Bone haemangiomas are uncommon lesions, occurring in the skull or spine. A solitary haemangioma in the diaphysis of a long bone is rare. We retrospectively investigated six patients who presented with a solitary haemangioma in a long bone diaphysis. After segmental bone resection, the bone defect was replaced by a bone autograft. Patients were reviewed clinically and with radiographs. The mean follow-up was 6 years (range: 1-20 years). At the time of latest follow-up, no patient had a recurrence. Postoperative complications were one wound necrosis and one superficial wound infection. Union of the gap filling graft with the host bone was achieved in all patients at an average of 4 months (range: 3-8 months). The average Musculoskeletal Tumor Society functional score was 77% (range: 53%-90%) of normal at 6 months postoperatively, and 97% (range: 95%-99%) at the last follow-up evaluation. Segmental resection for solitary haemangioma and reconstruction with autologous bone graft can be considered as a suitable treatment option.

Keywords: bone tumour; haemangioma; bone grafting; diaphysis.

INTRODUCTION

Bone haemangiomas are uncommon lesions, accounting for approximately 0.5-1% of all intraosseous tumours (1) and 1% of all primary bone tumours (2). The most frequent sites of involvement are vertebrae and calvarium (3). The involvement of long tubular bones is rare, the metadiaphysis being the most commonly affected (4-5). The diaphysis location for a long bone haemangioma is rare, and difficult in radiological and histological differential diagnosis (6). Orthopaedic oncologists consequently have limited experience in the management of a solitary haemangioma in long bone diaphyses. The purpose of the present review is to report on the functional and oncologic results in six patients who underwent segmental resection and reconstruction with autologous bone graft for a solitary haemangioma in long bone diaphyses.
PATIENTS AND METHODS

Six patients diagnosed with a solitary haemangioma in a long bone diaphysis during the period 1990-2010 were included in the study. There were four females and two males with an average age of 42 years (range: 27-58). The ulna was affected in two cases, the clavicle in two cases, the radius in one case and the humerus in one case. Two patients had pain and partial restriction of the range of motion of adjacent joints. The other four patients complained only of a painless swelling or soft tissue enlargement at the affected long bone diaphyses. All patients were investigated with X-rays, CT-scan and MRI scan of the involved long bone. Open biopsy was done in one case, while five patients underwent needle biopsy under CT guidance.

The surgical technique varied depending on the anatomical location of the tumour. The extent of resection of the tumour was measured on radiographs. Resection of the periosteum was not attempted. After tumour resection, an autograft was harvested from the iliac bone (if segmental loss < 5 cm) (Fig. 1) or fibula (if segmental loss ≥ 5 cm). A locking plate was used for autograft fixation.

Patients were followed with radiographs, and union was assessed postoperatively. The functional outcome was evaluated according to the Musculoskeletal Tumor Society score (MSTS) (7).

RESULTS

The outcomes are summarized in Tables I and II. At the time of review, all patients were alive with a mean follow-up of 6 years (1-20 years). There was no local recurrence. The mean operating time was 73 minutes (60 min to 85 min), and intraoperative blood loss was 133 ml (100 ml to 200 ml). Two patients had wound problems: one patient with wound necrosis required surgical debridement, and one superficial wound infection was treated conservatively with antibiotics. The mean length of bone resection was 5 ± 2 cm. The mean time to union was 4 months (3 to 8), and delayed union occurred at the distal junction in two of six patients. These delayed unions were not treated surgically and healed with time. The average MSTS functional score was 77% of normal (53%-90%) for all patients at 6 months postoperatively, and 97% (95%-99%) at the last follow-up evaluation.

DISCUSSION

Intraosseous haemangiomas are rare benign tumours of vascular nature (7). Females, aged 40-50 years, are more frequently affected than males, in the ratio 3:1 (i). Primary skeletal haemangioma of the extremities is uncommon, and occurs predominantly in the lower extremity (73%) usually in the long bones, with the tibia and femur being the
most common sites (8). In the current study, we present six patients with a solitary haemangioma in the diaphyses of upper limb bones.

Clinical presentation can vary from an incidental finding to local pain, swelling and/ or pathological fracture, according to localization, size and number of lesions (9). Plain radiographs show a classic coarse trabecular bone pattern or soap bubble appearance, and most lesions show cortical destruction (10). CT reveals lytic lesions that exhibit a spiculated pattern known as Irish lace (11). MRI displays hypo- to hyperintense appearance on T1-weighted sequence depending on the amount of adipose tissue, and hyperintense on T2-weighted sequence due to vascularity (12). Biopsy can provide a diagnostic orientation, but is better avoided for large haemangioma, because of the risk of haemorrhage. Differential diagnosis includes fibrous dysplasia, sarcoma, metastatic disease and multiple myeloma.

Management is controversial. Errani et al noted no local recurrence in 8 of 11 patients with epithelioid haemangioma (EH) in bone treated with intraleisonal curettage (13). Therefore, they concluded that EH of bone could be treated with intraleisonal curettage, whereas EH of soft tissue should be treated with marginal/wide excision. However, Satoh et al (14) believed that once bone haemangiomas have been diagnosed, the definitive treatment should be radical en bloc resection including surrounding intact tissue, to guarantee no recurrence and avoid the risk of massive bleeding. In the current study, six patients with a solitary haemangioma in long bone diaphyses underwent segmental resection of their tumours. Despite the fact that our series is relatively small, our findings were similar to those reported by Wu et al (15) and Hong et al (16). No recurrence was noted in any case with a follow-up period of 1 to 20 years.

