The use of polymethylmethacrylate in the management of hydatid disease of bone

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Hydatic disease of bone is rare. It probably represents between 0.5% and 4% of all human hydatid disease and, in about 60% of patients, affects the spine or pelvis. Between 1986 and 1998, we treated 15 cases of bone hydatidosis. Curettage, swabbing with povidone iodine and filling the defect with polymethylmethacrylate (PMMA) were carried out in ten patients. Three of these had a recurrence after five years, but seven had no signs of relapse during a mean follow-up of 52 months. We believe that the combination of antihelminthic therapy, wide resection and the use of PMMA gives the best outcome in the treatment of bone hydatidosis.

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Hydatid disease is caused by a parasitic tapeworm (*Echinococcus*). The disease was probably known to Hippocrates, but the complete life-cycle was not documented until the 19th century.¹ *Echinococcus granulosus*, the species most responsible for hydatid disease, is transmitted by sheep as the intermediate host. The precise incidence of human hydatid disease is not known, but it remains a serious health problem in the sheep-rearing districts of the Mediterranean countries, the UK, Australia, South America, Iraq and Iran.²⁻⁴

Bone hydatidosis is rare, and the incidence in relation to involvement of other organs varies from 0.5% to 4% in different countries.^{5,6} About 60% of cases of bone hydatidosis affect the spine and pelvis, 28% the long bones, and 8% the ribs and scapula. The lesions in bone may lie dormant for 10 to 20 years.⁷

The reported prognosis for bone hydatidosis is poor since

recurrence is frequent and remission rather than cure has been the aim.^{6,8-12} Antihelminthic drugs may improve the prognosis, but radical surgery is likely to remain the most effective treatment for the foreseeable future. We describe our experience of the surgical management of bone hydatidosis using polymethylmethacrylate (PMMA) as a space filler for bones destroyed by hydatid infection.

Patients and Methods

Between 1986 and 1998, we diagnosed and treated 15 cases of bone hydatidosis in our clinic. There were ten men and five women with a mean age of 38 years (24 to 68). In all patients plain radiography, ultrasonography and CT were used to establish the diagnosis. MRI was also used in ten patients. The diagnosis was confirmed by both gross and microscopic examination. Ten of the patients had had surgery previously in other clinics for bone and/or soft-tissue lesions, and suffered relapses which required further treatment. Two of these had an initial misdiagnosis of tuberculosis and had received antituberculous drugs before referral. Relapse had occurred three months to two years after the primary operation.

In all patients the presenting symptom was pain. In two there was a pathological fracture and in a further two there was a neurological complication. The mean duration of symptoms was 27.6 months (6 to 72). In nine patients (60%) the hydatidosis was localised in the spine and pelvis (Table I).

All received surgical treatment. Curettage and swabbing with povidone iodine were carried out and the defect was filled with PMMA in ten patients (Table I). In four (cases 11 to 14) PMMA could not be used because the lesion involved the spinal canal, but we undertook a wide excision of the involved bone. In the patient with talar involvement (case 15) only curettage and grafting were performed. Six of the ten patients in whom PMMA was used had at least one operation before referral to us, in which curettage and grafting were carried out. The mean time to recurrence for these patients was 11 months (3 to 24).

The administration of an antihelminthic drug, mebendazole, 60 mg/kg/day, was planned for 14 patients for a period of six months, but only two completed the course of treatment. The remainder stopped because of side-effects or

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Table I. Details of the 15 patients with hydatidosis of the bone

Case	Age (yrs)	Gender	Localisation	Symptoms	Duration of symptoms (mths)	Follow-up (mths)	Previous surgery*	Operation*	Duration of chemotherapy (mths)	Results†
1	33	M	Femur prox + iliac bone	Pain	24	127	1. Renal transplantaion 2. C+G (twice)	1. C+G 2. C+PMMA 3. Custom-made prosthesis+PMMA	–	NED
2	62	F	Femur prox	Pain+path fracture	35	119	C+G	1. Partial prosthesis+PMMA 2. Debridement 3. Custom-made total prosthesis+PMMA	6	NED
3	42	M	Femur prox	Pain	45	72	Intra-abd. cyst excision+C+G	1. C+PMMA+internal fixation	3	NED
4	29	F	Femur prox	Pain+path fracture	8	30	C+external fixation	1. Excision+custom-made partial prosthesis+PMMA	4	LFU
5	3	M	Tibia	Pain	24	81	C+G (twice)	1. C 2. C+PMMA	3	NED
6	40	M	Humerus	Pain	24	48	–	1. C+PMMA	4	NED
7	23	F	Acetabular roof	Pain	18	60	–	1. C+PMMA	3	NED
8	41	M	Iliac bone	Pain	30	65	–	1. C+PMMA	3	AWD
9	68	M	L5+iliac bone	Back pain	12	24	1. Nephrectomy 2. Abdominal cyst excision 3. C+G (twice)	1. C+PMMA	2	Died
10	60	M	L5+sacrum	Pain+neuro complications	48	68	Posterior decomp+fusion	1. Anterior decompression +PMMA	6	AWD
11	21	M	L5+sacrum	Pain+peroneal palsy	6	108	Discectomy	1. Posterior instrumentation	3	AWD
12	25	M	L5+sacrum	Pain	72	96	Discectomy	1. Posterior instrumentation	3	NED
13	30	F	T5-T6	Pain	20	28	1. Intrathoracic cyst excision 2. Epidural cyst excision	1. Anterior+fusion	3	LFU
14	20	F	T12-L2	Back pain	28	56	–	1. C+decompression+fusion 2. Posterior instrumentation	4	AWD
15	23	M	Talus	Pain	20	70	C	1. C+G 2. Amputation	3	NED

* C, curettage; G, grafting

† NED, no evidence of disease; LFU, lost to follow-up; AWD, alive with disease

lack of compliance. One patient (case 1) with chronic renal failure did not receive mebendazole.

