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CASE REPORT

Spinal hydatid disease: a pressing issue

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Summary
Hydatid disease is a zoonotic disease caused by the *Echinococcus* tapeworm. The primary hosts include livestock, cats and dogs. Humans can become accidental hosts when ingesting food products or water contaminated by these animals. The most common organs to be affected in humans include the liver and lungs. Bone involvement is rare, accounting for 0.5–3% of all cases. However, when there is spread to bones, the spine is involved in 50% of that subset of cases. This case report highlights an example of spinal cord compression due to partially treated hydatid disease, and the importance of routine follow-up for these patients.
Keywords: hydatid disease, hydatid cysts, spinal hydatid disease

Case report

A 28-year-old female from a semi-urban location in South Africa presented with an uncommon neurological complication as a result of hydatid disease. In 2017, she presented to a district hospital with shortness of breath, a productive cough and constitutional symptoms. She was noted to be GeneXpert negative for *Mycobacterium tuberculosis* but was confirmed to have positive *Echinococcus* serology. A computed tomography (CT) scan revealed that the patient had multiple cystic hydatid lesions in the lung, liver and spleen. HIV testing was negative on this admission. The patient had a lateral thoracotomy to address the disease burden in the thorax and was started on albendazole 400 mg 12 hourly orally. She then had two outpatient follow-ups with cardiothoracic surgery.

Thereafter the patient was lost to follow-up and next presented in 2023 with a similar clinical presentation as her first admission, namely, shortness of breath and constitutional symptoms. At that presentation she tested positive for HIV, with a CD4 count of 1055 cells/µL. She was GeneXpert negative for *Mycobacterium tuberculosis* and had a negative hepatitis screen. A repeat CT scan demonstrated persistence of the hydatid disease in the liver and spleen (Figure 1 - pictures A and B respectively). Over the next year, the patient's HIV disease was well-controlled, with consecutive viral load counts being lower than detectable limits. She was recommenced on albendazole 400 mg 12 hourly orally.

The patient was again lost to follow-up and next presented directly to a tertiary hospital in 2024 with progressively worsening weakness in her lower limbs. A magnetic resonance imaging (MRI) scan demonstrated a loculated cystic lesion anterior to the vertebral bodies of cervical and thoracic spine levels C7-T3 (Figure 2 - picture A). There was marked destruction of the T2 and T3 vertebral bodies, with

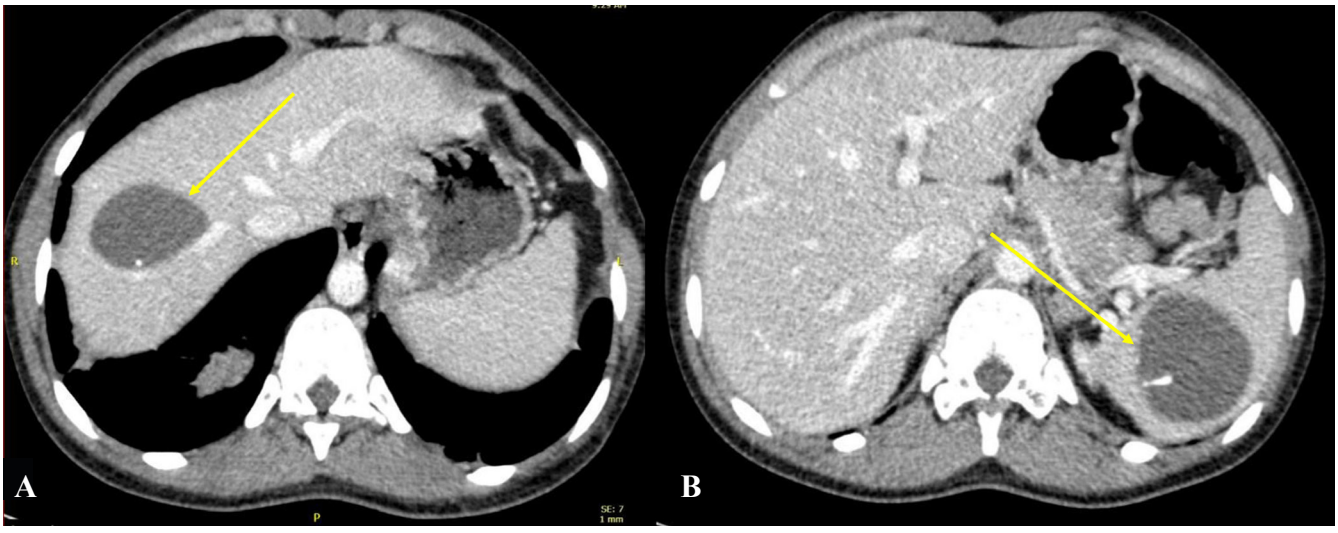

Figure 1: A CT scan from 2023 showing hydatid cysts in the liver (picture A) and spleen (picture B)

