



Spinal Hydatid Cyst Disease

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■ **OBJECTIVE:** A hydatid cyst (HC) is a zoonotic infection affecting the liver and lungs, with rare spinal involvement. We discuss the long-term results in 8 patients with spinal HC who were monitored at our clinic for 7 to 15 years.

■ **METHODS:** The demographic data and clinicopathologic characteristics of 8 patients with spinal HC diagnosed between 2000 and 2016 were evaluated for their contribution to recurrence, and the long-term follow-up results were examined.

■ **RESULTS:** Four male and 4 female patients with a median age of 30.75 years (range, 17–45 years) at the first surgery were included. Infections were localized in the thoracic (3), thoracolumbar (1), lumbar (1), sacral (1), cervicothoracic (1), and lumbosacral (1) regions. Two patients had secondary HCs that spread from another organ (lung and kidney). Patients underwent 2 to 5 surgeries during the study period, with an average follow-up time of 8.5 years (range, 7–15 years) after the first surgery. The surgical treatments included an anterior corpectomy and anterior plate for a patient with cervical localization and cystectomy for a patient with sacral localization; the other patients underwent cystectomy with corpectomy and stabilization with an anterior approach, cyst excision and laminectomy with a posterior approach, or additional posterior transpedicular screw stabilization. The patients were prescribed albendazole.

■ **CONCLUSIONS:** Spinal HC treatment is difficult, particularly in patients with vertebral and paraspinal involvement, spinal instability, and recurrence. Long-term follow-up is critical, and patients require medical and

surgical treatment, with regular clinical, radiologic, and serologic examinations. The cysts must be removed without rupture during surgery, and the surgical area must be irrigated with hypertonic saline solution to reduce the risk of recurrence.

INTRODUCTION

Spinal hydatid cyst (HC) disease is a rare but malignant form of a neglected tropical disease, and this zoonotic infection is caused by echinococcosis resulting from cestodes. Although the disease has been reported in all continents except Antarctica, HCs are more prevalent in warmer regions, such as South America, Mediterranean countries, the Middle East, New Zealand, South and Central Russia, Australia, China, and North and East Africa.¹

The HC disease is spread when eggs in the feces of echinococcosis-infected cats and dogs infect humans through fecal–oral transmission.^{2–6} The liver is the most affected organ (65%–70%), and the second most affected organ is the lung (20%–25%). Only 0.5% to 3.1% of patients experience bone involvement, and only 50% of these cases affect the vertebral column.^{5,7–13} Braithwaite and Lees¹⁴ reported that the ratio of vertebral and paravertebral localization of spinal HC disease was higher when classification was used.^{5,15–19}

Although advances in imaging methods enable easier diagnosis of the disease, the treatment of HC disease with spinal involvement is very difficult, and recurrence is common despite modern surgical and medical treatment methods.^{17,20,21} The treatment of choice for HC is surgery, and it is vital that cysts are not ruptured during the operation. If the cysts are

Key words

- Albendazole
- Hydatid cyst
- Long-term follow-up
- Recurrence
- Spinal
- Surgery

Abbreviations and Acronyms

ASIA: American Spinal Injury Association
CT: Computed tomography

HC: Hydatid cyst

MRI: Magnetic resonance imaging

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perforated, this may cause local seeding that in turn causes systemic spread of disease and, even more importantly, chronic recurrence. Curative surgery is difficult because of the involvement of bones and the infiltration of cysts into the bone. Thus, long-term results have shown high recurrence ratios.²² Other than surgery, the only other option for spinal HC is antiparasitic treatment with benzimidazole (mebendazole or albendazole) compounds.⁹

The evaluation of treatment results remains difficult because recurrence can emerge years and even decades after the initial treatment of spinal HCs. Therefore, there are very few retrospective and even fewer prospective long-term follow-up studies on spinal HC.

This study sought to identify the reasons for recurrence based on the long-term follow-up of 8 patients who received diagnoses of spinal cysts at our center.

