Wide *en bloc* extra-articular excision of the elbow for sarcoma with complex reconstruction

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We describe a consecutive series of five patients with bone or soft-tissue sarcomas of the elbow and intra-articular extension treated by complex soft tissue, allograft bone and prosthetic joint replacement after wide extra-articular *en bloc* excision. All had a pedicled myocutaneous latissimus dorsi rotation flap for soft-tissue cover and reconstruction of the triceps. Wide negative surgical margins were obtained in all five patients. No local wound complications or infections were seen. There were no local recurrences at a mean follow-up of 60 months (20 to 105). The functional results were excellent in four patients and good in one. Longer term follow-up is necessary to confirm the durability of the elbow reconstruction.

Approximately 1% of bone and soft-tissue sarcomas occur about the elbow,¹ a few of which originate in or extend into the joint. Amputation is an effective treatment but the functional, cosmetic and emotional burdens of the procedure are such as to encourage every effort at preservation of the limb. However, there is little information as to the ability to achieve wide margins of resection or the safety of extra-articular resection about the elbow. Most case series have described transarticular hemi-resection through the elbow joint and reconstruction rather than wide extra-articular excision.² ⁴ Constrained allograft, prosthetic, or allograft-prosthetic composite reconstructions are needed for these patients.

The long-term durability of total elbow replacement in osteoarthritis (OA) and rheumatoid arthritis (RA)⁵ ⁷ is well established, but little is known about its use after complex bone, joint, and soft-tissue resections which have sacrificed major bone segments and functional soft tissues. Total elbow reconstruction with allografts has been associated with a high risk of infection and Charcot degeneration.⁸ Few papers have reported the outcome following custom-made endoprosthetic replacement of the elbow for tumour.² ⁹

We describe our experience with wide *en bloc* extra-articular excision of the elbow for high-grade sarcomas.

Patients and Methods

We reviewed retrospectively five patients who had undergone wide, extra-articular resection of an elbow for high-grade bone or soft-tissue sarcoma between 1995 and 2005 (Table 1). These patients were followed for a mean of 60 months (20 to 105). The diagnosis and margins of excision were obtained from review of the pathology reports. Local recurrence and elbow function were assessed by clinical examination and plain radiographs every three months for the first year, and then every six months. Magnetic resonance imaging of the elbow was obtained every six months. Screening for distant recurrence was accomplished with plain radiographs of the chest every three months for the first year and CT of the chest every six months.

Three patients had a high grade intra-articular soft-tissue sarcoma, two with a synovial sarcoma and one a malignant fibrous histiocytoma. One patient had a Ewing’s sarcoma of the distal humerus with intra-articular soft-tissue extension and one had a Ewing’s sarcoma of the antecubital soft tissue with multiple recurrences and extensive anterior intra-articular involvement.

Three patients received neoadjuvant external beam radiation (mean 5966 cGy, 5000 to 6600). The patient with the multiple recurrent Ewing’s sarcoma had received intra-operative brachytherapy during a previous resection performed prior to her referral. Both patients with Ewing’s sarcoma received chemotherapy including vincristine, cyclophosphamide, ifosfamide, etoposide and doxorubicin. One patient with a synovial sarcoma received dacarbazine, ifosfamide and doxorubicin.
All patients had a wide, extra-articular resection. Four had an allograft prosthetic composite reconstruction and one an endoprosthetic reconstruction only, all using the Coonrad-Morrey prosthesis (Zimmer Inc., Warsaw, Indiana). One patient had the median nerve and brachial artery resected in continuity with the major tumour mass with simultaneous reconstruction using grafts from the sural nerve and saphenous vein. Two others had the radial nerve resected in continuity with the tumour mass. All patients had tendon transfers to optimise hand function.

Soft-tissue cover was achieved on all patients by means of a pedicled latissimus dorsi myocutaneous rotation flap at the time of the reconstruction. An effort was made to envelop the alloprosthetic composite with the latissimus muscle, while positioning the associated skin island to replace resected skin or to increase the thickness of cover over the posterior aspect of the elbow. The humeral reconstruction was shortened by 0 cm to 2 cm as needed to permit adequate soft-tissue cover. The latissimus muscle was sutured in continuity with the native proximal triceps and the distal insertion of the native or allograft triceps tendon to restore extensor continuity. The mean operative time was 12 hours (7 to 18). The post-operative care was similar for all patients. All elbows were immobilised for three weeks. Active assisted flexion exercises started at three weeks. Active extension began at six weeks. Assisted active and passive movement of the fingers started on the first post-operative day even in those patients with tendon transfers. Motor re-education for tendon transfers was initiated at three weeks with protective splinting of the wrist and fingers for a total of six weeks.

