Preliminary results after reconstruction of bony defects of the proximal humerus with an allograft-resurfacing composite

We retrospectively studied 14 patients with proximal and diaphyseal tumours and disappearing bone (Gorham’s) disease of the humerus treated with wide resection and reconstruction using an allograft-resurfacing composite (ARC). There were ten women and four men, with a mean age of 35 years (8 to 69). At a mean follow-up of 25 months (10 to 89), two patients had a fracture of the allograft. In one of these it was revised with a similar ARC and in the other with an intercalary prosthesis. A further patient had an infection and a fracture of the allograft that was revised with a megaprosthesis. In all patients with an ARC, healing of the ARC-host bone interface was observed. One patient had failure of the locking mechanism of the total elbow replacement. The mean post-operative Musculoskeletal Tumor Society score for the upper extremity was 77% (46.7% to 86.7%), which represents good and excellent results; one patient had a poor result (46.7%).

In the short term ARC effectively relieves pain and restores shoulder function in patients with wide resection of the proximal humerus. Fracture and infection remain significant complications.

Osteoarticular allografts have been used for biological reconstruction of large bone and cartilage defects, allowing restoration of bony and soft-tissue anatomy with optimum joint stability and function. However, rates of infection and instability can exceed 45%. Fracture, resorption and nonunion of the graft body, and fracture, fragmentation or collapse of the humeral head occur in between 25% and 67% of cases. Filling allografts with cement in order to prevent fracture has been proposed, but fractures still occur with subsequent complicated revision surgery. Joint incongruity has been a significant problem, especially in children, where size-matched small-diameter osteoarticular allografts are not easily available. Another concern is the potential for bacterial or virus transmission. Moreover, access to a good tissue bank is essential to provide sterile parts of the right size in a timely fashion. These factors tend to conspire against routine reconstruction using osteoarticular allografts.  

Alternatively, allograft-prosthesis composites (APC) may facilitate joint congruency and reduce the risk of fracture and nonunion while avoiding fragmentation of the humeral head. Higher Musculoskeletal Tumor Society (MSTS) scores have been reported with APC than with reconstructions using osteoarticular allografts and megaprosthesis. Function was found to be directly proportional to the amount of soft tissue that could be preserved or reconstructed, and allografts allowed more effective reattachment of soft-tissues such as the rotator cuff and deltoid muscles, thereby yielding better stability and function. On the other hand, reattachment of tendons to megaprosthesis is difficult, resulting in reduced function of the shoulder, instability and dislocation.

We became increasingly aware of the high complication rate associated with the APC technique and in 2003 we began to use proximal humeral osteoarticular allografts in selected patients following humeral resection. The aims of this paper are to present the preliminary results of a series of patients with tumours and disappearing bone (Gorham’s) disease of the humerus treated by wide resection and reconstruction using a humeral osteoarticular allograft-resurfacing composite (ARC), and to discuss the indications and contraindications, the short-term outcomes and the complications encountered.

Patients and Methods

The medical records of 14 patients with proximal and diaphyseal tumours and Gorham’s disease of the humerus treated between 2003 and 2010 were retrospectively analysed.
patients were treated by the senior authors (PR, MM) at a tertiary tumour centre. There were ten women and four men, with a mean age of 35 years (8 to 69) and a mean follow-up of 25 months (10 to 89); at the latest evaluation all patients were still alive and were therefore included (Table I). This study was approved by the Institutional Review Board, and all patients or their relatives gave written informed consent to be included in this study.

Pre-operative assessment included anteroposterior, axillary and lateral radiographs of both shoulders to measure the diameter of the humeral head and evaluate the glenoid; and standard oncological work-up including CT and MRI of the shoulder, CT of the chest, pre-operative percutaneous biopsy and staging.26 Pre- and post-operative chemotherapy was administered in all patients with osteosarcoma; the patient with Ewing's sarcoma had pre- and post-operative chemotherapy, and lung radiation therapy for metastases (Table I).

