Spine

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Echinococcosis of the spine

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- Echinococcosis or hydatid disease affecting the spine is an uncommon manifestation of *Echinococcus granulosus* infection of the spine.
- More commonly found in endemic areas, it causes significant morbidity and mortality as it grows slowly and produces symptoms mainly by compressing the spinal cord.
- As diagnostic methods are non-specific, diagnosis and management are usually delayed until the disease is advanced, thereby therapy is usually unlikely.
- Treatment is usually surgical, aiming at cyst excision, spinal cord decompression and spinal stabilization.
- This article summarizes the clinical findings of echinococcosis of the spine, discusses the specific laboratory and diagnostic findings, lists the current treatment options, and reviews the patients' outcomes.
- The aim is to prompt clinicians to be aware of the possibility of echinococcosis as a possible diagnosis in endemic areas.

Keywords: daughter cysts; decompression; echinococcosis; *echinococcus granulosus*; endemic; fusion; hydatid cyst; spine

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Introduction

Human echinococcosis is a zoonotic disease – a disease transmitted to humans by animals – caused by the tapeworm parasites of the genus *Echinococcus*. Echinococcosis occurs in four forms: the more common forms are the hydatid or unilocular echinococcosis caused by *Echinococcus granulosus* and the alveolar echinococcosis caused by *Echinococcus multilocularis*, and the far more rare forms are the neotropical echinococcosis including the polycystic echinococcosis caused by *Echinococcus vogeli* and the unicystic echinococcosis caused by *Echinococcus oligarthrus*. *Echinococcus vogeli* infections are similar to alveolar echinococcosis, whereas *Echinococcus oligarthrus* infections are less aggressive.[1]

Life cycle and disease transmission

The adult *Echinococcus granulosus* resides in the small intestine of the definitive host. The tapeworm consists of a head (scolex), a neck and a tail. The head has two or more suckers and, in some cases, a rostellum or knob of small hooks that are used by the parasite to attach to the wall of the host's intestine. The scolex is connected by a short neck to the lower portion of the tapeworm called the strobila that is a ribbon-like chain of independent but connected segments called proglottids. Each proglottid has both male and female sexual organs and is responsible for producing the parasite's eggs. Proglottids start to develop in the neck region of the parasite, where they mature and move downward in the strobila as new segments are added from above.[2]

The hermaphroditic proglottids become gravid and eventually are released from the tapeworm. Alternatively, gravid proglottids release eggs that are passed in the faeces and are immediately infectious.[2] When this contaminated waste is excreted into the environment, an intermediate host may contract the parasite. After ingestion, eggs hatch in the small intestine and release six-hooked oncospheres that penetrate the intestinal wall mucosa and migrate through the circulation into various organs, more commonly the liver and the lungs. In these organs, the oncospheres develop into thick-walled hydatid cysts that gradually enlarge and produce protoscolices and daughter cysts that fill the cyst interior.[2,3]

Hydatid cysts are round and are usually filled with clear fluid. The parasites evoke a granulomatous inflammatory reaction that leads to walling off of the cyst by fibrous tissue. To become infected, the definitive host must ingest the cyst-containing organs of the infected intermediate host. After ingestion, the protoscolices evaginate, attach to the intestinal mucosa, and develop into mature tapeworms in the lumen of the definitive host's intestine.[3]

Humans serve as inadvertent intermediate hosts for cestodes of *Echinococcus granulosus*. *Echinococcus granulosus* shows a considerable phenotypic and genotypic variability, with several strains having been identified.[4] Strains exhibit several discrepancies, however, a common feature is the utilization of dogs and other canids as definitive hosts.[5] Sheep, cattle, pigs, camels, horses and goats are known to serve as intermediate hosts, while canines such as dogs, wolves and foxes serve as definitive hosts. Typically, transmission involves dogs as definitive hosts and livestock as intermediate hosts. Dog infection occurs as a result of feeding with contaminated intestines after home slaughter and improper abattoir management, in which stray dogs have access to contaminated offal, or by herding dogs scavenging on livestock carcasses left on pasture.[6]