**Illustrative case.** A 23-year-old right handed woman was referred with a third recurrence of Ewing’s sarcoma of the soft tissues anterior to and involving the right elbow. She had received a full course of chemo- and brachytherapy. She was not a candidate for further adjuvant treatment. Plain radiographs showed soft-tissue calcification consistent with recurrence of the tumour. The T1-weighted MRI showed invasion of the elbow joint with encasement of the median nerve and brachial artery (Fig. 1). Amputation was recommended but was refused by the patient. After full consultation, wide en bloc excision of the elbow was offered.

An extended anterior incision was made with a wide resection incorporating the previous incision and the entire operative field (Fig. 2). The radial and ulnar nerves were mobilised away from the tumour and the median nerve transected after proximal and distal mobilisation. The brachial artery was divided proximally and distally just before completion of the wide excision and after the saphenous vein had been harvested for reconstruction (Fig. 3). The specimen was then removed as an extra-articular en bloc mass incorporating the median nerve and brachial artery as they traversed the tumour. The radial nerve was transposed from the posterolateral aspect of the humerus to an anterior position through the defect created by the resected specimen in order to reduce the risk of injury to the nerve in the event of future prosthetic revision from a posterior approach. The allograft prosthetic composite utilised for reconstruction

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**Table I.** Details of the five patients with extra-articular resection of the elbow for high-grade sarcoma with prosthetic reconstruction

<table>
<thead>
<tr>
<th>Case</th>
<th>Age (yrs)</th>
<th>Diagnosis</th>
<th>Radiation</th>
<th>Chemotherapy†</th>
<th>Reconstruction‡</th>
<th>Nerve or artery resection</th>
<th>Nerve or artery reconstruction</th>
<th>Tendon transfer</th>
<th>ISOLS§ score</th>
<th>Mayo elbow score§</th>
<th>Arcs of motion</th>
<th>Local recurrence</th>
<th>Lung metastasis</th>
<th>Death from the disease</th>
<th>Follow up (mths)</th>
</tr>
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<tbody>
<tr>
<td>1</td>
<td>40</td>
<td>Ewing’s</td>
<td>No</td>
<td>Ifos, Etop</td>
<td>APC</td>
<td>No</td>
<td>No</td>
<td>26</td>
<td>85</td>
<td>110</td>
<td>No</td>
<td>No</td>
<td>No</td>
<td>No</td>
<td>No</td>
</tr>
<tr>
<td>2</td>
<td>23</td>
<td>Ewing’s</td>
<td>Brachy-</td>
<td>Ifos, Etop,</td>
<td>APC</td>
<td>Median nerve</td>
<td>No</td>
<td>Yes</td>
<td>24</td>
<td>93</td>
<td>130</td>
<td>No</td>
<td>No</td>
<td>No</td>
<td>No</td>
</tr>
<tr>
<td>3</td>
<td>51</td>
<td>Synovial</td>
<td>6600 cGy</td>
<td>Dacar, Ifos,</td>
<td>APC</td>
<td>Radial nerve</td>
<td>Yes</td>
<td>24</td>
<td>90</td>
<td>95</td>
<td>No</td>
<td>Yes</td>
<td>Yes</td>
<td>No</td>
<td>No</td>
</tr>
<tr>
<td>4</td>
<td>27</td>
<td>Synovial</td>
<td>5000 cGy</td>
<td>No</td>
<td>APC</td>
<td>No</td>
<td>No</td>
<td>No</td>
<td>26</td>
<td>95</td>
<td>110</td>
<td>No</td>
<td>No</td>
<td>No</td>
<td>No</td>
</tr>
<tr>
<td>5</td>
<td>58</td>
<td>High grade pleomorphic soft-tissue sarcoma</td>
<td>6300 cGy</td>
<td>No</td>
<td>Prosthesis</td>
<td>No</td>
<td>No</td>
<td>No</td>
<td>26</td>
<td>95</td>
<td>85</td>
<td>No</td>
<td>No</td>
<td>No</td>
<td>No</td>
</tr>
</tbody>
</table>