Operative technique. The humeral tumour was widely resected and reconstructed with ARC; intra-articular resection aimed to achieve complete resection of the tumour without contamination during the excision. Under general anaesthesia, with the patient in the beach-chair position, the extended deltopectoral approach was used, incorporating the previous biopsy tract; if necessary, horizontal detachment of the deltoid from the acromion was performed to allow access to the glenoid and the superior and posterior aspects of the rotator cuff. Acromioplasty, with or without resection of the coracoacromial ligament, was performed in patients with hooked acromion and ligamentous impingement. After resection, the deltoid and rotator cuff were prepared for reattachment to the allograft. If resection of the deltoid, rotator cuff or axillary nerve was performed, or if the rotator cuff was irreparably torn, the humerus was reconstructed using a megaprosthesis rather than an ARC.

Table I. Details of the patients with proximal and diaphyseal humeral tumours and Gorham's disease treated in this series

<table>
<thead>
<tr>
<th>Case</th>
<th>Age, gender</th>
<th>Diagnosis</th>
<th>Surgery*</th>
<th>Adjuvant treatments†</th>
<th>Complications</th>
<th>Follow-up (months)</th>
<th>MSTS‡ score (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>8, F</td>
<td>Osteosarcoma (high grade)</td>
<td>Proximal and diaphyseal humeral resection, ARC§, elastic nailing</td>
<td>Pre-op and post-op CMT</td>
<td>Fracture of the allograft (at 12 months), revision with intercalary prosthesis</td>
<td>89</td>
<td>70.00</td>
</tr>
<tr>
<td>2</td>
<td>11, M</td>
<td>Osteosarcoma (telangiectatic, high grade)</td>
<td>Proximal humeral resection and ARC§</td>
<td>Pre-op and post-op CMT</td>
<td>Fracture of the allograft (at 12 months), revision with ARC</td>
<td>56</td>
<td>86.67</td>
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<tr>
<td>3</td>
<td>69, F</td>
<td>Gorham's disease</td>
<td>Total humeral resection, ARC§ and elbow arthroplasty</td>
<td>-</td>
<td>Failure of the locking mechanism of the arthroplasty, replacement of the locking mechanism</td>
<td>31</td>
<td>73.33</td>
</tr>
<tr>
<td>4</td>
<td>52, F</td>
<td>Chondrosarcoma (high grade)</td>
<td>Proximal humeral resection and ARC**</td>
<td>-</td>
<td>-</td>
<td>19</td>
<td>83.33</td>
</tr>
<tr>
<td>5</td>
<td>28, M</td>
<td>Chondrosarcoma (low grade)</td>
<td>Proximal humeral resection and ARC**</td>
<td>-</td>
<td>Fracture of the allograft (at 5 months), revision with ARC Infection, two-stage revision with megaprosthesis</td>
<td>19</td>
<td>46.67</td>
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<tr>
<td>6</td>
<td>55, F</td>
<td>Giant cell tumour (stage 3)</td>
<td>Proximal humeral resection and ARC**</td>
<td>-</td>
<td>-</td>
<td>18</td>
<td>86.67</td>
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<tr>
<td>7</td>
<td>40, F</td>
<td>Giant cell tumour (stage 3)</td>
<td>Proximal humeral resection and ARC**</td>
<td>-</td>
<td>-</td>
<td>17</td>
<td>73.33</td>
</tr>
<tr>
<td>8</td>
<td>26, F</td>
<td>Giant cell tumour (stage 3)</td>
<td>Proximal humeral resection and ARC**</td>
<td>-</td>
<td>-</td>
<td>17</td>
<td>76.67</td>
</tr>
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<td>9</td>
<td>36, F</td>
<td>Chondrosarcoma (low grade)</td>
<td>Proximal humeral resection and ARC**</td>
<td>-</td>
<td>-</td>
<td>16</td>
<td>73.33</td>
</tr>
<tr>
<td>10</td>
<td>60, M</td>
<td>Chondrosarcoma (myxoid, high grade)</td>
<td>Total humeral resection, ARC** and elbow arthroplasty</td>
<td>-</td>
<td>-</td>
<td>16</td>
<td>70.00</td>
</tr>
<tr>
<td>11</td>
<td>58, F</td>
<td>Giant cell tumour (stage 3)</td>
<td>Proximal humeral resection and ARC**</td>
<td>-</td>
<td>-</td>
<td>14</td>
<td>83.33</td>
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<tr>
<td>12</td>
<td>8, M</td>
<td>Ewing's sarcoma</td>
<td>Proximal humeral resection and ARC**</td>
<td>Pre-op and post-op CMT, lung RT</td>
<td>-</td>
<td>14</td>
<td>86.67</td>
</tr>
<tr>
<td>13</td>
<td>27, F</td>
<td>Osteosarcoma (osteoblastic, high grade)</td>
<td>Proximal humeral resection and ARC**</td>
<td>Pre-op and post-op CMT</td>
<td>-</td>
<td>12</td>
<td>80.00</td>
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<tr>
<td>14</td>
<td>13, F</td>
<td>Osteosarcoma (high grade)</td>
<td>Proximal humeral resection and ARC**</td>
<td>Pre-op and post-op CMT</td>
<td>-</td>
<td>10</td>
<td>86.67</td>
</tr>
</tbody>
</table>