Humans acquire echinococcosis by ingesting parasite eggs with their food; parasite eggs are distributed via local environmental contamination via faecal contamination from infected dogs. Parasite eggs are resistant to dehydration and remain viable for a long time, therefore allowing for delayed transmission to humans without a direct contact with vector animals. Once in the intestinal tract, the eggs hatch to form oncospheres that penetrate the intestinal mucosa, enter the circulation, and encyst visceral organs forming mature larval cysts.[7]

Epidemiology and classification

Echinococcosis appears in every region of the planet since *Echinococcus granulosus* is transmitted by domestic dogs in livestock-raising areas.[8] However, the incidence of echinococcosis differs from area to area depending on the presence in a respective country of nomadic or semi-nomadic sheep and goat flocks that serve as the usual intermediate hosts.[9] The higher incidence is reported in countries of the temperate zones, such as the Mediterranean region, southern and central Russia, central Asia and China, but also in Australia, South America and north and east Africa.[9,10] Currently, echinococcosis is considered endemic in the Mediterranean region.[11] Echinococcosis does not seem to have a gender predilection; both genders are affected equally, possibly with a slight male predilection.[12,13] Similarly, echinococcosis does not have an age predilection;[5] it appears to infect people of all age groups, however, a higher incidence in patients aged 30–36 years has been reported.[14–24]

Hydatid cyst formation is more common in the liver (approximately 70%), especially the right lobe. The second most common organ involved is the lungs (approximately 20–30%). Other less commonly involved organs are the brain, heart and bones.[17–19] The incidence of osseous echinococcosis is low (approximately 0.5–4%).[20–22] In osseous echinococcosis, spinal involvement is the most common form, though rare overall (approximately 0.2–1%).[22,23] The most common spinal location is the thoracic spine (approximately 50%), followed by the lumbosacral region (approximately 29%) and the lumbar spine (approximately 21%).[24–26] Most patients with thoracic spine echinococcosis had a history of extraspinal cystic echinococcosis, most commonly of the lungs, liver, kidneys and soft tissues in decreasing order of frequency, and some had a history of surgically treated lung echinococcosis; this may explain the increased rates of thoracic spine involvement.[27,28] The hydatid cysts are usually not confined to the vertebral bodies, often, they affect the intervertebral discs, the spinal cord and the posterior spinal elements, and they may grow in the spinal canal as well.[26,27,29,30]

Braithwaite and Lees classified spinal echinococcosis into five types: In type 1 the hydatid cyst is intramedullary, in type 2 the hydatid cyst is intradural and extramedullary, in type 3 the hydatid cyst is extradural and intraspinal, in type 4 the hydatid cyst is into the vertebral body, and in type 5 the hydatid cyst is paravertebral.[31] To our knowledge, only four cases of primary intramedullary hydatid cyst have been reported in the literature.[32–35] Type 2 disease or intradural extramedullary cyst is extremely rare as well; only 45 cases have been reported in the literature as of 2013,[36] one of them being of special interest because of multiple spinal intradural extramedullary cysts.[37] A distinct type of *Echinococcus granulosus* infection is the dumbbell formation in which the cyst begins to form in the spinal cord and follows the neural exit foramen, forming a paravertebral extension.[38,39] The dumbbell type is commonly a type 4 affecting the vertebral bodies, and only rarely a type 3 or 5 manifesting without bony involvement.[40]

Besides the aforementioned anatomical classification, spinal echinococcosis can also be classified according to the route of the spinal infection into primary and secondary, haematogenous and extension per continuitatem; primary haematogenous spinal echinococcosis (hematogenous infection of spinal structures at primary infection), secondary haematogenous spinal echinococcosis (haematogenous infection of spinal structures following spontaneous or iatrogenic seeding from extraspinal cystic echinococcosis), secondary extension per continuitatem spinal echinococcosis (direct invasion of spinal structures from extraspinal echinococcosis such as mediastinal and paravertebral soft tissue, pleura, lung, ribs, pelvis, posterior paravertebral muscles), and secondary extension

per continuitatem spinal echinococcosis (cerebral disease with spontaneous or iatrogenic seeding into the cerebrospinal fluid, leading to intradural spinal seeding).[37,38,41]

