination and laboratory test

Magnetic resonance imaging showed lesion clusters 60 × 8 mm in size at the L4/L5 level of the lumbar spine, isointense on T1W, hypointense on T2W, and irregular enhancement of the border with hyperenhancement of the arachnoid membrane, suggesting inflammatory lesions (Fig. 1).

Although the white blood cell count was normal (9.13 g/L), the eosinophil component was elevated (0.98 g/L). Cerebrospinal fluid analysis revealed increased protein (1.61 g/L) and monocytes (0.019 g/L). Other tests were within normal ranges. The ELISA test for tapeworm was negative.

Surgery

An L4 and L5 decompressive laminectomy was performed. Intraoperatively, the lesion was intradural and measured around 50 × 8 mm, extending from L4 to L5. The lesion caused arachnoid membrane thickening and adhesion to the nerve roots (Fig. 2). The results of an immediate biopsy suggested immune-mediated granulomatous responses against tapeworm larvae. We removed a portion of the lesion while leaving the tissue firmly attached to the nerve roots and performed duraplasty.

Histopathology result

The lesions were caused by the infiltration of inflammatory cells such as lymphocytes and multinucleated giant cells, interspersed with tapeworm larvae (Fig. 3).

Postoperative treatment

Following surgery, the patient was transferred to the National Institute of Malariology, Parasitology, and Entomology for further tapeworm disease treatment.

Discussion

According to the World Health Organization, more than 2.5 million people worldwide are infected with tapeworms [1]. The disease is common in developing countries, particularly in areas where pigs and cattle are raised in unsanitary conditions [1,2]. People in Vietnam's mountainous regions, particularly in the north and northwest, also habitually eat raw food, such as raw fish salad, animal blood soup, and raw pork [7,8]. Certain surveys conducted in Vietnam to study the risk factors associated with human taeniasis and cysticercosis have highlighted significant factors. These factors include the consumption of raw meat and unwashed vegetables, allowing pigs to roam freely, inadequate sanitation practices, absence of hygienic latrines, utilization of human feces as fertilizer, and the absence or insufficiency of meat inspection procedures [9].

T solium undergoes a 2-host lifecycle involving humans and pigs. The adult tapeworm thrives exclusively in human hosts, while both humans and pigs can serve as intermediate hosts that harbor the larval form [4]. *T solium* larvae exist as fluid-filled membrane vesicles containing a tapeworm scolex. Upon ingestion of cyst-contaminated pork, the scolex evaginates and attaches itself to the intestinal wall, transforming into a 2-4 meter-long ribbon-like tapeworm. The gravid proglottids release microscopic fertile eggs, each containing an infective embryo, into the environment via feces. In places with inadequate sanitation and roaming animals, pigs can access human feces containing *T solium* eggs. Upon ingestion, the embryos are released from the egg and actively traverse the intestinal mucosa to the bloodstream, where they are carried to the peripheral tissues, including the central nervous system and form cysticerci [10,11]. The pigs infected with the eggs become intermediate hosts, playing a critical role in the survival of *T solium*. The lifecycle concludes when humans consume undercooked pork infected with cysts [4].

Humans, like pigs, can contract cysticercosis by ingesting *T solium* eggs, which usually occurs through the fecal-oral route from close contact with a tapeworm carrier. The exact method and timing of transmission are unknown [1,4,12]. Cysticercosis infections are concentrated around tapeworm carriers, with person-to-person transmission being more prevalent than previously assumed, particularly in individuals with numerous cysts. This mode of transmission is likely the primary source of human contamination with *T solium* eggs rather than contamination from environmental sources. Although most investigations for *T solium* eggs in water or soil have yielded negative results, the possibility of environmental contamination as a means of infection cannot be entirely ruled out, and its role requires further study [4,1,12,13].