* ARC, allograft-resurfacing composite  
† Pre-op, pre-operative; Post-op, post-operative; CMT, chemotherapy; RT, radiotherapy  
‡ MSTS, Musculoskeletal Tumor Society  
§ Capica Implantcast surface replacement of the shoulder joint (Implantcast Italia S.R.L., Rome, Italy)  
¶ SMR-R Cup Lima-Lto shoulder resurfacing prosthesis (Lima Lto, Udine, Italy)  
** Global Cup Depuy resurfacing humeral head implant (DePuy Italia, Rome, Italy)
The next step was to prepare a resurfacing humeral osteoarticular allograft matched for size and length. The head of the allograft was shaped with reamers and a resurfacing humeral head was cemented on. The allograft was filled with cement, avoiding extrusion of the cement at the allograft-host bone junction by inserting the cement in the doughy state. Before the cement had hardened, the ARC was fixed to the remaining humerus with a plate and screws incorporating retroversion equivalent to that of the opposite humeral head (usually 30°) as assessed in preoperative radiographs of the opposite shoulder. Elbow arthroplasty with a long humeral cemented stem (Coonrad/Morrey Total Elbow Revision; Zimmer Srl, Milan, Italy) was performed in the patients with total humeral resection (patients 3 and 10). The rotator cuff tendons were reattached to the allograft with heavy, non-absorbable sutures (FiberWire; Arthrex Inc., Naples, Florida) without tension to avoid subluxation or stiffness. The deltoid was reattached to the allograft at its correct anatomical length. If preserved, the joint capsule was also sutured to the allograft and the wound was closed in layers with two deep suction drains.

Post-operatively, the arm was immobilised in a sling for two weeks; elbow, hand and wrist exercises were encouraged. Passive circumduction and pendulum movements were then started under supervision by a physical therapist, limited to 30° for the first four weeks and followed by passive and active-assisted full range of movement exercises. After eight to 12 weeks the patients were allowed to begin exercises against resistance, but heavy manual work and athletic activities were prohibited. Radiographs were performed monthly for the first three months, every three months for the first year, every six months for the following two years, and annually thereafter. Allograft-host bone union was evaluated and complications were recorded; routine oncological follow-up including CT of the chest was performed every six months for the first three years, and then annually. Function was evaluated using the MSTS score for the upper extremity, with values from 0 to 5 assigned for each of pain, function, emotional acceptance, hand positioning, dexterity and lifting ability; the score was presented as a percentage.