Clinical presentation

Spinal echinococcosis most commonly presents with symptoms stemming from cyst compression of adjacent spinal structures. The patients most commonly present with back pain; limb weakness occurs later. Other presenting symptoms include radiculopathy, myelopathy and pathological fractures.[25,36,42–57] However, the patient may be asymptomatic in the early stages of the disease.[42] In a study of 36 patients with spinal echinococcosis, 10 patients presented with backache, and 17 patients experienced variable degree; four patients experienced complete paraplegia, another four patients experienced urinary retention, and one patient experienced quadriplegia.[43] In a similar study of 84 patients with spinal echinococcosis, 61 patients experienced muscle weakness of the limbs, 36 patients experienced back pain, 27 patients experienced bowel and bladder dysfunction, and 20 patients experienced variable sensory disturbances.[44] In another study of 11 patients with spinal echinococcosis, the average time from symptom onset to hospital admission was 3.5 weeks; at admission, five patients were paraplegic and six patients were paraparetic.[45]

Unusual case presentations have also been reported.[36,48–50] A patient presented with fever and vomiting, altered mental status, and left-foot weakness of the last 30 days; imaging showed intraspinal, intradural, intramedullary, and epidural thoracic spinal echinococcosis.[49] Another patient presented with low back pain of increasing severity, bilateral lower extremity weakness and spastic paraparesis; imaging showed an arachnoid hydatid cyst of the lumbar spine.[36] Another patient presented with low back pain and left-leg pain and urge incontinence from recurrent lumbosacral echinococcosis 13 years after laminectomy with excision of lumbar spine hydatid cysts; imaging showed multiple loculated cysts in the left paraspinal area extending into the extradural space at S2 level causing thecal sac compression.[50]

Primary echinococcosis of the sacral spine is rare.[51–53] A 27-year-old man from India presented with low back pain, right sciatica and walking difficulty of one year; imaging showed a sacral hydatid cyst.[51] A 43-year-old woman from Turkey presented with left-leg pain and swelling without neurological symptoms. Fluid leakage was observed from a cutaneous fistula at the left hip; imaging showed bilateral multicystic echinococcosis that eroded the first sacral wing at the S1 level, extending to the left S1 foramen and paravertebral muscles.[52] Another patient presented with cauda equina syndrome from lumbosacral echinococcosis; imaging showed multiple, pearl-like, white capsular cysts, extending from L4 to S3 level with components in perisacral and sacroiliac joints.[53]

Rare cases of spinal echinococcosis in children have also been reported.[54–57] An eight-year-old boy with unremarkable past medical history, presented with back pain, progressive weakness, numbness in both legs and difficulty in walking, and urinary and faecal incontinence from 7 months; imaging showed lumbar echinococcosis located in the extradural space compressing the dural sac and caudal roots and expanding to the L3 and L4 neural foramina.[54] Another child presented with back pain and left leg monoparesis from the last month; imaging showed intradural extramedullary echinococcosis at the T7–T8 level.[55] A five-year-old boy from India presented with paraparesis, bilaterally decreased sensation below L4 level and loss of perianal sensation; imaging showed intradural echinococcosis from L4 to sacral region and extensive involvement of the spinal cord.[56]

Diagnosis

Diagnosis of spinal echinococcosis is predominantly done by imaging. Computed tomography (CT), and magnetic resonance imaging (MRI) are the imaging methods of choice for the diagnosis of spinal echinococcosis as they provide excellent imaging of the spinal cord.[25,30,31,42–47,58] Laboratory tests are less specific for spinal echinococcosis; however, they may detect an echinococcal infection in the patient or aid in the confirmation of the diagnosis if cysts are found on imaging.[3,58–60]

