The treatment of sacral giant-cell tumours by serial arterial embolisation

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Giant-cell tumours of the sacrum are difficult to treat. Surgery carries a high risk of morbidity, local recurrence and mortality. Radiation is effective in some patients, but has a risk of malignant change. We evaluated the effectiveness of serial arterial embolisation as an alternative to surgery. Five patients with giant-cell tumours of the sacrum which had been primarily treated by serial embolisation were retrospectively reviewed for changes in the size of the tumour. In four the symptoms resolved with full return of function and arrest in the growth of the tumour. They remained free from growth, recurrence, or metastases at follow-up (4 to 17 years). One patient died from metastatic disease within 18 months of the initial diagnosis.

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Giant-cell tumour of bone is an aggressive benign neoplasm consisting of a vascularised network of spindle-shaped stromal cells surrounding multinucleated giant cells. Although formally classified as benign, sarcomatous change has been reported, often in lesions which have previously been treated by radiation. The sacrum is the most common site for the tumour in the axial skeleton and the fourth most common site after the distal femur, the proximal tibia and the distal radius. In the Mayo Clinic series, giant-cell tumours of the sacrum represented 8% of 425 tumours seen over a period of 26 years. Although rare, such tumours present a difficult challenge to the surgeon and their treatment remains controversial.

Complete resection may be curative but technically difficult and is often complicated by extensive hemorrhage, infection and neurological complications. Curettage alone may be used in the conservative treatment of giant-cell tumours, but is not effective in preventing recurrence, and postoperative adjuvant therapies have limited application. Radiotherapy has marginal benefit and the risk of malignant change.

The treatment of pelvic tumours, and specifically giant-cell tumours, by arterial occlusion has mainly been used as a method of decreasing blood loss intraoperatively, as a palliative measure for inoperable lesions, and after exhaustion of other forms of treatment. This technique has been shown to devascularise tumours, reduce their size, cause calcification of their margins and alleviate pain. We present five patients with sacral giant-cell tumours which were treated primarily by serial embolisation.

Patients and Methods

We retrospectively reviewed the medical records of five patients with giant-cell tumours of the sacrum which had been primarily treated by serial arterial embolisation between 1984 and 1997. There were four women and one man, with a mean age of 27 years (19 to 42) at the time of diagnosis. All had presented with pain in the buttock or the back for a mean of six months. The lesions were identified by CT and/or MRI (Fig. 1). Three patients underwent open biopsy and two had needle biopsies, the diagnosis being confirmed by histological examination.

Arterial embolisation was used as the initial form of treatment in all patients and the sole treatment in four. They received serial embolisation of the main vessels feeding the tumour. These vessels were identified by angiography and embolised by steel coils, gelfoam, and polyvinyl alcohol (Fig. 2). The vessels which were embolised included the internal iliac, the lateral sacral, the lumbar, the iliohypogastric, and the median sacral arteries. Success of embolisation was confirmed by further angiography. In one patient the initial embolisation was not effective and re-embolisation was undertaken two weeks later. All patients had further angiography at four- to six-week intervals with embolisation of any new vessels until no new vessels were noted. Subsequent angiography was undertaken at six and 18 months.
Few angiograms were obtained after this time. The patients were thereafter closely monitored by MRI or CT of the pelvis and radiography or CT of the chest. The mean follow-up was 6.7 years (4 to 17).

**Results**

The patients were followed clinically and radiologically. The presence of pain, radiological changes in the size of the lesion, calcification of its margins and the development of pulmonary metastases were the criteria which were used to evaluate the outcome. In order to assess the size of the lesion the greatest dimensions in the sagittal, axial, and coronal planes were measured. Tumour volumes were not calculated since we could not obtain the exact corresponding measurements at different time intervals. Some patients had CT initially and MRI more recently. We compared the results with published data on the treatment of these lesions.

All patients had relief from pain after embolisation. The tumour increased in size in two patients by less than 1 cm. The maximum decrease in size in any one plane was 2.7 cm. Assessment of the remaining scans showed minimal or no change. Sclerotic margins were noted on MRI after embolisation in three patients within two years of treatment (Fig. 3).

Pulmonary lesions developed in one patient. One had sacroiliac instability and underwent fusion from L4 to the
sacrum two years after diagnosis. After surgery her symptoms improved. Four patients returned to full function and continue to pursue active lives (Table I). Two report mild discomfort after prolonged sitting and two have discomfort on lifting or exertion. All have annual clinical and radiological review. There was little change in the appearance of their tumours three to nine years after the most recent embolisation. They have remained free from metastases and symptomatic disease at a mean of 6.7 years (4 to 17) after diagnosis.

In the fifth patient resolution of pain was temporary. One month later she had further back pain. Six months after embolisation, growth of the tumour was seen on imaging studies and 500 Gy of external beam radiation were administered. This did not alleviate her symptoms or halt the progression of the tumour. At the time of initial presentation a chest radiograph was normal, but six months after irradiation she developed pulmonary metastases. Pathologists at several centres reported a further biopsy as benign giant-cell tumour. She received multiagent systemic chemotherapy, but developed multiple metastases and died 18 months after the initial diagnosis.

