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Vertebral hydatid cyst infection (*Echinococcus granulosus*): a case report

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Abstract Spinal echinococcosis is a rare but serious condition. Within bone tissue hydatid cysts enlarge by daughter cyst formation. The value of drug treatment in bone echinococcosis is questionable. The aim of surgery is therefore removal of all the cysts. The best way to achieve this is at the first operation early in the progress of the disease. An anterior or circumferential approach is

generally required to give the necessary accessibility. Owing to diffuse spread of the infection within the bone and the canal, recurrence is common. If neurological deterioration occurs, reintervention is necessary.

Key words Spinal hydatid cyst infection · Surgical treatment

Introduction

Two species of the parasitic tapeworm *taenia echinococcus* can affect man. *Echinococcus multilocularis* is manifested as a rare, more malignant alveolar form. *Echinococcus granulosus*, characterized by cyst formation, is more benign and remains a common condition in many rural parts of the world [4]. The disease most often affects liver and lung. Only 0.5–2% is located in the skeletal system [2], and in approximately 50% of these cases the spinal column is involved [9]. The purpose of the present paper is to draw attention to the formidable problem that arises if a hydatid infection is located in the spinal column, and to describe how this surgical challenge was handled in one case.

Case report

A 36-year-old male Iraqi Kurd presented in May 1990 at our Refugee Medical Centre with a history of progressive paraparesis. He had previously been operated on twice in Teheran, in 1985 and 1989, with decompressive laminectomies and excision of hydatid cysts in the thoracolumbar area. Pharmacotherapy with Albendazole 10 mg/kg bodyweight/day was started and has since been given more or less continuously. In August 1990, a large abdominal cyst was explored at the department of general surgery. The cyst was found to be adherent to the large vessels and to communicate with the spinal canal. The capsule wall was opened and the content was drained. A 20% NaCl solution was instilled. The cyst was then removed, leaving only the posterior part of the capsule. One month later the patient was referred to the spinal unit because of back pain and paraparesis. On admission he was wheelchair bound. On examination he was kyphotic in the thoracolumbar area. He had no voluntary control of the feet, but some control remained

Fig. 1 Lateral preoperative radiograph showing compression of T11 and T12 with a local gibbus of 45°

Fig. 2 Thoracolumbar MR image, T1-weighted sagittal view, shows narrowing of the spinal canal at the level of the destroyed vertebrae T11 and T12

Fig. 3 Lateral postoperative radiograph illustrating the instrumentation with Cotrel Dubousset rods from T7 to L5

Fig. 4 Lateral view of the thoracolumbar spine 5 years after surgery, showing that the anterior fusion has healed

in the knees and hips. Both knee and ankle jerks were absent, and sensation was impaired over the feet and lower legs. He complained of defective bladder control. Routine plain radiographs showed compression of vertebrae T11 and T12. A local gibbus of 45° was measured between T10 and L1 (Fig. 1). MRI showed destruction of T11 and T12 (Fig. 2). Because the laminae of T11–L3 had previously been removed, the deformity was certain to progress, resulting in paraplegia. At surgery it was obvious that the infection was more extensive and also involved L1, L2, and L3. Cotrel Dubousset rods were used for fixation from T7 to L5. Both the proximal and distal ends were fixated by claws. The bone was very osteoporotic and sublaminar wires were used to strengthen the fixation (Fig. 3). Bone transplants for fusion were taken from the iliac crest. The peroperative bleeding was 6000 ml and the anterior operation was therefore postponed. After 2 months an anterior decompression and fusion from T11 to L1 was performed using a fibula bone graft. The patient was mobilized in a brace for 6

months owing to the osteoporosis. Functionally he was able to walk on crutches. One year later the weakness of the legs had progressed, and he was no longer able to walk. Myelography showed extradural blockage at T12. Costotransversectomy was performed on T11 and T12. A great number of cysts welled up from T11, T12, and L1 and even from the inside of the canal. These were carefully removed. The posterior parts of the infected vertebral bodies were extirpated. The posterior fusion had not healed, and further bone transplants were added to both the anterior and the posterior fusions. The weakening after the removal of infected tissue justified the use of a brace for 6 months postoperatively. The patient regained and has retained until today, more than 6 years after presentation, his ability to walk on crutches. His muscular strength is 3–4 in all the muscle groups of the legs except for dorsiflexion of the feet, which is 0. He still has a neurogenic bladder with detrusor hyperreflexia. Routine radiographs show that the anterior fusion has healed (Fig. 4).