Laboratory tests

Common laboratory markers used in infections such as C-reactive protein (CRP), estimated sedimentation rate (ESR) and white blood cell (WBC) count are often within normal range and they are variable findings.[36,46,50,61] Eosinophilia, commonly seen in parasitic infections is not a consistent or reliable finding.[2,46,62] ELISA and Western Blot are the most commonly used serologic assays for echinococcal antibody detection in liver disease with an 80–100% sensitivity and 88–96% specificity.[2,58–60,63,64] ELISA is performed by obtaining antigens from fertile liver hydatid cysts, most commonly from sheep, horses and camels. Two kinds of antigens are used; whole parasite or parasite organelles and soluble antigens prepared from cyst fluid. In ELISA, an insoluble medium is coated with the antigen and successive incubations with the patient's serum follow. The last step involves incubation with an enzyme-conjugated anti-human IgG, which binds the patient's specific antibodies (if present) to the antigen. The enzyme colours it, enabling us to identify its presence.[58,59] In Western Blot, the antigens used are denatured and separated by electrophoresis and are subsequently transferred in a nitrocellulose membrane. The next step

involves incubation with the patient's serum and then successive incubation with an anti-human IgG conjugated to an enzyme; an enzymatic coloured reaction renders the test positive. One advantage of the Western Blot method is that it allows molecular weight analysis of the detected antigens.[58,64] However, serologic assays are less sensitive and specific for spinal echinococcosis with a 25% sensitivity and 56% specificity.[2,62] Additionally, children may present with normal serology, despite being infected.[54–56]

The Casoni intradermal skin test is a hypersensitivity-based skin test used to detect hydatid disease. Sterile fluid (0.25 mL) of hydatid cyst origin is injected into one arm whilst 0.25 mL of normal saline is injected into the other arm to act as a control. A wheal response occurring at the injection site within 30 minutes is considered positive (hypersensitivity reaction type I); care must be taken though for an anaphylactoid reaction.[65,66] Although once a major test in diagnosing hydatid disease, it has largely been superseded by the aforementioned serologic assays, which are more sensitive, specific and safer.[67,68] In a study of 36 patients with spinal echinococcosis, the Casoni intradermal test was found positive in six out of 14 patients in which the test was performed and haemagglutination tests were found positive in three out of four patients.[43]

Imaging

Radiographs are useful for echinococcal cysts in lung, bone and muscles.[69,70] Radiographic findings include single or multiple, moth-eaten osteolytic, expansile cavitatory areas without periosteal reaction or sclerosis; the vertebral bodies and posterior spinal elements are extensively involved.[25,27,43,44,46,47,62,69–75] Multiple osteolytic lesions resemble a bunch of grapes and contain trabeculae with cortical thinning and calcification of neighbouring soft tissues.[27,71] In most cases, the intervertebral disc is not affected, but the cartilage may be flattened or distorted by a vertebral fracture. Radiographic imaging of zones of multilocular osteolysis with a hazy image of the bone, without periosteal or osteophytic reaction and without process of condensation is a highly suggestive finding of echinococcosis.[72,74] 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 tumours such as metastases and chondroblastoma, or other infections such as tuberculosis, spinal or paraspinal abscess.[44,62,73,74] Moreover, in patients with extradural and intradural disease, radiographs do not show any abnormality.[44]

Ultrasonography is an excellent diagnostic modality for liver echinococcosis. It is safe, non-invasive, and cost-effective, which makes it especially useful considering that echinococcosis is endemic in certain poor and developing areas. However it is not helpful in the context of spinal echinococcosis per se.[2,3,42] Myelography use has declined nowadays due to the superiority of MRI in the visualization of abnormalities of the spinal cord and to the risks it carries.[62,76] Nevertheless, when performed, the most common finding is a complete blockage of the contrast material; other findings include a characteristic brush-border appearance, suggestive of extradural lesion, and multiple intradural round-shaped lesions masquerading as arachnoid cysts.[25,47,77]

