Spinal Manifestation of Hydatid Disease: A Case Series of 36 Patients

Abdulameer Kafaji[1], Tarek Al-Zain[2], Johannes Lemcke[3], Ferass Al-Zain[4]

Key words
- Echinococcus
- Hydatid cyst
- Hydatosis
- Spinal hydatid
- Vertebral hydatid

Abbreviations and Acronyms
CT: Computed tomography
MRI: Magnetic resonance imaging
PMMA: Polymethyl methacrylate

From the [1]Department of Neurosurgery, Nahraine Medical College, Baghdad, Iraq; [2]Orthopaedic and Neurological Rehabilitation, Naturana Hospital, Bad Salzschlirf, Germany; [3]Department of Neurosurgery, Unfallkrankenhaus Berlin, Berlin, Germany; and [4]Spine Centre, St. Joseph Hospital, Berlin, Germany

To whom correspondence should be addressed: Ferass Al Zain, M.D. [E mail: dr.ferass@al zain.de]

Citation: World Neurosurg. (2013) 80, 5:620 626. http://dx.doi.org/10.1016/j.wneu.2013.06.013

Journal homepage: www.WORLDNEUROSURGERY.org

Available online: www.sciencedirect.com

1878 8750/$ see front matter

OBJECTIVE: **Vertebral hydatid cysts are found in <1% of all cases of hydatidosis. The pathology has an infiltrative malignant nature, affecting the vertebral body with possible extension in the epidural space. This pathomechnism is associated with a high rate of morbidity, mortality, and relapse. Decompressive surgery combined with antihelminthic therapy is recommended to eradicate the disease and prevent recurrence.**

METHODS: **Between 1990 and 2007, 36 patients with a mean age of 31 years were diagnosed as having spinal hydatid disease and underwent surgery on several occasions for multiple recurrences. Combined chemotherapy with albendazole also was given.**

RESULTS: **Initial surgery and chemotherapy steadied clinical progression and functional deterioration, but with a recurrence rate of 89% and an average time to disease recurrence of 2.5 years, did not improve the known malignant course of disease in comparison to the literature.**

CONCLUSION: **The devastating and malignant course of this disease, which affects mostly young patients, demands continuous development of preventive care in endemic regions, the early detection and screening of the diseased patients, and eventually the advancement of the combined medical and surgical treatment.**

INTRODUCTION

Echinococcosis infestation in humans is a major health problem and an endemic disease in Iraq (31). The tapeworm *Echinococcus granulosus* accounts for the overwhelming majority of cases of hydatid disease. The manifestation of this disease in bone tissue ranges between 0.5% and 2%. The vertebral column is involved in 45% of the cases. The thoracic spine is the most affected, followed by the lumbar and cervical spine (23, 27). Dew in 1928 (10) and Dévé in 1948 demonstrated that the embryos of the Echinococcus are commonly lodged in the more vascular part of the spine in the vertebral body. The embryos produced a microvesicular polycystic lesion. The bony wall is then penetrated, and the cysts escape into the spinal canal, compressing the spinal cord and the intraspinal roots. The dura remains intact, and intrathecal cysts are very rare (1). Dew classified the spinal manifestations of hydatid disease as having intramedullary, intradural, epidural, vertebral, and paravertebral involvement (10).

Despite the modern development of spinal surgery and pharmacologic therapy, the infiltrative nature of spinal involvement in this disease reduces the probability of cure and increases the rate of recurrence. Although this pathology is rare, it is a serious disease with devastating consequences. A survey of case series and reports is the best way of developing management concepts to combat this malignant infectious disease.

MATERIALS AND METHODS

Clinical Presentation

Between 1990 and 2007, 36 patients (ages: 6–62 years with an average of 31 years and a male to female ratio of 1.35:1) were diagnosed and treated for spinal manifestation of hydatid disease in three Hospitals in Baghdad/Iraq (Department of Neurosurgery, Medical City teaching hospital; Department of Neurosurgery, Kadhmyia Hospital; and Nursing Home hospital). One patient (2.8%) presented with quadriplegia, 10 patients (27.8%) with backache (with/without radicular pain), 17 patients (47.2%) with varying degrees of paraplegia, 4 (11.1%) with complete paraplegia of the lower extremities, and 4 patients (11.1 %) with urinary retention.

Diagnostic Tools

In all patients, routine screening, including hematologic tests, was performed. The Casoni intradermal test was performed in 14 patients, and a positive result was found in six. Hemagglutination tests were performed in four patients and found to be positive in three. Unfortunately, because of the political events in Iraq, a large collection of patients' data was lost or destroyed.

All patients underwent X-rays of the spine, which showed the classic "moth-eaten" erosion of the vertebral body with spreading to the lamina, pedicle, and sometimes to the paravertebral and mediastinal soft tissue (Figure 1). Before 2000, all patients were investigated further with the use of computed tomography (CT). Myelography was performed only when further differentiation was needed. Before

Figure 1. Posteroanterior radiograph of the chest and cervical spine showing the erosion of the pedicle and lateral part of the second thoracal vertebral body spreading to the rib head and paravertebral tissue.