Discussion

Thirty-four percent of stray dogs in Iran are reported to be infected with *Echinococcus granulosus* [12]. The adult worm inhabits the small intestine of its definite host, usually a dog. The ova that are shed in its excreta are protected by a chitinous shell, and can survive in adverse climatic conditions for many weeks. Ingested by its intermediate host, preferably a sheep but exceptionally a human, the embryo loses its protective shell and penetrates the mucosa of the duodenum to enter the mesenteric venules and is carried with the portal stream to the liver. It may pass the liver sinusoidal system to be discharged directly into the venous return of the heart. The lungs are the second most common localization for the cysts. On occasion, a blood-born embryo can escape both the filters of the liver and lung and enter the arterial system, whence it can reach every organ including the skeleton. Within 3 weeks the embryo develops into a larva, and a cyst is formed. The natural history may involve calcification and death of the cyst, but more frequently it enlarges radially. Depending on nutritional support the parasite maintains a peaceful relationship with its host, causing no general systemic effects, and the growth can be measured over years and decades. The pericyst is the outermost of the three layers of the cyst wall, and contains exclusively cells of host origin. The acellular laminated membrane is always separable from the pericyst. The innermost germinal layer is the origin of the fertile worm heads. Endogenous vesiculation may result in formation of daughter cysts within the parental cyst. The fluid pressure inside the cyst is high, and even minor trauma is enough to make it burst. The outermost lamina of the laminated membrane is under the highest tension, so if the cyst bursts the whole membrane becomes everted exposing all the fertile elements to the host tissues. If remains from an infected sheep are eaten by a dog, the larvae develop into adult worms thus completing the life cycle of the parasite [13].

The rare cases of hydatid disease located in the skeletal system are exceptional owing to the resistant nature and lack of defensive reaction of the bone [2]. The pericyst layer is missing, and as the cyst penetrates between the bony trabeculae in the direction of least resistance, small diverticulae are formed by exogenous vesiculation and these can separate from the main cyst, resulting in a mass of cysts. As the parasite enlarges, the bone quietly erodes away. In time the process breaks through the cortex, and once beyond the bony tissues the parasite forms

well-circumscribed cysts with a pericyst layer, as in any other tissue [4]. The symptoms of bone echinococcosis are pain, although seldom severe, local swelling, and pathological fracture [12]. The basic manner of progression of bone echinococcosis in the vertebral column is similar to that in other bones, i.e. chronicity without systemic disturbances, but ultimately the disease becomes clinically manifest [8]. Pain, deformity, and weakness may result from either collapse of the spine or by extension of the process beyond skeletal confines. It is most serious, of course, if the disease breaks into the spinal canal. Cysts can migrate up and down the canal, explaining why compression of the cord can occur at a level distant from the primary focus [1]. Neurological deterioration is usually very slow, but will result in paraplegia in 25–50% of cases [15]. Though the parasite is thought to infect mainly during childhood, the onset of symptoms more commonly takes place between 21 and 40 years of age [2]. The radiographic features are not pathognomonic, but vertebral destruction, paraspinal extension, and particularly involvement of a contiguous rib are suggestive [3]. CT shows bone destruction as well as para- and intraspinal soft tissue masses. MRI is essential for evaluating the intra- and paraspinal extension of the disease and for delineating the spinal cord [11]. Myelography still has a place in postoperative cases. Tuberculous, metastatic, and other osteolytic bone states are among the differential diagnosis [6]. The most specific antigen is antigen 5 (ARC5) [14]. Serological sensitivities of 80–100% in liver cysts, 50–56% in lung cysts, and 25–56% in other organs are reported [10].

Albendazole is the current drug of choice. Its action is likely to be parasitostatic rather than paracitidal, and therapeutic failures have been reported. Cyst size is a main determinant for response to drug therapy, less important is cyst location, with the exception of bone. No data are available on drug penetration into bone tissue, but the clinical impression is that bone cysts respond less well to drug treatment than do other cysts [7].

The aim of surgery is total removal of all the infected tissue. The best chance of cure is at the first operation early in the course of the disease. Laminectomy is generally not sufficient, and anterior or circumferential decompression is advocated. In general, a remission of symptoms is achieved, but because of the difficulty in eliminating the diffusely spread cysts inside the bone and within the canal, the disease is more likely to be controlled rather than cured. If neurological deterioration occurs, reoperation is mandatory [5].

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