CT shows similar findings to radiographs but is more detailed; it may show even small cysts.[47] Typical CT findings include round or ovoid space-occupying lesions with 'double layer arcuate calcification', which is considered specific enough for echinococcosis rather than any other cystic disease; vertebral bodies and arches are eroded with multiple cysts.[27,43,46,47,73,75,77] The measurement of cyst density in CT offers a way to differentiate between parasitic and non-parasitic cysts.[46,62,78] Additionally, CT depicts the multiple cysts as osteolytic expansile lesions in vertebral bodies.[46,74,77–79]

MRI is considered a better imaging modality than CT, especially for the evaluation of recurrences after surgical treatment. MRI provides higher contrast between different types of soft tissues and can be displayed in any plane besides the three classical planes used in CT. MRI findings include multiple cystic fluid-filled lesions, septated with thin walls and irregular branching resembling a bunch of grapes at multiple levels; paravertebral muscles show multiple large, spherical, cystic lesions with smaller cysts inside.[20,46,47,62,70,71,73,74,77–80] The signal intensity of hydatid cysts is hypointense on T1-weighted MRI and hyperintense on T2-weighted MRI.[20,62,70,73,74,77–80] Multilocular cysts may represent daughter cysts; these are small spheres that contain protoscolices and are formed from the inner layer of the hydatid cyst.[20,46,70,73,77] Daughter cysts grow slowly, and the bone appears capsular and distensible with smooth and often sclerotic circumscription.[70,71,73]

Biopsy

When imaging and serologic tests are inconclusive and negative respectively, ultrasound- or CT-guided fine-needle aspiration is sometimes used; mostly in abdominal disease.[62,81] In spinal hydatid disease, due to high rates of bone involvement, cyst rupture is more common leading to anaphylaxis and further disease seeding. Therefore, cyst aspiration is rarely, if ever, performed.[48,62,82] Nevertheless, a few cases have been reported in which the cysts were drained for therapeutic purposes in patients in whom surgery was not an option.[83,84]

Treatment

Current treatment options for echinococcosis of the spine are surgical and pharmacological treatment. Surgery, either curettage or resection, is considered the treatment of choice; as most patients present with spinal cord

compression symptoms, urgent surgery is required in such instances.[24,62,85–87] Depending on the location and extent of the echinococcosis, the aims of surgery are decompression of the spinal cord and stabilization of a compromised spinal segment.[88] Although variable and combined surgical approaches have been reported, posterior decompression by laminectomy and spinal fusion with or without thoracotomy for thoracic spine echinococcosis is the most common.[24,25,43–46,85,86,89,90] The purpose of the surgery is total removal of the cyst without rupture. However, complete clearance is difficult due to the invasive diffuse spread within the bone and the spinal canal. Often, rupture of the cyst resulting in fluid spillage leads to recurrence. Thorough irrigation of the surgical area with hypertonic saline is recommended to prevent recurrences.[47,86,90] A novel type of drainage of spinal hydatid cyst was proposed by Caglar et al.[90] During surgery, the surgeon places a two-way drainage catheter inside the cyst; the distal end catheter is placed outside the patient. Postoperatively, the cyst is injected with chlorhexidine solution (0.04% Chx-Glu). After five minutes of washing, the solution and the cyst contents are emptied from the catheter exit. However, this method has not been tested in a clinical setting yet and studies should be carried out to verify its efficacy.[90] A palliative surgical operation is considered in patients with extensive disease that cannot be totally excised because of innumerable vertebral cysts.[44,46,91]