The parasite is known to frequently invade the central nervous system, leading to neurocysticercosis, a diverse clinical condition. Upon entering the central nervous system, the cysticerci maintain their viability and induce minimal inflammatory responses in the surrounding tissues. In this stage, the cysticerci can persist for extended periods, as they are shielded by the blood-brain barrier [7,10,13]. After an indeterminate period, the parasite undergoes degeneration accom-

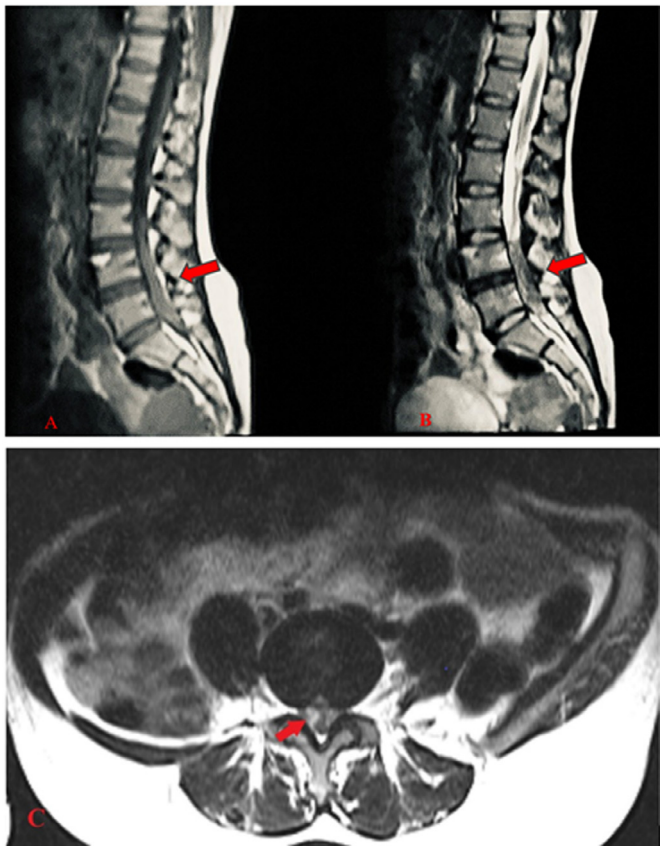

Fig. 1 – (A) Sagittal T1W showing a lesion with isointensity compared to cerebrospinal fluid (arrow). (B) Sagittal T2W showing a hypointense lesion(arrow). (C) Lesion on Axial T2W at the L4/L5 level.

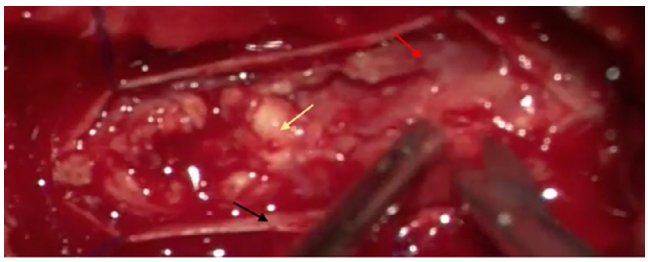

Fig. 2 – Intraoperative lesion. The lesion (yellow arrow) was intradural (dural - black arrow). Arachnoid membrane thickening (red arrow).

panied by inflammatory reactions mediated by the immune system. The presence of cysticerci leads to symptoms through mass effect or the obstruction of cerebrospinal fluid circulation. However, the majority of symptoms observed in neurocysticercosis stem directly from the inflammatory processes associated with cyst degeneration.. These clinical presentations are influenced by individual variations in the number, size, and location of lesions, as well as the host's immune response severity towards the parasites. Hence, the symptoms and signs exhibited in neurocysticercosis are multifaceted and lack specificity [4,13].

In the central nervous system, spinal cord lesions are very rare, accounting for only 1%-5% of cases [2,4]. The disease is most common in people aged 20-45 years, and it is uncommon in children and the elderly [2–4,6]. The 4 locations of injury in the spinal cord are extraspinal, epidural, subdural-extramedullary, and intramedullary [5]. Of these, subdural-extramedullary lesions are the most common [2]. Many authors have proposed 2 main mechanisms for tapeworm larvae transmission into the spinal cord. The first is hematogenous transmission. This frequently results in lesions in the medullary parenchyma and is common in the thoracic medulla, which has a rich blood supply network, explaining why the lesions are more common in the brain than the spinal cord. In the second route, the larval lesions on the brain move along the cerebrospinal fluid in the subarachnoid space to the spinal cord and cause local damage. This path leads to cystic lesions in the subdural-extramedullary region [2,3,6,14].