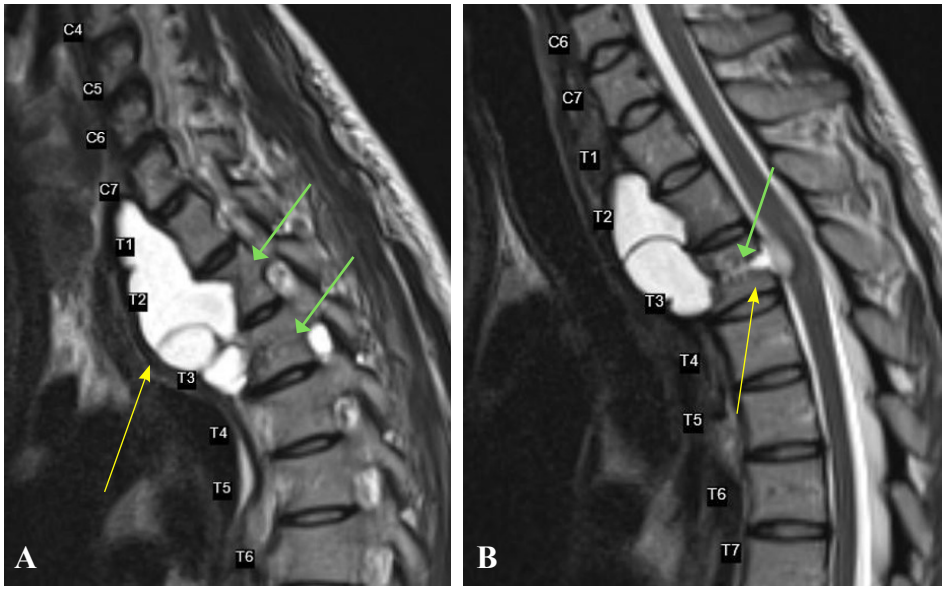

Figure 2: An MRI scan from 2024 indicating the cranio-caudal extent of the paraspinal hydatid cyst (yellow arrow A) and the protrusion of cystic contents into the spinal canal (yellow arrow B). The green arrows indicate the bone erosion at the T2/T3 vertebral body level (A) and T3 vertebral body (B)

cystic content protruding through the T3 vertebral body and into the spinal canal (Figure 2 - picture B). This subsequently resulted in spinal cord compression at the T3 level.

Neurosurgery was consulted and the patient was prepped for surgical intervention. Intraoperatively, laminectomies were performed at T2-T3 and the paraspinal cyst was aspirated. The patient was noted to have improved power in her lower legs shortly after the surgery. The histology of the cyst wall collected intraoperatively confirmed the diagnosis of a hydatid cyst. The patient was stepped down to an intermediate rehabilitation centre one month after surgery. She spent 3 months at the facility and was discharged successfully able to mobilise independently with the aid of an assistive device. After consultation with the infectious diseases department, it was advised that this patient remain on lifelong albendazole treatment.

Discussion

Hydatid disease is still prevalent in South Africa, particularly in sheep-farming communities.[1] The safety checks that are in place for meat sold in the urban commercial setting are not always available in more rural areas of South Africa. This leads to patients consuming meat that appears safe but may be harbouring parasites such as the *Echinococcus tapeworm*.

The most common site for hydatid disease is the liver, which is involved in 68–75% of cases.[2] In such cases, patients typically present with hepatomegaly and jaundice. The second most common site of infection is the lungs, with the right lung being more frequently involved. Pulmonary hydatid disease can result in a chronic cough, chest pain and shortness of breath. All cases of hydatid disease can present as anaphylaxis if there is a rupture of one or more of the cysts.