Pharmacologic treatment consists of administration of anti-parasitic drugs such as benzimidazoles albendazole and mebendazole; albendazole has replaced mebendazole due to lower dose requirements and better absorption.[2,92,93] A few reports advocate the use of an albendazole-praziquantel combination; however, the combination has not been extensively applied to verify its efficacy.[91,94–96] Many authors suggest that albendazole administration delays recurrences and reduces complications, thus it is commonly administered after surgery.[25,43,44,47,62,85,86,97] Despite that, drug efficacy in echinococcosis of the spine as monotherapy without concomitant surgery is debatable; available data are scarce and controversial.[44,46,47,93,96,98,99] Nevertheless, it remains the only treatment when surgery cannot be performed.[24,46,62,89,91,96,98,99]

Outcome

Spinal echinococcosis, despite available treatment options, carries a high rate of disease recurrence. This is primarily due to the infiltrative nature of the cysts and intraoperative cysts rupture and fluid spillage.[25,39,43,47,62] Other factors influencing outcome are disease extension (many cases arrive late in disease course), and anatomical location according to the Braithwaite and Lees classification. Intradural disease is less likely to recur.[44,45,62,85,86] The most common complication is recurrent symptoms of spinal cord compression and this occurs in 40–90% of cases which require reoperation.[25,47,62,100] Intraoperative death is the most serious complication caused by anaphylactic reaction during surgery due to cyst rupture; the organism is highly immunogenic.[85] Vertebral column instability is another postoperative problem encountered in patients with spinal echinococcosis extending in the vertebral column.[86,101] Finally, a rare cause of death occurred due to formalin injection; in this patient the dura was accidentally torn during surgery leading to the patient's death after formalin injection.[25]

In Kafaji et al's series of 36 patients with spinal hydatid disease, the patients were followed for one to 10 years (median follow-up, 3.65 years). Seven patients (19.44%) died of disseminated disease. All surgically treated patients, regardless of whether hydatid disease was primary or recurrent, were treated four months postoperatively with albendazole (10 mg/kg/day). Almost all recurrences were regional at the level of the previous surgery. Of the 29 patients from whom follow-up was available for at least one year after the initial surgery, 26 had a recurrence after one year (89%), and 10 (27.8%) patients were disease-free after a mean of 5.3 years.[43] Herrera et al studied 20 patients with spinal hydatid disease at a median follow-up of 4.8 years (range, 2–11 years). Eight patients did not receive medical therapy after surgery, seven patients received mebendazole and five patients received albendazole treatment. All but one of them experienced disease recurrence. Repeated surgery was necessary in 13 patients; in 12 of them because of recurrence of the hydatidosis and in one patient because of surgical wound infection. Two patients died in the early postoperative period and eight patients died because of secondary later complications of spinal cord injury increasing the death rate to 50%.[85]

Turgut reviewed 28 reports (84 patients) of spinal hydatid disease from Turkey; 82 patients were treated surgically; 38 patients had only surgery, 45 patients had only medical therapy, one patient had surgery and medical therapy, and one patient had radiotherapy and medical therapy. A significant difference in the recurrence rates was noticed in between spinal or paraspinal involvement and intraspinal disease; 32% and 4% respectively. Patients treated with surgery plus chemotherapy had 5% recurrence rates whereas those treated with only surgery had 32% recurrence rates.[44] Another study published in Turkey by Pamir et al examined the disease in question in 11 patients treated surgically. In two patients mebendazole was used postoperatively and patients remained symptom-free for two and five months respectively. In the early postoperative period seven patients manifested neurological improvement. Recurrence was seen in two patients; a correlation was noticed between cyst location and recurrence. In the epidurally located cysts, microvesicles are diffusely spread inside the bone. These multiple cysts are easily ruptured during surgery and recurrence is