## Discussion

The sacrum is the most common site for giant-cell tumours in the axial skeleton and these are very difficult to treat.\(^1,4,5\)

Wide resection carries a significant morbidity and mortality. Wang-Peng et al\(^11\) described a series of 87 primary sacral tumours which were treated surgically. There was an incidence of wound infection of 12.6% and a mortality rate of 11%. Simpson et al\(^8\) described 12 patients who underwent partial resection of a large sacral lesion. One died from massive haemorrhage and six suffered wound complications. Resection of a considerable portion of the sacrum

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**Table I.** Details of the four patients with giant-cell tumour of the bone treated successfully by embolisation

<table>
<thead>
<tr>
<th>Case</th>
<th>Gender</th>
<th>Age at diagnosis (yrs)</th>
<th>Number of embolisations</th>
<th>Follow-up (yrs)</th>
<th>Measurement of the maximum dimensions (cm) in the sagittal, axial and coronal planes</th>
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<td>F</td>
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</table>
has a high incidence of neurological complications which may affect bowel and bladder control and may lead to impotence in men.

More conservative surgical treatment has a high risk of local recurrence. Campbell and Bonfiglio reported a recurrence rate of 100% in five sacral giant-cell tumours of bone treated by curettage alone and a recurrence rate of 33% was reported by Tsurcote et al. Marcove et al reported a recurrence rate of 23% after curettage and cryotherapy.

Postoperative adjuvant therapy has a limited place in the treatment of giant-cell tumours. Radiotherapy has been abandoned since limited therapeutic benefit is combined with a risk of malignant change. Of 218 patients with giant-cell tumours of bone reported by Goldenberg et al, 46 received radiotherapy as either primary or secondary treatment and 29 developed recurrence or metastases. Three of these died from post-irradiation sarcoma at nine, 13, and 31 years after treatment. Tsurcote et al described 21 patients with sacral giant-cell tumours who received radiotherapy as either primary or secondary treatment. Three developed local malignant change. The mean interval between radiotherapy and the diagnosis of malignancy was ten years.

Chakravarti, Spiro and Hug used megavoltage radiotherapy for the treatment of inoperable giant-cell tumours. No malignant change was seen in the 20 patients who received a single course of high-voltage radiation. Of the five patients with sacral lesions there was a local recurrence in two, five and eight months after treatment. These patients were followed for a mean of 7.8 years and longer follow-up is required in order to assess the incidence of sarcomatous change after megavoltage radiation.

Embolisation has been used for inoperable tumours after the failure of other forms of treatment. Interest in arterial embolisation of pelvic tumours followed its success as a palliative measure, particularly after the resolution of pain in patients with renal-cell carcinoma. Feldman et al alleviated the pain of metastases in the right ilium by embolisation of the internal iliac and lumbar arteries. Also in 1975, Goldstein, Medellin and Beydoun used embolisation for renal-cell carcinoma. In 1979, Wallace et al described arterial occlusive treatment, using gelfoam and stainless-steel coils in nine patients with primary and secondary tumours of the pelvis including three with giant-cell tumours. Eight patients reported relief from pain and two with giant-cell tumours developed a calcified margin after embolisation and the tumours decreased in size by 15%.

Similarly, Chuang et al treated ten patients with inoperable giant-cell tumours and aneurysmal bone cysts by arterial embolisation using gelfoam and stainless-steel coils as a palliative measure. Four had previously failed to respond to both chemotherapy and irradiation and one to chemotherapy alone. In four patients, further embolisations were required to occlude the blood supply to the tumours completely. Seven patients had relief from pain after treatment. Increased calcification of the margin of the tumours was seen in five. Complications included one patient with a foot drop and one with paraesthesiae in the foot.

Walker et al treated a sacral giant-cell tumour in a 12-year-old girl by arterial embolisation using activated microfibrillar collagen. She had temporary relief from pain, but further embolisation was required after CT had shown growth of the lesion and angiography had demonstrated increased vascularity. The sacral area was irradiated three months after embolisation and at follow-up at one year she remained free from pain, without metastases and with regression and increased calcification of the tumour.

In our series, we examined the efficacy of serial arterial embolisation as curative treatment. Embolisation was the primary therapy after histological confirmation of the diagnosis which therefore obviated any exposure to chemotherapy or radiation. Four of five patients responded to this treatment alone, with relief from pain and arrested growth of the tumour.

The main vessels supplying the tumour were initially identified by angiography and embolisation was achieved using gelfoam, stainless-steel coils and polyvinyl alcohol. Repeat angiography was used to evaluate the success of the treatment. Persistent vascularity of the tumour was treated by further embolisation at monthly intervals until there was decreased blood flow on angiography, or the symptoms resolved. Patients were then followed closely by clinical and radiological assessment of the lesions and chest radiography at regular intervals. Any return of symptoms or radiological progression of the tumour was evaluated and treated by further angiography and embolisation. We have not experienced any of the occasional complications previously reported after embolisation secondary to ischaemic nerve injury.

The risks of nerve injury should, however, be described to patients before treatment.

One patient had progression of the disease with increase in the size of the tumour and persistent symptoms. She received radiation followed by chemotherapy and died from metastatic disease within 18 months of the initial diagnosis. Although repeated biopsies showed it to be benign, we suspect that this was a malignant lesion.

Four patients had relief from pain, increased mobility, and arrest of growth of the tumour after embolisation. They remain active with a full range of movement in their lower limbs and normal bowel and bladder function.

Giant-cell tumours of the sacrum are rare and treatment is difficult because of their site and aggressive behaviour. Our study demonstrates a minimally invasive technique for treating a challenging disease with good outcomes. Embolisation deserves consideration as a primary and possibly the sole treatment of giant-cell tumours of the sacrum. It requires an initial angiogram before embolisation, followed by further assessment by CT or MRI and also an angiogram and possible embolisation if there is persistent or renewed vascularity, at intervals of one or two months for a period of six months. Thereafter, angiography should be carried out if there are persistent...
symptoms. Treatment requires close clinical and radiological follow-up and a full commitment on the part of the patient and surgeon.

No benefits in any form have been received or will be received from a commercial party related directly or indirectly to the subject of this article.

References