*Ifos, Ifosfamida; Etop, Etoposide; Cyclo, Cyclophosphamide; Dox, doxorubicin; V in, Vincristine, Dacar, Dacarbazine
† APC, alloprosthetic composite
‡ ISOLS, International Society of Limb Salvage score

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**Fig. 1a** – Lateral radiograph of the elbow demonstrating a soft-tissue mass lying anteriorly. **Fig. 1b** – A T1-weighted axial MRI demonstrating the soft-tissue recurrence.
used a long stem Coonrad-Morrey semi-constrained prosthesis (Zimmer) (Fig. 4). This was cemented into the allograft humerus and ulna on a side table, and while the cement was not yet fully mature the composite was cemented into the humerus and ulna of the patient in a single stage. The allograft of the proximal radius was fixed to the recipient bone with a dynamic compression plate.

The sural nerve and saphenous vein were harvested from the ipsilateral leg and microsurgical reconstruction of the brachial artery and median nerve was performed. A pedicled latissimus dorsi myocutaneous rotation flap was used to envelop the alloprosthetic composite and to cover the anterior soft-tissue defect created by the resection (Fig. 5) with well vascularised soft tissue in order to improve antibiotic delivery and reduce the risk of post-operative wound breakdown with associated infection. Oral antibiotics were administered for three months.

The patient was reviewed 26 months after operation when the images in Figure 6 were obtained. When seen at 66 months after surgery, she had an excellent range of movement (0° to 130°) at the elbow and no evidence of local recurrence. She had a sensate hand with reasonable function, with a Mayo elbow score of 90°.

**Results**

Wide margins of resection were obtained in all five patients. There were no local recurrences at the last follow-up. Two patients developed distant metastasis, one of whom has since died.
The mean International Society of Limb Salvage Score\textsuperscript{11,12} was 25.2 (24 to 26), which is 84\% of the maximum. The mean Mayo Elbow Performance score\textsuperscript{6} was 91 (85 to 95). Four patients had an excellent result based on the Mayo elbow scoring system.\textsuperscript{1} The other had a good result. All patients could extend the elbow against gravity.

In one patient, further operations were required for failure of the humeral component, nonunion of the humeral allograft and failure of the prosthetic bushings. One patient with a partial radial nerve palsy after previous radiotherapy had a complete post-operative palsy which resolved after six months. There was no early or late infection. One patient developed radiolucent lines about the humeral component when reviewed at 26 months (Fig. 6), but these had not progressed, nor was there evidence of bone resorption or loosening during a further 40 months of follow-up. No other patient had evidence of loosening.

**Discussion**

These tumours required wide en bloc extra-articular excision of the elbow with extensive resection of soft tissue often incorporating major nerves. Of the five patients in our series, three had resection of a major nerve and/or artery requiring grafting or tendon transfers. The use of alloprosthetic composites in these circumstances has not been described previously. Immediate soft-tissue cover successfully avoided problems of wound healing and infection.

The compact nature of the functional tissues which include major vessels, nerves and tendons, with the limited soft-tissue envelope, thin dermis and limited subcutaneous fat, increases the difficulty of wide en bloc excision of intra-articular tumours of the elbow which commonly extend extra-articularly. In order to achieve adequate margins of resection substantial excision of soft-tissue, skin, triceps, brachialis and possibly nerves and vessels is required. This increases the technical difficulty of the procedure and the associated functional morbidity. Any contamination of skin flaps or functional tissues which may have occurred at the time of biopsy or during prior diagnostic arthroscopy, increases the complexity of the resection and magnifies the risk and morbidity. We have shown that negative margins can be reliably achieved with prevention of local recurrence often with the use of appropriate adjuvants.

The segmental bone defect produced by wide en bloc excision is typically greater than 10 cm. The upper and lower segments of the arm and forearm are then held in continuity only by relatively thin skin, fat, nerves and vessels (Fig. 2). There are no reconstructions which have been shown to have a predictable functional outcome. The durability of custom made prostheses is not known. The rates of failure for semi-constrained total elbow replacements have been established in patients with RA or OA, who are often older, with a lower functional demand.\textsuperscript{5,13,14} It is likely that failure of prostheses will be greater when used in reconstruction following tumours given the larger size of the defect and the greater needs of younger patients.