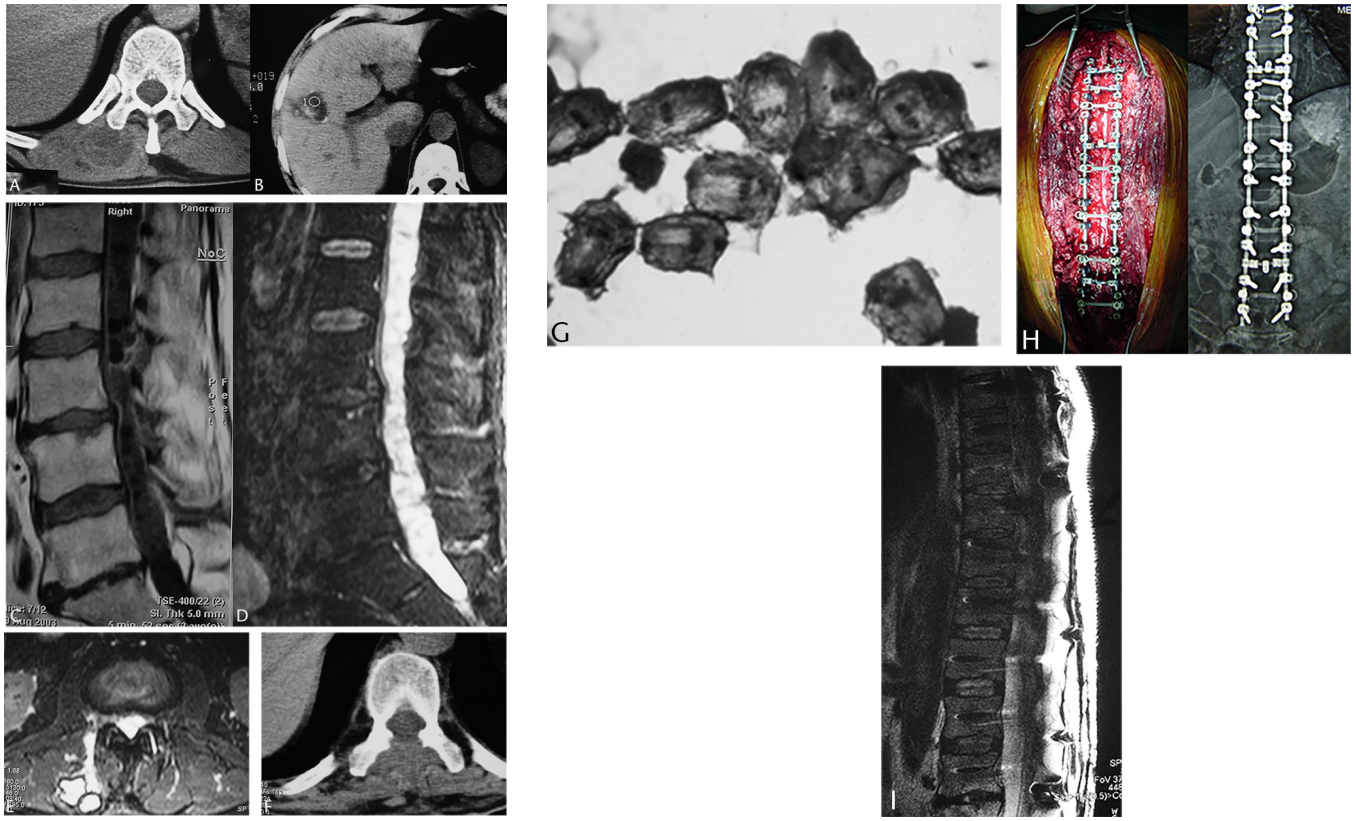

Fig. 1 A 63-year-old man with low back pain and paraparesis. Laboratory examination showed increased values of CRP, ESR, WBC and eosinophilia. Radiographs of the thoracic spine and chest were normal. (A) CT scan of the spine showed paravertebral cysts at the thoracic and lumbar spine, (B) similar cysts at the liver. Sagittal (C) T1-weighted and (D) T2-weighted, and (E) axial T2-weighted MRI of the spine showed paravertebral cystic lesions extending into the spinal canal. With the presumptive diagnosis of a parasitic infection, (F) extensive T6 to L5 laminectomy and cysts excision was carried out. Recovery was uneventful; cultures and (G) histology showed echinococcus scolices. Ten days later, (H) T2–L5 long spinal fusion for spinal stabilization was carried out. Postoperatively, the patient was administered mebendazole for 12 months. At the 15-year follow-up after treatment, (I) the patient did not experience any echinococcosis recurrences; he recovered paraparesis but did not recover urinary control and required intermittent bladder catheterizations.