Results

There were complications with the ARC that required revision in three (21%) patients, none of whom had coronary heart disease or diabetes mellitus, and there was no relationship with activity or weight-lifting. In one patient (patient 1) the ARC was stabilised with elastic titanium nails, which was a technical error as fracture of the allograft and nails occurred at 12 months post-operatively, and reconstruction was performed with a cemented intercalary prosthesis (Fig. 1). The second patient (patient 2) had a fracture of the ARC 12 months post-operatively that was revised with a similar ARC. At 56 months after the initial operation, ARC-host bone union was observed without evidence of loosening of the prosthesis. The third patient (patient 5) had a fracture of the ARC five months post-operatively that was also revised with a similar ARC. This second operation was complicated by infection, which was treated by two-stage revision of the proximal humerus using a megaprosthesia, without recurrence of the infection at the latest examination.
In the remaining 11 (79%) patients, there were no allograft- or resurfacing implant-related complications in the short term, and in all patients union with the host bone was observed. At the latest clinical examination no patient had instability of the shoulder, rotator cuff deficiency or pain from glenoid erosion. The patient with Gorham’s disease and total humeral ARC (patient 3) had failure of the locking mechanism of the elbow replacement at 31 months post-operatively (Fig. 2), which was treated with replacement of the locking mechanism. The mean post-operative upper extremity MSTS score was 77% (46.7% to 86.7%). Seven patients had an MSTS score > 80%, which represents an excellent result, six had an MSTS score > 70%, which represents a good result, and one had an MSTS score < 50%, which represents a poor result (patient 5).

Discussion

Resurfacing of the proximal humerus (hemi-resurfacing) consists of reaming the humeral head and fitting a cap that may be mated against a glenoid component (total shoulder resurfacing).28 The advantages of resurfacing over stemmed arthroplasty are preservation of bone stock, shorter operating times, lower risk of peri-prosthetic fractures and ease of conversion to a stemmed arthroplasty.28-32 However, there are no studies evaluating resurfacing osteoarticular allografts for reconstruction of the humerus.

A prerequisite for proximal humeral ARC is preservation of the deltoid and rotator cuff.33 In those of our patients in whom we could preserve the deltoid, rotator cuff and axillary nerve we used an ARC instead of a mega-prosthesis. Because osseo-integration and incorporation of the prosthesis by avascular allograft bone is highly unlikely, all resurfacing prostheses were cemented for stability and the prevention of fractures, and none had a hydroxyapatite coating. We filled the allografts with cement, avoiding extrusion of the cement at the junction between the allograft and the host bone, and then spanned the total length of the allograft with long plates; passage of the fixation screws would have been precluded if an APC had been used. Using ARC meant that if allograft head resorption or cup loosening occurred, revision to APC would be feasible, with subsequent union of the allograft to host bone. If allograft fracture occurred, revision with a similar ARC (as in patient 2), APC or mega-prosthesis (patient 5) could be performed.

Frozen allografts in humans are osteoconductive rather than osteo-inductive,4 functioning as biological spacers.5,34-36 Retrieval data has shown that only a small percentage of the allograft becomes revascularised, whereas the rest remains necrotic.4,5 Allograft union takes place slowly at cortical junctions by external callus from the host cortex, and more rapidly at cancellous junctions by internal callus. The deep portions of the graft retain their architecture, and where cement is used there is no evidence of bone resorption. The allograft cartilage becomes necrotic and covered by fibrovascular tissue, but functions well for as long as five years. Cartilage changes occur earlier and are more advanced in specimens retrieved from patients with an unstable rather than a stable joint.4,34 The soft tissues of the host become attached to the graft by a thin seam of new bone on the graft surface.4 After more than five years allografts exhibit zones of resorption or fragmentation, and following repeated loading the devitalised allograft is prone to stress fractures and further fragmentation.36 Infection leads to extensive osteomyelitis and septic arthritis.35

Figures 2a and 2b – radiographs of the left arm of a 69-year-old woman with Gorham’s disease (patient 3), in a) anteroposterior (AP) and b) lateral views. Figures 2c and 2d – radiographs in c) AP and d) lateral views at 31 months after total humeral resection and reconstruction using a total humeral allograft-resurfacing composite and a cemented long-stemmed elbow arthroplasty showing failure of the locking mechanism.
Irradiation has no notable effect on the mechanical quality of the allografts, but might favour resorption. Adjuvant chemotherapy increases the complication rate. Short-term administration of methotrexate and doxorubicin reduced trabecular bone volume and diminished bone formation by 60%; osteoblast toxicity was reflected by reduced volume and thickness of osteoid, without, however, affecting the number of osteoblasts and the percentage of trabecular surface covered by bone-forming cells. Combined doxorubicin, cisplatin and ifosfamide chemotherapy reduces mineral apposition in cancellous bone, without any differences in the porosity, osteonal activity and mineral apposition in cortical bone. Reduction in mineral apposition in cancellous bone, without any differences in the risk of fracture and osteoporosis.