More rare locations of hydatid disease include the spleen, adrenal gland, kidney, peritoneum, bone and brain. Non-spinal bony locations are involved in 1–4% of cases, whereas spinal hydatid disease occurs in < 1% of cases.[2] Depending on the exact location of the spinal hydatid cysts, symptoms can vary from back pain to decreased range of motion and, if the cyst extends into the spinal canal, compression of the spinal cord. In the latter scenario, neurological deficits can involve both the sensory and motor pathways.

Despite spinal hydatid disease being uncommon, several cases have been reported in the literature, both in South Africa and internationally. A 3-year-old boy in Iran presented with bilateral lower limb weakness and was noted to have an extradural hydatid cyst that extended from the lumbar and sacral spinal levels L4-S3.[3] Another report from South Africa documented cases involving the spine and lower limb weakness: one in a 6-year-old with a para-spinal cyst at thoracic spine level T8, and another in an 8-year-old with a para-spinal lesion at thoracic and lumbar spine levels T12/L1.[4] A report from India documented a 38-year man who had developed multiple extradural hydatid cysts extending from thoracic spine levels T2- T3.[5]

Diagnosis is aided by ultrasonography, which can detect floating membranes, internal debris and daughter cysts.[2] CT is better at highlighting calcifications within the cysts and bony hydatid disease. In cases of neurological involvement, MRI is preferred. Serological tests are also an important tool in diagnosing hydatid disease. Various immunological tests exist that detect specific serum antibodies. However, tests can be false-negative, or a low antibody response can occur in rare sites, such as bone or brain. Thus, a combination of clinical findings, radiological testing and a serology should be used to accurately diagnose hydatid disease.[6]

There are two ultrasonographic classifications of hydatid cysts – Ghabi and the WHO-IWGE (World Health Organization Informal Working Group on Echinococcosis). The latter describes the cysts as active (CE1 – unilocular anechoic cysts, CE2 – septated cysts with a honeycomb appearance), transitional (CE3A – daughter cysts with detached membranes/water lily sign, CE3B – daughter cysts with a solid matrix) and inactive (CE4 – absence of daughter cysts with hyperechoic content, CE5 – solid and calcified wall).[6]

Medical therapy, such as albendazole, is usually not sufficient to eradicate hydatid disease entirely. It is used

to prevent dissemination while awaiting surgery, in cases where patients are poor surgical candidates, or as secondary prevention post-operatively. Various surgical procedures exist depending on the location of the disease. If the cysts are uncomplicated (CE1, CE3A) and > 5 cm, a PAIR (puncture, aspiration, injection and re-aspiration) procedure can be performed. This involves the use of a scolicidal agent such as hypertonic saline and is injected into the cyst to destroy the *Echinococcus* parasites. If the hydatid disease is more complicated (CE2, CE3B), open surgery is preferred. Scolicidal agents are also used in these cases, with swabs draped around the cysts to prevent an anaphylactic reaction or dissemination.[6]

There is often significant morbidity and mortality associated with spinal hydatid disease. In the absence of a history or confirmation of hydatid disease elsewhere in the body, it can be difficult to diagnose. The erosion of vertebral bodies and spinal cord compression can result in paraplegia. Invasive surgery is required to alleviate the pressure on the spinal cord. A host of perioperative complications can occur, including anaphylaxis, sepsis and damage to the spinal cord. Complete remission is possible with a combination of medical and surgical treatment. However, this depends on the duration of symptoms and a timely diagnosis. In a third world setting, diagnosis and treatment may not be as readily available, resulting in greater morbidity and mortality.[7]

This case illustrates the importance of regular follow-up in patients with hydatid disease. The response to anthelminthic medication should be assessed with serial imaging and a decision should be made whether surgical intervention is required to help eliminate the disease burden. In more chronic or unique cases, infectious disease specialists can be consulted to guide medication regimens.

In conclusion, the clinical presentation of hydatid disease can vary based on the site involved. Although spread to bones is rare, one should have a low threshold to suspect spinal involvement in patients with established hydatid disease and new onset back pain or neurological symptoms. Early neurosurgical consultation will result in a better prognosis for the patient. In addition, more stringent safety measures need to be established in rural communities to allow for the safe consumption of meat products.

Conflict of interest

The authors declare no conflict of interest.

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Ethical approval

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