Note. CRP, C-reactive protein; ESR, estimated sedimentation rate; WBC, white blood cell; CT, computed tomography; MRI, magnetic resonance imaging.

common. In the intradural extramedullary form of disease recurrence is rarely noticed.[45]

Caglar et al performed posterior decompression with laminectomy and cyst excision in 12 patients with spinal echinococcosis; six of them needed to be stabilized. Hypertonic saline was administered topically to prevent further cyst spread in all cases during the operation before and after cyst excision. Albendazole was administered twice daily; treatment was initiated before the first surgery. Recurrence was seen in nine cases (75%).[90] Gezercan et al described eight patients with spinal echinococcosis followed for 7–15 years. All cysts ruptured during surgery and hypertonic saline irrigation was used; no anaphylactoid reaction occurred. Recurrence occurred invariably in all patients; two to five surgical operations were performed in each patient.[86] Işlekel et al reported on 13 patients followed from two months to 20 years (three patients were lost to follow-up early). Nine patients had one or more recurrences (90%). The shortest and longest time intervals between the recurrences were two months and 48 months (mean 25.2 months), respectively. Two patients died in that series; one patient died of complications of the renal hydatid disease and the other due to the formalin irrigation mentioned above (15.4%).[25]

Hamdan et al reported a series of nine patients treated with surgery; six patients had a posterior laminectomy, three patients had anterior decompression, one patient had a large mass at the L4 level eroding the aorta, and another patient had dissemination of a daughter cyst in both femoral arteries resulting in pulseless limbs (he died within 24 hours of surgery due to severe anaphylactic reaction). All patients presented at a very late stage for treatment; eight patients had a recurrence, with the only patient without recurrence having no bone involvement. Another cause for the high recurrence rate was the wide bone and neural tissue infiltration which made local resection with safety

margin practically impossible.[46] In the case series performed by Prabhakar et al, four patients with spinal echinococcosis had laminectomy through a posterior approach, followed by antihelminthic therapy with 400 mg of albendazole three times daily for one year. The imaging studies at one-year follow-up invariably showed residual or recurrent cysts. Two patients with thoracic spine involvement developed symptomatic recurrence with neurologic status deterioration; repeat surgery through the anterior approach was performed for thorough decompression and fusion with iliac crest bone grafts. At two to three years follow-up, they had deteriorated to paraplegia, but refused further surgical intervention; thus, succumbing to the disease and the complications of paraplegia. The patients with lumbosacral disease developed recurrence but, because they had no symptoms, refused to be treated and they were eventually lost to follow-up.[47]

Conclusion

Echinococcosis of the spine, although an uncommon manifestation of *Echinococcus granulosus* infection it is the most common form of skeletal echinococcosis. The thoracic spine is most commonly involved. The patients usually present with neurological symptoms secondary to spinal cord compression and pain secondary to bone destruction and spinal instability. Although there are not any pathognomonic imaging findings for echinococcosis, MRI is the modality of choice for the diagnosis of patients with spinal disease as it offers excellent imaging of the spinal cord and high contrast between different types of soft tissues. Cyst excision, and spinal decompression and fusion, combined with pharmacologic medication administration before and after surgery is the treatment of choice to delay or prevent recurrences. However, complete excision of all lesions is usually not possible due to the infiltrative nature of the disease and advanced stage at presentation. Therefore, recurrences after treatment are the rule in most cases. Clinicians should be aware of this entity, especially in endemic areas, and promptly recognize it in patients presenting with back pain and neurological symptoms associated with highly suggestive imaging findings.

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