The use of an allograft prosthetic composite may have several advantages. The allograft soft tissues facilitate reconstruction of the triceps and repair of the flexor-pronator and extensor origins. We consider that the healing of bulk allograft humerus or ulna to the native bone may increase the durability of the prosthetic stem and decrease loosening, since rotational forces on the stem in native bone are likely to be decreased compared with the prosthetic replacement alone. Modular prostheses may facilitate revision procedures and it may be necessary to augment the bushings in order to provide better long-term durability.

We did not encounter infections or complications with the wound. We attribute this to the use of a latissimus dorsi flap. Mastorakos et al\textsuperscript{10} noted a significant reduction in reoperation in patients who had primary flap cover for allograft reconstruction in limb salvage.
latissimus flap allowed repair of the triceps by sewing the proximal triceps in continuity with the rotated muscle, which was then sutured to the allograft triceps tendon on the ulna allograft.

Reports of prosthetic reconstruction after excision of tumours about the elbow have focused on trans-articular resections. Weber et al. described 23 patients, 15 with primary and eight with metastatic lesions, treated for malignant tumours, 11 of whom were reconstructed with a segmental total elbow prosthesis with a mean Musculoskeletal Tumour Society score of 83%.\textsuperscript{11,12} In a similar series, Kulkarni et al.\textsuperscript{15} gave details of ten patients with tumours about the distal humerus who were reconstructed with a constrained total elbow replacement. All had satisfactory function and pain relief. Complications were related to prosthetic loosening and wear of the bushings.\textsuperscript{15} Sperling et al.\textsuperscript{3} reported the experience at the Mayo Clinic (Rochester, Minnesota) with total elbow arthroplasty after resection of primary malignant bone tumours about the elbow, basing their results on the Mayo Elbow Performance Score.\textsuperscript{6} This paper focused on prosthetic reconstruction with no mention as to whether

Figure 6a – post-operative radiograph showing the allograft prosthetic composite reconstruction. Figures 6b and 6c – post-operative clinical photographs showing the range of movement of the elbow.
the tumours had intra-articular extension or whether the excisions were extra-articular. Two of their seven patients had an excellent result, and in five it was good. An update was recently published by Athwal et al,9 which included patients reconstructed with a linked, semi-constrained total elbow prosthesis. They found a mean Mayo Elbow Performance score of 75. They included patients with metastatic disease which may have contributed to the lower performance scores. Five patients had a nerve injury and four required revision because of failure of the reconstruction.5 This study comprised predominantly extra-articular tumours.

The use of total elbow allografts has been associated with significant complications and rates of failure. Dean et al8 used total elbow allografts in 23 patients for various indications, only one of which was a tumour. Only 14 patients retained their allograft after a minimum follow-up of one year; in ten the result was satisfactory and in four it was unsatisfactory. There were significant complications in 16 patients, the most common being nonunion, which occurred in seven cases. Instability was seen in six patients, radial nerve palsy in four, and major infection in three.8

The functional results in our series, with an International Society of Limb Salvage score of 83%, were similar to previous studies not limited to wide extra-articular resection and reconstruction.2-4 Four of our patients had excellent Mayo Elbow Performance Scores and one had a good score. All distal tendon transfer reconstructions were performed at the time of the resection. Strong tendon weaving and early active movement are necessary to maximise tendon gliding in these patients with very extensive surgical dissections which may promote adhesion formation. The immediate initiation of active movement of the tendon transfers improved the functional results and was not associated with any complications.

The indications for this complex resection and reconstruction are narrow and we do not have a comparison group of patients who were treated with amputation or an alternative treatment such as resection-replantation.16 The technical consistency achieved by a single surgeon is difficult to duplicate and may limit the ability to generalise the results. We do not define precisely which patients are candidates for this procedure. A general principle has been that if two major nerves to the hand can be spared, resection may be considered with the anticipation of a reasonable functional result following reconstruction.

Wide excision with negative margins can be achieved in a specific subset of high risk, technically-demanding resections for intra-articular bone and soft-tissue sarcomas of the elbow. These procedures require an experienced team of pathological and reconstructive limb-salvage surgeons. Treatment requires careful planning, specialised surgical skills and meticulous technique coupled with the use of effective adjuvant therapy when appropriate. Reconstruction using an elbow prosthesis or allograft prosthetic composite provides good or excellent function. Microsurgical technique is needed for the reconstruction of major vessels or nerves. The routine use of myocutaneous soft-tissue cover may help to prevent wound complications and infection. Further follow-up of this relatively young group of patients is needed to monitor the risk of local recurrence and to determine the durability of the prosthetic construct.

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References

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