The primary manifestations of human taeniasis encompassed various symptoms. These symptoms included an irritated anus (reported in 63.7%-80.2% of cases), the passage of proglottids through the anus or with the feces (observed in 82.9%-92.1% of cases), abdominal pain (experienced by 59.7%-62.1% of individuals), digestive disorders (noted in 45.2%-50.0% of cases), sleep disturbances (reported by 35.2%-39.5% of individuals), low blood pressure (observed in 10.4%-12.1% of cases), and the presence of Taenia eggs in stool samples (detected in 18.1%-22.5% of cases) [8,9]. According to Nguyen et al. [8]. Prominent symptoms of human cysticercosis comprised a

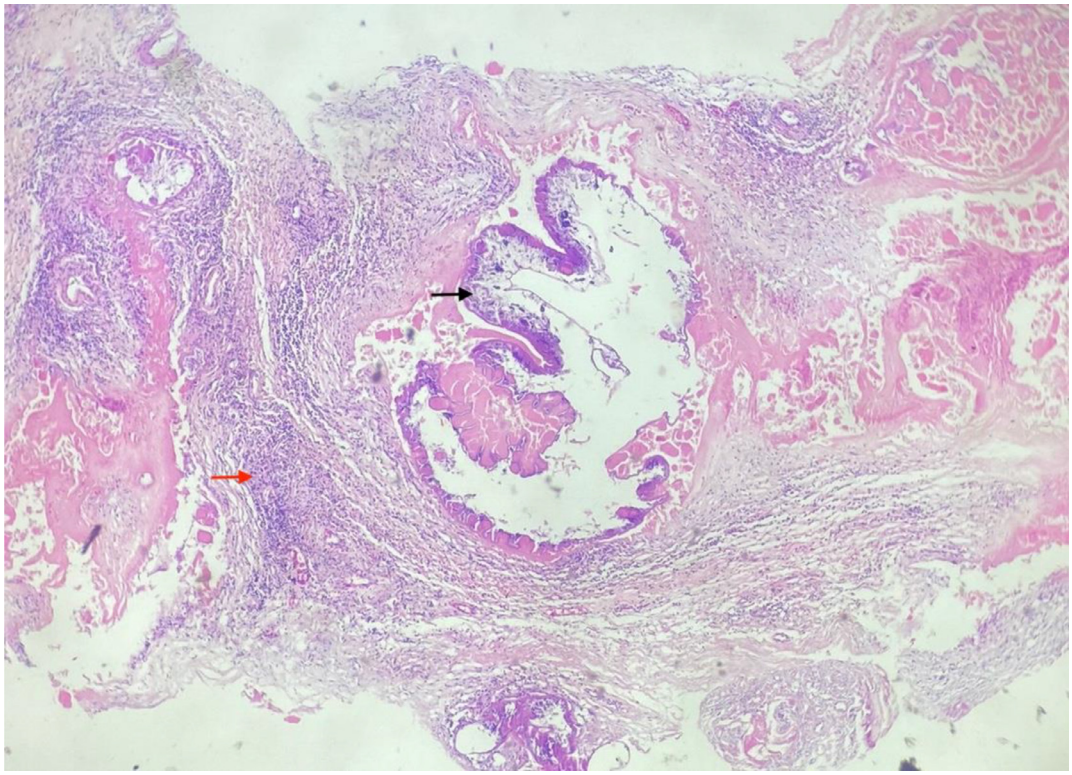

Fig. 3 – Histopathological image revealing tapeworm larvae. Black arrow: tapeworm larvae. Red arrow: infiltration of inflammatory.

range of manifestations. These included headaches, epilepsy, increased muscle activity, memory loss, sleep disturbances, hearing impairments, episodes of nausea/vomiting, paralysis, sensory disturbances, and positive biopsy results in subcutaneous tissues.