In this series we acknowledge a high rate of allograft-related complications, unsurprisingly in the form of fractures and infections. Despite cementation, bone resection and allograft length might be relevant risk factors for fracture, as the length of the lever arm increases the forces during weight lifting. Complications occurred in both of the patients with the longest follow-up, who had also received post-operative chemotherapy. Although the MSTS may not be the most appropriate score to evaluate shoulder function, we prefer to use it in tumour patients as it provides a comprehensive evaluation of factors such as pain, functional activity and emotional acceptance, which are pertinent to the patient as a whole, and includes factors specific to the upper limb, such as hand positioning, manual dexterity and lifting ability. The fact that 13 of the 14 patients had MSTS scores > 70% indicates good or excellent shoulder function.

Glenoid erosion after hemi-resurfacing occurs at a similar rate to that which follows stemmed hemiarthroplasty, appearing three to five years post-operatively. Age is a significant variable: the younger and more active the patient, the higher the chance of glenoid erosion. The size and placement of the cup and joint incongruency also have an effect on eccentric glenoid loading and progressive erosion. Even with a concentric glenoid, longer follow-up has shown progressive joint space loss and declining function. In this setting, hemiarthroplasty was found inferior to total shoulder arthroplasty. However, osteolysis and loosening of the glenoid prosthesis, polyethylene wear and difficult revision procedures are concerns when offering total shoulder arthroplasty to patients over 50 years of age with an uninvolved or minimally involved glenoid surface. Because of these concerns, many surgeons favour humeral head replacement alone.

In our practice we do not perform glenoid arthroplasty in tumour patients so as to avoid spread of the tumour to the opposite side of the joint, nor in patients without glenoid wear. Instead, to avoid eccentric glenoid loading, we tend to reduce subacromial contact by acromioplasty and coracoacromial ligament release, to reconstruct the offset to within 4 mm with an anatomical head size, and to carefully balance the soft tissues. If superior migration and subacromial contact of the humeral head, loosening of the resurfacing prosthesis or glenoid erosion are observed, revision to conventional or reverse total shoulder arthroplasty or biological glenoid arthroplasty are possible solutions.

Our study was a short-term evaluation. Previously we planned osteoarticular allograft humeral reconstructions with the objectives of preserving bone stock and improving function by reattaching the soft tissues. We then found that osteoarticular allografts were fraught with complications, and so decided to perform APC and then ARC reconstruction. The allograft restores bone stock, which is potentially beneficial for the patients, allows for soft-tissue reattachment and facilitates the use of a resurfacing prosthesis which results in congruent articulation. We now recommend ARC reconstruction in selected patients with deltoid, rotator cuff and axillary nerve-sparing resections. If the rotator cuff cannot be spared, an APC with a reverse prosthesis can produce better shoulder function. However, these allografts may fail in the long term and require revision, usually with a megaprosthesi, and shoulder function tends to deteriorate due to glenoid erosion.

We recognise several limitations in this retrospective study. Because this is a novel technique the number of patients is small, the follow-up short and the population heterogeneous. Mixing benign and malignant tumours, adding a non-tumour disease such as Gorham’s disease, and failing to control for confounding errors such as activity level and resection length, make it impossible to draw statistically valid conclusions. Moreover, because the choice of reconstruction in tumour patients remains controversial, our technique was not randomised. However, in the short term it has been effective in restoring function.

Our decision-making process should take into account the risk of glenoid wear complications. In order to improve outcome, important technical steps such as correct sizing, resurfacing and stable plate and screw fixation of the ARC are necessary, and in future a greater number of cases should be subjected to long-term follow-up to evaluate the survival of the reconstruction and its effect on the glenoid.

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


