Disseminated intraspinal hydatid disease

Case report

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Spinal echinococcosis is a rare entity, accounting for 1% of all cases of hydatid disease. The authors report the case of a 60-year-old man whom they treated for recurrent nerve root compression due to disseminated intraspinal echinococcosis (hydatid disease). Six years previously he had undergone surgery on an emergency basis at another institution after presenting with acute paraplegia due to a primary extradural hydatid cyst of the thoracic spine. Unfortunately, during surgical removal of the cysts, the echinococcosis disseminated into the spinal canal. This complication was documented by magnetic resonance (MR) imaging. In the 4 years before the authors treated him, he was hospitalized 4 times for 4 recurrences of nerve root compression.

The authors treated the disseminated disease successfully with total T7–8 corpectomy, grafting with titanium cage and Texas Scottish Rite Hospital instrumentation, and long-term administration of albendazole (400 mg daily). Early diagnosis, proper utilization of MR imaging, and radical resection of diseased vertebrae and soft tissues followed by anthelmintic treatment are essential to control disseminated spinal hydatidosis and prevent recurrence.

(DOI: 10.3171/SPI/2008/8/5/490)

KEY WORDS • echinococcosis • hydatid cyst • magnetic resonance imaging • paraplegia • spine

HYDATID disease is caused by infestation by a parasitic tapeworm, Echinococcus granulosus. Humans are the intermediate hosts and acquire the parasite by ingesting water or food contaminated with eggs that were excreted in the feces of an infected dog.7 The disease affects the liver and lungs in ~80–90% of cases.13 Bone hydatidosis is much less prevalent, accounting for only ~0.5–4% of the reported cases, and spinal involvement occurs in 50% of patients with this form of the disease.15

We present a case of disseminated echinococcosis in a patient who had previously undergone resection of primary extradural hydatid cysts after presenting with acute paraplegia. The condition had not been accurately diagnosed until the histopathological examination of the resected lesion, and the disease disseminated into the spinal canal during the initial surgical excision. Magnetic resonance imaging findings, medical and surgical treatments, and the outcome during an 8-year follow-up period are discussed.

Abbreviations used in this paper: MR = magnetic resonance; VB = vertebral body.

Case Report

This 60-year-old man presented to our hospital in October 2005, 6 years after undergoing emergency surgery at another university hospital. He had originally been brought to the emergency department of that hospital in September 1999, complaining of weakness and numbness in the lower limbs. At that time, he had been experiencing back pain of increasing severity for several months, and during the 5 days prior to his presentation at the hospital, he had experienced difficulty walking. Neurological examination had revealed bilateral lower-extremity weakness and paraparesis. The results of all hematological and biochemical tests had been normal. An MR imaging study of the spin had revealed multiple extradural heterogeneous lesions with severe spinal cord compression at the T6–8 levels and VB hyperintensity (image no longer available). Neither physical examination nor ultrasonographic examination of the abdominal and pelvic organs demonstrated any localization of disease other than in the spine. Due to rapid progression of the patient’s symptoms toward spastic paraplegia, he underwent an emergency surgical decompression procedure.
During the procedure, numerous extradural cysts (~ 60–100) were found. They had translucent walls and clear fluid content typical of hydatid cysts and had destroyed the posterior elements of the T-7 and T-8 vertebrae. The laminae of these vertebrae were also full of cysts. The spinal cord was compressed and pushed aside by 2 large cysts, which were located lateral to the pedicles of the T-7 and T-8 vertebrae. Laminectomy was performed at these 2 levels, and all cysts were removed from adjacent extradural spaces. Histopathological examination established the diagnosis of hydatid disease (Fig. 1). After the operation, the patient’s neurological status improved, and he was discharged with instructions to continue a regimen of anthelmintic treatment consisting of albendazole (400 mg twice a day) and praziquantel (600 mg daily).

During the next 4 years, the patient was admitted 4 times to the institution at which he had undergone surgery. Each readmission was due to recurrence of weakness and paresthesia of the lower extremities following a symptom-free period. The pattern of taking the prescribed medications had been irregular, and recurrences had occurred upon interruption of the medical regimen. During hospitalization, he was treated with conservative therapies such as intravenous corticosteroid therapy; he refused additional surgical intervention. Neuroimaging studies demonstrated disseminated hydatid disease (Figs. 2–4).

In October 2005, the patient presented to the emergency department of our university hospital with acute paraplegia. Because of the chronic nature of the disease and the lack of response to anthelmintic chemotherapy, he underwent a second complex operation. Using an anterior approach, we performed surgical drainage of numerous hydatid cysts and total corpectomy of T-7 and T-8 with excision of surrounding tissues. We irrigated the surgical field with hypertonic saline and placed a graft using a titanium cage and Texas Scottish Rite Hospital instrumentation. Anthelmintic therapy with albendazole (400 mg daily) was continued to prevent recurrences. The patient was symptom free at the last follow-up visit, almost 2 years after the second surgery (Fig. 5).