Results

The mean follow-up was 70.1 months (24 to 127). In those in whom we used PMMA (cases 1 to 10), it was 69.4 months (24 to 96). In two patients, cases 1 and 5, in whom we had used curettage with or without grafting, the disease recurred at two and six months, respectively. These patients were reoperated on with further curettage and insertion of PMMA. There was no subsequent evidence of disease in one patient (case 5) but in the other (case 1) 84 months after curettage, cementation and prophylactic internal fixation with an intramedullary nail (Fig. 1), there was recurrence, and wide resection and reconstruction with a modular tumour prosthesis were performed. He currently has no evidence of active disease. Late infection (*Staphylococcus aureus*) occurred in one patient (case 2) six months after the initial procedure and a two-stage revision with a custom-made prosthesis and PMMA was performed. She has no current active disease. Two patients (cases 8 and 10) are alive with active disease; the times to recurrence were 62 and 64 months, respectively. One patient (case 4, Fig. 2) was lost to follow-up after 30 months and another (case 9) died at 24 months. Both had no evidence of hydatid disease

at that time. The remaining three patients in the PMMA group (cases 3, 6 and 7) are free from disease.

In four patients (cases 11 to 14) the lesions were close to the spinal canal which was also involved, and PMMA was therefore not used. The results were not good in these patients. Two are alive with disease, one was lost to follow-up, but had persistent symptoms. One had no evidence of disease after 96 months. The mean duration to recurrence was 16 months.

In one patient (case 15), curettage and grafting were carried out because the lesion was small and closely related to the ankle. He was lost to follow-up after six months, but when he returned after six years the disease involved all the tarsal bones and distal tibia, with a fistula which necessitated below-knee amputation.

Discussion

Bone echinococcosis occurs in adults. Distortion of bone architecture is slow because of the resistance of bone.⁵ The mechanism of spread into and through the bone has not been fully determined. The parasites must pass through the liver and the lung filters to enter the arterial circulation. The clinical features of hydatid disease of bone may take 10 to 20 years to become manifest.¹³

The diagnosis may be easily overlooked unless there is a

Fig. 1

Radiograph of a 33-year-old man (case 1) who had a recurrence after attempted curettage and grafting for a hydatid cyst. He also had chronic renal failure. The cortical expansion and thinning as shown are characteristic features of bone hydatidosis.

strong element of suspicion, and usually is determined and confirmed only after surgery or biopsy. There are no specific characteristics to distinguish it from other more common causes of bone pathology. It may mimic spinal tuberculosis,¹⁴ pyogenic infection, and malignant disease. The most common radiological characteristic of the osseous lesion is a combination of multilocular cysts and reactive sclerosis as in a honeycomb, involving a large expanse of bone.¹⁵ Osteolysis is usually seen, sometimes associated with expansion of the bone and thinning of the cortex.^{16,17} These signs are not specific, but large lesions with soft-tissue calcification are highly suggestive of echinococcosis.^{16,18}

The only definitive treatment when bone is involved is complete resection of the involved area with a wide healthy margin. This may be difficult, but incomplete removal is followed by recurrence.^{6,11,14} Surgical excision and curettage can remove only macroscopic cysts, and most scolecidal agents such as formalin and hypertonic saline, do not kill all microscopic daughter cells. We decided to use PMMA to fill defects after excising the cyst. Although some surgeons recommend a combination of surgical resec-

tion and bone grafting,^{2,19} grafts can be invaded by hydatid extension or recurrence. PMMA is preferred because the elevation of temperature in the polymerising cement has a necrotising effect which may kill the daughter cysts.²⁰ Monomer released by PMMA is reported to be toxic to living cells as also are the free radicals which are released during polymerisation.^{21,22}

For the first five years of follow-up there was no recurrence in those patients in whom PMMA had been used and the results seemed to be excellent. There were, however, recurrences in three of seven patients who were followed up for more than five years, and thus curettage and PMMA may not eradicate bone hydatidosis. The use of PMMA has lowered the rate of recurrence and therefore its use is recommended if there is doubt, after resecting a cyst, that there may be small residual cysts. In a previous report³

Fig. 2

Radiograph of a 29-year-old woman (case 4) who presented with a history of pathological fracture treated elsewhere by external fixation. The multilocular cysts and reactive sclerosis were treated by resection of the infected bone and soft tissues, followed by implantation of a custom-made prosthesis and PMMA. The histopathological diagnosis was of a hydatid cyst. She was free from symptoms 30 months after operation.

surgical drainage alone gave a rate of recurrence of 50%, as did posterior decompression alone after one to six years.²³ In another report of 19 patients with bone hydatidosis each had surgery one to eight times, with 14 having at least two operations.²⁴

It is possible that a combination of chemotherapy and surgery could be more efficacious,²⁵ but the preoperative use of the drugs is not always possible, as the diagnosis may only be established after surgery.

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