The clinical symptoms of spinal cysticercosis typically develop gradually, but they can also progress rapidly in a short period. According to various reports, symptoms may develop between around 1 week and 10 years [5]. Pain, paralysis of the lower extremities, sensory disturbances, dysuria, and decreased sexual ability are common clinical symptoms [2,4–6]. During pathogenesis, cysts can exist in the central nervous system for many years without causing symptoms. However, with cyst degeneration, the body's inflammatory response may cause severe, acute symptoms [2]. According to Isidro et al. [5], 3 mechanisms lead to spinal cord compression and injury. First, an inflammatory response develops as a result of the presence of toxins in the cyst. Second, the mass effect causes spinal cord compression as the hydatid cyst grows. Third, the arachnoid's inflammatory response causes an increase in inflammatory cells and increased fibrous tissue, which causes compression and decreased blood supply to the spinal cord. The spinal cord or nerve root compression is determined by the form and size of the lesion, as well as the location and size of the spinal canal [2,6,8,9].

Histopathology is the gold standard for detecting spinal cord cysts [4]. In practice, however, it is not always possible to biopsy the lesion for histopathology. When a patient exhibits clinical signs of spinal cord compression, magnetic resonance imaging of the spinal cord is recommended. On magnetic resonance, the lesion is a cystic structure in the early stages: hypointensity on T1W, no enhancement, and hyperintensity on T2W, with a thin capsule hypointense on T2W [14]. At this stage, the hyperintensity on T1W of the scolex inside the cavity of the cyst may be visible. This is a typical image with high specificity, but it is rare in clinical practice [2,4,6,14]. As the contents of the cyst gradually transform into a colloidal cyst, the signal on T1W becomes hyperintense, making it difficult to see the scolex [3]. When the cyst degenerates and the cystic structure is broken, this activates the diffuse granulomatous inflammatory response, infiltrates inflammatory cells, and stimulates fibrous proliferation. The lesion image on magnetic resonance imaging is unclear at this stage, with hyperenhancement accompanied by adjacent arachnoid membrane hyperenhancement [14]. At this stage, the lesion image is not specific and is similar to other myelitis lesions, including tuberculous myelitis, granulomatosis, medullary sclerosis, and primary or metastatic tumors [2,6,14]. Live blood analysis is normal or shows elevated eosinophils. Cerebrospinal fluid testing typically reveals increased protein, primarily elevated lymphocytes or eosinophils, and decreased or normal glucose [2,3,5]. Patients in endemic areas should be ELISA tested. This method is highly specific: 98% [4,13]. However, because it is a local disease, the test kit is not always available, and the sensitivity is low, so many cases of tapeworm infection have negative results [2,4].

Sharma et al. [15] reported that surgery had good results in 60% of patients, whereas 25% did not improve and 15%

died. Later studies produced more positive results, such as Mohanty's study, which reported that 75% of patients had good outcomes after surgery [2,3]. With the development of microsurgery instruments and equipment, surgery is becoming a more effective treatment option. Surgery is indicated in patients with spinal tapeworm lesions for 2 reasons. First, this can surgically remove the lesion, release nerve structure compression, improve symptoms, and increase the patient's capacity for recovery. Second, in most cases, a surgical biopsy of the lesion helps confirm the diagnosis and allow treating patients with antiparasitic drugs after surgery [2–4,6]. Albendazole, an effective drug for spinal cysticercosis, was first introduced in 1996. When the diagnosis is confirmed, albendazole is used for pre and postoperative treatment at a dose of 15 mg/kg/d for 4-6 weeks [1,3,5]. Furthermore, corticosteroids should be used concurrently to avoid an excessive inflammatory response when the cysts degenerate, which can cause spinal cord damage [3,5].

Conclusion

In clinical practice, spinal cysticercosis is extremely rare. The clinical symptoms and magnetic resonance imaging features of the disease are commonly nonspecific, making it difficult to diagnose and easily confused with other diseases. As a result, if a patient lives in a high-risk endemic area, additional serological tests should be performed. Surgery is an effective and safe treatment that also aids in the confirmation of the diagnosis in difficult cases.

Author's contributions

Bui HM and Tran D: Case file retrieval and case summary preparation. Bui HM and Nguyen MD: preparation of manuscript and editing. All authors read and approved the final manuscript.

Availability of data and materials

Data and materials used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Ethics approval and consent to participate

Our institution does not require ethical approval for reporting individual cases or case series.

Patient consent

Informed consent for patient information to be published in this article was obtained.

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