MATERIAL AND METHODS

The demographic data, number of operations, localization, surgical procedures, surgical approach, classification of primary/secondary HC, medical treatment duration, drug use during the proposed time, achievement of infection control during the specified time, visits to the clinic regarding complaints, incidence of intraoperative cyst rupture, symptoms, and preoperative and postoperative American Spinal Injury Association (ASIA) Impairment Scale for 8 patients, who were monitored for 7 to 15 years with a diagnosis of spinal HC between 2000 and 2016 at our hospital, were evaluated together with their long-term follow-up results. Four of these patients received their initial surgeries at another clinic. Demographic data such as age, gender, symptoms, and the results of neurologic examination, direct imaging, computed tomography (CT) and magnetic resonance imaging (MRI) were used to determine localization in all patients. Spinal localization was evaluated according to the classification of Braithwaite and Lees.¹⁴ All patients underwent 2 to 5 surgical operations and were medically treated (albendazole). The patients' diagnoses were pathologically confirmed. All patients were examined in detail for the presence of HCs in other organs and systems.

RESULTS

Age and Gender

Four of the patients (50%) were male, and 4 (50%) were female. The median age was 30.75 years (range, 17–45 years) at the first surgery and 41 years (range, 24–52 years) at the time of most recent recurrence surgery.

Localization

Three (37.5%) patients had thoracic localization, 1 (12.5%) patient had thoracolumbar localization, 1 (12.5%) patient had lumbar localization, 1 (12.5%) patient had sacral localization, 1 (12.5%) patient had cervicothoracic localization, and 1 (12.5%) patient had lumbosacral localization. The HCs were vertebally and paravertebrally localized. Five patients (62.5%) showed pressure on the canal.

Foci

Two (25%) patients had secondary HCs that developed as a result of spread of disease from another organ (lungs and kidneys), and 6 patients had no other foci.

Symptoms and Neurologic Examination

Six (75%) patients had pain, 4 (50%) had loss of strength in the legs and could not walk, and 3 (37.5%) could not feel their urine or feces. Neurologic examination revealed paraparesis in 3 of the patients (37.5%), paraplegia in 1 (12.5%) patient, more pronounced quadriparesis in the lower extremity in 1 (12.5%) patient, and urinary and anal sphincter disorder in 3 (37.5%) patients.

Preoperative ASIA Impairment Scale

One patient (12.5%) was classified as ASIA A, 4 patients (50%) were classified as ASIA C, and 3 patients (37.5%) were classified as ASIA E.

Radiologic Examination

All patients underwent direct imaging and CT and MRI. Anterior pressure on the canal was detected in the patient with cervical localization; 3 of the 6 patients with thoracic and lumbar involvement had anterior pressure, and 2 had posterior pressure; the patient with sacral localization had posterior pressure; 4 patients (50%) had anterior pressure on the spinal canal, and 3 (37.5%) had posterior pressure. Two patients (25%) had kyphotic deformation, and 3 (37.5%) showed destruction of the disc gaps due to cysts.

Spreading

All cases were graded 4 or 5 according to the Braithwaite and Lees¹⁴ classification. All patients had paravertebral spreading. The thoracic localization in 1 (12.5%) patient at the time of surgery spread to the cervical area; the sacral localization in 1 (12.5%) patient spread to the lumbosacral area; and the thoracic localization in 1 (12.5%) patient spread to the thoracolumbar area.

Surgical Treatment

The patients underwent 2 to 5 surgical interventions. A total of 23 anterior, posterior, and combined operations were performed. These operations included only cyst excision by the use of anterior, posterior, or combined anteroposterior stabilization approaches. Two (25%) patients underwent posterior deformity surgery because of kyphotic deformations. According to the last examinations, 2 patients (25%) had anterior, 2 patients (25%) had posterior, and 3 (37.5%) patients had anterior plus posterior cyst excision and stabilization surgeries. One patient with cervicothoracic localization (12.5%) underwent anterior corpectomy plus anterior plate surgery (Figure 1A–E) at different times, and 1 patient with sacral localization (12.5%) underwent only posterior cystectomy. Patients with thoracic, lumbar, or thoracolumbar (Figure 2A–E, Figure 3A–F) localization underwent anterior cystectomy, corpectomy, and stabilization or posterior laminectomy and cyst excision (Figure 4) and posterior transpedicular screw stabilization or a combination of both procedures. All cysts ruptured during surgery. There

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