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**Discussion**

Following ingestion by humans, the *Echinococcus granulosus* ova lose their protective shell. The outer layer embryos (oncosphere) develop and penetrate the duodenal mucosa, enter the portal circulation, and disseminate to the liver and then to the lungs. Within 3 weeks, the embryo develops into a larva, and a cyst is formed. The center of the VB is the first part of the vertebra to be involved, and the infection subsequently extends extradurally or paravertebrally. As the cysts grow along the cancellous bone trabeculae, small diverticula are formed through exogenous vesiculation. These diverticula

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**Fig. 1.** Photomicrograph showing the outer acellular laminated layer and inner germinal layer characteristic of a hydatid cyst. H & E, original magnification × 50.

**Fig. 2.** Midsagittal T2-weighted MR image showing multiloculated hydatid cysts in the T7–8 VBs as well as disseminated cysts in the posterolateral extradural space of the canal at the T2–9 levels compressing the thecal sac and leading to spinal canal stenosis at the level of the destroyed T-9 vertebra.
may separate from the main cyst, resulting in a mass of
cysts.\textsuperscript{12,14} Intervertebral discs are usually preserved due to
the tendency of the disease to propagate beneath the periosteum and ligaments.\textsuperscript{7}

Patients may present with symptoms and signs related to
spinal cord compression, such as low-back pain, radicular
leg pain, and paraparesis that last for several weeks and
may culminate in acute paraplegia with rapid progression,
as was observed in our patient. Paraplegia results from the
cysts invading the spinal canal and compression or ischemic changes of the spinal cord or cauda equina.\textsuperscript{10}

Definitive diagnosis can be achieved by histopathological
examination of resected tissue. Myelography and fine-
needle aspiration biopsy are invasive procedures, and puncture of a cyst may lead to intradural dissemination and anaphylaxis.\textsuperscript{6,7} The best preoperative diagnostic procedure
is MR imaging,\textsuperscript{3,4,8} which provides comprehensive information about the location, extension, and anatomical relationships of hydatid cysts. Their characteristic appearance is
similar to that of a flattened sausage with 2 dome-shaped ends. They usually have thin single walls without septation. The intensity of their contents is similar to that of cerebrospinal fluid, and there is no debris in their lumens.\textsuperscript{4,8,17}

Radical surgery is the keystone of treatment, but complete removal of spinal hydatid cysts is rarely accomplished
in a single operation. This is mainly due to the extensive
bone invasion, spillage and implantation of live vesicles,

\begin{figure}
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\includegraphics[width=\textwidth]{fig3}
\caption{Axial T2-weighted MR image showing hydatid cysts in
the VB, right pedicle, and transverse process of T-8 with extension
in the right paravertebral space and also to the canal on the right
anterolateral side.}
\end{figure}

\begin{figure}
\centering
\includegraphics[width=\textwidth]{fig4}
\caption{Magnetic resonance myelograms showing multiple
hydatid cysts surrounding and compressing the thecal sac.}
\end{figure}

\begin{figure}
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\includegraphics[width=\textwidth]{fig5}
\caption{A T2-weighted MR image obtained in August 2007
shows no evidence of residual cysts.}
\end{figure}
Spinal hydatid disease

and high rate of local recurrence. Laminectomy has been introduced as the preferred operative procedure, but because of residual diseased VB and adjacent tissue, it is rarely curative.

Almost all authors who have written on the topic agree that isolated chemotherapy with albendazole is not sufficient to cure the osseous lesions of hydatid disease. This option should be only considered for management of inoperable lesions. For operable lesions, radical surgery must be followed by albendazole treatment to prevent or delay recurrences. Controversy remains, however, regarding the duration, schedule, and dosage of albendazole therapy in the treatment of hydatid cysts of the skeleton, especially those in the spine. In primary cervical spine hydatidosis, it has been shown that following surgical debridement, prolonged administration of albendazole (400 mg twice daily for ~9 years) is safe and effective in the prevention of recurrences. It should be noted, however, that whenever patients receive long-term albendazole therapy at a dosage of 10 mg/kg/day, their blood cell counts and liver enzymes (serum glutamic oxaloacetic transaminase [SGOT], serum glutamate pyruvate transaminase [SGPT], and serum alkaline phosphatase) should be monitored for signs of associated toxicities, including cholestatic jaundice and hepatitis.

In contrast to the report of successful treatment, a paper from India reported recurrences in 4 patients with spinal hydatidosis who received 400 mg of albendazole 3 times daily for 1 year after surgical excision. Clearly, the location of the lesions, their degree of extension, and the surgeon’s experience in complete removal of the cysts all contribute to the outcome of surgical treatment of hydatid cysts.

Conclusions

The diagnosis of hydatid disease should always be considered in patients with atypical presentation of vertebral lesions, particularly in endemic regions. In surgically treated cases, MR imaging is extremely useful for preoperative planning and postoperative follow-up. Hydatid disease has a high risk of recurrence and anterior surgical excision of the affected vertebrae and soft tissues combined with bone grafting followed by postoperative adjuvant chemotherapy is an appropriate treatment.

References