After segmental resection for solitary haemangioma in long bone diaphyses, a large defect is a challenge for the restoration of the biomechanics of the adjacent joints. Van Isacker et al (17) advocated

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### Table I. — Patients’ diagnoses and operative data

<table>
<thead>
<tr>
<th>Number</th>
<th>Age (years)</th>
<th>Gender</th>
<th>Location</th>
<th>Side</th>
<th>Operative Time (min)</th>
<th>Blood Loss (ml)</th>
<th>Length (cm)</th>
<th>Source</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>31</td>
<td>Female</td>
<td>clavicle</td>
<td>Right</td>
<td>75</td>
<td>150</td>
<td>4</td>
<td>iliac bone</td>
</tr>
<tr>
<td>2</td>
<td>40</td>
<td>Male</td>
<td>clavicle</td>
<td>Right</td>
<td>65</td>
<td>100</td>
<td>3</td>
<td>iliac bone</td>
</tr>
<tr>
<td>3</td>
<td>52</td>
<td>Female</td>
<td>humerus</td>
<td>Left</td>
<td>85</td>
<td>200</td>
<td>7</td>
<td>fibula</td>
</tr>
<tr>
<td>4</td>
<td>27</td>
<td>Male</td>
<td>ulna</td>
<td>Right</td>
<td>60</td>
<td>100</td>
<td>6</td>
<td>fibula</td>
</tr>
<tr>
<td>5</td>
<td>58</td>
<td>Female</td>
<td>ulna</td>
<td>Left</td>
<td>70</td>
<td>100</td>
<td>4</td>
<td>iliac bone</td>
</tr>
<tr>
<td>6</td>
<td>42</td>
<td>Female</td>
<td>radius</td>
<td>Left</td>
<td>85</td>
<td>150</td>
<td>4</td>
<td>iliac bone</td>
</tr>
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</table>

### Table II. — Functional outcomes and complications at follow-up

<table>
<thead>
<tr>
<th>Patient Number</th>
<th>Follow-up (ms)</th>
<th>Prox. union (ms)</th>
<th>Distal union (ms)</th>
<th>Functional Outcome 6 months follow-up</th>
<th>Functional Outcome last follow-up</th>
<th>Complications</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td>Overall MSTS Score (%)</td>
<td>Overall MSTS Score (%)</td>
<td></td>
</tr>
<tr>
<td>1</td>
<td>240</td>
<td>3</td>
<td>4</td>
<td>Excellent 81.0</td>
<td>Excellent 99.0</td>
<td></td>
</tr>
<tr>
<td>2</td>
<td>24</td>
<td>3</td>
<td>3</td>
<td>Good 76.5</td>
<td>Excellent 98.0</td>
<td>-</td>
</tr>
<tr>
<td>3</td>
<td>108</td>
<td>5</td>
<td>8</td>
<td>Fair 53.0</td>
<td>Excellent 96.0</td>
<td>Wound necrosis</td>
</tr>
<tr>
<td>4</td>
<td>30</td>
<td>4</td>
<td>8</td>
<td>Good 73.7</td>
<td>Excellent 95.0</td>
<td>-</td>
</tr>
<tr>
<td>5</td>
<td>24</td>
<td>4</td>
<td>3</td>
<td>Excellent 90.0</td>
<td>Excellent 96.0</td>
<td>-</td>
</tr>
<tr>
<td>6</td>
<td>12</td>
<td>3</td>
<td>4</td>
<td>Excellent 88.0</td>
<td>Excellent 97.0</td>
<td>-</td>
</tr>
</tbody>
</table>
the use of a bone allograft, because it allows for immediate restoration of the anatomy without harvesting or weakening another bone. The absence of incorporation in the long term has however limited its use (18). Recently, reconstruction with alloplastic materials, such as methylmethacrylate cement (1, 16), has been presented as another option. Yu et al (19) filled the bone defect with bone cement and K-wires, and after 12-month follow-up, there was no evidence of local recurrence of haemangioma or loosening of bone cement. Stoffelen et al (20) reported the reconstruction of a large ulnar defect due to tumour resection, using the Ilizarov technique of segmental transport. However, several drawbacks limit the wide application of this technique (21).

Autografts have been shown to provide better long-term outcomes (22) and are commonly used to bridge large bone defects after resection of a tumour (23-24). In our patients autogenous iliac bone (for segmental defects < 5 cm) or fibula (for segmental defects ≥ 5 cm) reconstruction was used and fixed rigidly with locking plates. This procedure is simple and definitive. In our series, all patients achieved union by a mean of about four months. The average MSTS functional score was 77% of a normal function (53%-90%) for all patients at 6 months postoperatively, and 97% (95%-99%) at the last follow-up evaluation.

In conclusion, segmental resection and reconstruction with autologous bone graft is a viable treatment option for solitary haemangioma of long bone diaphyses. Functional results are satisfying, as well as oncological results in terms of local tumour control are excellent.

REFERENCES