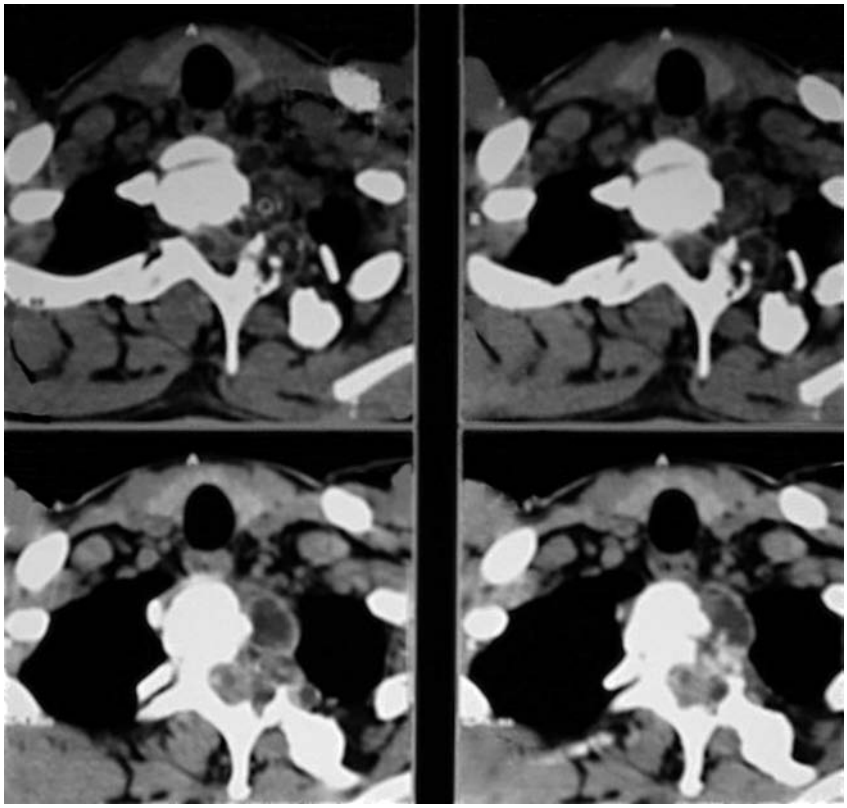

Figure 2. Axial view of a computed tomographic image of the second thoracic vertebral body showing cystic infiltration and destruction of the vertebral body with invasion of the spinal canal and involvement of the spinal cord. Note the intradural small hypodense cystic lesions.

myelography, CT was performed to localize the major focus of the disease to prevent puncturing of the cyst and the spread of the disease (Figure 2). After the year 2000 and the introduction of a magnetic resonance imaging (MRI) facility in Iraq, it became the tool of choice in diagnosing the disease and during the follow-up of patients (Figure 3).

The distribution of the spinal manifestation was found to be as follows: 1 patient (2.8%) with cervical spine affection, 23 patients (63.9%) with thoracic spine manifestation, 10 patients (27.8%) with lumbar spine, and 2 patients (5.5%) with lumbosacral affection. According to Dew's classification, all our patients had primarily a vertebral body involvement with epidural infiltration occurring later. An intradural spread of the disease was only found in a recurrent case after intraoperative duratomy and in one patient with multiorganic disease of cerebral, renal, and pericardial hydatosis.

To explore further hydatid metastasis, all patients underwent organ screening with chest x-ray and abdominal ultrasound. A CT scan of the thorax and abdomen was performed if suspected hydatid masses were found within initial diagnostic measures. CT scans of the head and neck were performed in special cases upon suspicion of central nervous system or orbital hydatid. In five patients, we found only one additional organ manifestation (three with pulmonary and two with hepatic hydatid cyst), three patients had two additional organ manifestations (one with hepatic and renal hydatid, one hepatic and orbital hydatid, and one with hepatic and ovarian hydatid). These patients all had thoracic spine involvement. In the patients with cervical, lumbar, and lumbosacral involvement, we found no other organ involvement on initial presentation.

TREATMENT

A total of 69 operative procedures were performed in these 36 patients; 18 of the 23 patients with thoracic hydatid manifestation underwent a ventral approach via thoracotomy with debridement of the affected vertebral body and defect filling with iliac bone autograft. Postoperative cast bracing was done in ambulatory patients with extensive debridement or resection. One thoracic case had a combined anterior and posterior approach for extensive vertebral infiltration with severe compression of the spinal cord but died within 1 year of the disseminated disease. The remaining four patients with thoracic spine hydatid disease were treated through a posterior approach because of the severe compression of the spinal cord with an acute paraplegia. All the lumbar and lumbosacral cases were approached from posterior by laminectomy and sometimes with additional partial dorsal transpedicular vertebral debridement. The only patient with cervical hydatid involvement underwent posterior decompression surgery four times in three successive years; after the last operation he developed instability of the spine at the treated level, and the initial paralysis deteriorated into complete quadriplegia. Occipital traction was applied then, but eventually the patient died of multiorgan failure as a consequence of severe neurologic deficits.

Almost all the recurrences were regional at the level of previous surgery and needed